CURRENT PERSPECTIVES ON DEVELOPMENTAL COORDINATION DISORDER (DCD)

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CURRENT PERSPECTIVES ON DEVELOPMENTAL COORDINATION DISORDER (DCD)

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EDITORIAL
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Editorial: Current Perspectives on Developmental Coordination Disorder (DCD)

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Keywords: developmental coordination disorder, DCD, co-occurring disorders, genome, motor skill, intervention—behavioral

Editorial on the Research Topic

Current Perspectives on Developmental Coordination Disorder (DCD)

INTRODUCTION

Developmental Coordination Disorder (DCD) occurs in approximately 5% of children (Blank et al., 2019) and describes a condition in which motor coordination is below the level expected given a child's age and opportunity for learning (APA, 2013). Children with DCD display motor difficulties which persist into adulthood and cannot be better explained by a medical or neurological condition (APA, 2013). The difficulties that individuals with DCD experience have a significant impact on activities of daily living, scholastic achievement, inter-personal relationships, and employment (Kirby et al., 2010). In addition, secondary consequences of DCD include higher anxiety (Harris et al., 2021), poorer levels of physical fitness (Schott et al., 2007) and negative self-perceptions (Piek et al., 2006). Despite significant growth in research into DCD over the last four decades, and international clinical practice guidelines being released (Blank et al., 2019), there are still pending questions regarding etiology, the influences of co-occurrence, movement behavior and ways in which change can be promoted. This Research Topic aimed to capture the breadth of the recent focus of research into DCD.

ETIOLOGY

Mountford et al. presented a genome-wide association study to examine potential biological drivers of early motor coordination in the context of motor difficulties. The authors looked for common genetic variants (single nucleotide polymorphisms) which could explain poor motor coordination and identified 59 genetic variants within five genes. Although some caution is necessary, as the inclusion of data was based solely on motor function rather than all of the DSM-5 criteria, this study is an exciting additional insight into the potential genes which may drive DCD.

CO-OCCURRENCE

Two studies within this topic considered co-occurrence. Meachon et al. considered inhibitory function in adults with ADHD, with DCD or with combined ADHD and DCD. Behaviourally the groups did not differ and performed as well as age-matched peers. However, event related potentials measured using EEG differed among the study groups and typical controls. In addition, differences were identified across co-occurrence groups. These data point toward compensatory mechanisms

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in simple tasks, resulting in typical behavior even in the face of atypical underlying mechanisms, with the latter differing among diagnostic subgroups. Izadi-Najafabadi and Zwicker used MRI to consider white matter microstructure before and after intervention using Cognitive Orientation to Occupational Performance (CO-OP) in children with and without co-occurring ADHD. Children with DCD showed significant alterations in white matter microstructure. In contrast, the children with DCD and ADHD showed no such white matter changes despite intervention improving motor performance. This suggests CO-OP has a distinctly different neurological effect on children with DCD and ADHD compared to those just with DCD and that some modifications to the CO-OP protocol may better address the needs of children with DCD and ADHD. Together these studies emphasize that the neurology underpinning behavior in those with and without co-occurrences can be very different.

MEASURED BEHAVIOR

When asked to perform a stepping task, Parr et al. found that children with DCD were more variable in their foot placement compared to children without DCD. However, neither gaze behavior nor state-anxiety differed across groups. In a second study, Parr et al. examined stair ascent and descent finding again that variability was higher in children with DCD and that these children looked further ahead than their typically developing peers, these factors may increase the likelihood of a fall. Children with DCD also reported significantly higher state-anxiety prior to stair descent. Switching to a manual task, Krajenbrink et al. considered motor planning within a grasping task, building on a plethora of research in the last 5 years which has raised the question of whether children with DCD *choose* not to or *cannot* plan for comfort. These authors used an established sword-task and a newly developed hammer-task. Differences between the two tasks led the authors to suggest that children with DCD can plan for comfort when it is demanded by task constraints. Finally, Blais et al. considered whether a specific auditory timing deficit contributes to DCD by exploring if children with DCD can synchronize motor output to sensory stimuli. Their data suggested a potential deficit in timing perception more generally in children with DCD, but that the learning of temporal motor sequences may be improved in DCD with the use of visual cues.

Three studies considered the experiences and abilities of adults with DCD on the road. Gentle et al. used a driving simulator to consider driving characteristics under three conditions of increasing complexity. The data suggest that young adults with DCD behave differently to their peers when driving in progressively more complex environments. Warlop et al. considered the hazard perception of cyclists with and without DCD and found that although eye fixation on hazards was significantly different among groups, no differences were observed in terms of reaction speed or identification of hazards. To explore the potential vulnerability of individuals with DCD as pedestrians, Wilmut and Purcell asked parents of children with DCD and adults with DCD about their roadside experiences. Individuals with DCD alongside ADHD/ASD were significantly more likely to engage in risky looking behaviors. Regardless of co-occurrence the vast majority of participants reported that motor difficulties influenced road crossing behavior. Although these studies are all very different, taken together they demonstrate clear difference between those with and those without DCD in high risk situations.

All of these studies are a timely reminder that the emerging movement we observe is a consequence of the *individual* we are measuring, the *task* we have set and the *environment* we have set up (Newell, 1986).

PROMOTING CHANGE

Finally, Smits-Engelsman et al. considered whether exergames could be used to improve physical fitness in children with DCD. A 10-week intervention improved both aerobic and anaerobic fitness as well as balance and coordination in children with DCD and controls. The findings highlight the potential for these games to do more than improve motor control and given the ease with which families can engage in exergames, without sustained input from a healthcare professional, their potential to be an affordable and accessible intervention is promising. In another study, Grohs et al. considered whether transcranial direct current stimulation (tDCS) of the primary motor cortex could boost motor learning. Although motor skill improved over the 5-day learning period, motor cortex tDCS did not enhance motor learning. The authors concede that targeting the cerebellum may have produced a different result. This is the first study to examine the therapeutic efficacy of tDCS on motor learning in children with DCD which future studies can draw upon. Again, although these are very different they highlight potential new avenues for how motor control can be influenced and changed.

AND FINALLY……

Included in this topic was a paper by Scott et al. which sits within Frontiers for Young Minds. This paper is not addressed to the scientific community but rather explains motor imagery and action observation interventions for children with DCD. Given the inaccessibility of academic research for certain stakeholders this is an excellent example of how scientific research can be disseminated to young people.

CONCLUSIONS

This Research Topic represents the reality of the broad range of diverse research being undertaken within the field of DCD. Within the DCD topic we see exciting progress in terms of understanding the underlying biology which is driving the behavior which we are all so familiar with. However, this is complemented by research aiming at understanding the individual experience of DCD and the impact it has on activities of daily life such as driving, crossing the road and walking down the stairs. This is a far cry from the very lab based work of 10 years ago. Finally, we also see work focusing on novel interventions and how we can help ensure movement is functional for people with

DCD and work modeling how our findings can be disseminated to a wide audience.

AUTHOR CONTRIBUTIONS

All authors listed have made a substantial, direct, and intellectual contribution to the work and approved it for publication.

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ORIGINAL RESEARCH
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Children With Developmental Coordination Disorder Exhibit Greater Stepping Error Despite Similar Gaze Patterns and State Anxiety Levels to Their Typically Developing Peers

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This study examined stepping accuracy, gaze behavior, and state-anxiety in children with (N = 21, age M = 10.81, SD = 1.89) and without (N = 18, age M = 11.39, SD = 2.06) developmental coordination disorder (DCD) during an adaptive locomotion task. Participants walked at a self-selected pace along a pathway, placing their foot into a raised rectangular floor-based target box followed by either no obstacles, one obstacle, or two obstacles. Stepping kinematics and accuracy were determined using three-dimensional motion capture, whilst gaze was determined using mobile eye-tracking equipment. The children with DCD displayed greater foot placement error and variability when placing their foot within the target box and were more likely to make contact with its edges than their typically developing (TD) peers. The DCD group also displayed greater variability in the length and width of their steps in the approach to the target box. No differences were observed between groups in any of the gaze variables measured, in mediolateral velocity of the center of mass during the swing phase into the target box, or in the levels of self-reported state-anxiety experienced prior to facing each task. We therefore provide the first quantifiable evidence that deficits to foot placement accuracy and precision may be partially responsible for the increased incidence of trips and falls in DCD, and that these deficits are likely to occur independently from gaze behavior and state-anxiety.

Keywords: developmental coordination disorder, fall-risk, gaze, kinematics, anxiety

INTRODUCTION

Developmental Coordination Disorder (DCD), also known as dyspraxia, affects around 5% of children and is characterized by difficulties in general motor skill learning and execution, which are independent of intellectual problems, visual impairments, and physical or diagnosed neurological disorders (American Psychiatric Association, 2013). The movements of children with DCD

are often described as awkward or clumsy and affect the ability to perform activities of daily living (ADLs). For example, children with DCD struggle walking around the environment safely (Van der Linde et al., 2015); an often overlooked skill that requires a complex interaction between the central nervous system, musculoskeletal system, sensory inputs from the visual, proprioceptive and vestibular systems, and environmental cues (Rossignol, 1996). Indeed, children with DCD appear to use shorter steps and a bent-forward posture to optimize safety when walking on a treadmill (Deconinck et al., 2006a) and display a reduced ability to control their momentum when crossing obstacles (Deconinck et al., 2010). Children with DCD also trip, fall, and bump into obstacles more frequently than their typically developing (TD) peers (Fox and Lent, 1996; Cleaton et al., 2020) which can negatively impact everyday life and the willingness to engage in sports and social activities (Kirby et al., 2011). Problems with walking can extend into adulthood, as exemplified by a recent study that showed adults with DCD reported falling more than 10 times over a 6-month period and tripping between one and five times per week (Scott-Roberts and Purcell, 2018).

Although laboratory studies have demonstrated that stability of gait is lower in children with DCD (Gentle et al., 2016; Speedtsberg et al., 2018) and that individuals with DCD fail to show key anticipatory adjustments when negotiating obstacles suddenly appearing in their walking path (Wilmut and Barnett, 2017), the mechanisms underpinning these differences have not been fully elucidated. Problems with internal (forward) modeling, balance control, rhythmic coordination, executive function, and aspects of sensoriperceptual function have been implicated as possible mechanisms of motor deficits in individuals with DCD (Wilson et al., 2013) but there is no direct evidence that these mechanisms can explain DCD-related changes in gait and posture. Therefore, there is a clear need for further exploration of the mechanisms underpinning walking problems of individuals with DCD so that effective interventions can be designed and implemented.

One potential mechanism of walking difficulties in DCD is the coupling between the visual and locomotor system. When navigating complex environments vision is critical for the acquisition of necessary information to guide safe stepping behavior. For example, when faced with stepping over a future obstacle, individuals typically look several steps ahead, fixating the obstacle and other task-relevant areas to plan future foot placement (Patla and Vickers, 2003; Marigold and Patla, 2007; Matthis et al., 2018). Additionally, when stepping onto a target, individuals tend to transfer their gaze toward the target prior to step initiation and maintain this fixation until around the time the step is completed (Hollands et al., 1995; Hollands and Marple-Horvat, 2001). It has been suggested that eye and stepping movements are programmed simultaneously as part of a coordinated eye-stepping movement (Hollands and Marple-Horvat, 2001), and that problems making accurate eye movements may lead to problems making accurate stepping movements (Hollands et al., 2017). It is, therefore, noteworthy that the oculomotor control of children with DCD differs from that of their TD peers. For example, children with DCD are less accurate during saccadic transitions to spatial targets (Katschmarsky et al., 2001) and struggle when faced with visually tracking a moving target (Robert et al., 2014; Sumner et al., 2018). Children with DCD also tend not to use predictive information to guide the planning of subsequent movements (Langaas et al., 1998; Wilmut and Wann, 2008; Ferguson et al., 2015), instead showing a preference to rely on visually guided online control (Smits-Engelsman et al., 2003; Wilmut et al., 2006; Debrabant et al., 2013) which has been shown to impair their ability to visually track and catch a ball (Miles et al., 2015). Importantly, these differences have recently been shown to persist in the context of walking (Warlop et al., 2020). Specifically, when faced with navigating sequential stepping targets, young adults with DCD walk slower and direct their gaze to the more proximal and immediate stepping targets compared to their TD peers. Difficulties using predictive control may therefore alter what is perceived to be the most task-relevant sources of visual information to guide action (Land and Lee, 1994; Land and Hayhoe, 2001) and encourage individuals with DCD to utilize slower (and online) sources of sensory feedback (Adams et al., 2014). Consequently, the extent to which vision is used to sufficiently identify and plan for subsequent stepping constraints may be limited. However, it is currently unknown whether these visuomotor deficits are also observed in children with DCD, and whether they contribute to decreased stepping accuracy and associated increased risk of falls.

Another potential mechanism for the movement problems in DCD, that has been hitherto unexplored, is the link between stepping accuracy and mental health issues, such as anxiety (Caçola, 2016). There is growing evidence that individuals with DCD have elevated levels of anxiety compared to their TD peers (Mancini et al., 2016, 2019; Omer et al., 2019), and that increased anxiety pertaining to mobility results in some adults with DCD exerting conscious effort to maintain balance and avoid tripping and falling (Scott-Roberts and Purcell, 2018). Whilst there is little known about any link between anxiety and fall risk in DCD, fear of falling is a known risk factor for falls in older adults and certain patient populations (Lord et al., 1993; Cumming and Klineberg, 1994; Delbaere et al., 2010) and can lead to changes to walking behavior that paradoxically increases the risk of tripping and falling (Young and Hollands, 2010, 2012b; Young et al., 2012; Young and Williams, 2015). For example, when approaching a stepping target followed by a series of obstacles, older adults with a high-risk of falling show a reduced tendency for feedforward and proactive visual search behaviors compared to their low-risk counterparts (Young et al., 2012). That is, they are more likely to only fixate the most immediate stepping constraints at the expense of sufficiently fixating the more distal and subsequent stepping constraints. This results in high-risk older adults, who also report heightened state-anxiety, sometimes looking away too early from the target box they are stepping onto which can result in inaccurate foot placement (Chapman and Hollands, 2006, 2007, 2010; Young and Hollands, 2012a; Young et al., 2012; Young and Williams, 2015). These findings therefore suggest that the increased likelihood of trips and falls in older adults are due, in part, to not looking in the right places at the right times; behavior shown to be directly linked to the effects of anxiety/fear of falling on attentional control processes (Young and Hollands,

2012b; Young and Williams, 2015; Ellmers and Young, 2019). Though it is currently unclear whether falls in older adults and children with DCD share common etiologies, the influence of anxiety on visuomotor control is a mechanism that may explain problems with effective gait in DCD populations.

The aim of the current experiment was to provide the first detailed account of the visuomotor control of stepping in children with and without DCD and to determine the extent to which deficits in stepping accuracy may be explained by anxiety and gaze behavior. Building upon recent insights to gaze behaviors during precision stepping in adults with DCD (Warlop et al., 2020), we report how children with and without DCD use gaze to preview a varying number of stepping constraints prior to precise foot placement within a floor-based target – providing the first quantification of foot placement error in children with and without DCD. We hypothesized that compared to their TD peers, children with DCD would display (1) greater foot placement error, (2) altered visual sampling during the approach to, and stepping into, our floor-based target, and (3) heightened levels of state-anxiety.

MATERIALS AND METHODS

Participants

Forty-seven participants aged between 8 and 15 years of age participated in the study, of which 28 were initially recruited for our DCD group. Participants in the DCD group were recruited using social media and from local DCD support groups, whilst participants in the TD group were recruited from the children of student and staff members of Liverpool John Moores University. The children in the DCD group satisfied the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) criteria (American Psychiatric Association, 2013). For example, the Developmental Coordination Disorder Questionnaire (DCDQ; Wilson et al., 2009) was completed by parents prior to testing to confirm that movement difficulties significantly interfered with their child's activities of daily living. Parents also confirmed that their child did not suffer from any general medical condition known to affect sensorimotor function (e.g., cerebral palsy, hemiplegia, or muscular dystrophy) and had no diagnosis of learning difficulties. Finally, participants in the DCD group were required to score below the 15th percentile on the test component of the Movement Assessment Battery for Children-2 (MABC-2; Henderson et al., 2007) carried out as part of the testing phase. This resulted in five participants' data being excluded from our analyses (min = 25th percentile). A further two participants were also excluded from the DCD group due to poor adherence to task instructions. Participants in the TD group were required to score above the 15th percentile, which resulted in the exclusion of one participant's data. This resulted in a net total of 21 participants in our DCD group (male = 12, female = 9) and 18 participants in our TD group (male = 10, female = 8). All participants were right footed. Parents also completed the Attentional Deficit/Hyperactivity Disorder (ADHD) Rating Scale —VI prior to testing (DuPaul et al., 1998) due to its high comorbidity with DCD (about 50% co-occurrence; American Psychiatric Association, 2013). None of the included children scored above the 98th percentile for inattention or hyperactivity, which is recommended to be the minimum cut-off used as an indication of ADHD in research (DuPaul et al., 1998). Ethical approval was granted by the Liverpool John Moores University Ethics Committee.

Kinematics

A 12 infra-red camera motion capture system (Qualisys, Gothenburg, Sweden) collected whole-body kinematic data at 80 Hz, with a total of 38 reflective markers placed on the feet, lower legs, thighs, pelvis, torso and head according to the conventional Plug-in Gait marker set. This included several additional markers to optimize segment tracking, one of which was placed on the "foot center" to guide each child's stepping behavior (see below). Finally, a triangular cluster of three reflective markers (14 mm diameter) were placed on each shoe over the forefront to track virtual landmarks created by a digitizing wand (C-Motion, Germantown, MD, United States) at the anterior-inferior (toe-tip) and posterior-inferior (heel-tip) point of each shoe. Marker trajectories were labeled and gap-filled using Qualisys Track Manager (QTM) before being exported as.c3d files to enable model application in Visual3D (C-Motion, Germantown, MD, United States). Finally, data were exported and analyzed in MATLAB (MathWorks, United States). All trajectories were smoothed using a bi-pass second order Butterworth low-pass digital filter with a 6 Hz cut-off.

Eye Tracker

Eye movements were recorded using a Pupil Labs binocular eye-tracking headset (Kassner et al., 2014) that featured two pupil cameras that recorded pupil movements at 60 Hz, and a scene camera to record the world view at 30 Hz. Prior to the task, children completed a 5-point screen marker calibration that was re-run every five trials or when the calibration accuracy had visibly been lost. If the child failed calibration after multiple attempts, or persistently lost calibration due to excessive movement of the eye-tracker, the task was run without the eye-tracker and their gaze data excluded. Participants were also only included in gaze analyses if they presented two or more usable eye-tracking trials per condition. In total, this resulted in the gaze data from 5 DCD children being excluded from the present study (age = 10.00 $\pm$ 0.71, Mabc-2 = 2.42 $\pm$ 3.75). Of the participants included in gaze analyses, an average of 1.68 trials (11.17%) in total (15 trials) were rejected from analyses (SD = 2.26, 15.24%). The characteristics of the DCD children included in the gaze analyses is presented in **Table 1**. The capture onset of the motion capture system provided a light emitting diode (LED) response that enabled synchronization between the eye tracker and the motion-capture system by identifying the frame in which this response was first seen.

Protocol

Data collection took place in a single session lasting approximately 2 h. Once fitted for kinematic and eye-tracking data collection, each child was permitted up to 2 min to walk freely around the lab to familiarize walking at their natural

TABLE 1 | Characteristics (mean ± SD) of all participants in the DCD and TD groups, and the characteristics of the subset of DCD participants whose gaze data were included in analyses.

	DCD	TD	DCD (gaze analyses)
Male (*n*)	12	10	10
Female (*n*)	9	8	6
Age (years)	10.81 ± 1.89	11.39 ± 2.06	11.06 ± 2.08
Height (cm)	152.25 ± 10.99	149.08 ± 13.06	153.50 ± 11.11
Weight (kg)	49.63 ±14.05	42.21 ± 14.69	51.20 ± 14.41
Mabc-2 (%)	1.60 ± 2.51	52.94 ± 31.03	1.34 ± 2.08

walking speed whilst wearing the testing equipment. Once their natural walking speed was agreed upon (confirmed by parent), the lab was marked out ready for the testing protocol. Baseline levels of state-anxiety were then measured using a child-friendly "fear thermometer"[1], which encompasses a 10-point "smiley-face" Likert scale ranging from 1 (low levels of anxiety) to 10 (high levels of anxiety). Specifically, each child was sat down on a chair and given a brief introduction to the thermometer. They were then asked how worried or anxious they currently felt about being in the laboratories and wearing our equipment. These simple scales have previously been validated against larger and more complex state-anxiety inventories (Houtman and Bakker, 1989).

The present study adopted a modified-version of the protocol previously used by Curzon-Jones and Hollands (2018) to investigate stepping safety in older adults. Specifically, participants were required to walk along a 7 m path, starting with their non-dominant foot, stepping accurately into a target box and over a varying number of obstacles until they reached the end of the course. The distance between the start-line, target box, and obstacles was personalized to each child's natural walking speed, such that their fourth step would intuitively place their dominant foot into the target box, and their sixth and eighth steps would place their dominant foot over the first and second obstacles, respectively (see **Figure 1**). To achieve this, each child walked along the entire pathway stepping onto a small sponge placed approximately where the target box center would later be located. The starting position was then adjusted until the above criteria were met.

The target box was a raised blue rectangular sponge outline with edges that were 5 cm high and 4 cm wide. Bespoke target boxes were created for each participant, ensuring that the length of the inside stepping area was 8cm plus the length at the longest part of the participant's right shoe, and the width was 8cm plus the width at the widest point of the participant's right shoe. The obstacles were formed using two 30 cm × 10 cm × 5 cm (height × depth × width) stabilizing wooden blocks positioned either side of a 4 cm × 4 cm × 65 cm polystyrene rectangular block. To also ensure the obstacles presented the same stepping constraint for each participant, the polystyrene block was attached to each stabilizing block using Velcro so it could be positioned at a height equating to 12% of body height. This height was chosen to closely match the constraints previously shown to induce fall-related anxiety in older adults (Curzon-Jones and Hollands, 2018).

Participants were informed that their goal was to reach the end of the course without knocking over the target box and/or the obstacles. Participants were also informed that they should step into the target box "as accurately and centrally as possible" – doing their best to minimize the distance between the additional "foot center" marker and what they perceived to be the center of the box. Three task difficulties were used: (1) no obstacles following the target box (Target only), (2) one (near) obstacle following the target box (One obstacle), (3) two (near and far) obstacles following the target box (Two obstacles). Participants completed five successive trials of each difficulty (one block). The order of each block was randomized (total 15 trials). Participants

[1] www.anxietycanada.com

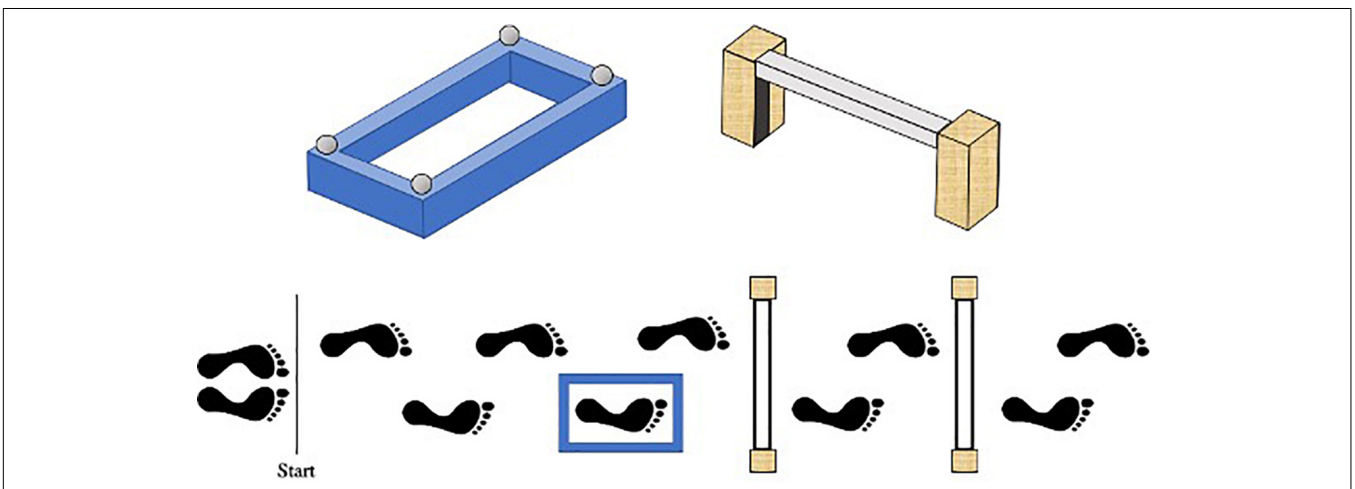

FIGURE 1 | Birds-eye schematic of our walking task. Starting with their left foot, participants had to walk along the path, step into the blue target box, and over either no obstacles, one (nearest) obstacle or two obstacles. The distance between the start line, target box, and obstacles was personalized to each child's preferred walking speed, such that their fourth step would naturally place their right foot into the target box, and their sixth and eighth steps would place their right foot over the first and second obstacles, respectively.

started each trial with their eyes closed, and after ten seconds, were verbally cued to "open" their eyes and initiate the trial. Starting each trial in this manner enabled route-previewing to be better standardized across participants and provide a point from which gaze data could be recorded. Immediately prior to each block of 5 trials, participants were again asked to report their levels of state-anxiety to determine how task difficulty influenced anxiety. Specifically, each child was asked how worried or anxious they currently felt about performing the upcoming set of trials.

Data Analysis

Foot Placement Variables

Foot placement error within the target box was determined as the relative distance between the foot center and the target center when the foot was placed inside the target. Foot center was calculated as the mid-point between the toe-tip and heel-tip. Target center was calculated as the mean of the four reflective marker (x, y) coordinates positioned on each corner of the target box. Both absolute error, constant error and variable error were calculated in the anteroposterior and mediolateral directions separately. Absolute error was defined as the mean scalar foot position distance (regardless of position) relative to the target center, reflecting foot placement accuracy. Constant error was defined as the mean vector foot position displacement (±) relative to the target, reflecting foot placement bias. Variable error was defined as the variability (one standard deviation) of the constant foot placement error across trial repetitions, reflecting precision of foot placement (Reynolds and Day, 2005; Chapman et al., 2012). Unlike absolute error, constant error captures directional foot placement bias as the mean vector foot displacement (±) relative to the target and is therefore better placed to measure variability. Positive values for anteroposterior and mediolateral constant error indicate the foot was positioned anterior and lateral of the target center, respectively. Finally, the experimenter manually recorded the total number of trials (out of 15 trials) that each participant accidentally contacted the target box. The lightweight design of the target box meant even a slight touch on its edges would result in a distortion to its rectangular shape and often knock it over. This allowed the experimenter to easily determine when the box had been contacted so it could be reset for the following trial.

Stepping Kinematics and Approach Speed

Heel-strike and toe-off gait events were determined using the local maxima and local minima of the heel and toe referenced to the pelvis segment, respectively (Zeni et al., 2008). Using these gait events, spatial step kinematics were calculated based on the position of the foot center (mid-point between the toe-tip and heel-tip). Step length was defined as the antero-posterior distance between the left and right foot centers at each heel-strike. Step width was defined as the medio-lateral distance between the left and right foot centers at each heel-strike. As these measures are highly dependent on body morphology, we chose to measure the variability (one standard deviation) in the length and width of the steps up to and including the final step into the target box, which can give insights to the ability to produce consistent movement patterns (Rosengren et al., 2009). Approach velocity was calculated as the mean horizontal velocity of the anterior trunk marker, from the first heel strike to the instant of touch-down within the target box. Finally, we examined balance control by measuring the maximal mediolateral velocity of the center of mass (CoM) during the swing phase of the targeting step into the box (Deconinck et al., 2010). Variability of this measure was also calculated as one standard deviation across each block of trials.

Gaze Variables

Gaze fixations were defined as a gaze stabilization on a location in the environment for three frames or longer (corresponding to ∼90 ms). Fixations were classified as being spatially located on one of three primary areas of interest: (1) immediate walkway (walkway preceding target box); (2) target box; (3) distal walkway (the sum of all fixations directed toward the path and/or stepping constraints following the target box). We chose to classify distal fixations as a single area of interest given their low summed-frequency, and to allow comparisons between the three task difficulties. These areas of interest were used to determine the duration spent fixating each location prior to stepping in the target box. Fixation durations were also normalized to individual trial length by presenting data as the percentage of time spent fixating each area of interest from the point when participants opened their eyes following the "open" cue, until the time when they stepped into the target box. We also measured the timing of the final gaze transfer toward the target box and the final gaze transfer away from the target box relative to foot contact within it, with a negative value denoting an early transfer of gaze. Other gaze variables included mean fixation duration, fixation rate (number of fixations per second), and number of gaze transfer between areas of interest.

Statistical Analyses

Kinematic and gaze variables were primarily analyzed using two-way mixed design repeated measures ANOVAs, with between-subject effects of group (x2; DCD; TD), within-subject effects of task difficulty (x3; Target only; One obstacle; Two obstacles), and interaction between terms. Significant effects were probed by polynomial trend analyses, and *post hoc* analyses were performed using pairwise comparisons with Sidak-corrections to account for the multiple comparison problem (Blakesley et al., 2009). ANOVA effect sizes were reported using partial eta squared (η_p^2), common indicative thresholds for which are small (0.01), medium (0.06) and large (0.14; Field, 2013). The results of univariate tests are reported, with the Huynh-Feldt correction procedure applied for analyses that violated the sphericity of variance. For step length variability, a natural-log transformation was applied to achieve a normal distribution. Where a normal distribution could not be achieved, within-participant effects were analyzed using Friedman's ANOVA with Bonferroni corrected Wilcoxon-signed rank tests used for *post hoc* analyses. Conversely, between-participant effects were analyzed using Mann-Whitney U tests. Non-parametric effect sizes were reported as $r = Z/\sqrt{N}$, for which common thresholds are small (0.1), medium (0.3) and large (0.5; Rosenthal, 1986). All statistical analyses were performed using IBM SPSS statistics (version 26) with an alpha level of ≤ 0.05.

RESULTS

Foot Placement Variables

Stepping Accuracy and Precision

There was a significant main effect of Group on absolute AP error, $F(1, 37) = 21.063$, $p < 0.001$, $\eta_p^2 = 0.363$, and constant AP error, $F(1, 37) = 7.020$, $p = 0.012$, $\eta_p^2 = 0.159$. Children with DCD had greater absolute AP error ($M = 2.6$ cm) compared to TD children ($M = 1.6$ cm) and tended to undershoot their foot placement ($M = -1.1$ cm) compared to TD children ($M = 0.2$ cm). A significant main effect of Group was also observed for AP, $F(1, 37) = 9.932$, $p = 0.003$, $\eta_p^2 = 0.212$, and ML variable error, $F(1, 37) = 10.011$, $p = 0.003$, $\eta_p^2 = 0.213$. Children with DCD exhibited greater AP ($M = 2.1$ cm) and ML ($M = 1.3$ cm) variable error compared to TD children ($M = 1.5$ cm and M = 0.9 cm, respectively). There was also no main effect of Difficulty, or interaction between Difficulty and Group, for all foot placement variables (**Figure 2**).

Total Box Contacts

Results from a Mann–Whitney *U*-test showed a significant difference between groups, $U = 118$, $z = -2.076$, $p = 0.038$, $r = -0.3329$, with more box contacts observed in the DCD group ($M = 1.82 \pm 1.41$) compared to the TD group ($M = 1.00 \pm 1.24$).

Stepping Kinematics and Approach Speed

Approach Speed

The main effect of difficulty failed to reach significance, $F(2, 74) = 2.968$, $p = 0.058$, $\eta_p^2 = 0.074$, but was significantly described by a linear polynomial trend ($p = 0.022$, $\eta_p^2 = 0.134$) with fastest approach speeds observed when faced with the target only ($M = 0.995$ m/s) and slowest approach speeds observed when faced with two obstacles ($M = 0.969$ m/s). There was no main effect of Group, $F(1, 37) = 3.273$, $p = 0.079$, $\eta_p^2 = 0.081$, and no Group × Difficulty interaction, $F(2, 74) = 0.174$, $p = 0.841$, $\eta_p^2 = 0.005$.

Stepping Variability

Results showed a significant main effect of Group for step length variability, $F(1, 37) = 6.423$, $p = 0.016$, $\eta_p^2 = 0.148$, with greater variability observed in the DCD group ($M = 10.6$ cm) compared to the TD group ($M = 6.7$ cm). There was also a main effect of Group for step width variability, $F(1, 35) = 4.958$, $p = 0.032$, $\eta_p^2 = 0.124$, with greater variability again observed in the DCD group ($M = 5.0$cm) compared to the TD group ($M = 3.8$ cm). There was no main effect of Difficulty and no Group x Difficulty interaction for either step length or step width variability.

Mediolateral CoM Velocity

There was no main effect of Group, $F(1, 37) = 0.128$, $p = 0.722$, $\eta_p^2 = 0.003$, no main effect of Difficulty, $F(2, 74) = 0.228$, $p = 0.796$, $\eta_p^2 = 0.006$, and no Group x Difficulty interaction, $F(2, 74) = 1.00$, $p = 0.373$, $\eta_p^2 = 0.026$, in the maximal mediolateral CoM velocity during the swing phase into the box. There was also no effect of Group, $F(1, 37) = 1.762$, $p = 0.193$, $\eta_p^2 = 0.045$, no effect of Difficulty, $F(2, 74) = 0.137$, $p = 0.872$, $\eta_p^2 = 0.004$, and no Group × Difficulty interaction, $F(2, 74) = 1.914$, $p = 0.155$, $\eta_p^2 = 0.049$, in the inter-trial variability (1 SD) of maximal CoM mediolateral velocity.

Gaze Behavior

Gaze fixations to task related areas of interest accounted for an average of 75.0, 73.7 and 73.5% of the total time taken to step into the target box for the Target-only, One obstacle, and Two obstacle conditions, respectively. There were no significant differences between groups for fixation duration, fixation rate, number of gaze transfers between AOI's, the total time spent fixating each AOI, or the onset of the final gaze shift toward the target prior to heel contact. These data are presented in **Table 2**.

Gaze Location

As data for gaze location were non-normally distributed, Friedman's ANOVA's was utilized to investigate within-participant effects and Mann–Whitney *U*-tests were utilized to investigate between participant effects. A Friedman's ANOVA showed the allocation of gaze to significantly differ between

TABLE 2 | Mean (± SD) values of gaze variables and state anxiety for both the DCD and TD groups for each of the three task difficulties.

	DCD			TD		
	Target only	**1 obstacle**	**2 obstacles**	**Target only**	**1 obstacle**	**2 obstacles**
Immediate walkway (%)	10.63 ± 14.00	11.75 ± 13.38	9.44 ± 12.23	11.12 ± 10.22	10.44 ± 10.73	11.72 ± 13.22
Target box (%)	61.00 ± 14.61	53.69 ± 15.13	55.56 ± 15.06	59.00 ± 17.12	56.61 ± 14.02	53.28 ± 16.51
Distal (%)	3.56 ± 3.79	8.94 ± 7.34	8.06 ± 8.15	5.13 ± 6.77	6.56 ± 6.92	8.28 ± 9.10
Immediate walkway (s)	0.45 ± 0.68	0.44 ± 0.55	0.42 ± 0.59	0.41 ± 0.38	0.39 ± 0.43	0.45 ± 0.54
Target box (s)	2.21 ± 0.59	1.97 ± 0.59	1.99 ± 0.54	2.00 ± 0.51	1.89 ± 0.49	1.79 ± 0.56
Distal (s)	0.20 ± 0.31	0.37 ± 0.39	0.24 ± 0.19	0.14 ± 0.16	0.22 ± 0.21	0.28 ± 0.28
Gaze shift toward box (s)	−2.22 ± 0.66	−2.06 ± 0.72	−2.14 ± 0.55	−1.84 ± 0.51	−2.01 ± 0.59	−1.85 ± 0.50
Gaze shift away from box (s)	0.13 ± 0.17	−0.03 ± 0.26	0.05 ± 0.15	0.11 ± 0.26	0.02 ± 0.20	0.01 ± 0.16
Gaze transfers	1.46 ± 0.96	1.96 ± 0.84	1.95 ± 0.98	1.85 ± 1.00	1.89 ± 0.98	2.01 ± 0.81
Fixation duration (s)	0.56 ± 0.14	0.54 ± 0.17	0.51 ± 0.20	0.53 ± 0.15	0.53 ± 0.14	0.50 ± 0.11
Fixation rate (fix per s)	2.19 ± 0.69	2.23 ± 0.59	2.11 ± 0.67	2.01 ± 0.65	2.09 ± 0.52	2.06 ± 0.60
State-anxiety	1.62 ± 1.12	2.05 ± 1.47	2.19 ± 1.75	1.42 ± 0.69	1.28 ± 0.46	1.28 ± 0.46

AOI's when faced with the target alone, $X(2) = 54.500$, $p < 0.001$, one obstacle, $X(2) = 52.757$, $p < 0.001$, and two obstacles, $X(2) = 47.197$, $p < 0.001$. Follow-up Wilcoxon tests with Bonferroni corrections (α adjusted to 0.0167) showed that for all task difficulties gaze-allocation was significantly greatest for the target-box ($ps < 0.001$), whilst there was no significant difference in gaze allocation between the immediate walkway and distal AOI's ($ps < 0.065$). A Friedman's ANOVA also showed distal fixations to significantly change across task difficulties, $X^2(2) = 11.123$, $p = 0.004$, with Wilcoxon tests with Bonferroni corrections (α adjusted to 0.0167) showing significantly greater distal fixations to occur when faced with either one obstacle, $Mdn = 7.00\%$, $z = -2.855$, $p = 0.004$, $r = -0.4694$, or two obstacles, $Mdn = 7.00\%$, $z = -2.812$, $p = 0.005$, $r = -0.4623$, compared to the target alone ($Mdn = 2\%$). Fixations to the immediate walkway, $X(2) = 2.279$, $p = 0.320$, and to the target box, $X(2) = 3.748$, $p = 0.153$, did not significantly change across task difficulties. Finally, separate Mann-Whitney U tests comparing the spatial allocation of gaze between TD and DCD groups for each AOI, and across each task difficulty, failed to show any significant differences between groups ($ps > 0.176$). These data are presented in **Table 2**.

Gaze Transfer From Target

There was a significant main effect of Difficulty, $F(2, 64) = 8.128$, $p = 0.001$, $\eta_p^2 = 0.203$, with *post hoc* comparisons revealing significantly earlier transfers of gaze when faced with one obstacle ($M = -5$ ms, $p = 0.003$) or two obstacles ($M = 30$ ms, $p = 0.031$) compared to when faced with the target box alone ($M = 121$ ms). There was no main effect of Group, $F(1, 32) = 0.004$, $p = 0.948$, $\eta_p^2 = 0.000$, and no Group × Difficulty interaction, $F(2, 64) = 0.986$, $p = 0.379$, $\eta_p^2 = 0.030$ (**Figure 3**).

Anxiety

State Anxiety

A Friedman's ANOVA showed anxiety to significantly differ across each instance of measurement, $X(3) = 12.854$, $p = 0.005$, with *post hoc* Bonferroni corrected ($\alpha = 0.012$) Wilcoxon signed ranks tests revealing significantly higher levels of anxiety at baseline ($M = 2.06$) compared to when faced with the target only ($M = 1.53$, $z = -2.635$, $p = 0.008$, $r = -0.388$) and when faced with one obstacle ($M = 1.73$, $z = -2.500$, $p = 0.012$, $r = -3.686$). Results from separate Mann–Whitney *U*-tests showed no significant differences between groups at any point of measurement ($ps > 0.075$). These data are presented in **Table 2**.

DISCUSSION

The present study is the first to quantify foot placement accuracy in children with DCD and to determine the underlying characteristics of gaze and anxiety. Our results show that children with DCD are less accurate than their TD peers when tasked with precisely placing their foot within a floor-based target and are more likely to accidentally contact its edges. The DCD group primarily showed lower foot placement accuracy than the TD group in the anteroposterior plane, which, when considering constant foot placement error (**Figure 2**), appeared to be a tendency to undershoot the target center (∼1.1 cm). However, the DCD group also displayed greater variable error in both the anteroposterior and mediolateral planes. We therefore provide the first quantifiable evidence that decreases in foot placement accuracy (increased AP error) and precision (increased AP and ML variable error) may be partially responsible for the increased incidence of trips and falls in DCD (Chapman and Hollands, 2007). Interestingly, no differences were observed between groups in the maximal mediolateral CoM velocity during the swing phase into the target box, suggesting that poor foot placement was, in this instance, not underpinned by decreased balance control (Deconinck et al., 2010). In addition to foot placement error, the children with DCD also exhibited significantly greater variability in the length and width of their steps preceding the stepping target. As variability in these gait parameters have previously been observed in adults with DCD (Du et al., 2015) and linked to fall-risk in older adults (Maki, 1997), our findings provide evidence of an inherent deficit in the ability to produce consistent and stable stepping movements in children with DCD.

Contrary to our hypotheses, no differences in gaze behavior were found between groups on any metric reported. Both groups allocated the majority of their gaze toward the target box during the approach toward it, with fixations to the distal pathway minimal, yet increasing when a future obstacle(s) had to be negotiated (∼7% of total gaze). The timing between looking away from the target box and stepping within it was also similar between groups, occurring approximately 120 ms after foot contact when faced with the target alone, and approximately at the instant of foot contact (∼10 ms) following the introduction of an obstacle(s). These similarities may be explained by the fact that our DCD participants did not experience heightened state anxiety pertaining to the completion of our task. Indeed, anxiety was generally low and highly variable in the DCD group, with at least 50% of the cohort reporting the lowest possible levels of anxiety prior to facing each of the three walking tasks. Anxiety was also highest at baseline for both groups, which suggests an anxiety response unrelated to the fear of falling, such as the fear of performing to unfamiliar people in unfamiliar surroundings (i.e., social phobia, see Beidel et al., 1995). Consequently, work is still needed to determine the extent to which gaze and stepping performance might be altered in the presence of heightened anxiety. To achieve this, researchers could explore time-pressure and/or dual-task situations as they have both been shown to induce anxiety (Uemura et al., 2012; Zult et al., 2019) and exacerbate motor difficulties in children with DCD (Wilson and McKenzie, 1998). Alternatively, researchers could explore ecologically valid tasks in which the cost of falling is much greater, such as when walking up and down a staircase. Yet, the similarities in gaze behavior between groups may also be attributable to the predictability of our tasks' dimensions prior to the target box. As gaze behavior is known to be driven by context complexity and task specificity (Aivar et al., 2005; Miyasike-daSilva et al., 2011), knowledge that the target would consistently be reached on the fourth step for all trials may have reduced between group differences in visual exploration.

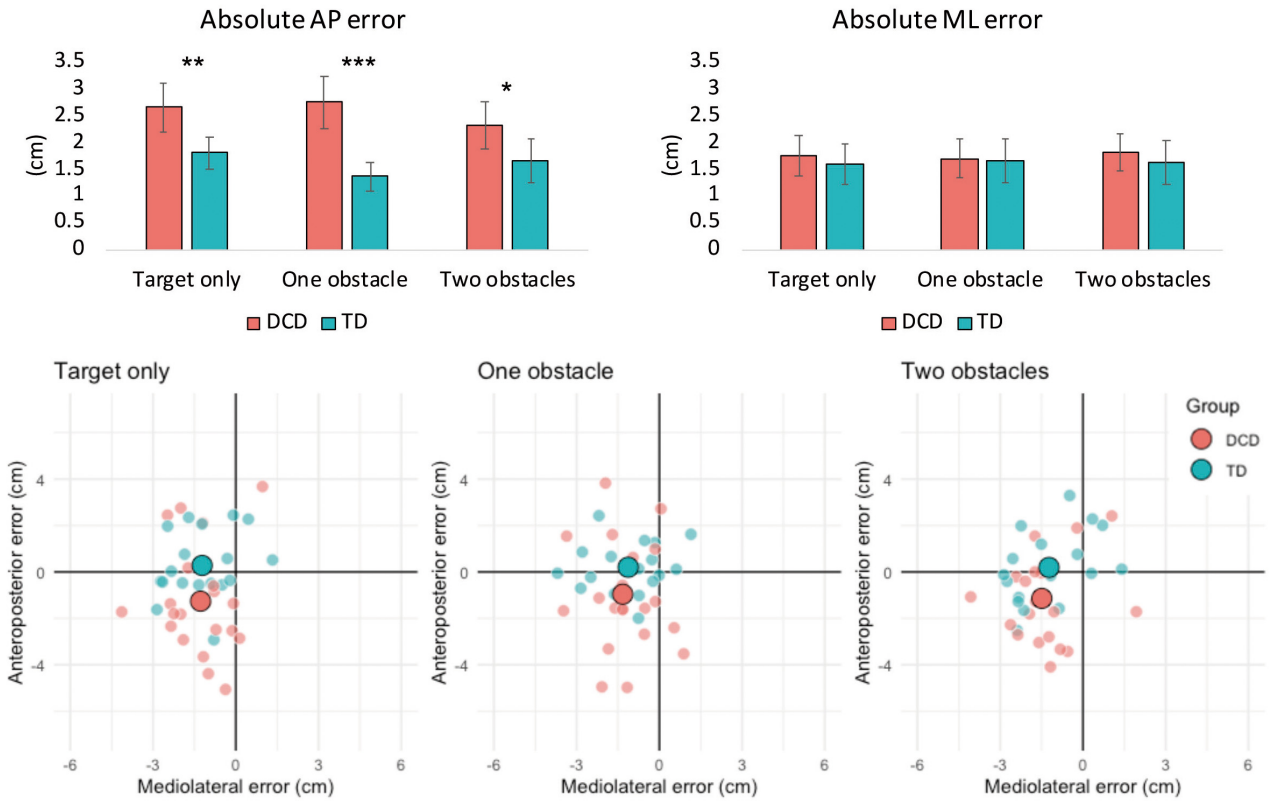

FIGURE 2 | Bar charts (top) representing mean (± 95% CI) absolute foot placement error in both the anteroposterior (left) and mediolateral (right) directions of movement. Asterisks signify between group differences at the < 0.05 (*), 0.01 (**), and 0.00 (***) levels. Constant foot placement error (bottom) for the DCD and TD groups for the target only, one obstacle and two obstacle task conditions. The large data points represent the group means, whilst the smaller data points represent the mean values of individual participants. Negative values on the horizontal and/or vertical axes indicate that the foot was positioned medial and/or posterior of the target center, respectively.

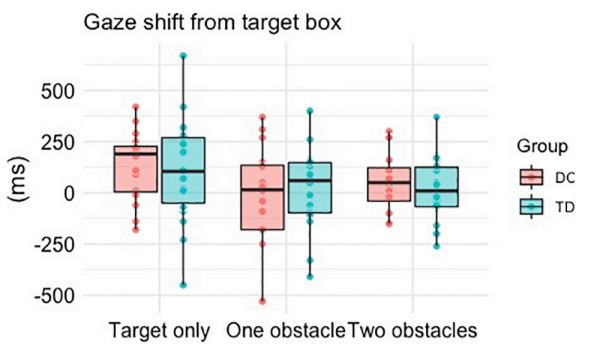

FIGURE 3 | Boxplots and individual mean values for the time taken (ms) to shift gaze away from the target box relative to stepping within it. Positive times reflect gaze to be shifted after foot contact within the box, whereas a negative time reflects an "early" shift of gaze prior to foot contact within the box.

Given the similarities in gaze behavior and anxiety between the DCD and TD groups, the results of the present study suggest that difficulties producing precise stepping actions in children with DCD occur independent of anxiety and overt attentional processes related to gaze behavior. As such, fall-risk in children with DCD may be better-explained by general deficits in neuromuscular control and the integration of acquired perceptual information during locomotion. For example, previous research has shown children with DCD exhibit greater variability in their shank and thigh movements during gait, suggesting an inherent difficulty controlling the lower limbs during locomotion (Rosengren et al., 2009). The extent of variability in the shank also appears to be greatest during the stance phase (Rosengren et al., 2009), which might explain increased variability when placing the targeting limb. Furthermore, when walking in dark conditions, children with DCD walk slower and sway more than TD children, suggesting a reduced ability to utilize proprioceptive and vestibular inputs to compensate for visual information and achieve a normal gait pattern (Deconinck et al., 2006b). Children with DCD also display slower and less accurate rapid online control, which is only achieved through the seamless integration of predictive models of movement and feedback mechanisms (Hyde and Wilson, 2011). Deficits in the ability to rapidly integrate information from the visual, vestibular and proprioceptive systems may therefore inhibit the extent to which children with DCD are able to accurately update and correct an ongoing stepping command whilst visually guiding the foot toward a

floor-based target (Gentle et al., 2016; Wilmut et al., 2016). However, it is worth acknowledging that, in the absence of any differences in overt attention (spatial location of gaze), differences may still exist in covert attention. Recent evidence has shown increased gait instability to be associated with an internal focus of attention (focusing on one's own movements) relative to an external focus of attention (focusing on the impact of the movement on the environment; Mak et al., 2019, 2020). Future work should therefore elucidate the covert attentional processes that underpin adaptive gait performance in children with DCD and its relative impact on stepping accuracy.

The results of this study may be limited by several factors. For example, it is important to acknowledge that our sample size is relatively small, and the age range of our participants is relatively heterogenous. Researchers should therefore take care when extrapolating our findings to children with DCD of all ages given evidence that the control of visually guided stepping goes through distinct changes throughout development (Mowbray et al., 2019) and that adaptations to walking on uneven terrain are better distinguished between DCD and TD individuals at childhood as opposed to adolescence (Gentle et al., 2016). Additionally, developmental aspects of emotional self-perception may question the accuracy of our simple self-report measure of state-anxiety (Smith et al., 2006). However, the similarity in gaze behaviors between groups may reinforce a similarity in their experienced anxiety, given the wealth of aforementioned research showing how anxiety can alter visual exploration during locomotor tasks. Regardless, future research would benefit from attempts to objectively capture physiological state-anxiety responses to complement additional measures of self-report. Finally, it should be reiterated that our findings only allow us to comment on the stepping performance of children with DCD in the *absence* of task-related anxiety. It is therefore important for future research to experimentally manipulate anxiety if we are to fully explore its role in fall-risk in children with DCD.

CONCLUSION

To conclude, our findings provide the first quantifiable evidence that children with DCD display reduced foot placement accuracy and precision compared to their TD peers. We also provide evidence that these reductions in foot placement accuracy are likely to occur independently of differences in gaze behavior and anxiety, suggesting a general deficit in neuromuscular control and a reduced ability to rapidly integrate perceptual information from the visual, proprioceptive and vestibular systems to guide stepping actions. However, as state anxiety was generally low, more research is needed to explore whether children with DCD may be more susceptible to anxiety-driven maladaptive gaze under more demanding situations.

DATA AVAILABILITY STATEMENT

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

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Liverpool John Moores Ethics Committee. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

MH, RF, and GW acquired the funding and edited the manuscript. MH, RF, GW, and JP designed the study. JP undertaken the data collection, completed the statistical analysis, figure preparation, and the first draft of the manuscript. All authors approved the final version of the manuscript for submission.

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The Lived Experience of Crossing the Road When You Have Developmental Coordination Disorder (DCD): The Perspectives of Parents of Children With DCD and Adults With DCD

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Pedestrians are one of the most vulnerable groups at the roadside, furthermore, previous research has demonstrated perceptual-motor limitations in individuals with DCD which may put these individuals at even more at risk in the context of road crossing. However, it is unclear whether this is the lived experience of these individuals at the roadside. Furthermore, difficulties with road crossing and safety have been found in other neurodevelopmental disorders but the impact this might have on an individual with co-occurring difficulties is unknown. Therefore, we utilized a questionnaire to survey the lived experience of adults with DCD and parents of children with DCD with the specific objectives of describing behaviors exhibited by adults and children with DCD (the latter reported by parents) at the roadside and to determine the how these individuals perceive road crossing actions. For each of these we compared different co-occurrence groups. We also had one final objective which was not focused on road crossing but more on the general perception of accidents and unrealistic optimism. Individuals with co-occurrences which have previously been linked to unsafe crossing behaviors (i.e., ADHD, ASD, and LD) reported a greater regularity of dangerous looking behavior (forgetting to look, running without looking) and visibility (crossing between cars, crossing when you can't see), these adults and the parents of these children were seemingly aware of the risky nature of these behaviors. When asked "why" crossing ability might be different, perceptual and motor difficulties alongside heightened awareness of risk and lowered awareness of risk were all cited by participants. Unrealistic optimism was not an explanation for the risky behavior in adults with DCD and in fact, these adults demonstrated a clear understanding of the likelihood of accidents. The findings of this study suggest that road crossing is perceived to be more challenging for both children and adults with DCD and this needs to be taken into account when considering remediation for this group.

Keywords: pedestrians, Developmental Coordination Disorder, co-occurrences, risky behavior, road crossing

INTRODUCTION

According to the World Health Organization, more than 1.25 million people die each year as a result of preventable road traffic accidents (World Health Organization, 2018). Furthermore, tens of millions of people are injured or disabled on the world's roads each year, with a high proportion of these people being pedestrians, i.e., in 2017 a total of 21% of all people killed on roads were pedestrians (European Commision, 2018). Although there is no accurate record of the economic cost of pedestrian deaths alone, the estimated cost borne from road traffic accidents is £12bn per annum in Great Britain (Department for Transport, 2019), or between 1 and 2% of gross national product (World Health Organization, 2013).

Clearly road traffic accidents bear both an economical and societal cost and pedestrians are considered to be a vulnerable road user. However, a population who might be at an even greater risk at the roadside is children and adults with Developmental Coordination Disorder (DCD). DCD occurs in 2–20% of the general population (Blank et al., 2019) and is an idiopathic condition characterized by marked impairments in motor coordination that negatively impact on activities of daily living which persist into adulthood (Kirby et al., 2013). No direct statistics exist regarding the number of roadside accidents for this group, however, recent research from our lab has highlighted a potential heightened vulnerability in this population (Purcell et al., 2011, 2012, 2017; Purcell and Romijn, 2017). For example, the ability to detect a vehicle as approaching (looming sensitivity) is consistently poorer in primary school aged children with DCD (Purcell et al., 2012) with children with DCD failing to detect vehicles approaching at speeds in excess of 14 mph in laboratory conditions, compared to 20 mph in primary school aged typically developing children (Wann et al., 2011). Assuming a vehicle is approaching above visual-perceptual threshold, and can be seen as moving, the child must then make a judgment regarding time-to-contact (TTC), previous evidence suggests further immaturities in children with DCD in TTC tasks as compared to their typically developing peers (Purcell et al., 2011). The studies mentioned above considered the perceptual component of road crossing, however, the ability to safely cross a road is a perceptual-motor skill which involves coordination between a pedestrian's perception of the approaching vehicle and their locomotive capability to execute the road crossing action. Previous evidence has demonstrated that children with DCD accept significantly shorter temporal crossing gaps as speed increases and tend to choose gaps which are too short given their walking speed (Purcell et al., 2017). Furthermore, although a DCD population was not included in their study, (Pitcairn and Edlmann, 2000) did find a correlation between poorer performance on a fine motor control task and errors in roadside perceptual judgments, both in typically developing children and adults. Taken together, the above program of research provides a clear indication that the perceptual-motor system in children with DCD may put them more at risk at the roadside. However, these limitations to the system could be overcome by: waiting a long time to cross; always using a signalized crossing and/or always crossing with another individual. These factors are difficult to determine in a lab based environment therefore, it is important to consider road crossing behavior and experience outside of a lab environment.

Two studies have attempted to explore the road crossing experiences of children with DCD. The first asked a small group of children with DCD to self-rate their perception of their ability and confidence in a road crossing environment and found no difference in either perceived crossing ability scores or confidence in their ability to execute a safe road crossing by themselves compared to their typically developing peers (Purcell, 2012). However, the children with DCD did perceive the task as significantly more dangerous. Likewise, a later study found no difference between children with DCD and their typical peers with regards to their self-reported knowledge of safe crossing places, their confidence in road crossing skills, their perceived road crossing ability, how often they felt they misjudged traffic gaps or having to wait a long time to cross (Purcell and Romijn, 2017). The mismatch here between the child's perceived judgment of their ability and previously identified limitations of the perceptual-motor system in children with DCD is striking and would suggest that this could place these children at even greater risk as they have less refined perceptual-motor skills and don't seem to recognize this limitation. These previous studies have made direct comparisons between how primary school aged children with DCD and their typically developing peers perceived their behaviors at the roadside (i.e., were they risky or not), but they didn't ask participants to rate the occurrence of specific behaviors. This is an important distinction as children may not self-report exhibiting risky behavior but they may often exhibit a behavior which would normally be considered as risky. For this reason the current study goes one step further and asks about specific behaviors. In addition, we also include a sample of adults with DCD. Despite the paucity of current literature we do know that DCD is a lifelong condition and adults with DCD continue to experience difficulties throughout their adult life (Cousins and Smyth, 2003). Which includes difficulties which are key in a road crossing context such as visual motor integration (de Oliveira et al., 2014), gait variability (Du et al., 2015), and executive functioning (Tal Saban et al., 2014). Furthermore, whereas a child may often be accompanied at the roadside either by friends, siblings or a parent this is rarely true for adults. Therefore, describing the experiences of a group of adults with DCD is an important first step. The current study, therefore, describes the lived experience of crossing the road in a large sample of individuals with DCD and their parents.

One interesting finding raised by Purcell and Romijn (2017) in their questionnaire was that children with DCD rated the act of crossing the road as more dangerous than their typically developing peers. The idea of how dangerous an activity is, has been considered in typically developing children and adults, if we see an activity as not posing any danger we would approach it differently from an activity which is seen as dangerous. However, our perception of danger can be biased, one such bias which lessens the appreciation of risk is unrealistic optimism or the "it won't happen to me" mentality (White et al., 2011; Shepperd et al., 2013). Studies which have demonstrated unrealistic optimism in typical adults include, but are not limited to, estimating

the risk of a heart attack (Radcliffe and Klein, 2002), the risk of experiencing severe alcohol problems in the future (Dillard et al., 2006), women's estimated risk for breast cancer (Waters et al., 2011), and smokers' estimated risk of cancer (Ayanian and Cleary, 1999). Unrealistic optimism has also been considered in children (Sissons Joshi et al., 2017) with participants comparing the likelihood of common childhood accidents happening to them compared to their peers, children consistently stated that an accident was "less likely" to happen to them. Children cited reasons such as "heightened skill" or "lack of exposure" as reasons for why these accidents were less likely. Therefore, this study suggests that typically developing children are unrealistic about the likelihood of accidents and so might take risks without perceiving them as risks. Whether this unrealistic optimism extends into individuals with DCD is unclear, however, studies have demonstrated that unrealistic conditional optimism is responsive to factors such as controllability and personal experience (Helweg-Larsen and Shepperd, 2001) and so might not be as prevalent in individuals with DCD if they perceive themselves as less in control because of their motor difficulties or have had previous experience of such accidents. The important factor here is that evidence suggests that people are less likely to take precautions if they perceive their absolute risk as low (Floyd et al., 2000) and so understanding the perception of risk in this population is key.

A final consideration in the current study is co-occurrences with other neurodevelopmental disorders. Often research studies might exclude participants on the basis of additional neurodevelopmental disorders in order to be sure that their research is describing the effects of a single disorder rather than looking at something else. However, in terms of the lived experience, an individual with multiple neurodevelopmental disorders experience the effects of all of them simultaneously. Research has shown that children with Attention Deficit Hyperactivity Disorder (ADHD, ADD) are more at risk at the roadside as pedestrians (DiScala et al., 1998; Brook et al., 2006), are less concerned about risk (Farmer and Peterson, 1995; Mori and Peterson, 1995) and accept crossing gaps which leave them with small safety margins (Clancy et al., 2006; Stavrinos et al., 2011). Furthermore, children with Autism Spectrum Disorder (ASD) show a poor understanding of how to use traffic signals (Josman et al., 2008) and adults with ASD display different looking behavior compared to peers (Earl et al., 2016, 2018; Cowan et al., 2018). Finally children with learning disabilities (LD) have been found to demonstrate difficulties in identifying safe crossing places (Anastasia, 2010). Although the studies cited above widely ignore other neurodevelopmental disorders it does suggest that co-occurrence of multiple neurodevelopmental disorders might place an individual at an even heightened level of risk as a pedestrian.

The current studies primary aim was to describe and explore the lived experience of adults with DCD and parents of children with DCD at the roadside and to highlight factors which may influence this, with a specific focus on the impact of co-occurrences. Within this primary aim we had two distinct objectives which focused on road crossing, the first was to describe the behaviors adults with DCD and children with DCD (as reported by their parents) exhibit at the roadside. The second was to determine the how these individuals perceive their road crossing actions (using both closed questions and an open question). We also had one final objective which was not focused on road crossing but more on the general perception of accidents and unrealistic optimism. Some of the accidents that participants were asked about have a clear motor component and so for these we expected adults with DCD to rate themselves as being more likely to experience an accident of that type and in fact they might be more at risk potentially making it a realistic judgment. However, whether this extends to accidents without an overt motor component is unclear. Within these research questions we considered the issue of co-occurrences by comparing a group with DCD and DCD plus one (or more) neurodevelopmental disorders which have not been found to have any potential road crossing difficulties (i.e., Dyslexia, Dyscalculia, DLD, etc.) to those with DCD plus one (or more) of the neurodevelopmental disorders shown to have potential road crossing difficulties (i.e., ADHD/ADD, ASD, LD).

MATERIALS AND METHODS

Participants

Participants were recruited via two methods, via the author's links on social media and via the author's personal contacts with individuals with DCD and parents of individuals with DCD. A total of 93 adults answering for themselves completed the questionnaire, however, six of these adults indicated no DCD (or Dyspraxia) related difficulties either diagnosed or undiagnosed and as such these adults were excluded, resulting in a total of 87 adult respondents with DCD. The majority of adults, 62.1%, with DCD reported crossing roads every day. Respondent demography is summarized in **Table 1**. A total of 75 parents completed the questionnaire, however, four of these indicated no DCD or Dyspraxia related difficulties in their child either diagnosed or undiagnosed and three participants indicated the age of their child to be over 18 years of age and so these participants were excluded leaving a final sample of 68 parent respondents. A total of 44.1% of the parents reported their child crossed the road every day, with 33.8% reporting that their child never crossed the road (either accompanied or unaccompanied). Respondent demography is summarized in **Table 1**.

TABLE 1 | Participant demographics.

	Adults	Parents
N	87	68
Age range	17–73 years	6–18 years
Mean age	32 years	11 years
Gender ratio	58 female, 24 male, 5 neither	16 female, 51 male, 1 neither
% from United Kingdom	82	75
DCD + ADHD (N)	20	30
DCD (N)	67	38
Previous accident (%)	21.8%	0%

Measures

The protocol for the study conducted was preregistered and is available online at the Open Science Framework (doi10.17605/OSF.IO/HWMS5), no major change were made to this initial protocol. The questionnaire used can be found in **Supplementary Material**.

Perception of Ability

All participants were asked to compare their (or their child's) road crossing behavior and ability to their peers, the format of these questions, i.e., comparing their behavior to other children their age comes from previous measures (Purcell, 2012; Purcell and Romijn, 2017). This section included five questions, the first two asked whether they felt they paid more or less attention or exhibited more or less risk compared to their peers, these were both measured on a five point Likert scale. The third question asked the participant to rate their confidence compared to their peers on a four point Likert scale. Finally participants were asked to state whether they felt their DCD, or their child's DCD, changed the way they crossed the road compared to their peers, based on a yes/no answer. If they answered yes they were asked to elaborate.

Road Crossing Behaviors

All participants were asked about the regularity with which they, or their child, exhibited certain behaviors, these questions were taken from previous measures (Chinn et al., 2004). Participants had to state regularity on a four point Likert scale (never, sometimes, often, always). The behaviors were: forgetting to look before crossing; running across without looking; seeing a small gap and going for it; crossing before the green man appears; crossing between cars; thinking there is enough time to cross but discovering there is not; looking both ways before crossing; keeping looking the whole way across; making traffic slow down so you can cross; getting half way across and having to run; crossing where there is no view and waiting a long time before crossing.

General Likelihood of Accidents

This section was only completed by adults with DCD (i.e., not the parents). These participants were shown a series of pictures taken from Sissons Joshi et al. (2017) and asked whether they felt the accident shown in the picture was more, the same amount or less likely to happen to them compared to their peers, if they provided an answer of more or less they were asked to provide a justification for their answer. The pictures depicted: an accident while cycling; an accident in the bath; an accident when pouring from a kettle; an accident on a trampoline; an accident while swimming; an accident in a thunderstorm; an accident with a dog and an accident when crossing the road.

Demography

Parents and adults were also asked a series of demographic questions regarding themselves or their child, including the regularity with which they crossed roads and whether this was accompanied or unaccompanied, whether they had been hit by a vehicle or bicycle in the past, the types of roads they crossed most often, i.e., one-way, two-way, etc., speed limit. We also asked about chronological age and gender and any confirmed diagnoses of DCD and other neurodevelopmental disorders.

Procedure

One questionnaire was generated using the online platform Qualtrics aimed at individuals over 16 years of age and initially asked participants to indicate whether they were answering for themselves (adult version) or their child (parent version). If they were answering for themselves (adult version) all questions were then addressed in that manner, if they were answering for their child (parent version) questions were addressed in that manner.

Statistical Analysis

Data are consistently reported from the two types of participants (parent, adult) within each section of the questionnaire. We also included co-occurrence as an additional factor, and as such split each group into two sub-groups, those with no co-occurrences or only co-occurrences for which there is no evidence of impact on road crossing (DCD) and those with co-occurrences of ADHD, ADD, ASD or LD (neurodevelopmental disorders which have previously been linked to difficulties with road crossing; DCD + ADHD), the values of N for these groups are provided in **Table 1**. The reported perception of confidence, attention and risk was analyzed with a two-way ANOVA (group × co-occurrence), *post hoc* tests with Bonferroni correction were used where appropriate. Prior to the ANOVA, assumption tests were conducted using Levene's test used to determine whether the data violated the assumption of homogeneity of variance and Q–Q plots were used to determine the nature of the distribution of the data. ANOVA was only conducted where data were found to meet these assumptions. A power analysis was undertaken to determine sufficient power to conduct a two-way ANOVA of this type, and assuming a medium effect size of 0.25 and a power of 0.85 a total sample size of 146 participants would be needed. Given that the sample exceeds this two-way ANOVA was undertaken. An exploratory factor analysis (EFA) was used to reduce data from the scales of reported behavior with parallel analysis used to determine the number of factors to extract. Bartlett's test of sphericity was conducted along with KMO tests for sampling adequacy and these are reported in text. Factor scores were created by taking average scores for questions within each factor and then these were subject to regression analyses. Prior to the regression analyses being conducted appropriate assumption tests were undertaken, i.e., Q–Q plots and residual plots were used to determine whether the residuals were normally distributed, Cook's distance was used to determine the influence of observations and variance inflation factor (VIF) values were compared to the square root of VIF to determine collinearity within the data. Our participant sample provided adequate power for this regression analysis with (Wilson Van Voorhis and Morgan, 2007) stating a number of different cut off points all of which were met. Unrealistic optimism was analyzed using Chi-squared to determine the frequency of more, same and less responses for each accident type. Prior to Chi-squared, assumption checks were carried out by checking a sufficient expected frequency count. Friedman analysis was also used to

determine the number of more, same and less responses with Durbin-Conover adjustments for *post hoc* tests. In all cases an alpha level for significance was set as 0.05.

Content Analysis

Content analysis was used to code the open responses to two parts of the question: (1) when participants described how their DCD/Dyspraxia altered their crossing behavior and (2) when stating why they felt an accident was more/less likely. In each case responses were coded by both KW and CP using published seven steps (Treadwell, 2013). An initial set of categories were developed by KW with responses then re-coded by CP. For the question asking how DCD/Dyspraxia altered crossing behavior there were 62 responses from adults and 63 from parents. Agreement between the coders was high, with coders assigning responses to the same category in 80.2% of cases, disagreements were resolved through discussions between the coders. The coding framework identified five categories: heightened awareness (including more cautious and more anxious); lowered awareness (including more risky, impulsive, distracted); motor difficulties; perceptual difficulties (such as judging speed) and not knowing. For the question asking about why an accident was deemed more or less likely there was again high agreement between coders at the initial stage (95.3% agreement). A total of eight categories were identified when a "more" response was provided: coordination difficulties; understanding cause and effect; spatial awareness difficulties; not understanding risk; impulsivity/lack of attention; lack of experience; lack of confidence and has happened to me. A total of five categories were identified when a "less" response was provided: cautious; good knowledge; no exposure; good skill and like risks.

RESULTS

Reported Behaviors

We asked participants to rate how often they displayed certain behaviors. Assumption checks revealed that KMO values were all above 0.63 with an overall level of 0.78. Bartlett's test of sphericity was $p < 0.001$ and as such this assumption was valid. An orthogonal, varimax, rotation was performed as the resulting factors were not correlated. Three factors were extracted using parallel analysis. The adopted solution explained 57% of variance, with the first factor explaining 24.9%, the second 16.8%, and the third 15.3%. All component loadings were above 0.3 and so all questions were included in the resulting solution, loadings can be found in **Table 2**.

The first factor includes questions which focused on looking behavior and visibility, whether an individual looks before crossing and whether they continue to look. A high scoring participant on this factor would be reporting that they often run across the road without looking, do not often keep looking while crossing, cross without good visibility of oncoming traffic, etc. Factor 2 is a measure of timing ability, thinking there is enough time when there isn't, making traffic slow down when crossing, having to run to get across in time. A high score on this factor would indicate that a participant often shows these timing misjudgments. Factor 3 describes impatience at the roadside, a high score on this factor would indicate a participant who crosses before the green man or waits at the roadside for very little time. Hence across all factors, high scores indicate dangerous behaviors.

TABLE 2 | EFA loadings for the three extracted factors.

	1	2	3
Looking both ways	−0.851		
Keeping looking all the way across	−0.794		
Forget to look	0.773		
Run across without looking	0.718		
Cross between cars	0.405		
Cross with no view	0.369		
Think there is enough time but there is not		0.745	
Start crossing and then have to run		0.672	
Make traffic slow so you can cross		0.346	
Cross before the green man			0.809
See a small gap and go for it			0.672
Wait a long time			−0.610

1 Looking behavior and visibility. 2 Timing ability. 3 Impatience at the road-side.

In order to determine what variables influence the behaviors described by those factors a regression analysis was conducted on each factor score using co-occurrence group membership and chronological age as potential predictor variables. Prior to regression analysis assumption tests were conducted. For all three factors residuals were normally distributed as determined via Q–Q plots and residual plots, Cook's distance indicated that there was no undue influence from a small sample of the data, with the maximum value always falling well below 1 (factor 1 = 0.25, factor 2 = 0.10, factor 3 = 0.50) and VIF values were very similar to the square root of VIF indicating no collinearity issues in the data (VIF value = 1).

A significant model was found for looking behavior and visibility [$F(2,151) = 18.5$, $p < 0.001$, $R^2 = 0.20$] and impatience at the roadside [$F(2,151) = 4.28$, $p = 0.016$, $R^2 = 0.05$], but not timing ability. Coefficients and p values for all predictors for each regression analysis can be found in **Table 3**. For looking behavior both chronological age and co-occurrence

TABLE 3 | Beta values, standard errors, *t* values and *p* values for the two significant regression models.

		Beta	SE	*t*	*p*
Factor 1: Looking behavior and visibility					
Co-occurrence group	DCD + ADHD vs. DCD	0.315	0.108	2.92	0.004*
Age		−0.017	0.003	−4.92	<0.001*
Factor 3: Impatience at the road-side					
Co-occurrence group	DCD + ADHD vs. DCD	0.025	0.125	0.201	0.841
Age		0.012	0.004	2.92	0.004*

*Significant effects are indicated by an *.*

were significant predictors. Where chronological age was higher we saw a reduction in dangerous looking behaviors (crossing without looking etc.) and visibility (crossing between cars). Furthermore, individuals with DCD + ADHD had higher scores on this factor and hence demonstrate more dangerous looking behaviors compared to DCD only. For impatience at the roadside, only chronological age was significant with an increase in age being related to an increase in impatience at the roadside and so an increase in risky behaviors, such as not waiting for the green man etc.

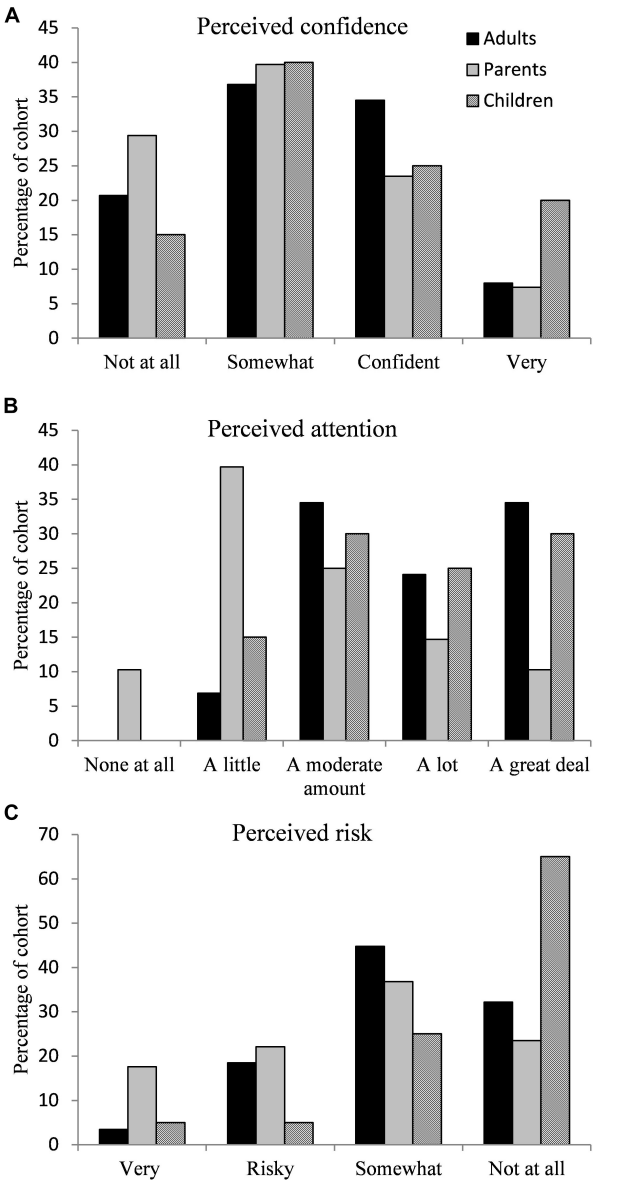

FIGURE 1 | An illustration of the percentage of responses to the questions asking about **(A)** confidence in road crossing skill, **(B)** attention paid when crossing the road, and **(C)** risk taking behavior.

Perceptions

Three questions focused on the perception of attention, risk and confidence when crossing the road. The percentage with which confidence, attention and risk was reported can be found in **Figure 1**. Only 42.5% of adults with DCD and 30.9% of parents of children with DCD rated themselves or their children (parents) as confident or very confident. Furthermore, 6.9% of adults with DCD stated they paid no or little attention while crossing the road while this was much higher for the parents of children with DCD (50%). Finally, 11% of adults with DCD stated their behavior at the roadside was very risky or risky while this figure was 19.8% of parents. Two-way ANOVAs (group × presence of co-occurrence) were carried out for confidence, attention and risk separately. All of the scales met the assumption of homogeneity of variance with Levene's test being non-significant (confidence $p = 0.88$, attention $p = 0.50$, and risky behavior $p = 0.16$). In addition, all of the scales met the assumption of normal distribution which was determined via Q–Q plots. No significant group or co-occurrence differences were found for confidence. For attention and risky behavior a significant effect of group was found [$F(1,151) = 33.29$, $p < 0.001$, $\eta_p^2 = 0.18$ and $F(1,151) = 6.87$, $p = 0.010$, $\eta_p^2 = 0.04$]. For both scales, this difference was due to parents stating less attention was paid and riskier behavior was apparent compared to the adults. In addition, a main effect of co-occurrence was found for attention [$F(1,151) = 6.96$, $p = 0.044$, $\eta_p^2 = 0.04$] and risky behavior [$F(1,151) = 5.23$, $p = 0.024$, $\eta_p^2 = 0.03$]. In both cases this was due to participants with DCD + ADHD showing significantly less attention paid and greater risky behavior compared to those with DCD. No significant interactions were observed for any of the three scales ($F < 1$). Data can be found in **Table 4**.

When asked whether they believed their motor difficulties meant that they crossed the road differently to their peers, 79.1% of adults with DCD (80% of the DCD and 75% of the DCD + ADHD group) answered yes, while 92.6% of the parents of children with DCD (92% of the DCD and 93% of the DCD + ADHD group) indicated that their child's motor difficulties meant they crossed the road differently to their peers. In each case the number of individuals with co-occurrences who answered yes to this question is equivalent to the proportion of the overall cohorts with co-occurrences, therefore, there is an equal representation and no further analysis of those with/without co-occurrences was made as this questions specifically asked about DCD. For those who answered *yes* to

TABLE 4 | Average responses for each group split across the co-occurrence groups.

		Confidence 1–4 scale	Attention 1–5 scale	Risk 1–4 scale
Adult	DCD	2.34 (0.11)	3.94 (0.13)	3.07 (0.11)
	DCD + ADHD	2.15 (0.20)	3.60 (0.23)	3.00 (0.20)
Parent	DCD	2.08 (0.15)	3.03 (0.17)	2.95 (0.15)
	DCD + ADHD	2.10 (0.16)	2.40 (0.19)	2.30 (0.16)

Standard error is given in brackets.

this question they were then asked how their motor difficulties changed the road crossing task for them. The categories from the content analysis and the frequency with which participants gave them as reasons can be found in **Table 5**. All groups reported perceptual difficulties as a key difference in their/their child's road crossing skill compared to peers. Heightened awareness was cited by both adult groups (DCD and DCD + ADHD) and parents of children in the DCD group. In addition, lowered awareness was commonly cited in the parent groups and in the DCD + ADHD adult group. Lowered awareness was the most commonly cited reason given by parents of the DCD + ADHD group. Some indicative quotes are provided in **Table 6**.

Unrealistic Optimism

The adults with DCD were asked to rate whether they felt an accident, depicted by an illustration was more likely, less likely or had the same likelihood to happen to them. The percentage of responses can be found in **Figure 2**. Chi-squared analysis revealed a difference in responses for all but accidents in the bath and drowning accidents. With accidents on the bike, on the road, with a kettle and on the trampoline being perceived as more likely (Bike $\chi^2 = 71.66$, $p < 0.001$, road $\chi^2 = 10.21$, $p = 0.006$, kettle $\chi^2 = 59.86$, $p < 0.001$, trampoline $\chi^2 = 50.14$, $p < 0.001$), accidents with a dog or with lightning (dog $\chi^2 = 28.76$, $p < 0.001$, lightning $\chi^2 = 41.66$, $p < 0.001$) being perceived as the same likelihood and bath and drowning accidents not showing a significant difference (bath $\chi^2 = 0.21$, $p = 0.90$, drowning $\chi^2 = 0.89$, $p = 0.639$). The assumptions of Chi-squared were met with expected frequencies greater than 5 in all cases and cases independent of each other.

In order to consider the frequency with which participants stated "more," "same" or "less" the count of each of these responses was taken across the eight accident types. Giving, for each participant, a score out of 8 for each response type, as this produced ordinal data a non-parametric test was used. Friedman analysis (response type) demonstrated a significant effect across these three response types [$\chi^2(2) = 29.2$, $p < 0.001$] with the "more" response given significantly more often than the "same" response and the "same" response given significantly more often than the "less" response. Finally, the percentage of "more" responses across the eight accident types were calculated and compared across co-occurrence group, no significant effect was found [$\chi^2(1) = 1.76$, $p = 0.19$].

The reasons provided for answers of an accident being more or less likely were subject to a content analysis with reasons given collated across "more" and "less" responses with different categories for the two responses. The frequency with which individuals provided these responses for each accident type are provided in **Supplementary Table 1**. Just considering the accidents which were perceived to be more likely (i.e., for bike, kettle, and trampoline) in adults with DCD, in all cases coordination difficulties were given as an explanation for an accident being more likely, in fact this was the most commonly cited answer. When the adults answered "less," the response they gave typically focused on having knowledge which would protect them from such an accident.

TABLE 5 | Frequency of responses to the content analysis categories regarding how motor difficulties changed the road crossing experience for adults with DCD and parents of children with DCD.

Category	Type of comment	Adults		Parents	
		DCD	DCD + ADHD	DCD	DCD + ADHD
Heightened awareness	More cautious	43.8%	33.3%	17.1%	10.7%
	More anxious				
Lowered awareness	More risky	14.6%	33.3%	28.6%	50.0%
	Not paying attention				
	Oblivious to rules				
Motor difficulties		6.3%		5.7%	10.7%
Perceptual difficulties	Judgment of speed/distance	35.4%	26.7%	45.7%	25.0%
I don't know/response uncodable			6.7%	2.9%	3.6%

Blank cells indicate no response fell within that category for that group.

TABLE 6 | Quotes regarding reasons as to why road crossing was perceived to be affected by DCD.

Group	Category	Quote
Adults	Judgment of speed/distance	My timing for crossing, and misjudgment of car distances is always way off
	Judgment of speed/distance	Can't judge distance/speed so juts have to guess a lot of the time
	More cautious	Being far, far more cautious
	More cautious	I wait longer, and only cross if I know that I'm totally safe
Parents	Judgment of speed/distance	He has trouble judging how far away the vehicle is and how long he might have to cross the road
	Judgment of speed/distance	No road sense, unable to judge distance and speed of traffic
	Not paying attention	My son is 9 and I have no little faith in his ability to safely cross roads unassisted so I walk him to and from school every day. In places where I do allow him to cross without guidance I have seen him cross without looking, stumble into the road, be unaware he is on a road, cross between park cars and walk out into traffic.

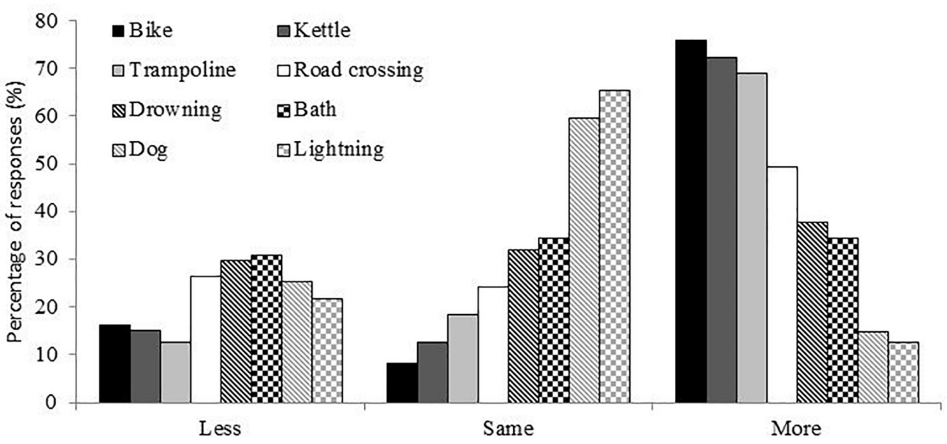

FIGURE 2 | Frequency of responses for the question asking about the likelihood, compared to peers, of non-road crossing accidents.

DISCUSSION

This study considered the lived experience of adults with DCD and parents of children with DCD as pedestrians at the roadside. We utilized a questionnaire which allowed us to survey exhibited behavior, perceived ability and unrealistic optimism in a large sample of these groups. We also specifically focused on the role of co-occurrences in road crossing behavior. In terms of our findings in relation to road crossing, behaviors were grouped into three factors: looking behavior and visibility, timing ability and impatience. Looking behavior and impatience varied with age, both showing a decrease in risk as age increased. Furthermore, participants in the DCD + ADHD group showed a greater level of risk in the looking behavior and visibility factor.

Previous evidence has shown us that children with DCD (Purcell et al., 2017) and children with ADHD (Clancy et al., 2006; Stavrinos et al., 2011) seemingly choose temporal crossing gaps which are too short to ensure a safe crossing or that children with DCD wait so long for a unnecessarily big crossing gap (Purcell et al., 2011). We also see children with ADHD being less concerned about risk (Farmer and Peterson, 1995) and children with LD being less able to judge the safety of crossing places (Anastasia, 2010). Finally, previous research has suggested that children with ASD may misunderstand rules of signalized crossings (Josman et al., 2008) and adults with ASD may show different eye gaze patterns when crossing (Earl et al., 2016; Cowan et al., 2018). These behaviors described in these previous findings show clear similarities to the factors from our analysis with *looking behavior and visibility* linking to eye gaze behavior (potentially atypical in ASD), identifying safe crossing places (potentially atypical in LD), and concern about risk (potentially atypical in ADHD). *Timing ability* links to choosing appropriately sized crossing gaps (potentially atypical in DCD and ADHD) and *impatience* links with waiting for a long time to cross (potentially atypical in DCD), understanding the rules of signalized crossings (potentially atypical in ASD) and concern about risk (potentially atypical in ADHD). In this way we can map previous findings in groups of children and adults with single neurodevelopmental disorders to those in the current study where we've considered children and adults with DCD and co-occurrences and compared these groups to each other rather than a typical group. What our findings demonstrated is that, across these factors only *looking behavior and visibility* differs between our co-occurrence groups, with those participants with DCD alongside ADHD, LD or ASD showing riskier behaviors. This supports previous research which has identified looking behavior and visibility as atypical among children and adults with ADHD, ASD and LD and from our data it would seem that these co-occurrences result in riskier behavior in individuals with DCD compared to those without those co-occurrences. In contrast, for timing ability and impatience these co-occurrences (ADHD, LD, ASD) do not impact on these areas over and above DCD. Therefore, previous laboratory based findings appear to be supported by the lived experiences, suggesting a level of awareness amongst these groups which might mitigate their risk at the roadside.

The elevated "risk" and lowered "attention" which is encapsulated within looking behavior and visibility in the DCD + ADHD group is also reflected where respondents were asked to report their perceptions of their behavior at the roadside, demonstrating that as well as reporting these behaviors these individuals (or their parents) are aware of the risky nature of some of the road crossing decisions which are made. Specifically, parents of children with DCD reported that their children paid less attention at the roadside and exhibited risky behavior. Furthermore, parents of children in the DCD + ADHD group (with co-occurrence of ADHD, ASD and/or LD) were reported to demonstrate the most risk and least attention at the roadside. It is worth noting that in Purcell and Romijn (2017) the primary school aged participants with DCD self-reported that they very much paid attention when crossing the road and didn't often take risks. The differences between these two findings could be because participants in the Purcell and Romijn (2017) study

underwent a selection process with some participants excluded due to IQ or co-occurrence, whereas the parents of those children would have remained in the current study. The differences could also be due to children with DCD not recognizing their inattention and risky behavior at the roadside, whereas their parents do. As well as asking about risk and attention, we also asked about confidence. Less than half of the adults with DCD and parents of children with DCD rated themselves or their children (parents) as confident or very confident. Again, this is in contrast to Purcell and Romijn (2017) where none of the primary school aged participants in either the DCD or TD group rated themselves as "not at all confident" and it would seem therefore, that the population in the current study are less confident than those included previously, again this could be an effect of asking parents rather than children or an effect of the more diverse nature of the sample in the current study. This type of self-reporting of perception or asking parents to report for children has not been done in populations with ADHD, ASD or LD and so comparisons with these groups cannot be made. This study grouped those without a co-occurrence and those with co-occurrences not known to cause issues at the road-side (Dyslexia etc.), however, it is not that we know that neuro-developmental disorders such as Dyslexia do not cause difficulties at the road-side, more that there is no evidence that they do. Future research is needed to pick this apart.

A key novel element of the current study was that we asked participants whether they felt their motor difficulties (adults) or their child's motor difficulties (parents) changed the way they or their child crossed the road. Both adults with DCD (79.1%) and parents of children with DCD (92.6%) overwhelmingly reported that their motor difficulties impacted upon their road crossing, and this was regardless of co-occurrence. Although we need to be slightly cautious, due to the self-selecting nature of the current study, this does suggest that this is an area which needs careful consideration in further research and any specific remediation. When asked why, participants provided a range of reasons, only some of which were specifically focused on the motor aspect of road crossing. All groups cited perceptual difficulties as a barrier to road crossing which sits well with the literature which has highlighted this as a potential source of error when crossing the road in children with DCD (Purcell et al., 2011, 2012, 2017) and in children with ADHD (Clancy et al., 2006; Stavrinos et al., 2011). The current study extends these findings into adulthood and also highlights that parents and adults are fully aware that this is an issue. Both adult groups and the parent DCD group also cited heightened caution, which is a factor seen in some simulated road crossing studies where primary school aged children with DCD were willing to wait up to 11 s for a "safe" crossing gap when presented with a simulated single vehicle on a straight stretch of road (Purcell et al., 2011). An interesting question is *why* an adult with DCD might show caution, the current data doesn't give us an insight into why caution is shown but it may be that they have experience of making poor road crossing decisions or struggling to safely cross the road and so have learnt to be cautious, this may explain why caution was not as commonly cited by the parents. Caution or anxiety has not previously been considered in other neurodevelopmental disorders, but the evidence from our study does not suggest that ADHD, ASD or LD consider these as a self-reported reason for difficulties with road crossing. The final factor which was commonly cited was lowered awareness (being more risky, more impulsive, more distracted), all but the adults in the DCD group cited this and it was the most common reason cited by parents in the DCD + ADHD group. It is unsurprising that where there is a majority of individuals with ADHD (regardless of other co-occurrences) impulsivity or lack of attention is cited as issues given that these characteristics are the hallmarks of ADHD. The citing of lowered awareness as a factor by parents of children without ADHD (in the DCD group) may simply be a consequence of childhood that has also been cited in studies of typically developing children at the roadside (Dunbar et al., 2001).

The final aim of this paper was to explore the issue of unrealistic optimism in adults with DCD. Anecdotal evidence would suggest that these adults are far more at risk of having very minor accidents, walking into objects, dropping things, tripping over, etc. However, how they perceive the likelihood of these accidents is unclear, i.e., do they show unrealistic optimism, i.e., the "it won't happen to me mentality," that we see in typically developing adults. For adults with DCD we see no evidence of unrealistic optimism, in contrast adults with DCD tended to report that they are more likely to experience an accident of any type compared to their peers, and this is regardless of their co-occurrence status. If fact adults with DCD stated they were more likely to have accidents which had a clear motor component, i.e., falling off a bike, a road traffic accident, falling from a trampoline or spilling water from a kettle and for these accident types they commonly cited "coordination difficulties" as the reason they were perceived as more likely. The literature focusing on unrealistic optimism in adults does explore mediating factors (Helweg-Larsen and Shepperd, 2001) with prior experience of an accident increasing risk estimates resulting in less optimistic bias (Helweg-Larsen and Shepperd, 2001). The findings of the current study point toward experience with accidents in the past mediating "unrealistic optimism" although it is difficult to draw firm conclusions about this as we didn't collect data on occurrence of accidents of specific types, nor did we collect data from children with DCD who would, have less experience and so may still show unrealistic optimism. Interestingly the lack of differences across our co-occurrence groups suggests that disorders such as ADHD do not increase unrealistic optimism even though it has been linked to a lack of concern about risk (Farmer and Peterson, 1995). Adults with DCD are seemingly very aware of the likelihood of accidents and in some cases may over-estimate these and so may be mitigating this elevated risk with compensations, i.e., waiting a long time at the roadside, extra caution, etc.

As mentioned above one limitation to this study is that the population who completed this questionnaire were a self-selected population, i.e., those individuals concerned about crossing the road or concerned about their children crossing the road might have selected to complete the questionnaire while other individuals who were not concerned about this aspect choose not to. Although this might influence the data on the number of

participants involved in accidents and the number of participants feeling that their road crossing was affected by their DCD it wouldn't change the pattern of the responses in terms of regularity of behaviors at the roadside, perception of behavior or unrealistic optimism. A secondary limitation is that we collected self-reporting of behavior and so we cannot asses the accuracy of this reporting, however, this method of self-report has been used previously in adults and children and furthermore, determining true naturalistic behavior at the road-side in individuals with neuro-developmental disorders would be vastly time consuming.

This questionnaire study has demonstrated that road crossing skill is something that adults with DCD and parents of children with DCD consider to be affected by their motor difficulties. They report that decision making behaviors are more dangerous and this may be linked to perceptual and motor difficulties. Individuals with co-occurrences which have previously been linked to unsafe crossing behaviors (i.e., ADHD, ASD, and LD) also report a greater regularity of dangerous looking behavior (forgetting to look, running without looking) and visibility (crossing between cars, crossing when you can't see), these adults and the parents of these children are seemingly aware of the risky nature of these behaviors. Unrealistic optimism was not an explanation for the risky behavior in adults with DCD and in fact, these adults demonstrated a clear understanding of the likelihood of accidents. Road crossing is clearly perceived as a different experience for adults with DCD and for parents of children with DCD and so should be recognized as an area in which remediation is needed for this population, with an understanding that those with specific co-occurrences show different behaviors.

DATA AVAILABILITY STATEMENT

Raw data supporting the conclusions of this article is available to view at https://doi.org/10.24384/nvwr-jc60.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the University Research Ethics Committee, Oxford Brookes University. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

KW designed the study and questionnaire, collected the data, conducted the analyses, and wrote the first draft of the manuscript. CP co-designed the study and questionnaire, contributed to the content analysis coding and to the writing and revising of the manuscript. Both authors contributed to the article and approved the submitted version.

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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.2020.587042/full#supplementary-material

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Children With Developmental Coordination Disorder Show Altered Visuomotor Control During Stair Negotiation Associated With Heightened State Anxiety

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Safe stair negotiation is an everyday task that children with developmental coordination disorder (DCD) are commonly thought to struggle with. Yet, there is currently a paucity of research supporting these claims. We investigated the visuomotor control strategies underpinning stair negotiation in children with (N = 18, age = 10.50 ± 2.04 years) and without (N = 16, age = 10.94 ± 2.08 years) DCD by measuring kinematics, gaze behavior and state anxiety as they ascended and descended a staircase. A questionnaire was administered to determine parents' confidence in their child's ability to safely navigate stairs and their child's fall history (within the last year). Kinematics were measured using three-dimensional motion capture (Vicon), whilst gaze was measured using mobile eye-tracking equipment (Pupil labs). The parents of DCD children reported significantly lower confidence in their child's ability to maintain balance on the stairs and significantly more stair-related falls in the previous year compared to the parents of typically developing (TD) children. During both stair ascent and stair descent, the children with DCD took longer to ascend/descend the staircase and displayed greater handrail use, reflecting a more cautious stair negotiation strategy. No differences were observed between groups in their margin of stability, but the DCD children exhibited significantly greater variability in their foot-clearances over the step edge, which may increase the risk of a fall. For stair descent only, the DCD children reported significantly higher levels of state anxiety than the TD children and looked significantly further along the staircase during the initial entry phase, suggesting an anxiety-related response that may bias gaze toward the planning of future stepping actions over the accurate execution of an ongoing step. Taken together, our findings provide the first quantifiable evidence that (a) safe stair negotiation is a significant challenge for children with DCD, and that (b) this challenge is reflected by marked differences in their visuomotor control strategies and state anxiety levels. Whilst it is currently unclear whether these differences are contributing to the frequency of stair-related falls in children with DCD, our findings pave the way for future research to answer these important questions.

Keywords: gaze, vision, kinematics, anxiety, fall-risk, fear of falling

INTRODUCTION

Developmental coordination disorder (DCD) is a neurodevelopmental disorder that is estimated to affect between 1.7 and 6% of children worldwide (American Psychiatric Association, 2013). DCD is characterized by motor skill learning and performance that is far below the expected level for an individual's age and cannot be better explained by intellectual delay, visual impairment, or other neurological disorders that affect movement (Blank et al., 2019). Motor skill difficulties in DCD significantly interfere with the ability to perform activities of daily living (ADL) requiring fine and/or gross motor coordination, such as handwriting or even walking. Indeed, the walking pattern of children with DCD is often described as awkward (Gillberg and Kadesjö, 2003) and is more variable than their typically developing (TD) peers (Rosengren et al., 2009). Children with DCD also adopt a safer walking strategy during treadmill walking (Deconinck et al., 2006) and display a reduced ability to control their momentum when crossing obstacles (Deconinck et al., 2010), both of which have been attributed to a deficit to dynamic balance control. Given that children with DCD are also less accurate when tasked with precise stepping actions (Parr et al., 2020), it is unsurprising that they trip and bump into things more frequently than their TD peers (Kirby et al., 2011; Cleaton et al., 2020).

Although the difficulties children with DCD have with walking are well-documented, there is currently a paucity of research exploring how difficulties with walking translate to difficulties walking up and down stairs. This is surprising, as stair negotiation is a fundamental task that children must overcome both in and outside the home, posing a serious threat to injury in the event of a fall. Difficulties climbing stairs are commonly cited as a physical characteristic of DCD (Henderson, 1992; Missiuna et al., 2011; NHS UK, 2017) and parents of children with DCD have reported their concerns when watching their child climb stairs (Kaufman and Schilling, 2007; Missiuna et al., 2007). It has even been suggested that teachers allow children with DCD to leave class early to avoid the hazardous task of going up and down stairs when busy and cluttered with other students (Ripley, 2001). Yet, research thus far has been limited to subjective teacher reports suggesting children with DCD are less functional going up and down stairs (Wang et al., 2009) and evidence that they cannot climb as many steps as TD children in 30 s (Ferguson et al., 2014). It therefore appears that difficulties children with DCD show with balance and stability may contribute to an inherent difficulty with negotiating stairs. Consequently, there is a need to elucidate the control strategies of stair walking in children with DCD and to determine the mechanisms that may contribute to a possible increased risk of tripping and falling.

Falls most often result from a trip on a stair edge or tread surface, which likely explains why smaller step-edge clearances (i.e., distance from foot to step edge), greater clearance variability, and greater misjudgements in foot placement are linked with an increased risk of falls (Simoneau et al., 1991; Hamel et al., 2005). Stair falls are also three times more likely to occur during stair descent than stair ascent (Startzell et al., 2000), possibly reflecting the greater challenge to postural dynamic stability (Mian et al., 2007). Indeed, to recover from a loss of balance when going down the stairs, individuals must rapidly reposition their limbs whilst controlling for downwards momentum (McFadyen and Winter, 1988; Novak and Brouwer, 2011). Difficulties negotiating stairs in children with DCD may, therefore, be associated with difficulties making accurate and consistent stepping actions (Rosengren et al., 2009; Parr et al., 2020) and deficits to dynamic stability (Deconinck et al., 2006, 2010).

Accurate foot placement is also generally dependent on the appropriate use of gaze to visually extract relevant environmental features at appropriate times to optimize the planning and control of movement. For example, both older and younger adults have been shown to use gaze in a feedforward manner when navigating stairs, looking approximately three steps ahead to control their stepping behavior approximately one or two strides in advance (Zietz and Hollands, 2009; Miyasike-daSilva et al., 2011). The retrieval of feedforward visual information is particularly important at the start of a stair ascent/descent, with evidence that falls are more likely to occur when a person fails to fixate these initial, transitioning steps (Archea et al., 1979). Difficulties children with DCD display navigating stairs may, therefore, be attributable to impairments in visuomotor control and the processing of task-relevant, visual information. In tasks such as throwing and catching (Wilson et al., 2013) and sequential stepping (Warlop et al., 2020) individuals with DCD tend not to use feedforward (predictive) control to guide the planning of subsequent movements (Ferguson et al., 2015). Instead, children with DCD show a dependence on visually guided online control (Debrabant et al., 2013) despite a reduced ability to use online information to rapidly adjust and correct ongoing action (Hyde and Wilson, 2011, 2013). Consequently, children with DCD may formulate inaccurate stepping actions due to the suboptimal use of gaze to extract relevant and timely information from the environment, and the inability to predict the consequences of the ensuing movement (Wilson et al., 2013). Though our recent findings suggest that stepping inaccuracies in children with DCD may also occur despite typical looking behavior (Parr et al., 2020), it is important to determine whether the visuomotor control strategies adopted by children with DCD may be contributing to difficulties with stair negotiation.

Another factor that may contribute to difficulties negotiating stairs in children with DCD is anxiety and the fear of falling. It is well-established that fear of falling can have a concomitant impact on the visuomotor processes described above. For example, when faced with a series of obstacles, fall-related anxiety causes individuals to prioritize gaze fixations to the most proximal stepping constraints at the expense of sufficiently previewing the entire walking environment prior to negotiating it (Young et al., 2012; Ellmers and Young, 2019). Consequently, anxious individuals sometimes look away from a stepping target prematurely which results in decreased stepping accuracy and an increased risk of falling. Fall-related anxiety is also proposed to decrease attentional processing efficiency, as cognitive resources are drawn toward consciously controlling ongoing movement as opposed to carrying out concurrent processes necessary for safe locomotion, such as feedforward movement planning (Gage et al., 2003; Young and Mark Williams, 2015). To compensate for

these maladaptive effects of anxiety, older adults have been shown to walk slower and increase their dynamic stability, allowing more time to plan appropriate foot placement and a heightened ability to counter forward momentum during a misstep (Novak et al., 2016; Thomas et al., under review). Evidently, anxiety is a critical factor that must be considered to fully understand the mechanisms underpinning the visuomotor control strategies of children with DCD during stair negotiation. Though we have recently shown that children with DCD do not experience heightened anxiety during over ground targeted stepping tasks (Parr et al., 2020), stair negotiation is likely to place greater demands on dynamic stability and increase the risk of significant injury in the event of a fall and may, therefore, be more likely to instill a fear response.

The aim of this study was to provide the first examination of the visuomotor control strategies that underpin stair negotiation in children with DCD and to explore the underlying influence of state anxiety. We hypothesized that the children with DCD would report heightened state anxiety and adopt a more cautious stair negotiation strategy, reflected by greater handrail use, slower walk times and more proximal gaze fixations to guide ongoing stepping commands in order to maintain stability. We also hypothesized that children with DCD would display smaller and more variable step-edge clearances compared to TD children, given their association with stair-related falls and recent evidence of decreased stepping accuracy and precision in children with DCD (Parr et al., 2020).

MATERIALS AND METHODS

Participants

Forty-one participants aged between 8 and 15 years of age participated in the study, of which 23 were initially recruited for our DCD group. Children in the DCD group were recruited via social media and local DCD support groups and satisfied the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) criteria (American Psychiatric Association, 2013). For example, parents completed the Developmental Coordination Disorder Questionnaire (DCDQ; Wilson et al., 2009) prior to testing to confirm that movement difficulties significantly interfered with their child's activities of daily living. Parents also confirmed that their child had no diagnosis of learning difficulties and did not suffer from any medical conditions known to affect sensorimotor function (e.g., cerebral palsy, hemiplegia, or muscular dystrophy). Finally, for inclusion in the DCD group, children were required to score below the 15th percentile on the test component of the Movement Assessment Battery for Children-2 (MABC-2; Henderson et al., 2007) carried out as part of the testing phase. This resulted in the data of five participants being excluded from the study (min = 25th percentile). Participants in the TD group were recruited from the family members of student and staff members at Liverpool John Moores University and were required to score above the "indication of DCD" zone of the DCDQ and above the 15th percentile of the MABC-2, resulting in one participants' data being excluded from the study. This resulted in a net total of 19 participants in the DCD group (male = 13, female = 6, age = 10.50 ± 2.04 years) and 16 participants in the TD group (male = 10, female = 6, age = 10.94 ± 2.08 years). All participants were right-foot dominant (**Table 1**). Parents also completed the Attentional Deficit/Hyperactivity Disorder (ADHD) Rating Scale-VI due to its high comorbidity with DCD (∼50% co-occurrence; American Psychiatric Association, 2013). No child scored above the 98th percentile for inattention or hyperactivity which is recommended to be the minimum cut-off used as an indication of ADHD in research (DuPaul et al., 1998). Ethical approval was granted by the Liverpool John Moores University Ethics Committee and written informed consent was obtained from each child participant and their legal guardian prior to testing.

Staircase Apparatus

Participants ascended and descended a custom-made seven-step instrumented staircase with handrails on either side and a top and bottom landing long enough to enable an entry and exit phase. The riser height (20 cm) and going length (25 cm) of each step was within the current UK building regulations for commercial and private properties (Gov, 2013). Force platforms (FP; Kistler), sampling at 1,080 Hz (subsequently down sampled 120 Hz), were embedded within the bottom four steps. All participants wore a passive overhead harness whilst on the staircase that was operated by a trained belayer (**Figure 1**). For a detailed layout of our staircase, see Thomas et al. (2020).

Kinematics

A 26-camera motion capture system (Vicon MX, Oxford Metrics, UK) collected whole-body kinematic data at 120 Hz, with thirty-eight reflective markers placed on the feet, lower legs, thighs, pelvis, torso and head, according to the conventional Plug-in Gait marker set. Participants wore flat footwear and tight clothing. A triangular cluster of three reflective markers (14 mm diameter) was placed on each shoe to track virtual landmarks created by a digitizing wand (C-motion, Germantown, MD, USA) at the anterior-inferior (toe-tip) and posterior inferior (heel-tip) point of each shoe. Marker trajectories were labeled, gap-filled, and exported as c3d files (Vicon Nexus 2.6, Oxford Metrics). The position of the whole-body center of mass (CoM) was estimated as the weighted sum of the various body segments using Visual3D (C-Motion, Germantown, USA). For further analysis, all kinetic and kinematic data were filtered using a phase-corrected low-pass fourth order Butterworth

TABLE 1 | Demographic information of the DCD and TD children included in the study.

	DCD	TD
Male (*n*)	13	10
Female (*n*)	6	6
Age (years)	10.42 ± 2.01	10.94 ± 2.08
Height (cm)	149.32 ± 10.12	146.98 ± 13.09
Weight (kg)	45.26 ± 11.41	38.95 ± 13.48
MABC-2 (%)	1.86 ± 2.75	51.31 ± 30.88

FIGURE 1 | Image displaying a child participant (face blurred) preparing to descend our custom-built instrumented seven-step staircase. The child can be seen wearing a safety harness that is attached to an overhead belay safety system. Each child was allowed to freely use the handrails that can be seen either side of the staircase. The Vicon cameras can also be seen, positioned around the staircase to optimize kinematic data capture.

filter with a cut-off frequency of 6 Hz. Initial foot contacts on the staircase included contacts on the bottom landing, on each step, and on the top landing. Initial contacts on the landings were determined using local maxima of the heel referenced to the pelvis segment (Zeni et al., 2008). For the steps with no FP, local minima in the CoM vertical velocity trace defined initial contacts, and local maxima in the trailing knee flexion angle trace defined toe offs (Foster et al., 2014). For the steps with FPs, >20 N defined initial contact (Zeni et al., 2008).

Eye Tracker

Eye movements were recorded using a Pupil Labs binocular eye-tracking mobile headset (Kassner et al., 2014) that featured two pupil cameras that recorded pupil movements at 60 Hz, and a scene camera to record the world view at 30 Hz. Prior to task performance, participants completed a 5-point screen marker calibration that was re-run when the calibration had been visibly lost. If the child failed calibration after multiple attempts, or persistently lost calibration due to excessive movement of the headset, the task was run without the eye tracker and their gaze data excluded. Participants' gaze data were also only included in the analyses of each condition if they presented at least two usable trials. Consequently, eye-movement data from two TD participants (male = 1, female = 1, age = 9.50 ± 0.71, MABC-2 = 25.00 ± 00.00) were excluded for the descent condition, and eye-movement data from four DCD participants (male = 2, female = 2, age = 9.50 ± 1.29, MABC-2 = 3.37 ± 3.82) were excluded from both the ascent and descent conditions. From the participants included in gaze analyses, an average of 7 ± 12% of trials were excluded for the stair ascent, and an average of 12 ± 18% of trials were excluded for the stair descent. Eye tracking footage was also used to determine whether a particular trial did or did not involve some use of the handrails.

Parental Confidence Questionnaire

All parents completed a 9-question survey designed to examine (a) the confidence they have in their child's ability to safely ascend and descend stairs, and (b) how frequently their child experiences stair-related falls in everyday life. The first eight questions consisted of a Likert scale ranging from zero (not confident at all) to one-hundred (absolutely confident) and probed how confident each parent was that their child could (Q1/2) go up/down the stairs without losing balance (typical stair negotiation), (Q3/4) go up/down the stairs rapidly without losing balance (rapid stair negotiation), (Q5/6) go up/down the stairs without a handrail and not lose balance, and (Q7/8) recover from a loss of balance going up/down the stairs to prevent a fall. The final question (Q9) asked each parent to provide an estimation of the total number of stair-related falls (going up or down) their child had experienced in the year prior to testing.

State Anxiety Questionnaire

State anxiety levels were measured using a child friendly "fear thermometer" (www.anxietycanada.com) which encompasses a 10-point "smiley-face" Likert scale ranging from 1 (low levels of anxiety) to 10 (high levels of anxiety). These simple scales have previously been validated against larger and more complex state anxiety inventories (Houtman and Bakker, 1989).

Protocol

Data collection took place in a single session lasting ∼2-h. Once prepared for kinematic and gaze data collection, baseline levels of state anxiety were collected. After a demonstration by the experimenter, participants were then instructed to ascend and descend the stairs at a steady self-selected pace and to avoid running or jogging up/down the stairs. All analyzed trials were performed in a step-over-step fashion, placing only a single foot on each step (as per the demonstration). A total of three trials were removed from analysis due participants exhibiting some step-by-step walking, placing each foot on a

single step. Participants performed 5 trials in each direction (ascent and descent), starting with ascent. At the beginning of each trial, participants stood facing the staircase on either the lower (ascent) or upper (descent) landing with their feet side-by-side whilst maintaining their gaze on a red light-emitting-diode (LED) positioned to the left of each starting position (**Figure 1**). This ensured standardization of visuomotor planning across participants prior to each trial. The red LED turned off at the onset of kinematic data collection (initiated by Vicon) and acted as a "go" signal for participants to begin each trial, at which point they could look where they wanted. To ensure ecological validity, participants were free to use the handrails throughout. Once participants had ascended/descended the staircase, they were instructed to continue walking along the landing before coming to a stationary position. Immediately prior to the first ascent and first descent trial, state anxiety was again measured to determine task-specific fluctuations in anxiety.

Data Analysis

Kinematic Variables

Stair ascent and descent durations were calculated as the interval (in seconds) between the foot contacts occurring on the first and seventh step steps. The interval between foot contacts occurring on subsequent steps ("step duration") was also measured to determine how particular sections on the staircase may contribute to overall stair ascent/descent durations. The variability in both ascent/descent durations and step duration were calculated as the standard deviation of values across trials. To characterize whether children with DCD showed lower postural stability and reduced stepping control during walking compared with TD children we measured margin of stability, foot-step-edge clearances and foot-step-edge clearance variability. These measures were identified due to their association with fall-risk on stairs. Margin of stability was expressed as the distance between the extrapolated CoM (xCoM) and the forward boundary of the base of support (in this instance, the toe-marker). When the toe-marker was overhanging the confines of the step-edge, the forward boundary was instead defined as the step-edge. Smaller (or more negative) margins of stability are considered to reflect a less dynamically stable pattern of stair negotiation (Bosse et al., 2012; Novak et al., 2016). Margin of stability has been shown to increase in older adults under conditions of poor lighting (Thomas et al., 2020) and ambiguous carpet patterns (Thomas et al., under review) when descending stairs, and can therefore detect safety-related adaptations to stair navigation strategy. *xCoM* was defined as

$$xCoM = pCoM + vCoM/\sqrt{(gl^{-1})}$$

where *pCoM* is the AP position of the CoM, *vCoM* is the instantaneous AP velocity of the CoM, g is acceleration due to gravity, and l is the absolute distance between the CoM and the ankle joint center. Margin of stability was calculated at initial contact on each of the seven steps (Debelle et al., 2020). For ascent, foot-step-edge clearance was defined as the minimum vertical distance between toe markers on the lead limb and the step edges (toe clearance). For descent, foot-step-edge clearance was defined as the minimum horizontal distance between heel markers on the lead limb and the step edges (heel clearance). Foot-step-edge clearance variability was measured as the standard deviation of step-edge clearances across trials on each step. Increased clearance variability is proposed to increase the risk of catching the foot on a step edge and thus cause a fall/loss of balance.

Gaze Variables

Frame-by-frame analysis of eye-tracking video footage was performed to identify gaze location from trial onset (identified by the LED "go" signal) until trial offset (foot contact on the seventh step in sequence). Gaze fixations were defined as a gaze stabilization on a location in the environment for three frames or longer (corresponding to ∼100 ms). Fixations were classified as being spatially located on one of the following areas of interest: (1) *bottom of the staircase (Bottom)*: one tread-length before the stairs and step 1; (2) *lower mid-steps (M1)*: steps 2 and 3; (3) *upper mid-steps (M2)*: steps 4 and 5; (4) *top of the staircase (Top)*: steps 6 and 7; (5) *far landing*: incorporating fixations on the far landing and on the back wall (relative to either the stair ascent or descent), and (6) *handrails*. To understand how gaze was allocated across each individual phase of movement (i.e., "gaze in action"), we expressed total time fixating each AOI during each phase of movement as a percentage of total phase duration (Miyasike-daSilva et al., 2011). We also measured the average number of steps the participants looked ahead during each phase of movement. In line with previous research (Zietz and Hollands, 2009), we considered participants to be looking one step ahead when they were fixating the next step in sequence. For example, a person who has just made foot contact on step 3 would be looking one step ahead if fixating step 4 but looking two steps ahead if fixating step 5. Other gaze variables included average fixation duration and fixation rate (number of fixations divided by the total stair ascent/descent duration, expressed as fixations per second).

Statistical Analyses

All analyses were performed and are presented separately for the stair ascent and stair descent. Independent samples *t*-tests were run to compare stair ascent/descent duration, fixation duration and fixation rate between groups. Effect sizes were expressed using Cohen's *d*, with common indicative thresholds reported as small (0.2), medium (0.5), and large (0.8). Kinematic variables characterizing risky stair behavior (step duration; foot clearances; foot clearance variability; margin of stability) were analyzed using a two-way mixed design repeated measures ANOVA, with between-subject effects of Group (x2 DCD; TD) and within-subject effects of Phase (x4; Bottom, M1, M2, Top). A two-way repeated measures ANOVA was also used to examine how many steps participants looked ahead throughout each trial, with between-subject effects of Group and within-subject effects of Phase (x4; Bottom; M1; M2; Top). Significant effects were probed by polynomial trend analyses, and *post-hoc* analyses were performed using pairwise comparisons with Sidak-corrections to account for the multiple

TABLE 2 | Mean (SD) levels of confidence reported by the parents of children with and without DCD across each stair-specific walking scenario during both stair ascent and stair descent.

	Confidence (%)			
	Ascent		Descent	
Stair walking scenario	**DCD**	**TD**	**DCD**	**TD**
Typical stair negotiation (Q1/2)	76.5 (21.32)*	96.80 (4.69)	67.68 (28.97)*	96.00 (5.63)
Rapid stair negotiation (Q3/4)	62.56 (27.59)*	92.66 (7.31)	51.5 (28.83)*	91.73 (8.28)
Without handrails (Q5/6)	47.94 (30.83)*	95.13 (5.84)	40.38 (29.42)*	94.07 (6.85)
Recover from loss of balance (Q7/8)	53.44 (30.33)*	90.2 (11.54)	44.38 (27.53)*	88.2 (14.64)

Asterisks indicate significant between group differences ($p < 0.001$).

comparisons problem (Blakesley et al., 2009). ANOVA effect sizes were reported using partial eta squared (η_p^2), common indicative thresholds for which are small (0.01), medium (0.06) and large (0.14; (Field, 2013). The results of univariate tests are reported, with Huyn-Feldt correction procedure applied for analyses that violated sphericity of variance. Where data were not normally distributed, within participant effects were analyzed using Friedman's ANOVA, and Bonferroni corrected Wilcoxon-signed rank tests for *post-hoc* analyses. Between-subject effects were analyzed using Mann-Whitney *U*-tests. Non-parametric effect sizes were reported as $r = Z/\sqrt{n}$, for which common thresholds are small (0.1), medium (0.3) and large (0.5; Rosenthal, 1986). All statistical analyses were performed using IBM SPSS statistics (version 26) with an alpha level of ≤ 0.05.

RESULTS

Parent Confidence Questionnaire

Results from separate Mann-Whitney *U*-tests showed the parents of children with DCD reported significantly lower confidence in their child's ability to use the stairs compared to the parents of TD children for all included scenarios ($ps < 0.001$). Compared to the parents of TD children (*Median* = 0.0, *Mean (M)* = 2.1 ± 5.2), the parents of DCD children (*Median* = 3.5, *M* = 12.3 ± 31.9) also reported significantly more stair-related falls in the last year ($U = 69.50$, $p = 0.034$, $Z = -2.123$; **Table 2**).

Stair Ascent

There were no significant Group x Phase interactions for any ascent outcome measures.

Anxiety

Results showed no significant difference between groups for levels of state anxiety prior to the first stair ascent ($U = 128.00$, $z = -0.874$, $p = 0.382$, $r = -0.148$).

Handrail Use

Children with DCD displayed greater frequency of handrail use than the TD children ($U = 81.00$, $p = 0.003$, $r = 0.495$). Two TD children used the handrails during the stair ascent, both using the handrail for all 5 trials. In comparison, 14 DCD children used the handrails, 10 using the handrails for all 5 trials.

Stair Ascent Duration

Children with DCD ($M = 4.78$ s) took significantly longer than the TD children ($M = 4.32$ s) to ascend the staircase, $t(33) = 2.596$, $p = 0.014$, $d = 0.861$. However, ascent duration variability did not significantly differ between the DCD ($M = 0.46$ s) and TD ($M = 0.36$ s) groups, $U = 109.00$, $z = -1.424$, $p = 0.154$, $r = -0.248$ (**Figure 2**).

Step Duration

There was a significant main effect of Group, $F_{(1, 33)} = 4.892$, $p = 0.034$, $\eta_p^2 = 0.129$, with greater step durations observed in the DCD children ($M = 679$ ms) compared to the TD children ($M = 625$ ms). There was also a significant main effect of Phase, $F_{(1.559, 51.447)} = 5.879$, $p = 0.009$, $\eta_p^2 = 0.151$, with *post-hoc* analyses revealing significantly longer step durations at the Bottom of the staircase ($M = 689$ ms) compared to the M2 phase ($M = 626$ ms, $p = 0.007$). There was no Phase x Group interaction, $F_{(3, 99)} = 2.187$, $p = 0.094$, $\eta_p^2 = 0.062$. For step duration variability, there was no main effect of Group, $F_{(1, 33)} = 1.399$, $p = 0.245$, $\eta_p^2 = 0.041$, no main effect of Phase, $F_{(2.132, 70.343)} = 1.887$, $p = 0.157$, $\eta_p^2 = 0.054$, and no Phase x Group interaction, $F_{(3, 99)} = 0.802$, $p = 0.496$, $\eta_p^2 = 0.024$ (**Figure 2**).

Kinematics

Vertical Toe Clearance

There was no effect of Group, $F_{(1, 33)} = 0.001$, $p = 0.971$, $\eta_p^2 = 0.000$, but there was a significant main effect of Phase, $F_{(1.656, 54.646)} = 118.584$, $p < 0.001$, $\eta_p^2 = 0.782$. Comparisons revealed greater vertical toe clearance at the Bottom ($M = 7.6$ cm, $ps < 0.001$) and Top (M = 4.7 cm, $ps < 0.017$) of the staircase compared to M1 ($M = 4.1$ cm) and M2 ($M = 4.0$ cm; **Figure 2**).

Vertical Toe Clearance Variability

There was a significant main effect of Group, $F_{(1, 33)} = 6.601$, $p = 0.015$, $\eta_p^2 = 0.167$, with greater variability observed in the DCD group ($M = 1.3$ cm) compared to the TD group ($M = 1.0$ cm). There was also a significant main effect of Phase, $F_{(1.934, 63.828)} = 9.673$, $p < 0.001$, $\eta_p^2 = 0.227$, with *post-hoc* comparisons revealing greatest variability at the Bottom of the staircase ($M = 1.5$ cm) compared to all other phases ($ps < 0.015$; **Figure 2**).

MoS Anteroposterior

There was no main effect of Group, $F_{(1, 33)} = 1.092$, $p = 0.304$, $\eta_p^2 = 0.032$. There was a significant main effect of Phase, $F_{(1.382, 45.599)} = 4.970$, $p = 0.021$, $\eta_p^2 = 0.131$, but comparisons did not reveal any significant differences across phase pairs.

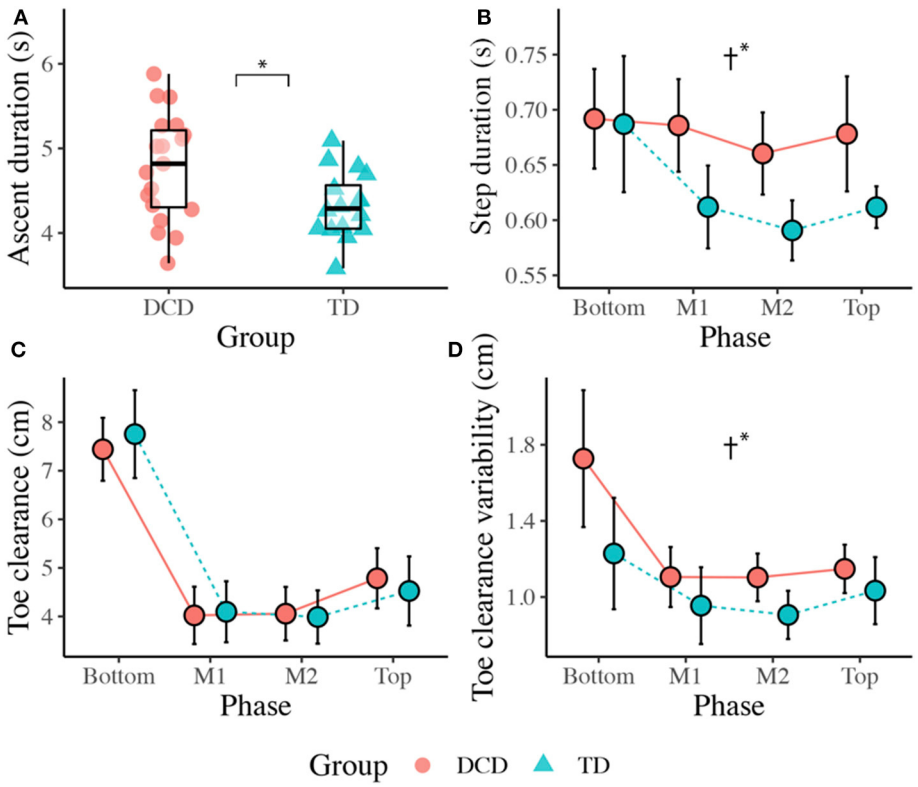

FIGURE 2 | Boxplots displaying the median, quartiles, and each individual's mean time taken to ascend the staircase **(A)**, and line plots displaying the mean (±95% CI) time taken to complete each step **(B)**, the mean (±95% CI) vertical toe clearance **(C)**, and the mean vertical toe clearance variability **(D)** for both DCD and TD children across each phase of the staircase. † Significant main effect of Group (*$p < 0.05$).

Gaze Behavior

Fixation Rate/Duration

Independent-samples *t*-tests showed no significant difference between the TD ($M = 2.21$) and DCD ($M = 2.08$) groups for fixation rate, $t(31) = 1.076$, $p = 0.290$, $d = 0.37$, and no difference between the TD ($M = 309.03$ ms) and DCD ($M = 319.12$ ms) groups for mean fixation duration, $t(31) = -0.818$, $p = 0.420$, $d = 0.28$.

Steps Looked Ahead

There was no main effect of Group, $F_{(1, 31)} = 0.025$, $p = 0.874$, $\eta_p^2 = 0.001$, but a significant main effect of Phase, $F_{(2.137, 66.233)} = 41.635$, $p < 0.001$, $\eta_p^2 = 0.573$, that was best described by a quadratic linear trend, $F_{(1, 31)} = 102.692$, $p < 0.001$, $\eta_p^2 = 0.768$, indicating that participants looked fewer steps ahead during the initial Bottom phase ($M = 2.20$), more steps ahead during the M1 ($M = 3.12$), and M2 ($M = 3.204$) phases, and then fewer steps ahead during the final Top phase ($M = 2.05$).

Gaze in Action

Children with DCD allocated more gaze time than the TD children to the handrails during the initial Bottom phase, $U = 85.00$, $z = -2.259$, $p = 0.024$, $r = -0.419$, and during the M1 phase, $U = 96.00$, $z = -2.301$, $p = 0.021$, $r = -0.429$; **Figure 3**).

Stair Descent

Anxiety

A Mann-Whitney *U*-test showed that the ranks of state anxiety for the DCD children (Median = 3.00) were significantly greater than the ranks of state anxiety for the TD children (Median = 1.00, $U = 91$, $z = -2.131$, $p = 0.033$, $r = -0.360$).

Handrail Use

The children with DCD displayed significantly greater handrail use compared to the TD children, $U = 91.50$, $p = 0.010$, $r = 0.423$. Specifically, three TD children used the handrails, two of whom used the handrail for all 5 trials. In comparison, 15 of the DCD children used the handrails, 11 of whom used the handrails for all 5 trials.

Stair Descent Duration

The DCD children ($M = 4.35$ s) took significantly longer than the TD children ($M = 3.77$ s) to descend the staircase, $t(33) = -2.547$, $p = 0.016$, $d = 0.876$. The DCD children ($M = 0.55$ s) also showed greater variability than the TD children ($M = 0.27$ s) in stair descent durations, $U = 58.00$, $z = -3.113$, $p = 0.002$, $r = -0.526$. A Spearman's rank correlation, performed to determine the relationship between state anxiety and stair descent duration, showed state anxiety to display a positive correlation with both

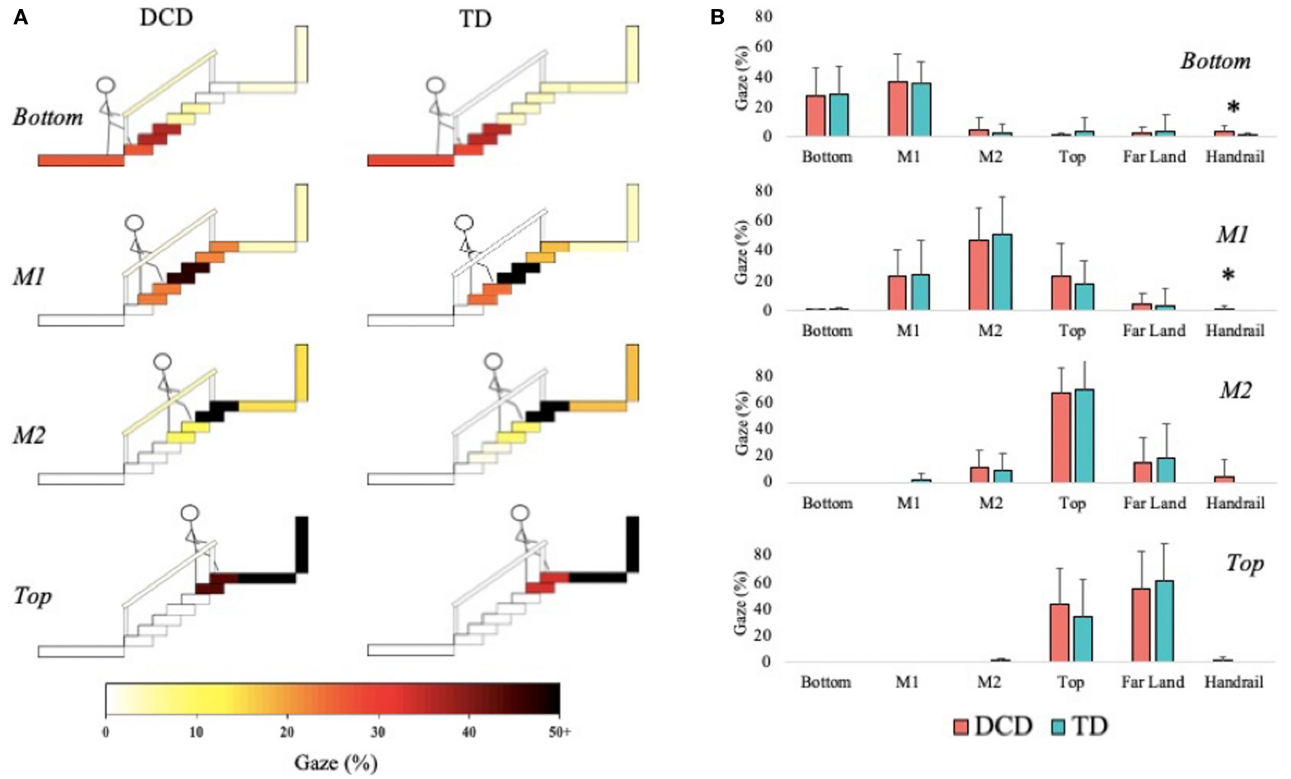

FIGURE 3 | Distribution of gaze fixations on stair features relative to the phase of action for both DCD TD children. **(A)** Adapted from Miyasike-daSilva et al. (2011), each set of stairs shows the participants stepping location (stick figure) and the respective percentage of gaze fixations directed from that location to each of the AOI's. Darker shaded AOI's represent the most fixated regions. **(B)** The mean (±95% CI) percentage of fixations to each AOI and across each task phase are further presented using bar charts for children with and without DCD. Analyses revealed the DCD children to display a greater percentage of fixations toward the handrails than the TD children during the Bottom and M1 task phases (*$p < 0.05$).

stair descent duration ($r_s(35) = 0.495$, $p = 0.002$) and stair descent duration variability ($r_s(35) = 0.409$, $p = 0.015$; **Figure 4**).

Step Duration

There was a significant main effect of Group, $F_{(1, 33)} = 6.343$, $p = 0.017$, $\eta_p^2 = 0.161$, with longer step durations observed in the DCD children ($M = 687$ ms) compared to the TD children ($M = 599$ ms). There was also a significant main effect of Phase, $F_{(2.187, 72.149)} = 76.038$, $p < 0.001$, $\eta_p^2 = 0.697$, with significantly longest step durations observed during the Top of the staircase ($M = 773$ ms, $ps < 0.001$). There was no Phase x Group interaction, $F_{(3, 99)} = 0.100$, $p = 0.960$, $\eta_p^2 = 0.003$. A main effect of Group was also observed for step duration variability, $F_{(1, 33)} = 17.244$, $p < 0.001$, $\eta_p^2 = 0.343$, with greater variability observed in the DCD children ($M = 104$ ms) compared to the TD children ($M = 60$ ms). There was also a significant main effect of Phase, $F_{(3, 99)} = 9.485$, $p < 0.001$, $\eta_p^2 = 0.223$, best described by a quadratic linear trend ($\eta_p^2 = 0.493$). Comparisons showed greater step duration variability over the Top ($M = 9.6$ cm) and Bottom ($M = 8.9$ cm) phases of the staircase compared to the M1 ($M = 7.1$ cm) and M2 ($M = 7.3$ cm) phases ($ps < 0.050$). There was no Phase x Group interaction, $F_{(3, 99)} = 0.952$, $p = 0.419$, $\eta_p^2 = 0.028$ (**Figure 4**).

Kinematics

Horizontal Heel Clearance

There was no main effect of Group, $F_{(1, 33)} = 0.005$, $p = 0.942$, $\eta_p^2 = 0.000$, but there was a main effect of Phase, $F_{(3, 99)} = 66.347$, $p < 0.001$, $\eta_p^2 = 0.668$. Comparisons revealed greatest clearance at the Bottom of the staircase ($M = 15.5$ cm, $ps < 0.001$) and smallest clearance at M1 ($M = 7.1$ cm, $ps < 0.001$) compared to all other phases (**Figure 5**).

Horizontal Heel Clearance Variability

There was a significant main effect of Group, $F_{(1, 33)} = 13.372$, $p = 0.001$, $\eta_p^2 = 0.288$, with greater variability observed in the DCD children ($M = 2.5$ cm) compared to the TD children ($M = 1.9$ cm). There was also a main effect of Phase, $F_{(1.433, 47.284)} = 44.423$, $p < 0.001$, $\eta_p^2 = 0.574$, with greatest variability observed at the Bottom of the staircase compared to all other phases ($M = 3.8$ cm, $p < 0.001$; **Figure 5**).

MoS Anteroposterior

There was no main effect of Group, $F_{(1, 32)} = 0.246$, $p = 0.623$, $\eta_p^2 = 0.008$. There was a main effect of Phase, $F_{(1.827, 58.461)} = 28.200$, $p < 0.001$, $\eta_p^2 = 0.468$, with lowest MoS values observed at the Top of the staircase ($M = -8.9$ cm) and

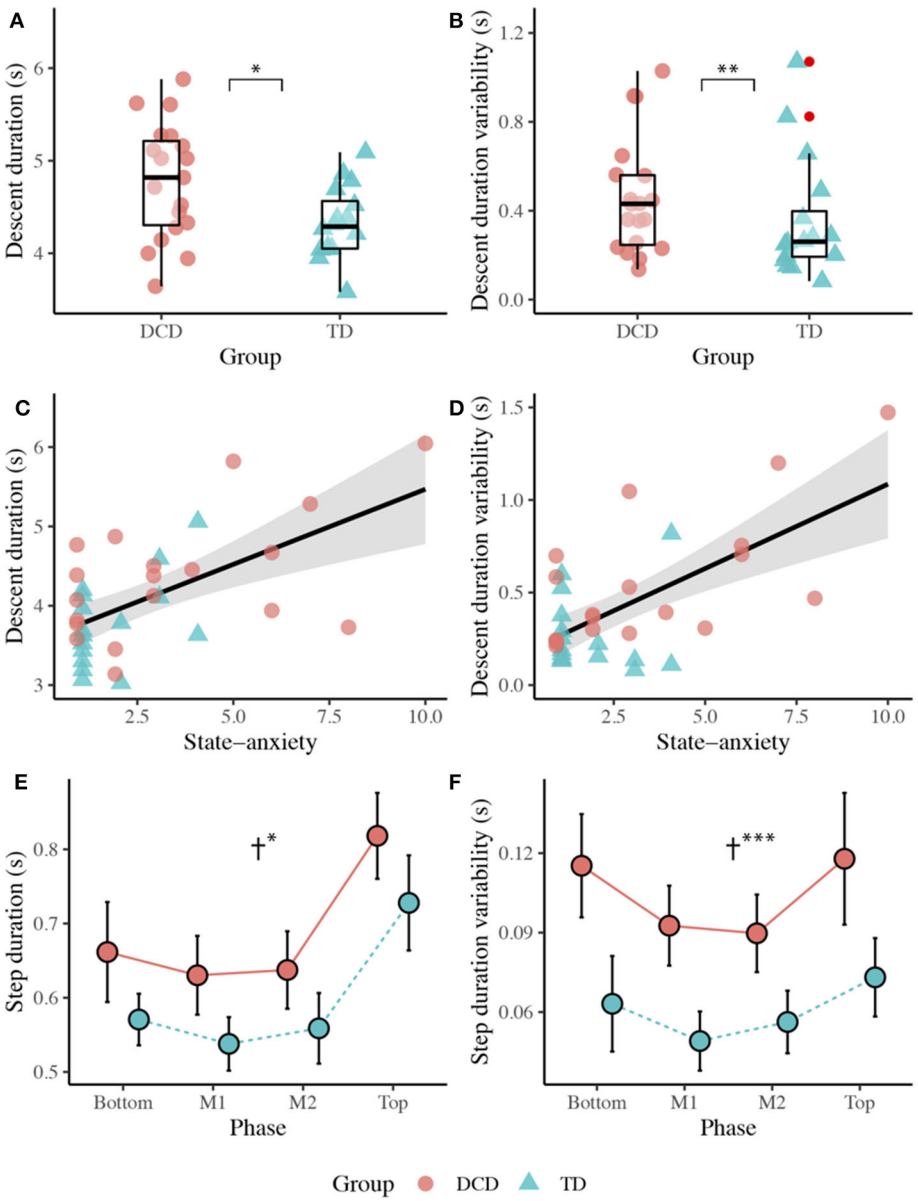

FIGURE 4 | Top panel: Boxplots displaying the median, quartiles, and individual mean time taken to descend the staircase **(A)**, and variability (1 SD across trials) in the time taken to descend the staircase **(B)** for both the DCD and TD groups. Middle panel: Scatter plots displaying the relationship between state anxiety and mean stair descent duration **(C)**, and the relationship between state anxiety and stair descent duration variability **(D)**. Bottom panel: Line plots displaying the mean (±95% CI) time taken to complete each step **(E)** and the variability (1 SD across trials) in time taken **(F)** to complete each step for both the DCD and TD children. † Significant main effect of Group (*$p < 0.05$, **$p < 0.01$, ***$p < 0.001$).

highest values observed at the Bottom ($M = -4.0$ cm) and M1 ($M = -4.2$ cm) phases.

Gaze Behavior

Fixation Rate/Duration

Independent-samples *t*-tests showed no significant difference between TD ($M = 2.01$) and DCD ($M = 1.82$) groups for mean fixation rate, $t(27) = 1.530$, $p = 0.138$, $d = 0.571$, and no significant difference between TD ($M = 367.48$ ms) and DCD ($M = 385.15$ ms) groups for mean fixation duration, $t(27) = -0.621$, $p = 0.540$, $d = 0.232$.

Steps Looked Ahead

There was no main effect of Group, $F_{(1, 26)} = 2.409$, $p = 0.133$, $\eta_p^2 = 0.085$. There was a significant main effect of Phase, $F_{(2.145, 55.770)} = 16.815$, $p < 0.001$, $\eta_p^2 = 0.393$, with participants looking more steps ahead at the Top ($M = 2.44$) and at M2 ($M = 2.82$) compared to when at the Bottom ($M = 1.89$, *ps* <

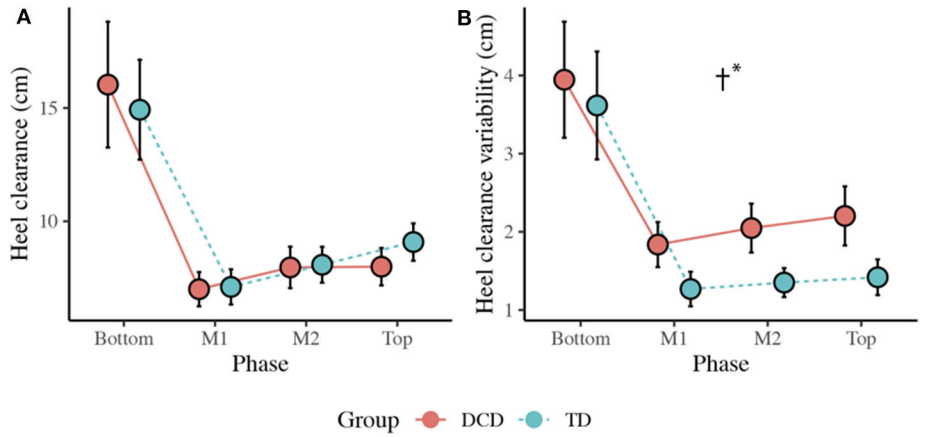

FIGURE 5 | (A) Line plots displaying the mean (±95% CI) horizontal heel clearance and mean horizontal heel clearance variability **(B)** for both the DCD and TD children. † Significant main effect of Group (*$p < 0.001$).

0.013) of the staircase. There was also a significant Group x Phase interaction, $F_{(2.145,\ 55.770)} = 6.233$, $p = 0.003$, $\eta_p^2 = 0.193$. *Post-hoc* comparisons with Sidak corrections showed that the DCD children ($M = 2.86$) looked significantly more steps ahead than the TD children ($M = 2.02$) during the initial Top phase ($p = 0.015$). A Spearman's rank correlation, performed to determine the relationship between the number of steps looked ahead and state anxiety, showed a positive relationship between state anxiety and the number of steps looked ahead during the M2 phase ($r_s(28) = 0.418$, $p = 0.027$; **Figure 6**).

Gaze in Action

The DCD children allocated more gaze ($M = 4.67\%$) than the TD children ($M = 1.86\%$) toward the Bottom of the staircase when they were positioned on the Top phase of the staircase, $U = 64.50$, $z = -2.086$, $p = 0.037$, $r = -0.387$. During the M2 phase, the TD children (45.71%) allocated more gaze than the DCD children (33.33%) to M1, $U = 59.00$, $z = -2.008$, $p = 0.045$, $r = -0.373$, whereas the DCD children ($M = 30.67\%$) allocated more gaze than the TD children ($M = 9.79\%$) to the Bottom, $U = 58.00$, $z = -2.074$, $p = 0.038$, $r = -0.385$; **Figure 7**).

DISCUSSION

The aim of the present study was to provide the first detailed account of the visuomotor control strategies that underpin stair negotiation in children with DCD and account for the possible influence of state anxiety. From our parental confidence survey, we provide the first quantifiable evidence that the parents of children with DCD report significantly lower confidence in their child's ability to maintain balance when walking up and down the stairs—with confidence lowest in their child's ability to navigate the stairs without using the handrails. We also provide evidence that children with DCD experience significantly more stair-related falls than children without DCD. These findings reinforce stair negotiation as a significant issue for children with DCD and reinforce the importance of the present investigation.

Stair Ascent

When ascending the stairs, the children with DCD walked more slowly (e.g., ∼500 ms longer to ascend stairs), displayed greater variability in their walk and step durations, displayed greater frequency of handrail use, and displayed greater toe clearance variability. State anxiety did not differ between groups, nor did their gaze behavior, with both groups maintaining gaze between two and three steps ahead of stepping location, aligning with previous work in older adults (Zietz and Hollands, 2009; Miyasike-daSilva et al., 2011).

More variable walking patterns have been observed in children with DCD previously (Rosengren et al., 2009; Wilmut et al., 2016; Parr et al., 2020) and is commonly taken as a sign of impaired motor control reflecting intrinsic neuromotor noise (Moe-Nilssen and Helbostad, 2005; Smits-Engelsman and Wilson, 2013). However, previous studies have found no difference in walking speeds between DCD and TD children during over ground gait (Wilmut et al., 2016), obstacle crossing (Deconinck et al., 2010) and an adaptive locomotion task (Parr et al., 2020). The slower walking speed adopted by the DCD group, combined with the greater reliance on handrail use, may therefore reflect a protective adaptation to minimize destabilizing momentum (Menz et al., 2004) and explain how the children with DCD were able to maintain similar margins of stability as their TD peers. Indeed, children with DCD have been shown to display difficulties with balance control during walking (Deconinck et al., 2006) and when crossing obstacles (i.e., when no handrails are available; Deconinck et al., 2010). Adopting this more cautious approach may therefore act as an important compensatory strategy that not only promotes stability, but also alleviates concerns related to the fear of falling. However, this adaptive behavior could also reflect the constraints children with DCD experience in lower limb strength and power (Raynor, 2001; Yam and Fong, 2018), given the increased mechanical demands placed

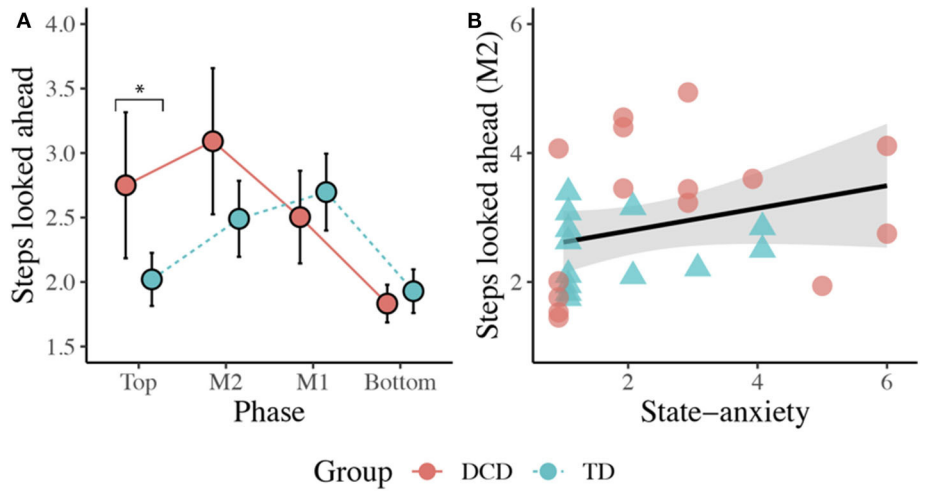

FIGURE 6 | (A) Line plot displaying the mean (±95% CI) number of steps looked ahead during each task phase for both the DCD and TD children. **Post-hoc* analyses revealed the children with DCD looked significantly more steps ahead than the TD children during the initial Top phase (*$p < 0.05$). **(B)** Scatter plot displaying the significant positive relationship observed between state anxiety and the mean number of steps looked ahead during the M2 phase ($p = 0.027$).

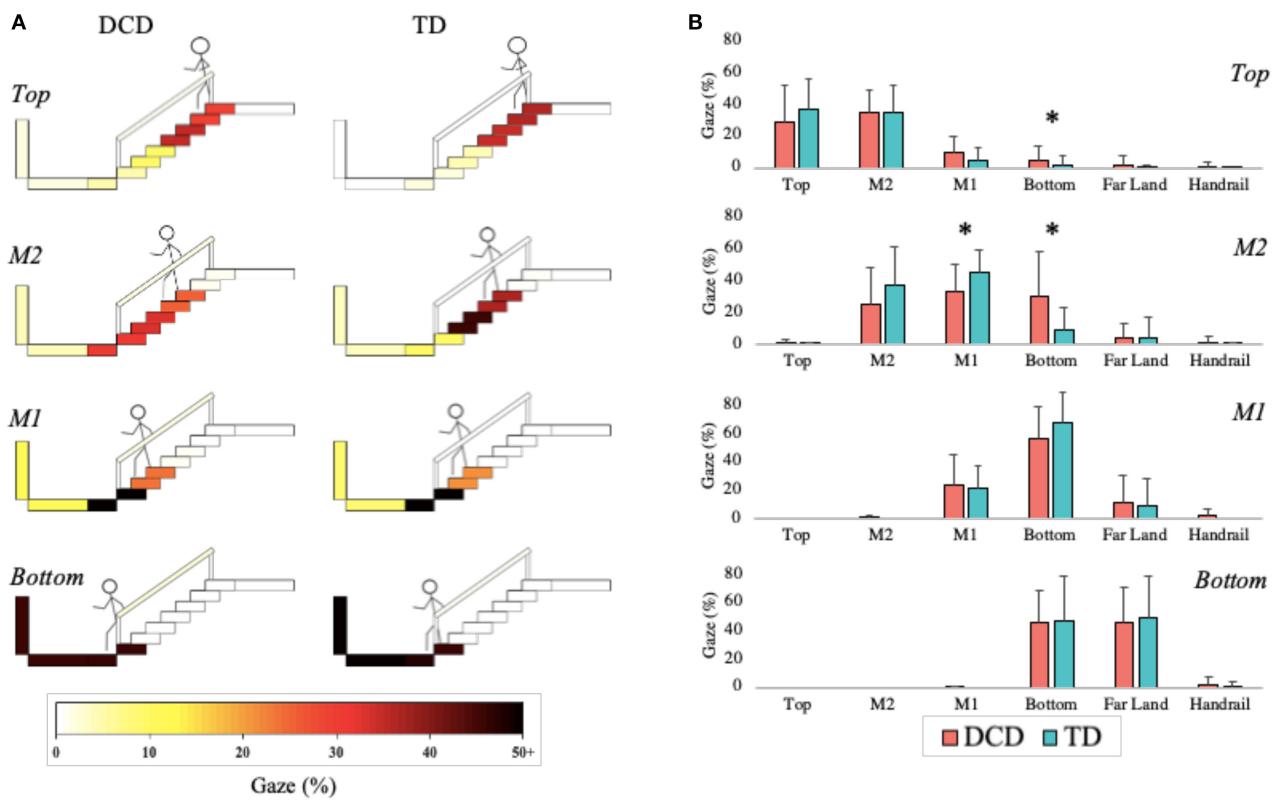

FIGURE 7 | Distribution of gaze fixations on stair features relative to the phase of action for both the DCD and TD children. **(A)** Adapted from Miyasike-daSilva et al. (2011), each set of stairs shows the participants stepping location (stick figure) and the respective percentage of gaze fixations directed from that location to each of the AOI's. Darker shaded AOI's represent the most fixated regions. **(B)** The mean (±95% CI) percentage of fixations to each AOI and across each task phase are further presented using bar charts for both DCD and TD children. Analyses showed that the children with DCD allocated significantly more gaze than the TD children toward the Bottom of the staircase when stood at the Top of the staircase and during the M2 phase (*$p < 0.05$). In comparison, the TD children allocated significantly more gaze than the DCD children toward M1 during the M2 phase (*$p < 0.05$).

on the lower extremities during stair ascent compared to level ground walking (Aldridge et al., 2012).

Despite similar margins of stability, children with DCD still displayed significantly greater toe clearance variability, which may contribute to stair-related falls by increasing the likelihood of accidentally contacting step edges (Hamel et al., 2005). Given the similarity in gaze behaviors between groups, inaccurate stepping in children with DCD during stair ascent is likely to occur regardless of appropriate looking behavior. This finding is consistent with our recent work in over ground precision stepping (Parr et al., 2020) and further implicates an inherent deficit to neuromuscular control of the lower limbs during locomotion in children with DCD (Rosengren et al., 2009). It is also possible that children with DCD are less effective at using the acquired visual information to guide safe and consistent stepping actions (Parr et al., 2020). By adopting a similar "look-ahead" gaze strategy observed in the TD children, the children with DCD are placing similar demands on feedforward and predictive control mechanisms they have previously shown to struggle with (Adams et al., 2014). Maintaining gaze several steps ahead during stair ascent could therefore be detrimental to stepping performance in children with DCD and may increase the risk of stair-related falls.

Descent

When descending the stairs, the children with DCD again walked more slowly (e.g., ∼600 ms longer to descend the stairs), displayed greater variability in their walk and step durations, displayed greater frequency of handrail use, and displayed greater heel clearance variability. However, unlike stair ascent, the children with DCD reported significantly higher levels of state anxiety and utilized a different gaze strategy than the TD children, looking significantly more steps ahead during the initial Top phase of the staircase.

Descending the staircase poses a greater challenge to postural dynamic stability and a greater threat to injury in the event of a fall, compared to stair ascent (Mian et al., 2007). Problems with balance control may therefore increase the fear of falling in children with DCD during stair descent despite compensatory behaviors (e.g., walking slower and using handrails) to maintain stability. As heightened anxiety was not observed during stair ascent, it is possible that motor difficulties in children with DCD may only increase state anxiety when it interferes with the ability to (safely) meet the demands of the task. It is also possible that increased state anxiety may be driven by ruminative thoughts and worries (Ellmers and Young, 2019) given the increased frequency of stair-related falls reported by the parents of children with DCD in the present study. Either way, our results suggest that state anxiety may have influenced the way children walked down the stairs. For example, state anxiety was positively correlated with stair descent duration and variability, suggesting that more anxious individuals walked slower and at more variable speeds when descending the stairs. This is in line with previous research in older adults and may reflect a "stiffening" strategy that is used to avoid potentially destabilizing motor patterns that might inflate the risk of a fall (Young and Mark Williams, 2015). Slower descent speeds have also been observed under conditions that increase the difficulty of visually identifying stair features, such as poor lighting (Thomas et al., 2020) and when faced with ambiguous stair surface patterns (Thomas et al., under review). Slower walking speeds may, therefore, serve to both improve stability and counteract anxiety-related decreases in attentional processing efficiency, allowing more time to extract and process acquired information to guide safe stepping.

State anxiety was also associated with group differences in the spatial allocation of gaze. The children with DCD looked significantly more steps ahead than the TD children during the initial entry (Top) phase which, when considering gaze in action, was seemingly underpinned by a greater tendency to fixate the bottom of the staircase. During the upper-mid-step (M2) phase, the DCD children again spent longer fixating the bottom of the staircase and the number of steps looked ahead was positively correlated with state anxiety. This tentatively suggests an anxiety-specific response that may bias gaze toward the planning of future stepping actions over the accurate execution of ongoing stepping commands (Chapman and Hollands, 2006) and may reflect a hypervigilance toward distant aspects of the environment that are perceived to pose a threat to balance (Young and Mark Williams, 2015). Maintaining gaze further along the travel path may, therefore, better serve balance in children with DCD by simplifying the extraction of pertinent information from optic flow and providing peripheral vision of the lower limbs and stairs (Zietz and Hollands, 2009). However, looking further ahead is likely to place an increased reliance on an internalized representation of stair dimensions and the use of predictive motor control. Given substantial evidence that children with DCD have difficulties generating and implementing predictive models of action (c.f. Adams et al., 2014), it is possible that this anxiety-driven gaze response may be contributing to increased heel clearance variability and the risk of falls.

Practical Implications to Improve Stair Safety

Taken together, our results highlight significant differences in the visuomotor control strategies that underpin stair negotiation in children with and without DCD. However, it is unclear at this point whether the visuomotor control strategies observed in the DCD group are actually mitigating or contributing to their increased frequency of stair-related falls. Future attempts to answer this question could, therefore, have practical implications for the optimisation of stair safety in children with DCD. For example, eye-movement training has been used to improve the coordination and performance of throwing and catching in children with DCD (Słowiński et al., 2019) and increase stepping accuracy in older adults when navigating obstacles (Young and Hollands, 2010). Manipulating eye-movement behavior during stair negotiation could therefore help determine an "optimal" gaze strategy that could subsequently be trained to aid stair negotiation. Similarly, movement training interventions have been used to improve functional strength and balance in children with DCD (Ferguson et al., 2013; Jelsma et al., 2014; Bonney et al., 2017) and to improve balance and reduce the fear of falling in older adults (Li et al., 2005; Schmid et al., 2010). Understanding how improved functional strength and balance

influence visuomotor control and anxiety would shed light on the mechanisms that underpin stair problems in children with DCD. Furthermore, evidence suggests that focusing attention internally, toward the conscious online processing of motor commands, can result in slower, less efficient and more unstable locomotion (Mak et al., 2020; Young et al., 2020). Future research could therefore consider the interplay between overt (spatial allocation of gaze) and covert attentional processes to provide a more holistic understanding of the attentional strategies that may differentiate DCD and TD stair navigation. Finally, it would be interesting to determine whether difficulties and/or anxieties with stair negotiation in children with DCD are related to the reduced confidence of their parents. It is possible that some parents may overcome concerns relating to injury by preventing each child from being exposed to potentially destabilizing situations (i.e., stairs without handrails), thus hindering the development of these task-specific skills.

LIMITATIONS

The results of this study should be considered with respect to several limitations that may stimulate the questions to be addressed in future work. For example, our sample includes a relatively wide age range (8–15 years) that is likely to encompass children of varying developmental maturation. As the development of visually guided stepping goes through distinct changes throughout these developmental years (Mowbray et al., 2019) we invite caution when extrapolating our findings to children of all ages. We also acknowledge the limitations of self-reported state anxiety in children given developmental aspects of emotional self-perception (Smith et al., 2006). However, the positive relationship we observed between anxiety and stair descent duration is consistent with previous literature and reinforces the utility of these simple inventories. Yet, future work would still benefit from attempts to objectively capture a physiological state anxiety response to compliment measures of self-report and overcome the limitations of ordinal data. Similarly, our binary measure of handrail use fails to quantify the precise handrail onset, duration, laterality, and contact force, each of which are required to determine the full extent of handrail dependency in children with DCD. Furthermore, it is important to recognize that handrails are not always available to aid stability. Future work should, therefore, explore how the manipulation of handrail use influences the risk/fear of falling in children with DCD during stair negotiation. It is also interesting to acknowledge that our task lacks the environmental complexity children are likely to face when navigating the stairs in the real world. For example, navigating a busy staircase at school will likely require the foveation of other people's walking behavior to avoid collision (Jovancevic-Misic et al., 2007). Similarly, using a mobile phone will draw attention away from the stairs and place greater demands on peripheral vision (Ioannidou et al., 2017). Understanding how a concurrent task affects stair safety could therefore have significant implications for clinicians managing children with DCD. Finally, whilst margin of stability provides a comprehensive assessment of dynamic balance, this measure has not been routinely used in the DCD literature. Future work may therefore benefit from complimenting margin of stability with more familiar measures of balance control (for review, see Verbecque et al., in press) to triangulate issues with stability.

CONCLUSION

In conclusion, the results of this study show that (a) safe stair negotiation is a significant and anxiety-inducing task that children with DCD struggle with, and (b) that there are clear differences in the visuomotor control strategies that underpin stair negotiation in children with and without DCD. Overall, it appears that children with DCD overcome difficulties with balance control, and successfully maintain stability, by walking slower and relying heavily on handrail use. However, children with DCD still display evidence of significantly greater step-edge clearance variability than TD children, which possibly increases the risk of a fall. Unlike stair ascent, children with DCD report heightened anxiety prior to stair descent and look further along the staircase during the initial entry phase. However, it is unclear at this point whether these anxiety related alterations to gaze are detrimental to stair negotiation safety and contribute to the frequency of falls.

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 human participants were reviewed and approved by Liverpool John Moores Ethics Committee. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin. Written informed consent was obtained from the minor(s)' legal guardian/next of kin for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

MH, RF, and GW acquired the funding. MH, RF, GW, and NT edited the manuscript. MH, RF, GW, and JP designed the study. JP and NT undertook the data collection. JP completed the statistical analysis, figure preparation, and the first draft of the manuscript. All authors approved the final version of the manuscript for submission.

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Effects of Transcranial Direct Current Stimulation on Motor Function in Children 8–12 Years With Developmental Coordination Disorder: A Randomized Controlled Trial

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Background and objectives: Developmental coordination disorder (DCD) is a neurodevelopmental motor disorder occurring in 5-6% of school-aged children. It is suggested that children with DCD show deficits in motor learning. Transcranial direct current stimulation (tDCS) enhances motor learning in adults and children but is unstudied in DCD. We aimed to investigate if tDCS, paired with motor skill training, facilitates motor learning in a pediatric sample with DCD.

Methods: Twenty-eight children with diagnosed DCD (22 males, mean age: 10.62 ± 1.44 years) were randomized and placed into a treatment or sham group. Anodal tDCS was applied (1 mA, 20 min) in conjunction with fine manual training over 5 consecutive days. Children's motor functioning was assessed with the Purdue Pegboard Test and Jebsen-Taylor Hand Function Test at baseline, post-intervention and 6 weeks following intervention. Group differences in rates of motor learning and skill transfer/retention were examined using linear mixed modeling and repeated measures ANOVAs, respectively.

Results: There were no serious adverse events or drop-outs and procedures were well-tolerated. Independent of group, all participants demonstrated improved motor scores over the 5 training days [$F_{(69.280)}$, $p < 0.001$, 95% CI (0.152, 0.376)], with no skill decay observed at retention. There was no interaction between intervention group and day [$F_{(2.998)}$, $p = 0.086$, 95% CI (−0.020, 0.297)].

Conclusion: Children with DCD demonstrate motor learning with long-term retention of acquired skill. Motor cortex tDCS did not enhance motor learning as seen in other populations. Before conclusions of tDCS efficacy can be drawn, additional carefully designed trials with reproducible results are required.

Clinical Trial Registration: ClinicalTrials.gov: NCT03453983

Keywords: neuromodulation, transcranial direct current stimulation, motor learning, developmental coordination disorder, randomized controlled trial

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INTRODUCTION

Developmental coordination disorder (DCD) affects 5–6% of school-aged children and is characterized by early onset of motor impairment, manifesting as clumsy, slow and inaccurate performance of motor tasks (American Psychiatric Association, 2013). Reduced motor competency interferes with activities of daily living (American Psychiatric Association, 2013), posing a threat to the physical literacy and mental health of affected children (Castelli et al., 2014; Harrowell et al., 2018; Blank et al., 2019). In addition to deficits in motor execution, children with DCD may also display difficulties learning new motor skills and/or tasks (Bo and Lee, 2013; Gomez and Sirigu, 2015; Biotteau et al., 2016a).

Children with DCD are encouraged to practice tasks that they find difficult, in the hope that movement repetition will improve performance (Levac et al., 2009; Smits-Engelsman et al., 2013; Blank et al., 2019). Although successes have be reported with practice, improvements are often variable and more commonly observed with intensive practice (Smits-Engelsman et al., 2013; Jane et al., 2018). The need for high doses of repetition could be attributed to a slower rate of motor learning among children with DCD (Biotteau et al., 2016b; Jane et al., 2018). However, such training may be an anathema to most children, highlighting the need for more efficient therapies.

The use of transcranial direct current stimulation (tDCS), a form of non-invasive brain stimulation, in motor rehabilitation is rapidly expanding (Bikson et al., 2016; Palm et al., 2016). TDCS, through the application of a subthreshold electrical current (1–2 milliamps), alters neuronal excitability and spontaneity, facilitating the brain's endogenous mechanisms of neuroplasticity (Stagg and Nitsche, 2011; Kronberg et al., 2017). When paired with motor skill training, multi-session tDCS is shown to augment motor learning in adults (Reis et al., 2009), typically developing children (Ciechanski and Kirton, 2017; Cole et al., 2018) and children with motor impairment such as cerebral palsy (Finisguerra et al., 2019; Grohs et al., 2019; Saleem et al., 2019). Recent reviews highlight the growing body of research investigating the therapeutic potential of tDCS in children with neurodevelopmental disorders (Finisguerra et al., 2019; Grohs et al., 2019; Saleem et al., 2019); preliminary evidence has supported tDCS enhanced motor functioning in balance, gait, hand function, reaction time and in inhibitory control. Safety and tolerability of tDCS is well-established in adults (Bikson et al., 2016) and is growing in children (Zewdie et al., 2020).

Given support for the safety, feasibility and efficacy of tDCS in pediatric populations with motor impairment, tDCS may provide an avenue to modulate motor learning and strengthen the effects of current therapies in children with DCD. However, the application of tDCS in a pediatric population with DCD has yet to be examined. Here, we present results of the first randomized controlled trial (NCT03453983) investigating the effects of multi-session motor cortex tDCS on motor learning in children with DCD; the primary motor cortex (M1) is a logistical initial target given its direct role in movement production (Sanes and Donoghue, 2000) and evidence showing that plastic changes within M1 are associated with early phases of motor learning (Dayan and Cohen, 2011). Based on previous evidence, it was hypothesized that enhanced rates of motor learning would be observed in children with DCD when fine manual skill training was paired with tDCS.

MATERIALS AND METHODS

Enrollment

This study was carried out between July 2018 and November 2019 at the Alberta Children's Hospital, Calgary, Canada. Participants were recruited through developmental and community pediatricians, psychologists, physical/occupational therapists and via social media. Written informed consent from participants' legal guardians and child assent were obtained at enrollment. The University of Calgary Conjoint Health Research Ethics Board approved this study (REB18-0183).

Eligibility

Inclusion criteria were: (1) age 8 to 12 years, (2) current diagnoses of DCD by a registered health care provider and (3) right-handed (i.e., hand used for writing). Children 8–12 years were recruited as DCD is commonly diagnosed in elementary school. Those with pre-term birth (<36 weeks' gestation) or any neuropsychiatric, neurological and/or chronic disorders were excluded. Children with a diagnosis of attention deficit/hyperactivity disorder (ADHD), learning disorder (LD), or generalized anxiety disorder (GAD) were included given the high co-occurrence with DCD (Dewey et al., 2002; Dewey, 2018).

Participants were screened to ensure they met clinical criteria for DCD outlined in the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) (American Psychiatric Association, 2013). Children demonstrated motor deficits (criterion A) with Total Test scores below the 16th percentile on the Movement Assessment Battery for Children 2nd Edition (MABC-II) (Barnett et al., 2007). Motor deficits interfered with daily functioning (criterion B), began early in development (criterion C), and were not better explained by an intellectual disability, visual impairment or neurological condition (criterion D). Diagnostic criteria B and C were confirmed by a parent questionnaire developed by the investigators, which included questions about difficulties experienced in three domains, (1) motor (i.e., handwriting, riding a bike, self-care tasks, motor planning, learning new motor tasks, etc.), (2) social (play and social skills, physically tired, lack of energy, etc.), and (3) academic (reading, writing, math skills, etc.), as well as the age at which motor difficulties were first observed. Criterion D was confirmed by questions on the parent questionnaire regarding all prior and current diagnoses as well as visual impairments, and children obtaining a Full-Scale IQ score >79 on the Wechsler Abbreviated Scale of Intelligence 2nd Edition (WASI-II) (Wechsler, 2011).

Study Design

A randomized, double-blind, sham-controlled trial was conducted in accordance with CONSORT guidelines (Schulz et al., 2010), including pediatric-specific considerations (Saint-Raymond et al., 2010). After screening, children were randomly assigned without stratification to one of two parallel intervention groups: (1) active tDCS or (2) sham. A simple

randomization procedure was used. Allocation (1:1) was concealed in sequentially numbered, opaque, sealed envelopes. Corresponding envelopes were opened by the investigator (MNG) immediately before intervention. Participants were blinded to their assigned group throughout the study and completed a post-intervention questionnaire that asked them to guess which intervention they received and why. Investigators were blinded at data analysis.

Sample Size

Previous evidence of tDCS enhanced motor learning in typically developing children and adolescents, using a similar protocol, reported a moderate-to-large effect size (Cohens $d > 0.65$) (Cole et al., 2018). To estimate the sample size required for the primary linear mixed model analysis in the current study, the smpsize_lmm command in RStudio was used with the above effect size and a two-sided type 1 error of 0.05. It was estimated that 16 participants per group would have 90% power to detect group differences in the primary outcome measure.

tDCS Intervention

The right M1 was localized using the 10–20 electroencephalography method (Steinmetz et al., 1989). A saline soaked 25cm^2 sponge electrode was placed over the right M1 (active anode electrode), with a second identical electrode placed on the contralateral supraorbital region (reference cathode electrode), held in place by adjustable head straps consistent with established methods (Ciechanski and Kirton, 2017).

Electrodes were attached to a conventional 1x1 tDCS system (Soterix, NY). Current was ramped up to 1 mA over 30 seconds (s). After 120 s, the current was maintained for 20 min (active tDCS group) or ramped back down to 0 mA over 30 s (sham group). The initial ramp-up produces transient scalp sensations and has been established as a valid sham technique (Ambrus et al., 2012). Following each stimulation session, participants completed a safety, and tolerability questionnaire (Garvey et al., 2001), documenting symptoms (i.e., headaches, burning, itching, tingling, and nausea), their severity and duration, as well as tolerability compared to seven common childhood experiences.

Outcome Measures

The primary outcome was left hand Purdue Pegboard Test (PPT) performance, a validated assessment of fine motor coordination and hand dexterity (Tiffin and Asher, 1948). The PPT consists of four subtests: left hand [PPT_L], right hand [PPT_R], bimanual [PPT_{LR}] peg placement, and bimanual assembly [PPT_A]. The peg placement tasks involved placing as many pins as possible into a pegboard in 30 s. The assembly task involved building as many copies of a demonstration structure using pins, collars and washers in 60 s. Scores were the highest total number of placed pegs or assembled items. The PPT_L (non-dominant left-hand performance) was used for motor skill training and as the primary outcome measure of motor learning, as it is a challenging fine manual task for children to learn without reaching a learning "ceiling" effect.

Motor skills may be acquired by two modes of learning: online and offline. Online learning refers to skill learning that occurs within a training period. Offline learning refers to skill learning that occurs after the training session has ended and is often referred to as consolidation. TDCS may differentially affect online and offline learning (Reis et al., 2009). In the current study, online effects (within-day training) were determined by comparing baseline to final PPT_L scores each day. Offline effects (between-day consolidation) were quantified by comparing baseline PPT_L scores each day to final PPT_L scores from the previous day. Daily effects were summed to obtain total online and offline effects.

Secondary outcomes included PPT_R, PPT_{LR}, and PPT_A performance, to examine intervention effects on the untrained hand and bimanual skills, as well as Jebsen-Taylor Test of Hand Function (JTT) performance (Jebsen, 1969), an upper extremity test of unimanual motor skills. The JTT included five subtests: card turning, picking up/placing small objects, stacking checkers, moving light objects, and moving heavy objects. Left and right hands were tested independently. An overall score for each hand was obtained by summing the completion times for each subtest [JTT_R, JTT_L].

Motor Training

A schematic of the study protocol is shown in **Figure 1**. On day 1, baseline motor tests (PPT, JTT) were administered, followed by tDCS intervention (active or sham). During the intervention, three PPT_L trials were performed at 5, 10, and 15 min as well as after intervention. Participants repeated this protocol for 4 consecutive days (days 2–5). Following training on day 5, all motor tests were repeated. Participants returned 6 ± 1 weeks later to repeat all motor tests. Assessments were video recorded and blindly scored offline.

Analysis

Statistical analysis was performed in RStudio (RStudio Team, V1.3.1093) (R Core Team, 2017) and SPSS (IBM SPSS Software, V25) (SPSS Inc., 2017). Shapiro-Wilk tests assessed normality of each measure. As appropriate, independent samples t-tests, Mann-Whitney *U*-tests or chi-square tests compared participant characteristics, clinical and motor scores at baseline and tolerability ratings between intervention groups. The primary analysis was intention-to-treat and involved all participants.

Our statistical approach was based on previously established methods (Cole et al., 2018). A linear mixed effects model was chosen for the primary analysis as this approach offers advantages for longitudinal data sets with more data points and non-linear outcomes (Gibbons et al., 2010); our primary outcome parameter (change in PPT_L score) was measured at six timepoints and previous findings from studies using similar protocols (Ciechanski and Kirton, 2017; Cole et al., 2018) showed non-linear changes in PPT_L performance over multiple training days. The linear mixed effects model examined changes in the primary outcome (PPT_L) between groups from pre- to post-intervention with fixed effects for Group, Day, the interaction of Group and Day, and random effects for participants including the intercept to account for repeated measures.

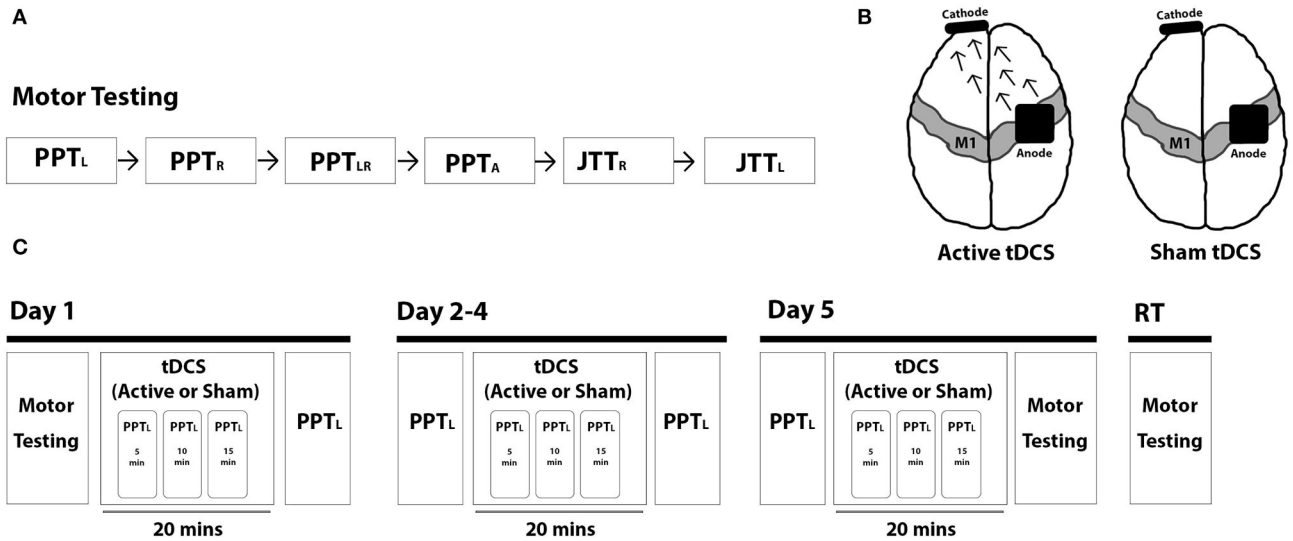

FIGURE 1 | Trial protocol. **(A)** Motor skill testing included the Purdue Pegboard Test (PPT) (left-handed: PPT_L, right-handed: PPT_R, bimanual: PPT_{LR}, assembly: PPT_A) and the Jebsen-Taylor Test of Hand Function (JTT) (right-handed: JTT_R, left-handed: JJT_L). **(B)** Intervention groups included 1 mA anodal tDCS (left; arrows represent the direction of current flow from anode to cathode) and sham tDCS (right). **(C)** Study protocol is shown broken down by each intervention day (day 1–5) and for retention testing (RT) at 6-weeks post-intervention.

As secondary motor outcomes were measured at fewer timepoints and motor learning curves were not being generated, two-way repeated measures ANOVAs were used to investigate changes in secondary outcomes between intervention groups. Independent samples *t*-tests examined between group differences in performance at each time point. Paired samples t-tests examined within group differences in PPT (PPT_L, PPT_R, PPT_{LR}, and PPT_A) and JTT (JTT_L, JTT_R) performance between timepoints (i.e., baseline to post-intervention, baseline to retention) and potential skill decay between the final training block (day 5) and retention testing. Online and offline learning effects were explored within and between groups using paired and independent samples t-tests.

RESULTS

Population

Twenty-eight children with DCD [10.62 ± 1.44 years; 22 (79%) male] were randomized (14 active, 14 sham). All participants completed baseline motor skill testing, the 5 consecutive intervention days and post-intervention motor skill testing. Six participants (3 active, 3 sham) did not complete retention motor skill testing due to travel or family factors (**Figure 2**, CONSORT recruitment flow diagram). Group demographics, clinical scores and baseline motor scores are shown in **Table 1**. No group differences were observed for age [$t_{(26)} = 0.637$, $p = 0.530$], sex [$x^2(1) = 0.848$, $p = 0.357$] or clinical scores (MABC-II: $U = 79$, $p = 0.374$; WASI-II: $t_{(26)} = -0.586$, $p = 0.563$). No group differences in baseline motor scores were observed (all $p > 0.7$). Fifteen participants had ADHD [$n = 6$ (43%) active, $n = 9$ (64%) sham], 11 had a LD [$n = 6$ (43%) active, $n = 5$ (36%) sham], and 5 had GAD [$n = 3$ (21%) active, $n = 2$ (14%) sham] (**Table 2**). Proportions did not differ between groups (all $p > 0.2$). Thirteen of the 28 participants were taking medications for ADHD (i.e., Vyvanse, Biphentin, and Clonidine) and/or anxiety (i.e., Prozac, Zoloft, and Citalopram) (**Table 2**). Proportions did not differ between groups ($p > 0.7$).

Motor Learning

PPT_L learning curves by group are shown in **Figure 3**. Curves were generated by plotting mean change in score from baseline to each training point. Linear mixed effects modeling showed that, independent of intervention, all participants demonstrated motor learning over 5 training days [$p < 0.001$, 95% CI (0.25–0.41), **Table 3**]. No interaction effect of Day and Group on rate of motor learning was seen; therefore, the interaction term was removed from the final mixed model [$PPT_L \sim$ group + day + (1|subjects)]. Average PPT_L performance was higher on post-intervention day 5 compared to baseline in both groups [active: $t_{(13)} = -5.824$, $p < 0.001$, Cohen's $d = 1.557$; sham: $t_{(13)} = -2.820$, $p = 0.014$, Cohen's $d = 0.754$]. No group differences were observed in average PPT_L performance at any time point (all $p > 0.1$).

Retention

Learning effects were retained in both groups, with no skill decay in PPT_L scores between post-intervention day 5 and retention testing at 6-weeks (**Figure 3**). In the active group, PPT_L scores at retention did not differ from post-intervention day 5 [$t_{(10)} = -1.966$, $p = 0.078$, Cohen's $d = 0.593$]. Within the sham group, higher PPT_L scores were observed at retention compared to post-intervention day 5 [$t_{(10)} = -4.989$, $p = 0.001$, Cohen's $d = 1.504$]. This difference may relate to lower scores in the sham group on day 5 (see above). In both groups, PPT_L scores

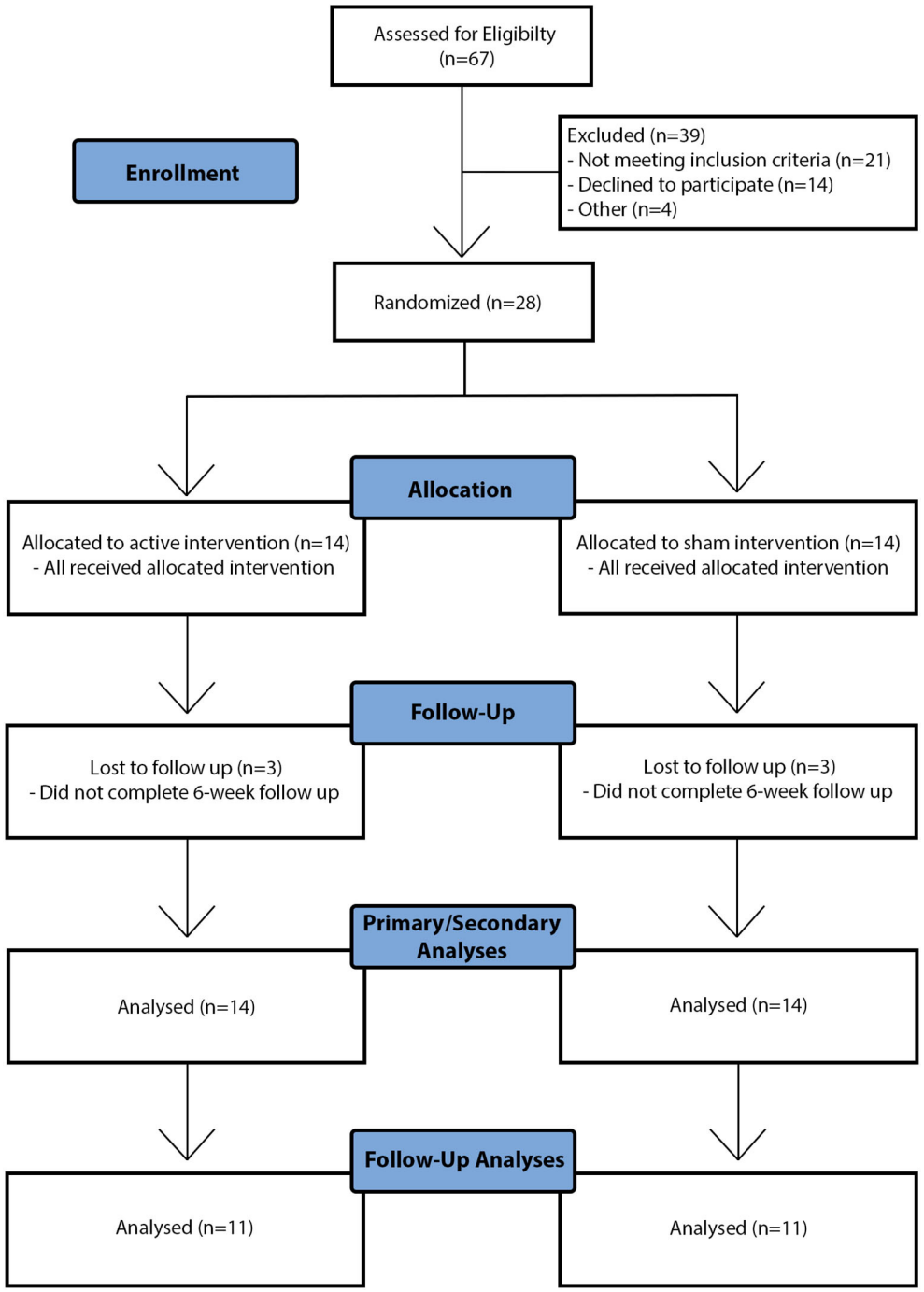

FIGURE 2 | CONSORT recruitment flow diagram. Visual schematic of participant recruitment, screening, data collection, and analysis. Note that "other reasons" for exclusion of children at eligibility screening and follow-up included travel and family factors.

at retention were higher than baseline [active: $t_{(10)} = -3.585$, $p = 0.005$, Cohen's $d = 1.080$; sham: $t_{(10)} = -6.037$, $p < 0.001$, Cohen's $d = 1.820$], with no group differences [$t_{(20)} = -1.025$, $p = 0.321$].

Online and Offline Learning Effects

There was more online learning compared to offline learning in both the active [$t_{(13)} = 2.545$, $p = 0.024$] and sham [$t_{(13)} = 5.488$, $p < 0.001$] groups (**Figure 4**). No group differences in online

TABLE 1 | Participant demographics, clinical scores and baseline motor scores by intervention group.

Group	Age (years)	Sex (M:F)	WASI-II FSIQ	MABC-II total test	Baseline Purdue Pegboard Test (PPT) scores				Baseline Jebsen-Taylor Test (JTT) scores	
					PPT_L	PPT_R	PPT_{LR}	PPT_A	JTT_L	JTT_R
Active ($n = 14$)	10.80 (±1.42)	10:4	100.29 (±9.88)	3.5 (±0.51)	11.52 (±2.10)	12.05 (±1.93)	8.93 (±1.67)	20.14 (±4.77)	29.48 (±5.16)	27.78 (±3.97)
Sham ($n = 14$)	10.45 (±1.50)	12:2	102.93 (±3.66)	2.86 (±0.38)	11.77 (±2.79)	12.26 (±2.42)	8.98 (±2.25)	20.50 (±6.49)	29.19 (±4.24)	27.68 (±4.98)
Mean	10.62 (±1.44)	22:6	101.61 (±2.23)	3.18 (±0.32)	11.64 (±2.43)	12.15 (±2.15)	8.96 (±1.95)	20.32 (±5.59)	29.33 (±4.64)	27.73 (±4.42)
Between group (*p*-value)	0.530	0.357	0.563	0.374	0.791	0.798	0.950	0.869	0.871	0.953

Note: Values are shown as group means ± standard deviation. Test statistics are shown for between group and within group comparisons. M:F, ratio of males to females; WASI-II, Wechsler Abbreviated Scale of Intelligence 2nd Edition; MABC-II, Movement Assessment Battery for Children 2nd Edition; PPT_L, Purdue Pegboard Test left-hand; PPT_R, Purdue Pegboard Test right-hand; PPT_{LR}, Purdue Pegboard Test bimanual; PPT_A, Purdue Pegboard Test assembly; JTT_L, Jebsen-Taylor Test left-hand; JTT_R, Jebsen Taylor Test right-hand.

TABLE 2 | Distribution of co-occurring attention, learning and anxiety disorders by participant and group.

Participant (DCD)	Attention disorder	Learning disorder	Anxiety disorder	Prescribed medications
Active tDCS Group				
S1	–	X	–	–
S2	–	–	X	–
S3	X	–	–	–
S4	X	X	X	X
S5	X	–	–	X
S6	–	–	–	–
S7	X	X	–	X
S8	–	X	–	X
S9	–	X	–	X
S10	X	–	–	X
S11	–	–	–	–
S12	X	X	X	–
S13	–	–	–	–
S14	–	–	–	–
Active total	6	6	3	6
Sham tDCS Group				
S1	X	–	–	X
S2	–	X	–	–
S3	X	–	–	X
S4	–	–	–	X
S5	X	X	–	–
S6	X	–	–	–
S7	–	–	–	–
S8	X	–	–	X
S9	X	–	–	X
S10	X	X	X	X
S11	X	X	X	X
S12	–	–	–	–
S13	X	X	–	–
S14	–	–	–	–
Sham total	9	5	2	7
Between group (*p*-value)	0.256	0.699	0.622	0.705

Note: The presence of a diagnosis by a registered health care provider is denoted with an X. Total within group numbers and test statistics for between group differences in distribution are also shown.

$[t_{(26)} = -0.669, p = 0.509]$ or offline $[t_{(26)} = 0.866, p = 0.395]$ learning were observed.

Secondary Motor Outcomes

Effects of intervention on secondary untrained PPT and JTT measures are shown in **Figure 5**. Learning effects were observed for PPT_R ($F = 32.346$, $p < 0.001$, partial $eta^2 = 0.554$), PPT_{LR} ($F = 32.795$, $p < 0.001$, partial $eta^2 = 0.558$) and PPT_A ($F = 28.041$, $p < 0.001$, partial $eta^2 = 0.519$). These were independent of group, with no interaction effects of Time and Group (all $p > 0.2$). In both groups, compared to baseline, PPT_R scores were higher on post-intervention day 5 [active: $t_{(13)} = -4.535$, $p = 0.001$, Cohen's $d = 1.212$; sham:

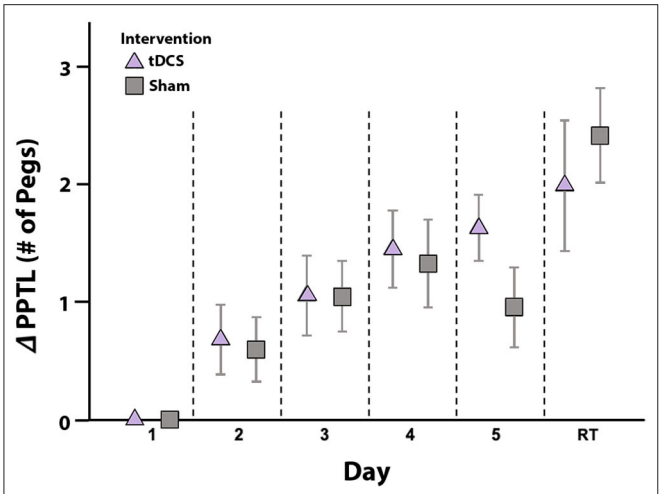

FIGURE 3 | Motor learning by intervention group. Mean daily change in PPT_L scores from baseline (y-axis) are shown for the active (triangle) and sham (squares) intervention groups. Error bars represent standard error. In both groups, scores improved from baseline to post-intervention testing on day 5, with no skill decay at retention testing (RT) 6-weeks post-intervention. No between group differences in PPT_L scores were noted at any timepoint. ΔPPTL: change in left-hand Purdue Pegboard Test scores from baseline.

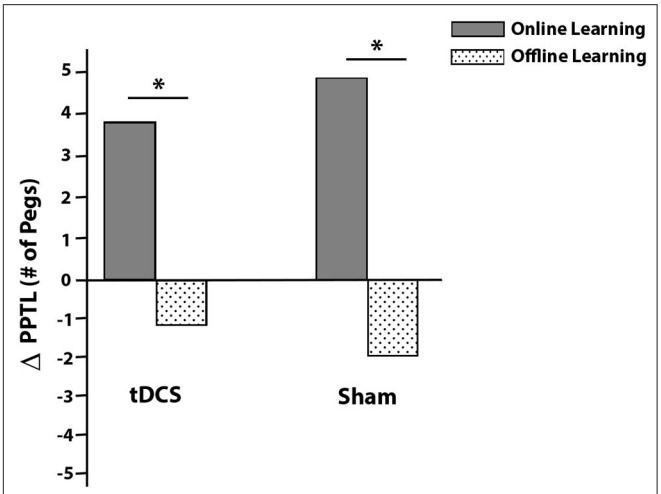

FIGURE 4 | Average PPT_L online (solid gray) and offline (dotted) learning effects by intervention group. Online effects are within session improvements while offline effects are improvements that occur between sessions (consolidation). Daily online and offline effects were summed to obtain total online and offline changes in scores (y-axis ΔPPTL). *$p < 0.05$.

TABLE 3 | Results of linear mixed effects model examining motor learning over 5 days of skill training.

	PPT_L score		
	Estimates	**CI**	***P***
Fixed effects			
Intercept	11.41	8.91–13.90	**<0.001***
Group	0.07	−1.50–1.64	0.927
Day	0.33	0.25–0.41	**<0.001***
Random effects			
Subjects	4.41	-	-
ICC	0.90	-	-
Marginal R^2	0.044	-	-

**Bold values represent statistically significant findings with a p < 0.001. CI, Confidence interval.*

$t_{(13)} = -3.863$, $p = 0.002$, Cohen's $d = 1.032$] and at retention [active: $t_{(10)} = -6.297$, $p < 0.001$, Cohen's $d = 1.899$; sham: $t_{(10)} = -4.856$, $p = 0.001$, Cohen's $d = 1.464$]. PPT_{LR} scores were higher by day 5 [active: $t_{(13)} = 4.436$, $p = 0.001$, Cohen's $d = 1.186$; sham: $t_{(13)} = 3.721$, $p = 0.003$, Cohen's $d = 0.994$] and at retention [active: $t_{(10)} = -4.730$, $p = 0.001$, Cohen's $d = 1.426$; sham: $t_{(10)} = -4.351$, $p = 0.001$, Cohen's $d = 1.312$] for both groups. PPT_A scores demonstrated a similar pattern at day 5 [active: $t_{(13)} = -4.200$, $p = 0.001$, Cohen's $d = 1.122$; sham: $t_{(13)} = -3.727$, $p = 0.003$, Cohen's $d = 0.996$] and retention [active: $t_{(10)} = -4.139$, $p = 0.002$, Cohen's $d = 1.248$; sham: $t_{(10)} = -4.967$, $p = 0.001$, Cohen's $d = 1.498$]. Finally, no skill decay from post-intervention day 5 to retention testing was observed in either group for PPT_R [active: $t_{(10)} = -2.015$, $p = 0.072$, Cohen's $d = 0.608$; sham: $t_{(10)} = -0.586$, $p = 0.571$, Cohen's $d = 0.177$], PPT_{LR} [active: $t_{(10)} = 0.379$, $p = 0.712$, Cohen's $d = 0.114$; sham: $t_{(10)} = -2.036$, $p = 0.069$, Cohen's $d = 0.614$], or PPT_A [active: $t_{(10)} = -1.919$, $p = 0.084$, Cohen's $d = 0.579$; sham: $t_{(10)} = -0.576$, $p = 0.578$, Cohen's $d = 0.174$].

Independent of intervention, learning effects were observed for JTT_L ($F = 11.476$, $p = 0.002$, partial eta$^2 = 0.306$) and JTT_R ($F = 7.887$, $p = 0.009$, partial eta$^2 = 0.233$). In the active group, JTT_L and JTT_R performance was faster on post-intervention day 5 [JTT_L $t_{(13)} = 3.150$, $p = 0.008$, Cohen's $d = 0.842$; JTT_R $t_{(13)} = 2.700$, $p = 0.018$, Cohen's $d = 0.722$] and at retention [JTT_L $t_{(10)} = 4.397$, $p = 0.001$, Cohen's $d = 1.326$; JTT_R $t_{(10)} = 3.348$, $p = 0.007$, Cohen's $d = 1.009$] compared to baseline. In the sham group, improved JTT_L performance was not seen on day 5 [$t_{(13)} = 1.722$, $p = 0.109$] but was present at retention testing [$t_{(9)} = 3.769$, $p = 0.004$, Cohen's $d = 1.136$]. JTT_R scores at day 5 [$t_{(13)} = 1.164$, $p = 0.265$, Cohen's $d = 0.311$] and retention [$t_{(9)} = 2.033$, $p = 0.073$, Cohen's $d = 0.613$] did not differ from baseline in the sham group. There was no evidence of skill decay from day 5 to retention testing on the JTT_L [active: $t_{(10)} = 2.093$, $p = 0.063$, Cohen's $d = 0.631$; sham: $t_{(9)} = 1.589$, $p = 0.147$, Cohen's $d = 0.479$] or JTT_R [active: $t_{(10)} = 1.521$, $p = 0.159$, Cohen's $d = 0.459$; sham: $t_{(9)} = 0.301$, $p = 0.770$, Cohen's $d = 0.091$].

Safety, Tolerability, and Blinding

In total, 140 tDCS sessions were performed with no serious adverse events and sessions were well-tolerated. Reported sensations included itching (89%; 44% mild, 48% moderate, 8% severe), tingling (68%; 79% mild, 5% moderate, 16% severe), and burning (54%; 73% mild, 27% moderate), which did not differ by group. Seven participants reported a mild headache lasting for the first few minutes of stimulation and five participants felt mildly

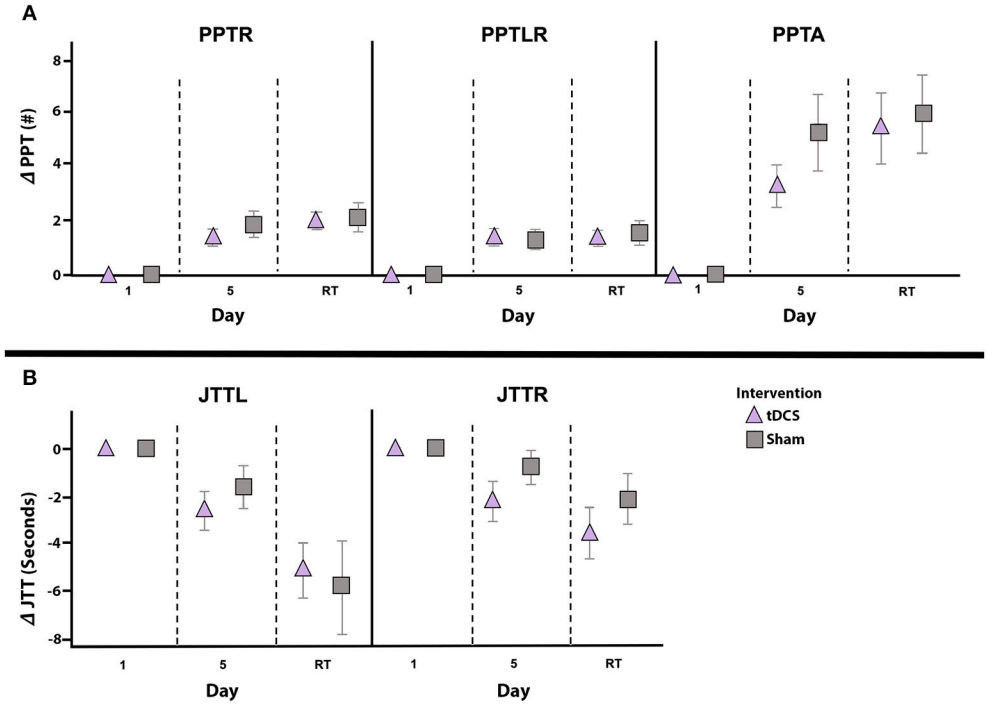

FIGURE 5 | Change in secondary outcomes measures by intervention group. Mean daily score change from baseline (y-axes) for PPT_R, PPT_{LR}, and PPT_A **(A)** as well as JTT_L and JTT_R **(B)**, shown for the active (triangle) and sham (squares) groups. Error bars represent standard error. Independent of intervention group all scores significantly improved from baseline (day 1) to post-intervention testing on day 5, and from baseline to retention testing (RT) 6-weeks post-intervention. No between group differences in performance on any secondary measures were noted at any timepoint. Note that negative JTT scores indicate improved performance (i.e., reduction in completion time) and positive JTT scores indicate worse performance (i.e., increased time required to complete tasks). PPT_L, Purdue Pegboard Test left-hand; PPT_R, Purdue Pegboard Test right-hand; PPT_{LR}, Purdue Pegboard Test bimanual; PPT_A, Purdue Pegboard Test assembly; JTT_L, Jebsen-Taylor Test left-hand; JTT_R, Jebsen-Taylor Test right-hand.

nauseated in a single session. TDCS tolerability rankings, on an 8-point scale, were similar for the active (4.1 ± 1.1) and sham groups (4.1 ± 1.2; p=0.974) and were comparable to watching TV (2.6 ± 0.9) or a long car ride (5.1 ± 1.3). Participants were unable to predict their treatment group (44% accuracy, 50% indicates chance).

DISCUSSION

The current trial is the first to examine the therapeutic efficacy of tDCS on motor learning in children with DCD. Independent of intervention, all children's motor performance improved over the 5 training days and skill improvements were retained for 6 weeks. Contrary to our hypothesis, excitatory stimulation of the right primary motor cortex did not enhance motor learning.

The research literature suggests that poor motor performance in children with DCD may be associated with deficits in motor learning (Bo and Lee, 2013; Biotteau et al., 2016a). However, research concerning the presence of motor learning deficits in DCD is inconsistent, with some studies reporting limited skill improvement following practice (Kagerer et al., 2004; Gheysen et al., 2011; Zwicker et al., 2011) and others reporting positive effects of practice (Ferguson et al., 2013; Lejeune et al., 2013; Mombarg et al., 2013; Smits-Engelsman et al., 2015). Studies supporting the latter emphasize that children with DCD are able to acquire motor skills, though they may display slower rates of motor learning, requiring more intensive practice to reach desired levels of motor competence. In the current trial, fine motor performance of the non-dominant limb improved significantly with practice, independent of intervention. This finding supports the capacity of children with DCD to learn novel motor skills.

Motor learning involves both online and offline processes. Online learning includes skill gains obtained during active training, whereas offline learning includes gains occurring between training sessions (i.e., consolidation). Within both groups, the majority of motor learning took place online. This suggests that children with DCD may show less efficient offline motor learning, or consolidation, which has been previously suggested in the DCD literature (Zwicker et al., 2011) and warrants further study.

Motor skill retention and transfer to untrained tasks are also features of successful motor learning (Muratori et al., 2013). We show no evidence of skill decay in either group between the final training day and retention testing at 6-weeks. Moreover, motor skill improvements were not restricted to the trained hand or task as improvements on all secondary motor

outcomes were observed. Learning effects were generalized to the untrained dominant hand. Taken together, these results suggest that children with DCD display intact motor skill acquisition, adaptation, retention, and transfer following practice.

Contrary to previous evidence of tDCS enhanced motor learning in typically developing children (Ciechanski and Kirton, 2017; Cole et al., 2018) and children with motor impairment (i.e., cerebral palsy) (Grohs et al., 2019), tDCS did not enhance the rate of motor learning in children with DCD relative to practice alone. The limited efficacy of tDCS could be reflective of stimulation parameters including cortical target (Thibaut et al., 2017) or montage (i.e., anode and cathode arrangement) (Woods and Martin, 2016). Although M1 is a common target to modulate motor learning due its direct role in motor production (Todorov, 2003), other structures may be better suited to the DCD population. For instance, dysfunction in cerebellar networks has commonly been identified in DCD (Biotteau et al., 2016a). Given the role of the cerebellum in motor control and learning (Manto et al., 2012), as well as positive findings from trials implementing cerebellar tDCS for motor impairment (Celnik, 2015), it may be a promising target in DCD.

Regarding montage, different anode/cathode placement uniquely modulates cortical excitability. Anodal tDCS involves placement of the anode over a target region and generally produces excitatory effects within the cortex, whereas in cathodal tDCS the cathode is placed over the target region producing an overall inhibitory effect. Although anodal stimulation was chosen here based on previous evidence (Cole et al., 2018), cathodal stimulation has also been shown to enhance motor learning in children (Ciechanski and Kirton, 2017). Neurophysiological research has reported reduced interhemispheric inhibition of M1 activity in DCD (He et al., 2018). It is, therefore, possible that inhibiting cortical activity via cathodal stimulation may produce favorable outcomes in children with DCD. Future studies that characterize baseline cortical excitability and neurometabolites, using techniques such as transcranial magnetic stimulation (TMS) and magnetic resonance spectroscopy (MRS), could help in refining application (i.e., stimulation intensity, montage, and target).

It is also possible that there are no effects of tDCS in children with DCD. However, given this is the first study to examine effects of neuromodulation in children with DCD, future studies using well-supported protocols that target different cortical regions and/or examine different montages are highly encouraged. Finally, given that tDCS enhances motor learning via facilitating endogenous neuroplastic mechanisms, the absence of response to tDCS observed here could also suggest disordered neuroplastic mechanisms in individuals with DCD. Future studies utilizing techniques such as TMS could help to elucidate plasticity mechanisms in DCD.

Limitations

Our sample size calculation estimated that 16 participants per group would provide us with 90% power to detect group differences; however, our final groups consisted of 14 participants. As a result, our sample size may have decreased our ability to detect potential group differences, or efficacy, and may have limited the generalizability of our findings. There was also a high degree of variability in performance on our outcome measures, which may have decreased our ability to detect group differences given the sample size. Thirteen participants were on medications that influence neurotransmitter systems and could have impacted tDCS efficacy (McLaren et al., 2018). Another limitation was the demanding nature of the trial, which required children to maintain their attention and motivation over 5 consecutive days. This may have been difficult, particularly for our sample with co-occurring attention, learning and anxiety disorders, and may have contributed to performance variability. Co-morbidities and the fact that children with DCD are a heterogeneous group who display many different types of motor skill deficits, constitutes a significant challenge for future trials.

CONCLUSION

Children with DCD demonstrated motor learning as measured by the PPT with retention of acquired skill at 6-weeks. The addition of motor cortex tDCS during training did not enhance motor learning, as seen in other populations. Procedures were well-tolerated and appear safe. Before conclusions can be made regarding the efficacy of tDCS in DCD, additional carefully designed trials with reproducible results are required. Establishment of an optimal tDCS protocol in DCD is essential, including stimulation target and montage.

DATA AVAILABILITY STATEMENT

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

ETHICS STATEMENT

This study, involving human participants, was reviewed and approved by the University of Calgary Conjoint Health Research Ethics Board (Ethics ID: REB10-0183). Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin. Written informed consent was obtained from the minor(s)' legal guardian/next of kin for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

MG, AK, and DD conceptualized and designed the study. MG collected, analyzed and interpreted the data, and took the lead in writing the manuscript. BC assisted with the data analysis. DD and AK secured grant funding for this study, provided, administrative, technical, material support, and supervised the project. BC, AK, and DD provided critical feedback and helped to shape the final manuscript. All authors contributed to the article and approved the submitted version.

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BRIEF RESEARCH REPORT
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Eliciting End-State Comfort Planning in Children With and Without Developmental Coordination Disorder Using a Hammer Task: A Pilot Study

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The end-state comfort (ESC) effect refers to the consistent tendency of healthy adults to end their movements in a comfortable end posture. In children with and without developmental coordination disorder (DCD), the results of studies focusing on ESC planning have been inconclusive, which is likely to be due to differences in task constraints. The present pilot study focused on the question whether children with and without DCD were able to change their planning strategy and were more likely to plan for ESC when demanded by complex object manipulations at the end of a task. To this end, we examined ESC planning in 18 children with and without DCD (aged 5–11 years) using the previously used sword-task and the newly developed hammer-task. In the sword-task, children had to insert a sword in a wooden block, which could be relatively easily completed with an uncomfortable end-posture. In the hammer-task, children had to strike down a nail in a wooden pounding bench, which required additional force and speed demands, making it relatively difficult to complete the movement with an uncomfortable end-posture. In line with our hypothesis, the results demonstrated that children with and without DCD were more likely to plan for ESC on the hammer-task compared with the sword-task. Thus, while children with and without DCD show inconsistent ESC planning on many previously used tasks, the present pilot study shows that many of them are able to take into account the end-state of their movements if demanded by task constraints.

Keywords: motor planning, end-state comfort, developmental coordination disorder, children, task constraints

INTRODUCTION

When selecting a grip in order to perform a grasping movement, several strategies can be used. Healthy adults show a consistent tendency to end movements in a comfortable posture, even if this comes at the expense of an uncomfortable start-posture, which is called the *end-state comfort (ESC) effect* (Rosenbaum et al., 1990). The results of studies in children are, however, inconclusive with regard to the onset of this ESC effect during development evidenced by varying percentages of ESC planning across varying age groups (Wunsch et al., 2013, for a review).

A group of children in which ESC planning appears to be comprised, are those with a developmental coordination disorder (DCD; Adams et al., 2014, for a review). While the majority of studies found that children with DCD are less likely to plan for ESC compared to typically developing (TD) children (e.g., van Swieten et al., 2010; Wilmut and Byrne, 2014a; Fuelscher et al., 2016; Adams et al., 2017), mixed results regarding the differences between children with DCD and TD children are also reported (e.g., Smyth and Mason, 1997; Noten et al., 2014). These equivocal results, both among TD children and between TD children and children with DCD, seem to be due to differences in task constraints that are evident in the different studies (Jongbloed-Pereboom et al., 2016; Bhoyroo et al., 2019). This has led to the discussion as to whether optimizing ESC is the preferred strategy for children on all tasks (e.g., Wilmut and Byrne, 2014b; Krajenbrink et al., 2020). The role of task constraints has been recently highlighted in a multi-component account of motor skill performance and development in children with DCD (Blank et al., 2019). Central tenet of this account is the mutual interaction between individual, environmental, and task constraints that determines the resulting behavior. With regard to ESC planning, depending on the biomechanical costs of the start- and ensuing end-posture, children may use alternative strategies to achieve a task goal. Following this reasoning, children are expected to change their strategy to plan for ESC if demanded by complex object manipulations at the end of the task compared to simple object manipulations. In the present pilot study, we examined this expectation using two tasks that required a similar start-posture but differed with regard to the task demands.

One of the tasks on which performance of both TD children and children with DCD has been described as relatively poor with regard to ESC is the sword-task (Craje et al., 2010). In this task, children are asked to pick up a wooden sword and to subsequently stick it into a tight-fitting hole in a wooden block. For the so-called critical trials, the sword needs to be rotated first before the blade can be inserted into the wooden chest. Jongbloed-Pereboom et al. (2013) examined performance across age on the sword-task among 3–10 years old TD children and found that the percentage of ESC on critical trials increased from about 20% for the youngest age group to about 60% for the oldest age group. When compared to the overturned cup task (i.e., turning an upside-down cup upright) and the bar transport task (i.e., placing a horizontal bar in a target standard), percentages of ESC on the sword-task were the lowest, both for TD children, adolescents, and even adults (Jongbloed-Pereboom et al., 2016). Adams et al. (2016) and Adams et al. (2017) assessed the sword-task among a group of 6–11 years old TD children and children with DCD and found lower percentages of ESC on the critical trials in the DCD group compared with their TD peers. This decreased tendency to plan for ESC has been interpreted as either a deficit or a developmental delay in motor planning in children with DCD.

However, these relatively low ESC percentages on the sword-task can be understood if we take a closer look at the way the task is set up. The sword-task can be relatively easily completed with a comfortable start-posture that results in an uncomfortable end-posture. At the same time, however, the postural demands of the initial uncomfortable start-posture that is necessary in order to achieve ESC are relatively high (Jongbloed-Pereboom et al., 2016). Thus, based on the relative (dis)comfort of the start- and end-posture, children may as well use the easiest initial grip to complete the task goal. This could be particularly true for children with DCD as the costs related to a biomechanically uncomfortable start-posture may be higher for them due to their motor difficulties (Wilmut and Byrne, 2014a). In other words, striving for ESC may not always be the most efficient strategy. Indeed, studies that focused on varying strategies used by children to solve motor planning tasks, found that next to ESC planning, children with and without DCD also use planning strategies based on start-state comfort, minimal initial rotation, or repetition of the previous movement (Wilmut and Byrne, 2014b; Bhoyroo et al., 2018).

It is, therefore, interesting to examine whether children may change their strategy to strive for ESC if the relative weight of the costs and benefits of an uncomfortable start- or end-posture change. It is assumed that by ending in a comfortable posture with the joints in a mid-range position, subsequent object manipulations can be performed with more precision (Short and Cauraugh, 1999). Thus, if the precision demands of the to-be-performed manipulation at the end of the task are higher, it is expected that it is more beneficial to end the movement in a comfortable posture in order to complete the required task goal. Following this reasoning, we developed a hammer-task in which children needed to pick up a hammer to strike down a nail in a wooden pounding bench (**Figure 1**). A hammering task has been used before to measure ESC in children (Comalli et al., 2016). The postural demands of the start of the movement are equal to the sword-task as well as the demanded end point precision. Importantly, however, the hammer-task incorporates additional complexity since sufficient force and speed needs to be exerted while hammering. This combination of precision, force, and speed demands for the hammer-task was hypothesized to lead to a higher degree of ESC planning compared with the sword-task, as a non-ESC

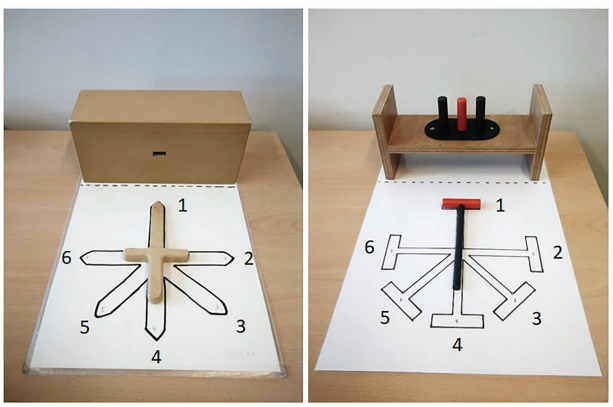

FIGURE 1 | Sword-task (left) and hammer-task (right) with the sword/hammer in orientation 1.

planning strategy would result in poor precision and power when striking the nail. In addition, we hypothesized that this would be particularly true for children with DCD compared with TD children.

MATERIALS AND METHODS

Participants

Participants were nine children (six boys and three girls) with DCD and nine gender and age-matched (within 8 months, except for one child that was matched within 14 months) controls that also participated in a larger study on motor planning as reported in Krajenbrink et al., in prep. Children were 5–11 years old ($M = 8$ y0 m, $SD = 2$ y0 m). The children with DCD met the following inclusion criteria based on the DSM-V criteria: a Movement Assessment Battery for Children 2 (Henderson et al., 2007) total score ≤16th percentile or component score ≤5th percentile (criterion A), treated or have been treated for a motor coordination problem by a pediatric physical therapist and interference of the motor difficulties with daily activities, measured using two parent questionnaires, namely the Developmental Coordination Disorder Questionnaire (DCD-Q, Dutch translation; Schoemaker et al., 2008) and the DCDDaily-Q (van der Linde et al., 2015; criterion B), early onset of symptoms (criterion C); and no report of any cognitive impairment, visual impairment, or neurological deficit that would explain the child's motor difficulties (criterion D). Comorbid disorders were Attention Deficit Hyperactivity Disorder (2) and Attention Deficit Disorder (1) as reported by parents. All TD children had a mABC-2 score >16th percentile. In addition, parents completed an ADHD-questionnaire as a descriptive measure of ADHD symptoms (AVL; Scholte and van der Ploeg, 2004). Child characteristics including the questionnaire scores are presented in **Table 1**. The study was approved by the Ethics Committee of the Faculty of Social Sciences at Radboud University (ECSW-2019-122).

Materials

The previously administered sword-task (Craje et al., 2010) and the newly developed hammer-task were used as a measure of second-order motor planning. Both tasks are depicted in **Figure 1**. In the sword-task (left picture), children were asked to pick up the sword and to subsequently stick it into the hole of the wooden block. In each trial, the experimenter placed the sword on the template board in one of the six sword orientations. In the hammer-task (right picture), children were asked to pick up the hammer and to subsequently strike down the middle nail in the wooden pounding bench. Here, each trial, the hammer was placed on a similar template board with six hammer orientations. The other two nails were added to increase precision demands. As can be seen in **Figure 1**, for both tasks, two orientations were critical orientations (i.e., orientations 2 and 3 for right-handed children and orientations 5 and 6 for left-handed children) for which children had to sacrifice comfort of their start grip in order to end the task in a comfortable position (i.e., critical trials). The other four orientations served as control orientations for which a comfortable start grip resulted in a comfortable end position (i.e., non-critical trials). For both tasks, each orientation was repeated three times in a pseudo-random order with all six rotations appearing every six trials, resulting in a total of 18 trials per task. A score of 1 (i.e., action ended in an ulnar deviation, with the thumb toward the blade/hammerhead) or 0 (i.e., action ended in a radial deviation, with the thumb away from the blade/hammerhead) was assigned for each trial for each child. The proportion comfortable end postures were the outcome measure.

TABLE 1 | Child characteristics for DCD group and TD group.

	DCD group	TD group
Age in years (*SD*)	8 y,0 m (2 y,1 m)	8 y,0 m (2 y,0 m)
Sex (male/female)	6/3	6/3
Dominant hand (left/right)	0/9	3/6
mABC-2 *M* (*SD*)	2.30 (2.93)	50.00 (19.92)
AVL *M* (*SD*)	28.83 (11.58)	13.17 (8.61)
DCD-Q *M* (*SD*)	28.56 (6.84)	64.11 (9.58)
DCDDaily-Q participation *M* (*SD*)	47.83 (7.28)	34.56 (3.64)
DCDDaily-Q activities *M* (*SD*)	52.22 (6.76)	31.89 (6.88)
DCDDaily-Q learning *M* (*SD*)	18.11 (3.26)	0.89 (1.69)

Procedure

The hammer-task was appended to the study procedure of a larger data collection reported in Krajenbrink et al., in prep. As part of this larger data collection, three second-order motor planning tasks were examined in counter-balanced order, one of which being the sword-task. For the final 9 children that were included in the DCD group and the final 29 children in the TD group, the hammer-task was added at the end of the protocol. The data from these children with DCD and a gender and age-matched selection of 9 TD children (children were selected randomly in case of multiple options) were included in the present study. Children were seated and could comfortably reach the experimental materials. Before examining the second-order motor planning tasks, hand preference was determined by asking children to write their name down on the session form. For most TD children, data collection took place at their school and for most children with DCD, data collection to place at their home. Completing the sword-task and the hammer-task took about 5 min in total.

Data Analysis

In order to examine whether performance differed between the sword-task and hammer-task for children with DCD and TD children, a generalized linear mixed-effects model with a binomial link function was performed using the glmer function of the lme4 package (Bates et al., 2015) in R (R Core Team, 2020). It was decided to use this analysis instead of a more traditional approach as it is most suitable for binomial data. In the model, performance (proportion of ESC on the critical orientations, included as the number of critical trials ending in ESC and the number of critical trials not ending in ESC) was predicted as a function of the fixed effect of group (DCD or TD),

the fixed effect of task (sword or hammer), as well as the interaction thereof. A random intercept for participant was included in order to control for individual variances across measurements. The model ran without warnings and provided a good fit of the data. Model diagnostic plots (i.e., a distribution of the residuals and a plot of the residuals as a function of the fitted values) yielded no indication of violations of the assumptions of normality, homoscedasticity, and linearity. Finally, there were no standardized residuals with values below −2.0 or above 2.0. Values of p are based on confidence intervals that were calculated with the confint function using bootstrap resampling. The beta coefficients that resulted from the model were converted into odds ratios (ORs). It should be noted here that the results of this model must be interpreted with caution as the small sample size has inherent limitation with respect to the reliability of the estimates and the generalizability of the results.

RESULTS

Descriptive statistics of both the critical and the non-critical trials of the sword-task and the hammer-task are represented in **Table 2**. The main variable of interest was the proportion of ESC on the critical trials of the sword-task and the hammer-task, which is represented in **Figure 2** for children with DCD and TD children separately. Fourteen children performed better on the critical trials of the hammer-task compared with the sword-task. The other four children performed equal on both tasks, with three of them having the maximum score on both tasks. When looking at both groups separately, on the sword-task, six TD children ended half or more of the trials in ESC, but the other three TD children ended none of the trials in ESC. In the DCD group, only one child ended half or more of the trials in ESC and six children ended none of the trials in ESC. On the hammer-task, seven TD children ended all trials in ESC, but the other two children still ended less than half of the trials in ESC. Two children in the DCD group ended all of the trials in ESC and another four children ended half or more of the trials in ESC. Three children with DCD ended less than half of the trials in ESC.

Results of the generalized linear mixed-effects model with a binomial link function showed a significant main effect of task, indicating that the average proportion of ESC was higher on the hammer-task compared to the sword-task, $OR = 0.02$, $b = -3.78$, $SE = 0.88$, $z = -4.32$, $p < 0.05$, 95% CI [−6.42, −2.16]. The main effect of group, $OR = 14.76$, $b = 2.69$, $SE = 1.85$, $z = 1.45$, $p > 0.05$, 95% CI [−0.48, 12.51], and the interaction between task and group, $OR = 0.75$, $b = -0.29$, $SE = 1.51$, $z = -0.19$, $p > 0.05$, 95% CI [−7.82, 2.60], were not statistically significant. This indicates that the difference in performance on both tasks was not statistically different between children with DCD and TD children.

TABLE 2 | Proportion of trials ending in ESC for the critical and non-critical orientations for the DCD group and TD group.

		DCD group	TD group
Sword-task	Critical trials *M* (*SD*)	0.17 (0.33)	0.52 (0.43)
	Non-critical trials *M* (*SD*)	0.89 (0.10)	0.98 (0.04)
Hammer-task	Critical trials *M* (*SD*)	0.59 (0.30)	0.80 (0.41)
	Non-critical trials *M* (*SD*)	0.98 (0.04)	1.0 (0.00)

DISCUSSION

The aim of this pilot study was to examine whether children with and without DCD are more likely to plan for ESC when demanded by complex object manipulations at the end of the task. To this end, children with and without DCD performed the newly developed hammer-task after completing the previously used sword-task. In contrast to the sword-task, completing the hammer-task requires sufficient force and speed to hammer the nail down, making it more difficult to complete the goal of the task with an uncomfortable end-posture. We found that both children with DCD and TD children were more likely to strive for ESC on the hammer-task compared with the sword-task. Below, we will discuss these results in more detail.

In line with our expectation, we found that almost all children were more likely to sacrifice comfort of the start-posture and end the movement in a comfortable posture when completing the hammer-task as compared with the sword-task. Clearly, the additional force and speed demands in the hammer-task elicited more planning for ESC. This supports the multi-component account proposed by Blank et al. (2019) which stresses the role of task constraints, in interaction with environmental and individual constraints, to explain the behavior of children with DCD. In addition, previous studies on ESC planning also suggested that performance is task dependent (Knudsen et al., 2012; Jongbloed-Pereboom et al., 2016; Bhoyroo et al., 2019). In contrast to these previous studies, however, in our study, an increase in task demands was associated with increased percentages of ESC. In the hammer-task, additional force and speed demands led to a higher use of the ESC optimization strategy. Thus, it seems that the relative costs and benefits of an uncomfortable start- and end-posture determine what strategy children use. Although we assume that the benefits of ending in ESC are higher in the hammer-task compared with the sword-task, our paradigms did not provide an objective measure to support this claim. Future research is, therefore, warranted in which a measure that reflects the efficiency of task completion is included (e.g., accuracy, speed, or force) in order to test whether a comfortable end-posture from an adult perspective, is also beneficial for children with and without DCD.

Collectively, the results suggest that if children, both TD and children with DCD, fail to show a high percentage of ESC in a certain task, this does not necessarily mean that they are unable to take into account the end-state of their movement when first planning their movements. Rather, they employ alternative planning strategies (Wilmut and Byrne, 2014a,b). In line with this argumentation, previous studies that used an octagon task in which children had to rotate a knob,

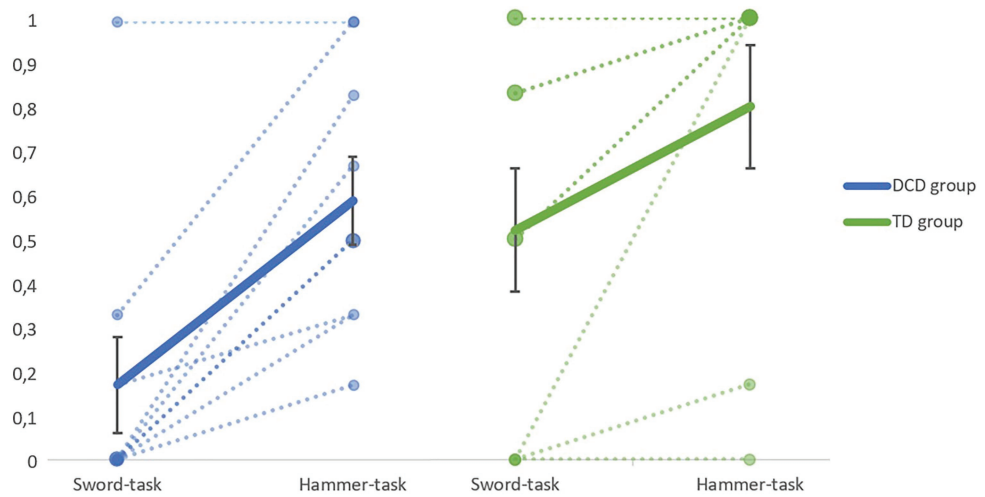

FIGURE 2 | Proportion of end-state comfort (ESC) on the critical trials of the sword-task and hammer-task, developmental coordination disorder (DCD) group on the left, and typically developing (TD) group on the right. Dotted lines represent individual data, with the diameter of the end caps being proportional to the number of cases. The fat lines represent the group average and error bars represent SEs.

found that children used varying strategies including optimization of start-state comfort, reduction of initial rotation, and repetition of the previous movement (Wilmut and Byrne, 2014b). Additional research showed that children with DCD were more likely to use a strategy with minimal initial rotation of the hand and arm if compared with TD children (van Swieten et al., 2010; Wilmut and Byrne, 2014a). It was already mentioned in these studies that this pattern of results does not necessarily imply a lack of motor planning ability in children with DCD, but rather that these children may plan their grasps commensurate with their motor ability (van Swieten et al., 2010; Wilmut and Byrne, 2014a). Our results extend this argument by showing that both, children with DCD and TD children, are more likely to plan for ESC if task demands are more complex, as in the case of the present hammer-task.

We did not find statistically significant differences between children with DCD and TD children. At the individual level, however, the pattern of results was in line with our hypothesis that the difference in performance between both groups would be smaller on the hammer-task than on the sword-task. On the sword-task, six TD children ended more than half of the trials in a comfortable posture, while this was the case for only one child with DCD. On the hammer-task, seven TD children completed all trials in a comfortable posture, but two children ended less than half of the trials in a comfortable posture. For the children with DCD, two children completed all trials in a comfortable posture and there were three children that ended less than half of the trials in a comfortable posture. The lack of statistically significant differences between both groups is likely due to the small sample size of the present study which results in low power. This is supported by the results of the larger study including 26 children with DCD and 26 matched controls, where we did find that TD children demonstrated a higher percentage of ESC on the sword-task than children with DCD (Krajenbrink et al., in prep). The findings in the present study warrant fully powered follow-up research to test whether the pattern of results can be replicated in larger groups of children with DCD and TD children in order to draw strong conclusions. In addition, the order of the tasks should be counter-balanced to more systematically assess the probable confounding effects of practice and attention.

In sum, our small scale pilot study is the first to clearly show that both, children with DCD and TD children, are more likely to plan for ESC when high end-precision demands are combined with speed and force demands, as is the case in the hammer-task. These additional task demands were considered to increase the benefits to use an uncomfortable start-posture in order to end the movement in a comfortable posture. Indeed, our results indicate that while children with and without DCD plan their movements less consistently than adults on many previously used motor planning tasks, they are able to take into account the end-state of their movement and plan for ESC if demanded by task constraints.

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 human participants were reviewed and approved by the Ethics Committee Social Science of the Radboud University. Written informed consent to

participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

All authors have contributed to the work in a meaningful way. HK, JL, and BS designed the experiment. HK conducted the experiment, analyzed the data, and wrote a first draft of the manuscript. JL and BS critically reviewed the manuscript. All authors agree with publication of the final version of the manuscript.

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frontiers in Human Neuroscience

ORIGINAL RESEARCH
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Driving Skills of Individuals With and Without Developmental Coordination Disorder (DCD/Dyspraxia)

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Learning to drive is a significant event for the transition to adulthood and delay or avoidance may have social, practical, and psychological implications. For those with Developmental Coordination Disorder (DCD/Dyspraxia), driving presents a considerable challenge, and the literature shows that there are differences in driving ability between individuals with and without DCD. The aim of the current research is to further our understanding of the mechanisms underlying the driving experiences of individuals with DCD. Nineteen participants with DCD (10 drivers and 9 non-drivers) and 36 controls (17 drivers and 19 non-drivers) aged 18–57 years took part in this study. Participants completed standardized tests, questionnaires and a driving simulation task designed to measure speed, road positioning, and rate of change of steering in three conditions with increasing perceptual complexity. Results indicate that behaviors for all participants changed as the perceptual demands of the task increased. However, drivers with DCD were more affected than all other groups, driving more slowly, and driving further to the right. These findings illustrate how the impact of both internal and external constraints negatively affect the success of the driving task for individuals with DCD compared to their TD peers.

Keywords: developmental coordination disorder/dyspraxia, driving, motor coordination, perception, experience, real-world skills

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INTRODUCTION

Driving is a core skill necessary for access to many activities of daily living. For example, drivers have independence and flexibility, increasing access to opportunities for education, employment, and leisure compared to non-drivers (Barkley, 2004). Furthermore, learning to drive is a significant event for the transition to adulthood and delay or avoidance may have social, practical and psychological implications (Barkley, 2004; Kirby et al., 2011). However, driving is not a simple task. Drivers must have good spatial perception (de Oliveira and Wann, 2011) and well-coordinated visuomotor control (Marple-Horvat et al., 2005) to be able to manage multiple stimuli simultaneously and react quickly in an emergency when completing the driving task (Reger et al., 2004).

In order to interpret environmental constraints and elicit skilled actions (e.g., to steer a car around obstacles in the pathway), humans are heavily dependent upon accurate perception (Hulme and Snowling, 2009), particularly vision and proprioception. Additionally, cognitive input is

necessary to process the feedback and feedforward information in response to the environment and to allow predictions about the expected consequence of a motor command (Wolpert et al., 2003). Research by Newell (1986) considers how constraints within the individual (flexibility, strength, motor, or sensory systems), the task (moving/stationary, accuracy/speed, and seated/standing) and the environment (environmental stability, lighting, and surface type) affect behavior. Thus, movement is viewed as an emergent process arising from dynamic interaction between the individual, the task and the environment. When all these systems integrate efficiently with motor control mechanisms, the individual can adapt to dynamic environmental constraints.

However, whilst driving the ability to accurately process sensory information from the environment may be compromised as this information has to be perceived at much higher speeds than humans are designed to travel. Additionally, cars are being designed with ever-more luxurious interiors such as improved soundproofing (to reduce external noise) and improved suspension (to increase smoothness of the ride, Davies, 2017). It is therefore possible that whilst these advances certainly make the driving experience more pleasurable, they may negatively affect the ability to use sensory information effectively and make informed judgments about the external environment. This could have a particular impact on individuals with sensorimotor difficulties, as is outlined below.

Driving With Developmental Coordination Disorder

Developmental Coordination Disorder (DCD/Dyspraxia) is a neurodevelopmental movement disorder that affects the development of motor control and coordination but is unexplained by a neurological condition. Prevalence rates are estimated to be around 5% of children aged 5–11 years (American Psychiatric Association [APA], 2013) and the motor impairments continue to negatively impact everyday activities into adulthood (Purcell et al., 2015). Individuals with DCD are known to have difficulties with sensori-perceptual function (visual form detection, motion detection, visuospatial processing, and tactile perception), forward modeling (Wilson et al., 2013), perception-action couplings (Wann et al., 1998), and learning new motor tasks (Missiuna et al., 2008). It is therefore unsurprising that learning to drive has been identified as an area of particular difficulty for this population (Cousins and Smyth, 2003; Missiuna et al., 2008; Kirby et al., 2011; Blank et al., 2019).

There is a paucity of research investigating the effects of sensori-perceptual processing on environmental interactions for individuals with DCD. However, research investigating behaviors of pedestrians show that adults with DCD are slower than controls when negotiating obstacles in their pathway (Cousins and Smyth, 2003; Wilmut et al., 2015; Gentle et al., 2016). The additional time taken to adjust to environmental constraints could indicate slower perceptuo-motor integration, i.e., individuals with DCD need more time to perceive relevant information and adopt the appropriate motor actions to avoid collisions (Wilmut et al., 2015; Gentle et al., 2016). Indeed, a real-time study investigating road crossing skills found that children with DCD did not allow enough time to safely cross the road. The authors argue if this outcome were translated to a real-world environment, a collision would have occurred (Purcell et al., 2017). These findings reflect anecdotal evidence of collisions with obstacles for pedestrians with DCD (Geuze, 2007) and have important implications for driving, where the time to process relevant information is shorter due to the higher speed of travel. Another observation of pedestrian behaviors relevant for driving is that individuals with DCD turn more often and to a greater degree than their typical counterparts when passing through a narrow aperture (Wilmut et al., 2015). The authors argue that these adaptations accommodate motor control difficulties, and this behavior ensures a wider margin when passing through the aperture thus avoiding a collision. Exactly how this adaptation transfers to a real-world driving task where narrow roadways are a common occurrence, has yet to be investigated for individuals with DCD.

Driving research shows that fewer individuals with DCD learn to drive compared to their typically developing (TD) peers (Kirby et al., 2011), and those who do succeed take longer to pass their test (Missiuna et al., 2008). Individuals with DCD also perceive themselves to be less competent at driving and report particular difficulty with more complex skills, such as parking or reversing (Missiuna et al., 2008; Kirby et al., 2011). Research using an automatic car simulator found that individuals with DCD used significantly more steering adjustments when maintaining a straight course at a controlled steady speed, used twice as many steering adjustments as necessary when negotiating a bend and had slower responses to pedestrians in their pathway than their age-matched TD peers (de Oliveira and Wann, 2011, 2012). de Oliveira and Wann (2012) further identified that those with DCD showed a larger variance in heading when turning bends but not when driving along straight roads compared to their TD peers. The authors explain their findings in terms of deficient mapping between visual information and steering actions (Fajen, 2008). Finally, de Oliveira et al. (2014) used a steering task to investigate the use of advanced visual information. They found that, whilst TD individuals showed a linear improvement as duration of visual information increased, individuals with DCD showed behaviors described as U-shaped, where optimal performance occurred with 750 ms of advance information. However, studies from the de Oliveira group collected data from young (mean age 17.4 years, 2011; 18.6 years, 2012; and 19 years 2014) and inexperienced drivers. Furthermore, the driving simulator used by de Oliveira and Wann comprised a simulator chair and steering wheel which is perhaps more indicative of a computer game than a real car. It is unclear whether older drivers would show similar behavior in a more ecologically valid simulator.

The aims of the current study were to extend the small body of previous research by investigating competencies of participants with and without DCD when negotiating everyday driving scenarios in conditions not previously tested. The use of a real car driving simulator (compared to the use of a chair, steering wheel and pedals used by de Oliveira and Wann, 2011, 2012) enhances ecological validity and the generalizability of the findings.

Based on the previous research we aimed to design an ecologically valid set up, using frequently experienced driving settings, to address three research questions: (1) Do drivers and non-drivers (defined by driving test status) with and without DCD behave differently when processing dynamic sensory information in progressively complex environments? (2) How do drivers and non-drivers with and without DCD negotiate narrow apertures in a car of fixed width? (3) Do individuals with DCD collide more with obstacles in the pathway compared to controls?

This experiment consisted of three conditions comparing speed, road positioning and steering adjustments between the groups and in scenarios of increasing perceptual load. *Condition 1* was a low load condition as drivers negotiated a clear, straight road; *Condition 2* increased the perceptual load as participants negotiated a clear, straight road, with the addition of stationary objects in the pathway; *Condition 3* increased the perceptual demands further as participants negotiated a clear, straight road, with the addition of an oncoming moving vehicle.

Research question 1 was addressed through all three conditions. We hypothesized that across all conditions, compared to typical drivers, individuals with DCD would drive more slowly, have less appropriate road positioning and use more steering adjustments and compared to typical drivers, typical non-drivers would drive more slowly throughout. Furthermore, we predicted that these differences would be more pronounced in conditions with higher perceptual load. Research Question 2 was examined in Condition 1 as participants drove along a road with cars parked either side. We hypothesized that, compared to typical drivers, non-drivers,and individuals with DCD would drive more slowly, have less appropriate road positioning and use more steering adjustments during this condition. Finally, research Question 3 was investigated in Conditions 1 and 2. We hypothesized that, compared to typical controls, participants with DCD would have more collisions during the drive.

METHOD

This research was approved by the University of Surrey Research Ethics Committee and informed consent was obtained from all participants.

Participants

Table 1 presents demographic information for the current sample. Nineteen individuals with DCD (10 drivers and 9 non-drivers; mean age: 26.5 years) and 36 controls (17 drivers and 19 non-drivers mean age: 21 years) participated in this study. Participants with a valid driving license were assigned to the Drivers group, participants without a valid drivers license were assigned to the Non-drivers group. All participants with DCD were recruited in line with the DSM-5 (American Psychiatric Association [APA], 2013) and the United Kingdom guidelines for assessment of adults with DCD (Barnett et al., 2015). Participants with DCD were recruited through a charitable foundation which supports individuals with DCD and contacts known to the researchers. Additionally, an advertisement was placed in the university setting to recruit those individuals with a diagnosis of DCD who have not taken part in previous research with the research team as well as TD controls.

Materials

Measures to Assess DCD

A range of assessments were used to ensure that the four DSM-5 diagnostic criteria for DCD were met. To assess coordinated motor skills (Criterion A) participants completed the Movement Assessment Battery for Children (MABC-2, Henderson et al., 2007), which is a standardized measure of motor skill suitable for ages 3–16 years. Due to the lack of appropriate motor assessments in the United Kingdom for adults with DCD, the 11–16-year age band was used, reflecting common practice in DCD research with adults (Cousins and Smyth, 2003; Cantell et al., 2008; Purcell et al., 2015). Individuals scoring below the 5th percentile demonstrate severe motor difficulties, and those scoring at or below the 15th percentile demonstrate moderate motor difficulties.

The Adult DCD checklist (ADC, Kirby et al., 2010) was used to assess Criterion B (motor deficit significantly interferes with daily living activities). The ADC is a standardized screening tool for those over the age of 16 to aid identification of DCD in adults. A score of at least 17 in section 1 of the ADC and a total score of at least 56 is required to meet DSM-5 criteria and demonstrates a significant effect of motor difficulties on everyday life which has been present since childhood (Criterion C). Participants were asked whether they had any visual impairment or neurological condition that would explain any movement difficulties (Criterion D). Participants were also tested using the Wechsler Adult Intelligence Scales (WAIS-IV, Wechsler, 2010) to provide a measure of verbal IQ. Participants were assigned to the DCD group if they scored above the cut-offs identified above on the ADC, below the 15th percentile on the MABC-2, had no visual impairment or neurological condition, and a verbal IQ score in the typical range. Participants were assigned to the control group if they scored within the typical range on the ADC, MABC-2, and verbal IQ, with no visual impairment or neurological condition.

Finally, participants were asked to complete the Conners Adult ADHD Scales, Short version (CAARS-S:SV, Conners et al., 2000). The CAARS-S:SV consists of 30 statements which relate to symptoms or behaviors associated with Attention Deficit-Hyperactivity Disorder (ADHD), which often co-occurs with DCD. Participants rate themselves in relation to each of these statements on a scale of 0–3, with a higher number corresponding to a higher frequency of the particular symptom (0 = not at all, 1 = just a little, 2 = often, and 3 = very frequently). Participants scoring highly on the CAARS (T-Score > 60) were not excluded as running the statistical analysis with and without them did not affect the results.

The Driving Simulator

As data were collected in the United Kingdom, speed limit signs and measurements are in miles per hour. Metric equivalents

TABLE 1 | Means of demographic data for participants in this study.

Measure	DCD group (*N* = 19)	DCD (*SD*)	TD group (*N* = 36)	TD (*SD*)	*p* value
Age	26.5y	(2.38)	21y	(0.92)	$p = 0.017$*
Gender ratio M: F (17:38)	9:10		8:28		$p = 0.027$*
Drivers: Non-drivers (27:28)	10:9		17:19		$p > 0.005$
M-ABC-2 (percentile)	6.07	(1.68)	46.9	(3.43)	$p < 0.001$*
ADC	75.2	(15.29)	17.2	(18.19)	$p < 0.001$*
CAARS	53.83	(2.96)	48.7	(2.12)	$p > 0.005$
WIAS-IV Vocab	27.6	(4.51)	39.1	(4.42)	$p = 0.112$
WIAS-IV Block design	33.7	(2.97)	42.9	(1.68)	$p = 0.005$*

Significant group differences (TD, DCD) for each measure are also reported.

FIGURE 1 | Photo of the car simulator.

are provided with United Kingdom measurements provided in brackets throughout.

The driving task was presented in a driving simulator (SIM. Systems Technology Incorporated STISIM Drive; see **Figure 1**). This method allows participants to drive in a real car on a virtual road and creates a naturalistic experience where drivers can turn their head to look into the car's wing mirrors and rear-view mirror to peruse their environment. The rear view was provided by a combination of a back projection screen behind the cab (rear view mirror) and small monitor screens (door mirrors). The driving simulator vehicle parameters are; 1.67 m (5.5 ft) wide (center line of car ±, 0.83 m; 2.75 ft), 3.66 m (12 ft) long with a maximum speed of 177 km/h (110 mph), information was captured at 60 frames/second. Engine sound effects (braking, cornering tyre screech, and horn) were all turned on. The crash alarm was turned off, with the drive continuing as normal without reposition or reset of speed if participants collided with objects in their pathway. The triggering of events was carefully programmed to occur at the same distance into the drive for all participants, regardless of their speed.

The Drive

The drive (**Figure 2**) was 10.20 km (33,456 ft) in distance and took approximately 8–10 min to complete. The drive began in an 80.47 km/h (50 mph) zone with edge and center line markings on a single carriageway (2.42 m, 8-ft lanes) with steep grass banks. There are several shallow bends before the road widens (3.66 m, 12-ft lanes) and returns to being straight. Next, roadworks to the left of the road appear and some on-coming traffic. The road enters a town (48.3 km/h; 30 mph speed limit), passing between buildings with cars parked on either side of the road, opposite each other but separated by 3.66 m (12-ft). Next, the road exits the town between steep hills trees and enters a tunnel, curving to the left. On exit, the participants experience the same set of events they experienced previously but with no center line markings (80.47 km/h, 50 mph speed limit), starting with the 3.66 m (12 ft) wide lane and then 2.42 m (8 ft) wide lane. Photos of these different sections of the drive are presented in **Figure 3**.

Procedure

Each participant completed the Adult DCD checklist and the Conners Adult ADHD Scales at home. Once seated in the driving simulator, participants were informed (a) that the car was automatic (accelerator and brake pedals indicated), (b) there was no need to use the handbrake or indicators, (c) they should adjust the rear-view mirror (with help if necessary), (d) the location of the speedometer (checked for recognition), and (e) they should treat all drives as a real driving situation and obey driving laws. Following answering any participant's questions, the orientation drive was performed (3.3 m/10,800 ft in distance lasting 3–4 min). Participants then completed the main drive (10.2 m/33,456 ft in distance, lasting approximately 10 min). Upon completion of the main drive, participants completed the MABC-2, and WAIS-IV. Breaks were given as required throughout the testing procedure, which lasted approximately 2 h in total.

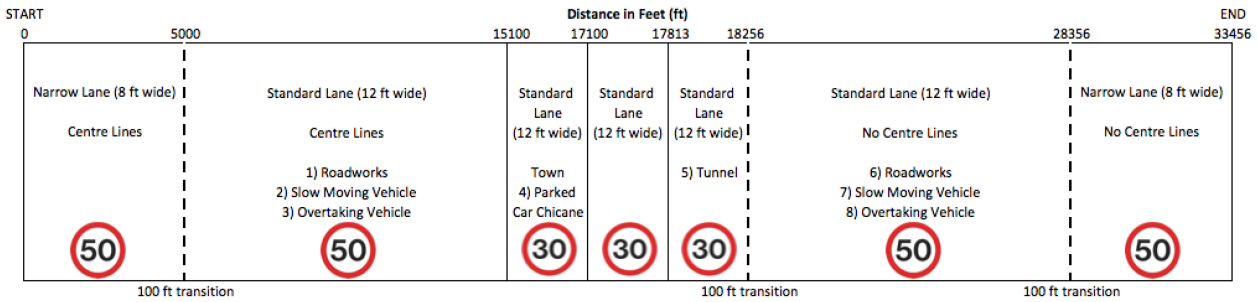

FIGURE 2 | Visual representation of the main drive (not to scale) based on distance (in feet) including width of lane, road markings, event number (in order of which it occurred) and speed limit.

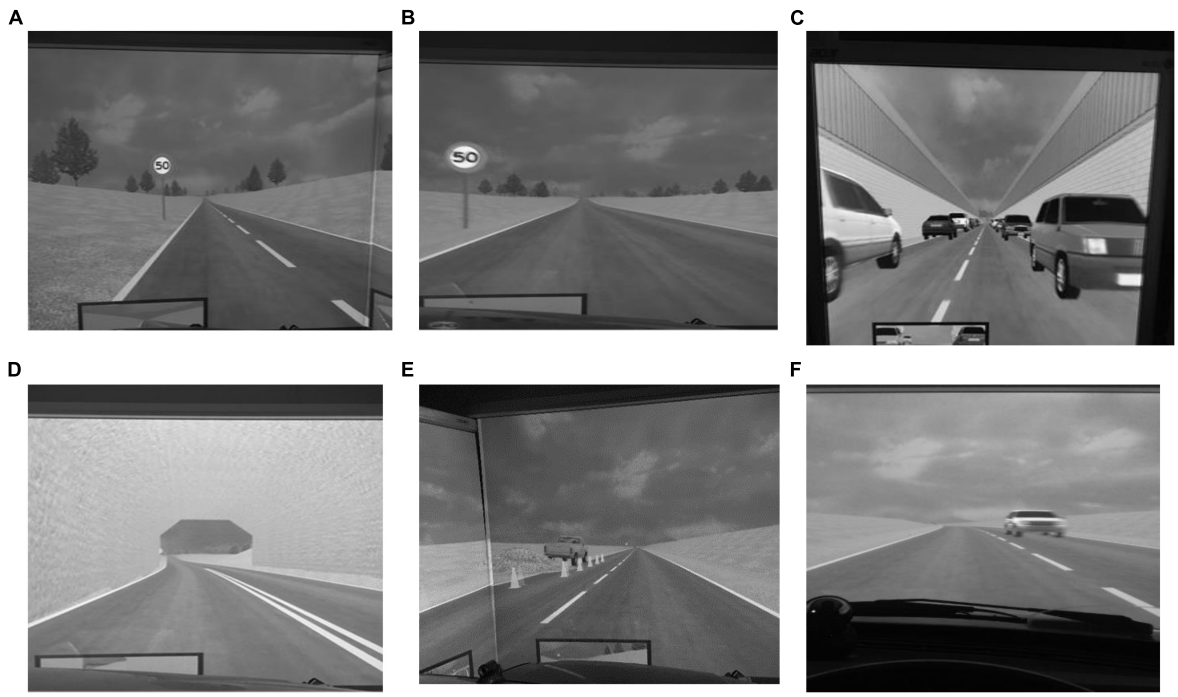

FIGURE 3 | Photos of sections of drive for experiment 1 **(A–D)**, experiment 2 **(E)**, and experiment 3 **(F)**. **(A)** Photo of section of drive with central road markings. **(B)** Photo of section of drive without central road markings. **(C)** Photo of section of drive through narrow aperture. **(D)** Photo of section of drive through tunnel. **(E)** Photo of section of drive past roadworks. **(F)** Photo of section of drive past on-coming vehicle.

Variables Measured

Longitudinal Speed

Longitudinal speed measured in miles per hour (mph) and converted to kilometers per hour (km/h).

Steering Adjustments

Steering adjustments were calculated by transforming the "Steering raw counts" to standardize the starting point for all participants to be zero. Steering raw counts is the raw output of the Analog to Digital converter on the steering sensors, this is presented in arbitrary units. The zero point is set when the simulation software is launched, but "straight on" is set as whatever position the steering wheel is in when the simulation run is launched. As, in almost all cases, the participant will move the steering wheel when seating themselves in the car, and although they will be asked to center the wheel before the simulation run is started, they will almost certainly not return the wheel to exactly the same place, hence the offset. The differences between each captured frame were calculated (as a series of points at 0.03 s intervals, which correspond to the video frames generated by the simulator), with any negative numbers transformed to the absolute value of the number before calculating the mean (the greater the rate of change of steering, the greater movement of the steering wheel).

Road Positioning

Road positioning measured in feet (ft), converted to meters (m) and taken from the mid-point of the car, in relation to

the center line of the road (center line value = 0). Participants start the drive at −1.8 m (−6 ft) left of center line in left-hand lane). The left-hand curb is −3.65 m (−12 ft) and a positive result indicates the participant has crossed the center line onto right-hand side of road.

Collisions Number

Collisions number of times the driving simulator virtually "hit" an object in the pathway (taken for Conditions 1 and 2 only).

Latitudinal Movement

Latitudinal movement mean speed of latitudinal movement, measured in feet per second (ft/s) and converted to meters per second (m/s) (taken for Conditions 2 and 3 only).

CONDITION DESCRIPTION AND MEASUREMENT PARAMETERS

Condition 1 (Clear, Straight Road, and No Oncoming Traffic)

Condition 1 investigated behaviors of drivers and non-drivers with and without DCD as they negotiated a series of 4 driving scenarios of low but increasing perceptual load (1: straight road/central lines, 2: Straight road/no central lines, 3: Parked car, and 4: Tunnel). Please see **Figures 3A–D** for visual representation of this condition. Mean data were collected for each Scenario as follows; Scenario 1; 0–4.6 km (0–15,100 ft); Scenario 3 = 4.6–5.2 km (15,100–17,100 ft) Scenario 4 = 5.4–5.6 km (17,813–18,256 ft); Scenario 2 = 5.6–10.2 km (18,256–33,456 ft).

Question

Do drivers and non-drivers with and without DCD differ in their behaviors when negotiating driving scenarios with a low, but increasing, perceptual load?

Condition 2 (Clear, Straight Road, No Oncoming Traffic, Stationary Obstacle in Pathway – Roadworks Straddling the Left Curb)

Condition 2 built upon the results of Condition 1 by increasing the motoric demands of the driving task whilst maintaining a low perceptual load. The task chosen reflected a scenario frequently encountered during the driving task and participants drove around roadworks positioned on the curb side of the road. The roadworks were positioned 1.98 km (6,500 ft) from the beginning of the drive and ended 2 m (6,570 ft) into the drive, extending 1.8 m (6 ft) from the curbside into the main carriageway. Pictorial representation of this scenario can be seen in **Figure 3E**.

Question

Do drivers and non-drivers with and without DCD differ in their behaviors when negotiating obstacles at the side of the road?

Condition 3 (Clear, Straight Road, With Dynamic Object – Oncoming Traffic in Right Hand Lane)

Condition 3, further, increased the perceptual load as participants negotiated safe passage alongside an approaching vehicle on the opposite side of the road. The oncoming car was 3.05 m (10 ft) long and 1.83 m (6 ft) wide and traveled at a fixed speed of 48.3 km/h (30 mph) along the center of the righthand carriageway. It was created (and started traveling along the carriageway) when the driver reached 1.52 km (5,000 ft) into the drive, 0.5 km (1,500 ft) ahead of the driver. Please see **Figure 3F** for visual representation of this condition.

Question

Do drivers and non-drivers with and without DCD differ in their behaviors as they pass an oncoming vehicle on the opposite side of the road?

Data Extraction for Condition 2 and 3

For Condition 2, the data were extracted $\pm$1 s from the point where the driver encountered the roadworks on a participant-by-participant basis in order to account for differing velocities. This was accomplished by specifying the point on the drive when the object was created, the distance from the driver, and the velocity at which the object was moving toward the driver (in this case 0 m/s; O ft/s). Using these variables, it was possible to calculate the distance of the object from the driver as they navigated the simulation and identify when they passed each other. The data for Condition 3 were extracted in the same way as in Condition 2, but in this case the obstacle was not static and so the velocity at which it approached the driver was set at a constant 9.1 m/s (30 ft/s).

DATA ANALYSIS

For all analyses, significant interactions were explored using simple main effects, and significant main effects were investigated using planned comparisons. Bonferroni corrections were applied to protect against Type I error. Mauchly's test of Sphericity was violated and Greenhouse-Geisser correction was therefore applied to all interactions. Please see **Table 2** for mean values by condition, driving experience and group.

Condition 1

Four separate repeated measures Analysis of variance (ANOVA) were used to investigate the effect of Group (DCD; TD), Driver Status (driver; non-driver) and Scenario (1: Lines; 2: No lines; 3: Narrow aperture/parked cars; and 4: Tunnel) on speed, road positioning, steering adjustments, and collisions.

Conditions 2 and 3

Separate ANOVAs were conducted to investigate the effect of Group (DCD; TD), and Driver Status (driver; non-driver) on speed, road positioning and steering adjustments, collisions as well as latitudinal movement for the period of time when the car was driving past the roadworks (Condition 2: static obstacle)

and vehicle driving toward them (Condition 3 dynamic obstacle, velocity at which it approached the driver was set at a constant 33 km/h (30 ft/s).

RESULTS

Condition 1 Behaviors of Drivers and Non-drivers With and Without DCD as They Negotiate a Clear, Straight Road in 4 Scenarios of Increasing Perceptual Complexity

Speed (km/h/mph)

There was a significant effect for Scenario [$F(3,81)$ 296.86, $p \leq 0.001$, $\eta p^2 = 0.853$]. Parameter estimates and contrast analysis reveal that participants drove slower in both the parked car (39 km/h/24 mph) and the tunnel (42 km/h/26 mph) scenarios compared to the lines (69 km/h/42 mph) and no-lines scenarios (69 km/h/43 mph). A Scenario-by-Driver Status interaction [$F(3,81)$ 6.956, $p = 0.003$, $\eta p^2 = 0.120$] revealed that drivers drove faster than non-drivers in all scenarios except parked cars, where they were slower. However, observations of the mean values in **Table 2** highlight that this significant interaction is mainly due to the drivers with DCD and a lack of a group interaction here might be due to low power. To investigate this further, planned comparisons using Mann-Whitney-U were run and showed that drivers with DCD were significantly slower in the parked car Scenario (M = 31 km/h/19.32 mph) compared to TD drivers (M = 40.2 km/h/24.99 mph), $U = 129$, $p = 0.027$.

Road Position (ft/m)

A significant effect for Scenario [$F(3,129)$, 292.027, $p < 0.001$, $\eta p^2 = 0.851$], together with mean values and pairwise comparisons revealed that road positioning was significantly different between each scenario ($p < 0.001$, M No-lines = −1.25 m/−4.11 ft; M Lines = −1.54 m/−5.04 ft; M Parked car = −0.6 m/−1.97 ft; M Tunnel = −1.9 m/−6.23 ft).

TABLE 2 | Mean values (*SD*) for speed (km/h/mph), road position (m/ft) steering adjustment, lateral speed (mps/fps) and collisions by condition (1, 2, and 3), and driving status (driver: non-driver) and group (DCD:TD).

		DCD		TD	
Speed km/h (mph)	**Condition**	**DCD:D**	**DCD:ND**	**TD:D**	**TD:ND**
1. No central lines	1	67.9 (42.2) (7.4)	65.5 (40.7) (2.6)	72.1 (44.8) (3)	68.8 (42.7) (3.4)
2. Central lines		68.7 (42.7) (6.3)	65.7 (40.8) (3.1)	73.1 (45.4) (3.6)	66.5 (41.3) (4.5)
3. Parked cars		31.1 (19.3) (6)	43.0 (26.7) (9.3)	40.2 (25) (5.3)	40.1 (25) (8.2)
4. Tunnel		44.8 (27.8) (2.5)	39.3 (24.4) (2.7)	43.5 (27.0) (2.3)	40.5 (25.2) (2.8)
5. Roadworks	2	61.9 (38.5) (4.6)	61.4 (38.2) (3.0)	64.7 (40.2) (3.6)	61.9 (38.5) (3.6)
6. Oncoming vehicle	3	69.3 (43.1) (4.9)	64.2 (39.9) (3.2)	73.5 (45.7) (3.8)	65.8 (40.9) (4.85)
Road Position m (ft)		**DCD:D**	**DCD:ND**	**TD:D**	**TD:ND**
1. No central lines	1	−1.28 (−4.2) (1.1)	−1.06 (−3.5) (1.7)	−1.31 (−4.3) (0.6)	−1.28 (−4.2) (1.2)
2. Central lines		−1.49 (−4.9) (0.5)	−1.46 (−4.8) (1)	−1.58 (−5.2) (0.4)	−1.58 (−5.2) (0.8)
3. Parked cars		−0.57 (−1.9) (0.8)	−0.67 (−2.2) (0.9)	−0.49 (−1.6) (0.7)	−0.67 (−2.2) (0.8)
4. Tunnel		−1.85 (−6.1) (0.8)	−1.8 (−5.9) (1)	−1.89 (−6.2) (0.6)	−1.95 (−6.4) (1.1)
5. Road works	2	−1.85 (−6.1) (0.88)	−1.89 (−6.2) (1.47)	−1.92 (−6.3) (0.66)	−2.1 (−6.8) (0.81)
6. Oncoming vehicle	3	−2.28 (−7.5) (1.18)	−2.24 (−7.4) (1.78)	−2.34 (−7.7) (0.57)	−2.43 (−8) (1.04)
Steering Adjust		**DCD:D**	**DCD:ND**	**TD:D**	**TD:ND**
1. No central lines	1	3.8 (1.3)	4.6 (3.2)	3.8 (1.2)	3.7 (1.8)
2. Central lines		4.2 (0.6)	4.5 (1.8)	4.2 (0.9)	4.3 (2.1)
3. Parked cars		8.4 (11.8)	9.2 (4.4)	6.6 (2.9)	6.7 (7.3)
4. Tunnel		26 (8.1)	17.4 (5.9)	20.5 (8.7)	20.5 (10.4)
5. Road works	2	4.7 (2.4)	4.9 (2.1)	4.2 (1.7)	4.03 (1.4)
6. Oncoming vehicle	3	31 (26.7)	42.7 (50.8)	22.8 (18.4)	26.6 (34.8)
Lat Speed m/s (fps)		**DCD:D**	**DCD:ND**	**TD:D**	**TD:ND**
5. Road works	2	−0.4 (−1.2) (1.0)	−0.5 (−1.6) (1.2)	−0.4 (−1.3) (0.9)	−0.3 (−0.9) (1.2)
6. Oncoming vehicle	3	0.03 (0.11) (0.4)	−0.04 (−0.13) (0.5)	−0.04 (−0.13) (0.3)	0.3 (0.9) (0.4)
Collisions		**DCD:D**	**DCD:ND**	**TD:D**	**TD:ND**
	1	0.2 (0.4)	3.9 (7)	0.1 (0.3)	2.1 (3.4)
	2	0 (0)	0 (0)	0 (0)	0.6 (1.2)

Steering Adjustment

A significant effect for Scenario [$F(3,96)$, 119.317, $p < 0.001$, $\eta p^2 = 0.701$], together with mean values and pairwise comparisons revealed that all participants used more steering adjustments in the parked car scenario (M Parked car = 7.37) compared to the other scenarios (M No-lines = 3.91, $p = 0.004$; M Lines = 4.29, $p = 0.001$; M Tunnel = 4.37, $p = 0.010$).

Collisions

There was a significant effect of Driver Status [$F(3,51)$ 8.490, $p = 0.005$, $\eta p^2 = 0.142$] showing that non-drivers had significantly more collisions ($M = 2.68$; $SD = 4.78$) than drivers ($M = 0.14$; $SD = 0.36$) throughout the drive. Further investigation of mean values reveals that this effect is mainly due to non-drivers with DCD who had more collisions ($M = 3.89$; $SD = 6.99$) than the TD non-drivers ($M = 2.10$; $SD = 3.38$).

Summary of Findings for Condition 1

Condition 1 investigated the effects of Group, Driver Status and Scenario on speed, road positioning, steering adjustments and collisions whilst driving under a low, but increasing perceptual load along a level, straight roadway. Analyses revealed that all drivers responded to increases in perceptual load, reducing their speed as they negotiated the parked car and tunnel scenario and used more steering adjustments whilst driving through the narrow aperture created by the parked car scenario. However, it appears that drivers with DCD may have been more affected by the increase in perceptual load as the Scenario-by-Driver Status interaction shows that drivers with DCD drove more slowly than TD drivers in the parked car scenario. Analysis also revealed that non-drivers had significantly more collisions than drivers. Finally, all participants adjusted their road position to accommodate the task demands. For example, whilst it is unsurprising that participants drove more centrally in the parked car scenario (−0.6 m/−1.97 ft), they also positioned the car more centrally in the left-hand lane in the tunnel scenario (−1.9 m/−6.23 ft) compared to the other scenarios (No-lines = −1.25 m/−4.11 ft; M Lines = −1.54 m–5.04 ft).

Condition 2: Behaviors of Drivers and Non-drivers With and Without DCD as They Negotiate Static Obstacle (Roadworks) in the Pathway

All Measures

Condition 2 investigated the effects of Group and Driver Status on speed, road positioning, steering adjustments, collisions and latitudinal movement whilst negotiating roadworks on the curb side of a level, straight roadway. There were no significant main effects or interactions for Speed (all Fs < 1.6, all ps > 0.2), Road position (all Fs < 2.6, all ps > 0.1), Steering adjustment (all Fs < 2.4, all ps > 0.1), Collisions (all Fs < 2.2, all ps > 0.1), and latitudinal movement (all Fs < 1.3, all ps > 0.2). Drivers and non-drivers with and without DCD drove at a similar speed and road position, they used a similar number of steering adjustments, collisions and latitudinal movement as they negotiated the roadworks. These findings show that increasing the motoric demands of a task with a low perceptual load for drivers and non-drivers with and without DCD had little effect on behavior.

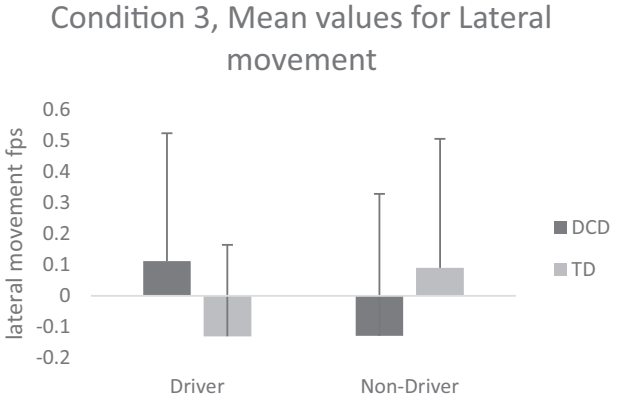

FIGURE 4 | Mean values for lateral movement condition 3.

Condition 3: Behaviors of Drivers and Non-drivers With and Without DCD as They Negotiate Oncoming Traffic

Speed (km/h/mph)

There was a significant effect for Driver Status [$F(3,51)$ 8.334, $p = 0.006$, $\eta p^2 = 0.140$] showing that drivers drove faster (71.3 km/h/44.30 mph) than non-drivers (65.1 km/h/40.45 mph) when passing a moving vehicle on the opposite side of the road.

Road Position (m/ft) and Steering Adjustments

There were no significant effects for Road position (all Fs < 1.9, all ps > 0.1) or Steering adjustment (all Fs < 2, all ps > 0.1),

Latitudinal Movement (mps/fps)

A significant Group-by-Driver Status interaction [$F(3,51)$, 4.388, $p = 0.041$, $\eta p^2 = 0.079$], mean values and pairwise comparisons show that lateral movement was significantly different between drivers with and without DCD ($p = 0.045$). Drivers with DCD drove further to the right, veering toward the oncoming vehicle (+0.03 m/s/ +0.112 fps) whereas TD drivers drove further to the left, veering away from the oncoming vehicle (−0.04 m/s/-0.130 fps). This effect was not evident when comparing non-drivers in the DCD and TD groups. Please see **Figure 4**. for illustration.

Summary Condition 3

Condition 3, investigated the effects of Group and Driver Status on speed, road positioning, latitudinal movement, and steering adjustments while passing an approaching vehicle on the opposite side of the road. Drivers drove faster than non-drivers when passing a moving vehicle on the opposite side of the road and the Group-by-Driver Status effect for latitudinal movement, showed that drivers with DCD veered toward (to the right) the oncoming car, whereas the TD drivers veered away from it (to the left).

DISCUSSION

The main aim of this study was to investigate behaviors of drivers and non-drivers with and without DCD when negotiating everyday scenarios. We used a series of three conditions to gradually increase the perceptual load and answer the following research questions; (1) Do drivers and non-drivers with and without DCD behave differently when processing dynamic sensory information in progressively complex environments? (2) How do drivers and non-drivers with and without DCD negotiate narrow apertures in a car of fixed width? (3) Do individuals with DCD collide more with obstacles in the pathway compared to controls? Given previous research (Cousins and Smyth, 2003; de Oliveira and Wann, 2011, 2012; Wilmut et al., 2015; Gentle et al., 2016), we expected that, across all conditions compared to controls, individuals with DCD would drive more slowly, have less appropriate road positioning, use more steering adjustments, and have more collisions. We also predicted that group differences would be more pronounced in conditions with higher perceptual load.

Whilst there were no main effects for group for any of the conditions, and our main hypothesis cannot be fully supported, several significant interactions show that individuals with and without DCD respond differentially to changing perceptual loads when negotiating everyday driving scenarios. For example, in condition 1, drivers with DCD were significantly slower in the parked car scenario compared to all other groups (non-drivers with DCD, TD drivers, and TD non-drivers). Furthermore, in Condition 3 drivers with DCD (but not non-drivers) drove further to the right, veering toward the oncoming vehicle whereas TD drivers drove further to the left, veering away from the oncoming vehicle. These behaviors suggest that TD drivers allow a larger gap between themselves and the oncoming vehicle to accommodate the additional demands of the task and maximize the opportunity for safe passage alongside the vehicle. The drivers with DCD do not adopt this 'safer' strategy and actually put themselves in greater danger by veering toward the oncoming vehicle.

It is beyond the remit of this study to identify causality, but differences in behaviors whilst negotiating oncoming traffic could be related to quality of visuo-motor integration. There is much evidence in the literature suggesting that vision and the motor control needed for steering are strongly interconnected (Land and Lee, 1994). Visual information of the road informs the arm-motor system controlling the steering wheel (Mars and Navarro, 2012), providing a direct link between gaze direction and steering (Robertshaw and Wilkie, 2008). Wilkie et al. (2008) showed that we steer where we look, and look where we steer (Land, 1998). These findings are supported by professional-but-anecdotal advice given in the Police Drivers Handbook and Experienced Rider Course (for motorcyclists) which warns against looking directly at a hazard to prevent steering toward it. Thus, if drivers with DCD are failing to inhibit the link between eye gaze direction and steering, this could explain why they veer toward the oncoming vehicle, placing them in a less optimal road position compared to the other groups in this study.

There may also be a biomechanical explanation related to postural control which is a fundamental skill necessary for every movement we make. Like all movement, postural control is heavily dependent upon the efficient integration of visual, vestibular, and proprioceptive information (Oie et al., 2002), an area of known difficulty for individuals with DCD (Wilson et al., 2013). There are two different strategies to control posture; "enbloc" (movement of head and trunk together) and 'articulated' (head is stabilized at the neck separately from the trunk, Assaiante and Amblard, 1995) argue that. The articulated mode of head stabilization occurs developmentally as sensitivity to the orientation of the head about the trunk increases, together with increased ability to control the degrees of freedom about the neck and head (Lund and Broberg, 1983). Assaiante and Amblard (1995) argue that this model predicts that any impairment in sensorimotor or biomechanical systems may prevent or postpone development. If the drivers with DCD are adopting the "en bloc" strategy to stabilize visual and vestibular information, this might explain why they veer toward the oncoming car. As the individuals with DCD move their eyes toward the oncoming vehicle, the head and trunk (and arms) follow "en bloc," steering the car toward the oncoming vehicle.

It is of note that in several scenarios, the non-drivers with DCD behave more like the TD groups than the drivers with DCD. For example, in Condition 1 the non-drivers with DCD drove at a similar speed to the TD drivers in the more challenging scenario when negotiating the parked cars. This behavior is replicated in Condition 3 where non-drivers with DCD and the TD controls veer away from the oncoming vehicle. There is no evidence to suggest that the non-drivers with DCD can perceive the dynamic information more accurately than the drivers with DCD, so what is happening in these scenarios to explain these inter-group differences?

An explanation can be sought after reviewing the data in **Table 2**. Drivers with DCD have higher standard deviations, particularly for speed and steering adjustments, suggesting more variance in their driving ability. The standard deviations for the non-drivers with DCD are lower and so they are more consistent. Given the age profiles of the participants for this study, it is possible that non-drivers with DCD and the TD groups have similar levels of driving experience as they have similar mean ages (21 years) compared to the drivers with DCD who had a higher mean age and wide age range (32 year, SD = 11.7). Thus, it could be that the wide age range reflects differences in driving experience which implicates performance. Clearly, there is a need for more investigation to tease apart the mechanisms behind differing behaviors for drivers and non-drivers with DCD.

A lack of significant effects for any of the measures taken for Condition 2 suggests that a small increase in motoric (but not perceptual) load of the task does not implicate behaviors for drivers or non-drivers with and without DCD. Whilst non-significant group effects are positive in terms of perceptions of the driving task, they need further discussion given previous work

in this area. Firstly, individuals with DCD drove alongside the roadworks at a similar speed to their TD peers, supporting the findings in both the de Oliveira and Wann (2011, 2012) driving studies. Furthermore, all participants slowed down (compared to the low perceptual load scenarios 1& 2 in Condition 1) as they negotiated the roadworks, suggesting appropriate awareness of the obstacle in the pathway.

However, the lack of group difference in road position was not expected, as previous work investigating navigation of obstacles in the pathway (Wilmut et al., 2015) suggests that individuals with DCD allow a higher "margin of error" to accommodate issues with visuo-motor integration and avoid collision. Methodological differences between studies may account for these unexpected results. For example, the Wilmut et al. (2015) study involved a narrow aperture which was constrained on both sides rather than only one side as in the current experiment. Had there been any oncoming traffic to limit the aperture width, behaviors of the DCD group may have been different. We also expected that individuals with DCD would use more steering adjustments, compared to the TD group when negotiating the roadworks (de Oliveira and Wann, 2011, 2012). Differences here can be explained in terms of task complexity as it could be argued that the motoric demands to safely negotiate a bend, as in the de Oliveira studies, are higher than avoiding roadworks protruding only 1.8 m (6 ft) into a 3.6 m (12 ft) carriageway found in the current study.

It must also be noted that, the measures taken in Condition 2 may not have been sensitive enough to reflect the subtlety of the sensory and motoric processes needed to successfully negotiate the roadworks (Cloete and Wallis, 2009). Future work could focus on a more detailed analysis of the specific demands of the task as a driver initiates steering to avoid the object in the pathway, then center's the vehicle on the straight trajectory, and finally repositions the vehicle on the main carriageway after completing the maneuver (Hildreth et al., 2000). Findings from this work would inform the literature on the contribution of group and driver status to navigational skills in complex, but real-world environments.

In terms of Driver status, we expected that non-drivers would drive more slowly and have more collisions than drivers. We can accept this hypothesis as results from Condition 3 show that non-drivers drove more slowly than drivers as they negotiated the oncoming vehicle and in Condition 1, non-drivers had more collisions than drivers. These findings reflect many studies in the literature of a greater incidence of crash rate for low mileage/inexperienced drivers compared to higher mileage drivers (e.g., Langford et al., 2006; Antin et al., 2017). Indeed, McCartt et al. (2009) show that when the relative importance of age and experience are investigated, it is experience that has the greatest effect on crash frequency. However, as this study did not measure driving experience, we need to interpret these findings with caution. Indeed, future work investigating the effect of experience on the success of the driving task would benefit the literature on driving with DCD. We know that individuals with DCD take longer to pass their test (Missiuna et al., 2008) and therefore gain more driving experience as they learn to drive compared to their TD peers. However, we do not know how this experience translates to driving skill.

Implications of This Research

We know that inexperienced drivers are more susceptible to road traffic accidents (Langford et al., 2006; McCartt et al., 2009; Antin et al., 2017) and for individuals with DCD, the additional perceptual constraints add a substantial cognitive load to the driving task (Wann et al., 1998; Missiuna et al., 2008; de Oliveira and Wann, 2011, 2012; Wilson et al., 2013). Thus, an individual's sensitivity to information from the environment needs to be considered alongside personal constraints that may affect how that information is used to inform movement and the driving task. One of the main implications for this work is to recognize the impact of both internal and external constraints on the ability of individuals with and without DCD to successfully interact with the environment. There is still much work to be explored in this area to fully understand the mechanisms behind skillful driving.

Interventions to support individuals with DCD when learning to drive would benefit from an environment with a low perceptual load such as off-road provision or an empty carpark to allow the individual with DCD to learn the motoric demands of the driving task. This strategy will provide a safe, yet effective approach and the individual can then slowly be introduced to a more dynamic environment as ability and confidence increase. Furthermore, the use of an automatic car would reduce the motoric demands of gear changing whilst driving to allow more attention to be given to the perceptual demands of the task.

CONCLUSION

We argue that drivers and non-drivers with and without DCD do behave differently when driving in progressively complex environments. However, the data presented here suggests differences within the DCD group need further investigation to fully understand the mechanisms that contribute to driving behaviors for this population. This study highlights how drivers and non-drivers with and without DCD apply a variety of strategies to accommodate their personal constraints depending on their perception of the task and environmental conditions. It is therefore important to consider both group membership, and driver status when evaluating behaviors whilst driving. The evidence suggests that even within a very safe and predictable environment (level, uncluttered road, and no unexpected hazards), as the perceptual demands of the tasks increased, individual constraints (lack of experience/difficulties in perceiving and responding to the environment) influenced the efficiency with which participants can respond to the dynamic environment when driving. These findings reflect previous suggestions of a deficient mapping between visual information and steering actions for individuals with DCD (Fajen, 2008; de Oliveira and Wann, 2012) and provide quantitative evidence to support work by Missiuna et al. (2008) and Kirby et al. (2011) who found that individuals with DCD self-reported particular difficulty with more complex driving skills, such as parking or reversing.

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 below: Open Science Framework (https://osf.io/frv29/).

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the University of Surrey FHMS Research Ethics Committee. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

JG, SC, and NW designed the study and undertaken data collection. NW and DB extracted driving data. JG and DB completed statistical analysis. JG completed Table and Figure preparation and first draft of the manuscript. JG, HL, and DB edited the manuscript. All authors approved the final version of this manuscript.

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Evoked Potentials Differentiate Developmental Coordination Disorder From Attention-Deficit/Hyperactivity Disorder in a Stop-Signal Task: A Pilot Study

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Developmental Coordination Disorder and Attention-Deficit/Hyperactivity Disorder are unique neurodevelopmental disorders with overlaps in executive functions and motor control. The conditions co-occur in up to 50% of cases, raising questions of the pathological mechanisms of DCD versus ADHD. Few studies have examined these overlaps in adults with DCD and/or ADHD. Therefore, to provide insights about executive functions and motor control between adults with DCD, ADHD, both conditions (DCD + ADHD), or typically developed controls, this study used a stop-signal task and parallel EEG measurement. We assessed executive performance via go accuracy and go reaction time, as well as motor response inhibition via stop-signal reaction time. This was complemented with analysis of event-related potentials (ERPs). Based on existing investigations of adults with DCD or ADHD, we expected (1) groups would not differ in behavioral performance on stop and go trials, but (2) differences in ERPs, particularly in components N200 (index of cognitive control) and P300 (index of attention and inhibition) would be evident. The sample included $N = 50$ adults with DCD ($n = 12$), ADHD ($n = 9$), DCD + ADHD ($n = 7$), and control participants ($n = 22$). We replicated that there were no between-group differences for behavioral-level executive performance and motor response inhibition. However, on a physiological level, ERP components N200 and P300 differed between groups, particularly during successful response inhibition. These ERPs reflect potential endophenotypic differences not evident in overt behavior of participants with ADHD and/or DCD. This suggests a disorder specific employment of inhibition or general executive functions in groups of adults with DCD, DCD + ADHD, ADHD, or control participants.

Keywords: dyspraxia, executive function, response inhibition, electroencephalography, ERPs

INTRODUCTION

Developmental Coordination Disorder (DCD) and Attention-Deficit/Hyperactivity Disorder (ADHD) are lifelong neurodevelopmental disorders known to co-occur in up to 50% of cases (Blank et al., 2019). The primary symptoms of DCD are difficulties in learning and executing coordinated fine and gross movements, while primary symptoms of ADHD include inattention, hyperactivity, and impulsivity (American Psychiatric Association, 2013). In the last decade, evidence for substantial symptomatic overlaps between the two disorders has been observed. This includes children with DCD displaying hyperactivity (Harrowell et al., 2018), and having deficiencies in executive functions (Bernardi et al., 2015; Sartori et al., 2020) with the latter potentially persisting overtime (Bernardi et al., 2018; Wilson et al., 2020). These are typically observed as core symptoms of ADHD. Conversely, impaired fine and gross motor skills, a primary symptom of DCD, have been found for children with ADHD in comparison to typically developing individuals (Kaiser et al., 2015). This overlap is not limited to children, as, for instance, adults with DCD have also expressed difficulties with executive functions (e.g., Tal Saban et al., 2014), while those with ADHD have shown weakened visuo-motor adaptation (Kurdziel et al., 2015). Despite considerable overlap between DCD and ADHD, researchers have often supported the notion that they are unique disorders (e.g., Martin et al., 2006; Sergeant et al., 2006; Goulardins et al., 2015). In their critical review, Goulardins et al. (2015) pointed out that further research is needed to identify the possible sources of symptomatic overlap in DCD and ADHD. This research gap was also documented in a recent international consensus on DCD, in which the authors also highlight a generally growing body of research on adult populations (Blank et al., 2019). To expand on this literature, a better understanding of the mechanisms involved in executive functions and motor skills for adults with DCD versus ADHD is pertinent.

An obvious target for such research is to examine how inhibitory control and related underlying mechanisms differ in DCD and ADHD. First, inhibitory control is central to executive functioning (Miyake and Friedman, 2012; Matzke et al., 2018). Second, inhibitory deficits have often been observed in those with ADHD compared to typically developing individuals, with some evidence also emerging for DCD (Wodka et al., 2007; DCD versus controls: Bernardi et al., 2015; Sartori et al., 2020). Given the prominence of work on response inhibition with ADHD (e.g., Pauli-Pott and Becker, 2011) along with a dearth of work on inhibition more generally for DCD, this is an apt starting point for examining unique features of inhibition between both conditions. Thus, the purpose of this paper is a unique and necessary investigation of the differences in motor inhibition between adults with DCD and ADHD and both conditions.

To this end, we used the Stop-Signal Task (SST), as it places particularly high demands on motor response inhibition (Rubia et al., 2001). Arguably, such increased demands on inhibitory performance should render the SST sufficiently sensitive to reveal capacity limits in inhibitory control (i.e., avoid the risk of a ceiling effect with optimal inhibitory performance), and thus permit observing differential effects across groups. Nonetheless, potential differences between DCD and/or ADHD could go undetected at the behavioral level alone (Mandich et al., 2003; He et al., 2018a). Therefore, we included parallel neurophysiological measurement. Event-related P300 and N200 components were examined at the neurophysiological level based on their high relevance in inhibitory control in previous research of the SST, especially with ADHD versus control groups (e.g., Bekker et al., 2005; MacLaren et al., 2007; Senderecka et al., 2012). We examine differences in these components between groups of adults with DCD, ADHD, both conditions, and those of typical development.

Behavioral Performance: Response Inhibition in DCD and ADHD

The SST is an opportune method for a closer look into inhibitory control and related executive processes (e.g., attention, Miyake and Friedman, 2012; Matzke et al., 2017, 2018), relevant to several disorders (e.g., ADHD; Verbruggen and Logan, 2008b; Nigg, 2017). The SST typically involves an ongoing binary selection process across go trials (e.g., left or right). On a small number of trials, a stop-signal cues participants that the response they are about to execute should be inhibited (stop trial). Stop trials involve a brief presentation of a go stimulus before the stop-signal appears after a variable delay. Owing to this delayed signal onset, the SST measures top-down response inhibition rather than automatic inhibition (Verbruggen and Logan, 2008a).

Various forms of the SST have been used to compare and contrast individuals with ADHD to unaffected individuals (e.g., Rubia et al., 2005; MacLaren et al., 2007; Pauli-Pott and Becker, 2011; Senderecka et al., 2012; Congdon et al., 2014). It has even been considered that impairments in task performance among those with ADHD versus typically developing individuals on a SST could be indicative of a prefrontal lobe dysfunction (Homack and Riccio, 2004), however, this may be specific to child populations. There is some evidence for potentially impaired inhibition in those with DCD compared to typically developing individuals as well (e.g., Mandich et al., 2003; He et al., 2018a) but when adults with DCD performed worse, these differences were subtle. One study has recently examined SST performance in a group of young adults with DCD versus typically developing individuals and found only a trend toward significantly different stop-signal reaction times (SSRTs; He et al., 2018a). This study also examined Go/No-Go task performance (automatic inhibition) (Verbruggen and Logan, 2008a), and found the DCD group had significantly reduced performance compared to typically developing individuals, showing some inhibitory differences in adults with DCD at the behavioral level (He et al., 2018a). More research is needed to determine if these inhibitory differences are consistent for adults with DCD and/or ADHD. Therefore, in the present study, we used the SST to capture the top-down processing of motor response inhibition for insight into executive functioning differences at both behavioral and neural levels.

ERP Evidence for Inhibitory Differences in ADHD and DCD

Adults with DCD and ADHD may employ advanced learned compensatory mechanisms (Kysow et al., 2017; Wilmut, 2017) which may in turn obscure their true differences

based on overt behavior alone. Therefore, it is important to consider the diverse endophenotypes of DCD and ADHD with parallel neurophysiological assessment. Endophenotypes, which are sometimes referred to as mechanisms, are the processes by which a phenotype is expressed (Rommelse et al., 2008). There is little research that has examined potential differences in both behavior and endophenotypic expressions (e.g., neural activity via EEG) in adults with DCD versus ADHD.

In fact, to date, explorations of neural activity via EEG versus behavioral performance have been rare for DCD versus control participants in general. To our knowledge, studies which have examined inhibition for individuals with DCD versus typically developing individuals have not yet included EEG to examine potential compensatory mechanisms, or in a more general brain-behavior comparison with a SST. However, there are some studies that examine inhibitory performance between individuals with ADHD versus typically developing individuals during a SST using EEG to capture event-related potentials (i.e., measurements of neural activity during discrete events; e.g., Bekker et al., 2005; MacLaren et al., 2007; Senderecka et al., 2012).

Among the studies examining inhibitory performance, those which included adult populations often found SST performance did not differ at the behavioral level for those with ADHD when compared to typically developing individuals, but variations have been found at the neural level (e.g., Bekker et al., 2005; MacLaren et al., 2007). More specifically, one study showed no differences on go trials, but revealed significantly longer SSRTs in the ADHD group (versus a typically developed individuals) coupled with significantly lower P300 ERPs (interpreted as an index of inhibition; Bekker et al., 2005). However, in another study, adults with ADHD did not differ in general behavioral performance on a simple SST compared to typically developing individuals, but instead showed significant differences in ERPs for P300 and N200 during stop trials (MacLaren et al., 2007). While the precise neural substrates of P300 and N200 have often been debated, both are thought to relate to aspects of inhibition, attention, and other executive functions in the context of a SST (Bekker et al., 2005; MacLaren et al., 2007; Huster et al., 2020). Taken together, these findings highlight the importance of testing the underlying neural responses to the SST in adult clinical populations. This may help elucidate the distinctions between those with ADHD and typically developing individuals.

The Current Study

The present study examined both behavioral and neural levels of performance of participants with DCD, ADHD, DCD + ADHD, and typically developing control participants in a motor inhibition task. We aimed to improve the understanding of brain-behavior differences in these adult groups in order to better inform the co-occurrence of DCD and ADHD, as well as to highlight differences between the occurrence of DCD versus ADHD alone. We expected that, due to compensatory mechanisms, no differences would be present at the behavioral level in general go accuracy, and mean reaction times for all trial types (particularly: Go RT, RT of unsuccessful stop trials, and SSRT). We further hypothesized that behavioral compensation among adults would relate to more robust differences in the EEG signals between groups, and more specifically, that they would be present in components P300 and N200 in line with symptoms of ADHD, DCD, or both conditions versus typically developing adults.

Due to insufficient data in the DCD literature to make specific assumptions of the direction of amplitudes in P300 and N200 we aimed to examine effects reported in the literature comparing ADHD and typically developing individuals. Furthermore, we explore and report all differences found in the complete set of electrodes to build insights into patterns when comparing DCD and DCD + ADHD groups. In addition, we explored all possible distinctions between the ERPs in the DCD and ADHD groups. These comparisons provide important pilot evidence in relation to group differences in inhibition and related executive functioning processes as well.

MATERIALS AND METHODS

Sample

A total of N = 59 participants were recruited at two sites (Germany and United Kingdom). Following EEG pre-processing and our criteria for the removal of outliers (see "EEG Pre-processing" and "Statistical Analysis" sections below), a final sample of N = 50 was included in the present analyses with the same participants across behavioral and neurophysiological levels (see **Table 1**). This sample included n = 30 from Germany and n = 20 from the United Kingdom. Overall participants were 67% female, 76% right handed, and M = 25.5 (SD = 7.9) years old. Groups included those with an existing diagnosis of ADHD (n = 9), DCD (n = 12), both ADHD and DCD (n = 7), and a control group (n = 22). In order to run more reliable analyses, we combined the participants from both sites to result in adequate group sizes (see **Table 1**).

In the clinical groups, all participants reported previous diagnoses of ADHD and/or DCD, and reported no history of brain damage or other developmental impairments (e.g., cerebral palsy). The control group reported no history of any psychiatric or other health conditions. Additionally, participants with ADHD were asked to not take ADHD-relevant medication on the day of the study session if they had the option. None of the participants included in the final sample reported taking such medication on the day of testing. The protocol was reviewed and approved by ethics committees at both sites (University of Mannheim and Oxford Brookes University).

Measures

We administered the Adult Dyspraxia/DCD Checklist (ADC; Kirby et al., 2010), it yielded good reliability in the present study via Cronbach's Alpha in overall scores (α = 0.950) and in its standard three subsections (A: difficulties in childhood, α = 0.918; B: current difficulties, α = 0.883; C, current difficulties manifested by others, α = 0.851). In addition, we compiled the Adult Self Report Screening (ASRS v.1) for ADHD (Kessler et al., 2005), which also yielded good reliability overall (α = 0.875) as well as

TABLE 1 | Group classification and testing location comparisons.

Groups: overall (*N* = 50)	**Sample size**	**Average ADC score**	**Average ASRS v.1 score**	
DCD	12	113.1 (14.1)	42.5 (9.3)	
ADHD	9	87.8 (12.0)	59.0 (9.0)	
DCD + ADHD	7	108.1 (11.7)	52.9 (11.7)	
Control	22	66.7 (12.8)	44.0 (8.2)	
Participants from Germany (*n* = 30)	**Sample size**	**Average ADC score**	**Average ASRS v.1 score**	
DCD	1	119.0	48.0	
ADHD	6	88.2	60.8	
DCD + ADHD	2	104.5	64.0	
Control	21	67.6	45.1	
Participants from United Kingdom (*n* = 20)	**Sample size**	**Average ADC score**	**Average ASRS v.1 score**	**Median MABC-2 percentile**
DCD	11	112.9	42.0	5th
ADHD	3	85.3	55.3	25th
DCD + ADHD	5	109.6	48.4	2nd
Control	1	50.0	22.0	63rd

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 [$F(3,46) = 38.37$, $p < 0.001$]. Post hoc tests revealed all group comparisons were significant ($p < 0.05$) aside from the DCD and DCD + ADHD group comparison. There was also a significant effect by group on ASRS v.1 scores [$F(3,45) = 7.78$, $p < 0.001$]. Post hoc tests revealed this effect was driven by all group comparisons except for ADHD and DCD + ADHD; DCD and DCD + ADHD; DCD and the control group; DCD + ADHD and the control group ($p > 0.05$).

in subsections A (adult ADHD symptom overview, $\alpha = 0.687$) and B (adult ADHD specific symptom probes, $\alpha = 0.835$).

Stop-Signal Task Features

The SST began with a black fixation cross at the center of the screen appearing for 500 ms. Next, a black shape cue (circle or square) was presented in the center of the screen surrounded by a black frame. On go trials, participants needed to press the key corresponding to the shape shown (go stimulus; counterbalanced "c" or "v" keys). On stop trials (25% of all trials) participants were instructed to refrain from pressing a key. Here, the black frame was replaced by a blue frame after a variable delay known as stop-signal delay (SSD). The SSD ranged from 250 to 1,250 ms with an adaptive up-down staircase method (Levitt, 1971) based on performance in steps of ±25 ms. The SSD was increased with successful inhibition and decreased with failed inhibition to maintain a 50% stop accuracy rate for each participant. Participants were informed of this tracking procedure and subsequently were to respond as fast and accurately as possible without waiting for the stop-signal to appear. The entire SST was presented against a gray background, and the SST was based on that of Gajsar et al. (2020; see **Figure 1**).

Participants completed a total of 768 trials across six blocks with open-ended breaks after each block. There were 128 trials per block, with 64 trials (48 go, 16 stop) per shape. The stimuli were presented on computers with MATLAB® (The MathWorks, Inc., Natick, MA, United States) using the Psychtoolbox extensions (Kleiner et al., 2007) on a 16 inch screen in Germany and a 24 inch screen in the United Kingdom. Viewing distances were approximately 5 and 3 feet, and visual angles of 43 and 67°, respectively.

Following recent guidelines on the SST set out by Verbruggen et al. (2019), this visual stop-signal task was designed with all recommended features aside from practice trials and block-based feedback of performance. In lieu of practice trials, the researchers checked in with participants after the first block to ensure that, to their knowledge, they completed the task properly. Block-based feedback was not included in order to reduce any external influences on performance given the novel DCD and DCD + ADHD groups.

Procedure

Prior to the main task, participants completed the ASRS v.1 for ADHD (Kessler et al., 2005), the ADC (Kirby et al., 2010), as well as questions about their demographics and health history. Participants in the United Kingdom also completed the MABC-2 age band 3 (Henderson et al., 2007). These measures were used to confirm preexisting diagnoses of DCD and/or ADHD for group assignment, and to ensure members of the DCD group did not have signs of ADHD, and vice versa (see **Table 1**). Next, participants were prepared for EEG measurement and then completed the stop-signal task. As a part of other studies, a subgroup of participants completed the SST and additional computer tasks in random order.

EEG Data Pre-processing

A 64 channel system was utilized for electrophysiological recoding in a standard 10–20 system. FCz was used as the reference electrode and AFz as the ground (for setup see Gerdes et al., 2013). Scalp impedances were maintained at 15 kΩ and below. Recordings were made at a sampling rate of 1 kHz at one testing location and 500 Hz at another; therefore, all data were adjusted to a 500 Hz sampling rate. The average of all channels was used as the reference in the data processing phase. A 50 Hz notch filter was implemented to remove any confounding high frequency noise. Moreover, a band pass filter was set from 0.5 to 70 to reflect the more cautious approach used in other studies with a SST and related clinical groups (e.g., Senderecka et al., 2012). Eye blink artifacts were removed with an Independent Component Analysis (ICA). Segments during responses were set with windows from −100 ms before the event to 400 ms after the event. For some participants, electrodes with uninterpretable signals were corrected with a topographic interpolation. An average of $M = 0.94$ ($SD = 1.21$, Range: 0–5) electrodes per participant needed to be corrected with topographical interpolations.

Trigger recoding was performed for all participants to tag trials as correct versus incorrect. In this process, three participants had insufficient or poor quality EEG data for which triggers could not be recoded, such that the recalculated triggers displayed a significant discrepancy between the original and

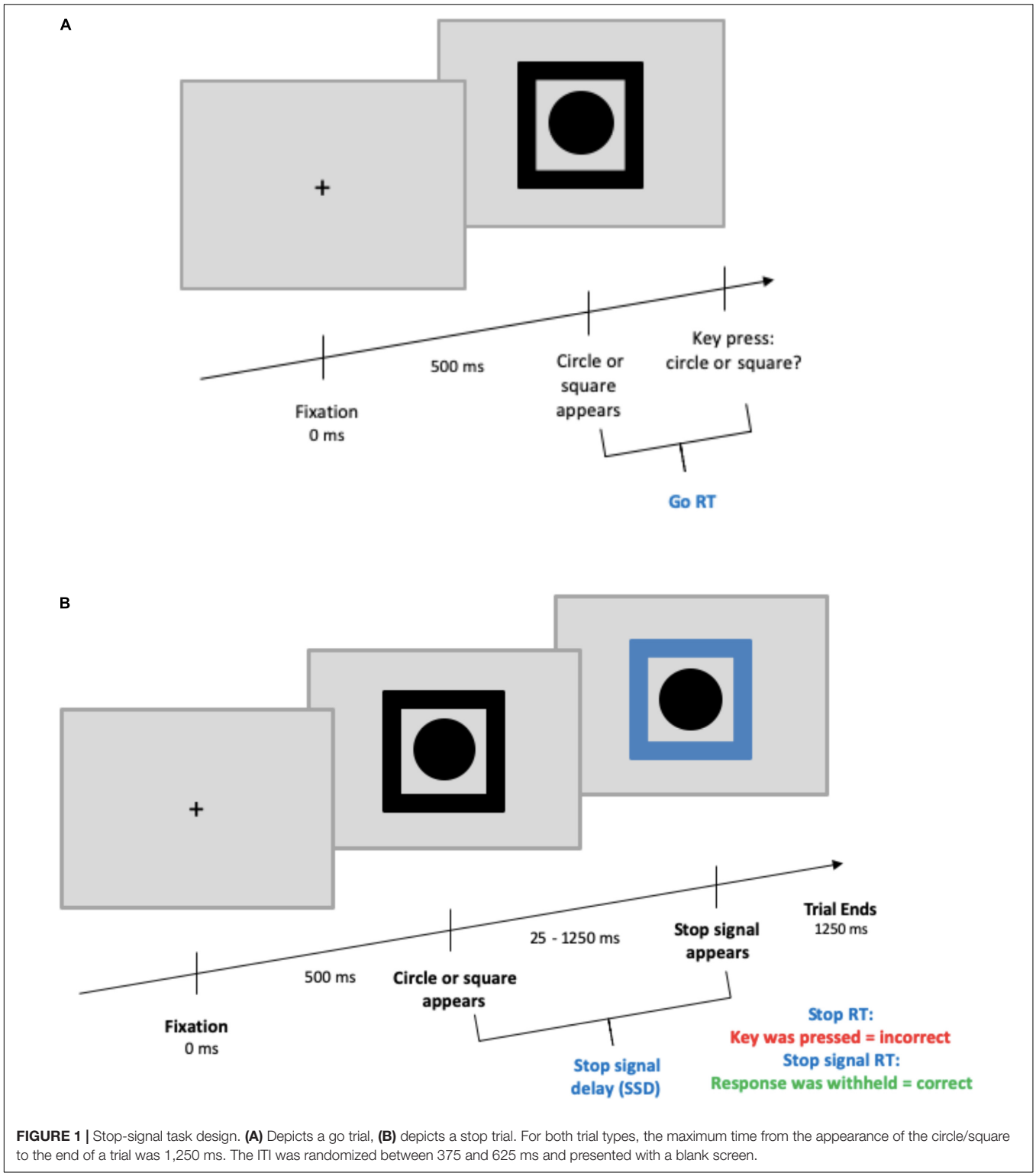

FIGURE 1 | Stop-signal task design. **(A)** Depicts a go trial, **(B)** depicts a stop trial. For both trial types, the maximum time from the appearance of the circle/square to the end of a trial was 1,250 ms. The ITI was randomized between 375 and 625 ms and presented with a blank screen.

new fixation trigger time points. This led to the exclusion of $n = 3$ participants.

Following other studies that have used the SST with ADHD and control groups (e.g., Kok et al., 2004; Ramautar et al., 2006; Johnstone et al., 2007; Senderecka et al., 2012), electrodes were explored with time windows set at 200 ms −310 ms for N200, and 230 ms −400 ms for P300. Epochs were defined at 100 ms pre-stimulus and 400 ms post-stimulus. We report any significant differences in amplitudes for individual electrodes.

Statistical Analysis

Participants were excluded if their average reaction times on go trials were larger than unsuccessful stop trial reaction times, thereby violating the independence assumption of the race model of the SST (Verbruggen et al., 2019). Following Verbruggen et al. (2019), mean RTs in this comparison included all trials with a key press (i.e., responses may also be incorrect or premature. This led to the removal of *one* participant with co-occurring DCD and ADHD from consecutive analysis. In an additional step, within-subjects outliers, i.e., extreme trial-level raw go RT and unsuccessful stop RT scores, were removed (on average, 4% an 33%, respectively) to exclude premature and late responses based on the criteria recommended by Leys et al. (2013; i.e., median $\pm$ 2.5 $\times$ absolute median score).

Average go reaction times were then computed for correct go trials and unsuccessful stop reaction times, with the latter including only responses in which a key was incorrectly pressed. The stop-signal reaction time (SSRT), or the estimated amount of time required to inhibit a response about to be executed, represents response latencies that were estimated for stop trials (i.e., in which a key was not pressed). The SSRTs were calculated with the block-based integration method (Verbruggen et al., 2013; see Gajsar et al., 2020 for a detailed description of this procedure). However, mean SSDs involved in estimating SSRTs were used to align with actual screen presentation times and are referred to simply as "SSDs"; see Verbruggen et al., 2019). This method is preferable when including clinical groups, such as individuals with ADHD (Verbruggen et al., 2013

Following the calculations above, performance was further screened to increase reliability by removing outlying individuals with sub-optimal performance based on the lenient outlier criteria set by Congdon et al. (2012), slightly adapted for clinical populations (see below). Based on this, participants were excluded if, on average, they violated one or more of the following criteria: (1) a proportion of successful inhibition on stop trials greater than 25% and less than 75%, (2) a proportion of go response greater than 60%, (3) an estimated SSRT which is positive and greater than 50 ms, and (4) a proportion of go errors less than 15%. The fourth criterion required slight modification to 15% (instead of 10%) to account for outliers in the DCD + ADHD group. Screening for these four criteria resulted in the removal of an additional n = 5 participants (n = 3 in control group; n = 1 with DCD; n = 1 with ADHD).

Behavioral and neurophysiological results were compared with between-subjects One-Way ANOVAs and Tukey's LSD *post hoc* tests. We also conducted independent samples *t*-tests to compare the amplitudes of ERPs between participants with DCD and ADHD in particular. Kolmogorov–Smirnov tests revealed the control group had a non-normal distribution (D = 0.20, p = 0.02) for SSRTs, which prevents its comparison to the other groups for correct stop trials. All aforementioned group comparisons held before and after outlier removal. Statistical analysis was conducted in R (v. 3.6.2).

RESULTS

Group Confirmation

As mentioned, participants were grouped based on their reported diagnostic history for DCD, ADHD, both conditions (DCD + ADHD), or no health conditions, and this was confirmed by self-reported symptoms. The average ADC scores for each group from highest to lowest were: DCD (M = 113.1, SD = 14.1), DCD + ADHD (M = 108.1, SD = 11.7), ADHD (M = 87.8, SD = 12.0), and the control group (M = 66.7, SD = 12.8). Scores of 90 and above signify probable DCD, and scores over 80 signal potential risk for DCD (Kirby et al., 2010). The ASRS v.1 scores from highest to lowest per group were: ADHD (M = 59.0, SD = 9.0), DCD + ADHD (M = 52.9, SD = 11.7), controls (M = 44.0, SD = 8.2), and the DCD group (M = 42.5, SD = 9.3) where a score of 47 or higher is indicative of likely ADHD. Classifications by group and testing site are listed in **Table 1**.

Stop-Signal Task Behavioral Parameters

Stop accuracy was not significantly different between groups, however, there was a significant difference for groups on go accuracy [$F(3,44)$ = 4.15, p = 0.011]. As revealed in *post hoc* testing, this difference was driven by a significant lower accuracy in the DCD + ADHD (M = 0.93, SD = 0.19) group compared to the control group (M = 0.97, SD = 0.11). The average reaction times for go trials, unsuccessful stop trials, and correct (i.e., successful) stop trials (SSRTs) were not significantly different between groups, whereby SSRTs were only compared between clinical groups (see **Table 2** for the descriptive statistics of the dependent measures of SST performance for all participants).

ERP Results

N200: All Group Comparisons

For component N200, there were amplitudes of several electrodes for which a significant group effect was found. This included C2 [$F(3,46)$ = 3.47, p = 0.024], C4 [$F(3,46)$ = 3.78, p = 0.017], C6 [$F(3,46)$ = 3.15, p = 0.034], FC2 [$F(3,46)$ = 3.25, p = 0.030], P4 [$F(3,46)$ = 3.00, p = 0.040], and P6 [$F(3,46)$ = 3.93, p = 0.014] during successful inhibition (correct stop trials). *Post hoc* tests revealed several of these differences were driven by the distinction in amplitudes between the ADHD and control groups (C2, ADHD: M = 0.71, SD = 1.74, Control: M = −2.27, SD = 2.88, p = 0.016; C4, ADHD: M = −0.28, SD = 1.75, Control: M = −3.04, SD = 2.69, p = 0.018; FC2, ADHD: M = 1.10, SD = 1.59, Control: M = −2.12, SD = 3.32, p = 0.020). A further difference in C6 was driven by marginally significant differences between the DCD and ADHD groups (DCD: M = −2.93, SD = 2.31, ADHD: M = −1.02, SD = 0.94, p = 0.056) and ADHD and control groups (Control: M = −2.64, SD = 1.43, p = 0.076; see **Figure 2**). Furthermore, an effect in amplitudes for electrodes P4 and P6 was driven by differences between the DCD + ADHD and control groups (P4, ADHD: M = −2.82, SD = 1.44, Control: M = −5.29, SD = 2.03, p = 0.048; P6, ADHD: M = −3.86, SD = 1.82, Control: M = −6.78, SD = 2.08, p = 0.010). Finally, one electrode was implicated in unsuccessful inhibition (incorrect

TABLE 2 | Descriptive statistics of dependent measures for stop-signal task behavioral data overall and per group.

Dependent Measure	Group	*M*	*SD*	Range
Probability of go omissions (no response)	DCD	0.02	0.03	0–0.07
	ADHD	0.04	0.04	0–0.14
	DCD + ADHD	0.03	0.04	0–0.1
	Control	0.02	0.02	0–0.08
	Overall	0.03	0.03	0–0.14
Probability of choice errors on go trials	DCD	0.04	0.03	0–0.1
	ADHD	0.04	0.04	0–0.13
	DCD + ADHD	0.07	0.05	0.02–0.14
	Control	0.03	0.02	0–0.09
	Overall	0.04	0.03	0–0.14
RT on go trials (mean)	DCD	663.64	107.25	491.25–842.05
	ADHD	618.46	167.22	402.91–837.31
	DCD + ADHD	654.59	197.17	496.53–988.59
	Control	672.49	151.75	407.1–904.31
	Overall	656.73	147.31	402.91–988.59
Intra-subject variability of correct go trials	DCD	133.40	35.80	67.43–191.7
	ADHD	120.17	47.27	49.42–192.59
	DCD + ADHD	114.68	35.26	80.69–167.44
	Control	122.37	39.98	54.47–184.19
	Overall	122.76	39.12	49.42–192.59
Probability of responding on a stop trial	DCD	0.50	0.03	0.45–0.59
	ADHD	0.49	0.02	0.45–0.51
	DCD + ADHD	0.48	0.03	0.43–0.51
	Control	0.48	0.03	0.44–0.56
	Overall	0.49	0.03	0.43–0.59
Average stop-signal delay	DCD	377.79	124.27	143.18–574.17
	ADHD	359.58	155.08	170.18–557.71
	DCD + ADHD	374.36	204.97	141.18–674.2
	Control	413.00	148.26	141.17–656.19
	Overall	389.00	147.96	141.17–674.20
Stop-signal reaction time	DCD	286.24	72.18	228.29–479.07
	ADHD	256.02	30.75	226.54–320.42
	DCD + ADHD	275.51	47.19	231.08–358.23
	Control	254.34*	43.42	200.4–413.9
	Overall	264.13	50.29	200.40–479.07
RT of go responses on unsuccessful stop trials	DCD	569.30	70.75	458.94–690.85
	ADHD	545.09	149.42	373.3–811.73
	DCD + ADHD	575.60	188.01	424.23–903.2
	Control	583.06	129.95	379.44–813.99
	Overall	570.77	126.75	373.30–903.20

Measures listed as recommended by Verbruggen et al. (2019), including measures for each group and overall. The SSRT was not estimated for n = 5 participants exhibiting suboptimal performance. An additional table is included in ***Supplementary Materials*** *listing descriptive measures for these participants. When outliers were removed on a trial-by-trial basis, group level differences did not change. *Not normally distributed.*

stop trials): C2 [$F(3,46) = 3.01$, $p = 0.040$], for which *post hoc* testing revealed differences between the ADHD ($M = 0.59$, $SD = 1.14$) and control groups ($M = -2.44$, $SD = 3.43$, $p = 0.037$; see **Figure 3**).

P300: All Group Comparisons

When considering component P300, there was a significant effect on the amplitudes of various electrodes based on group, including Fz during successful inhibition [$F(3,46) = 3.06$, $p = 0.038$], FC1 during unsuccessful inhibition [$F(3,46) = 3.05$, $p = 0.038$], and T7 during correct go trials [$F(3,46) = 3.39$, $p = 0.026$]. *Post hoc* testing via Tukey's HSD revealed these findings were driven by differences between the DCD + ADHD and control groups (Fz, DCD + ADHD: $M = 4.83$, $SD = 2.98$, Control: $M = 8.89$, $SD = 4.57$, $p = 0.092$; FC1, DCD + ADHD: $M = 4.95$, $SD = 3.01$, Control: $M = 9.37$, $SD = 4.58$, $p = 0.047$; T7, DCD + ADHD: $M = 2.63$, $SD = 2.48$, Control: $M = 0.32$, $SD = 1.68$, $p = 0.016$). However, in Fz this group difference was just marginally significant (see **Figures 2**, **3**).

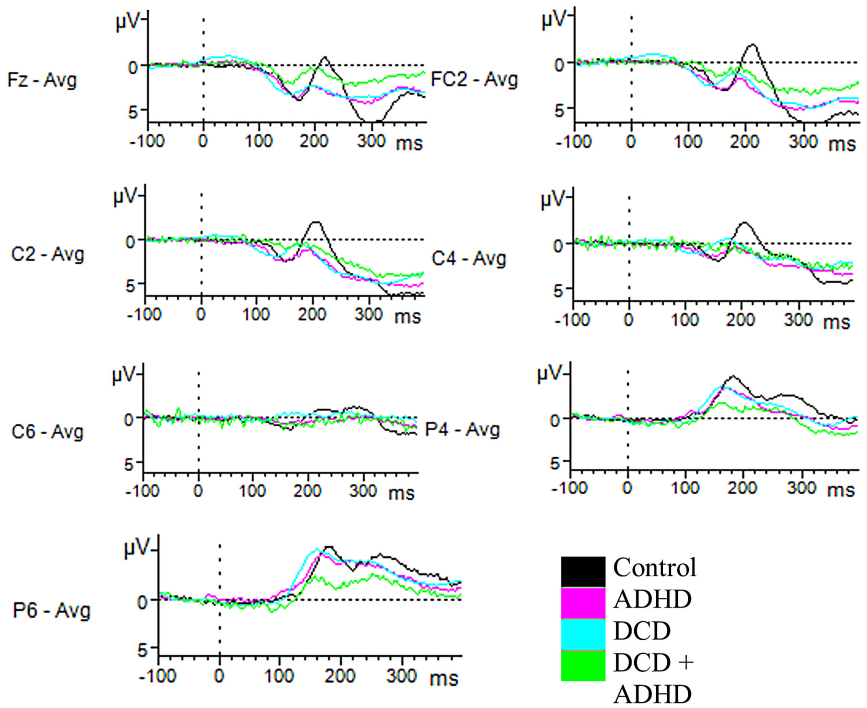

FIGURE 2 | Successful-stop ERPs. Depicted are all electrodes with significant differences in amplitudes for N200 and P300 for correct stop trials. Time windows were set at 200–310 ms for N200, and 230–400 ms for P300. For N200, significant differences in amplitudes in C2, C4, and FC2 were driven by differences in the ADHD and control groups, while differences for C6 were driven by differences between the control and DCD groups, and the control and ADHD groups. Differences in P4 and P6 were driven by differences in the control and DCD + ADHD groups. For P300, a significant difference between groups was indicated for electrode Fz, driven by the difference between the DCD + ADHD and control groups.

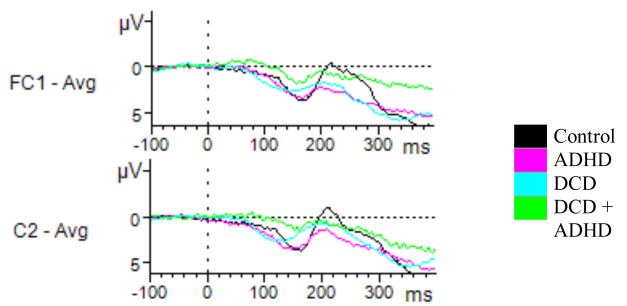

FIGURE 3 | Unsuccessful stop ERPs. Depicted are all electrodes with significant differences in amplitudes for N200 and P300 for incorrect stop trials. Time windows were set at 200–310 ms for N200, and 230–400 ms for P300. For N200, a significant effect of group on amplitude was found for C2, driven by a difference between the ADHD and control groups. For P300, a significant difference was found for FC1, driven by the DCD + ADHD and control groups.

Exploratory Comparisons: DCD Versus ADHD

No significant differences between the ADHD and DCD groups were found during go trials, however, for P300 and N200 on stop trials, there were several noteworthy differences between the DCD and ADHD groups in particular. During successful inhibition, significantly higher amplitudes were present in P300 for the DCD group compared to the ADHD group in fronto-temporal electrodes AF7 [$t(19) = 3.29$, $p = 0.004$] (DCD: $M = 4.30$, $SD = 1.84$; ADHD: $M = 1.88$, $SD = 1.40$), F7 [$t(19) = 2.99$, $p = 0.008$] (DCD: $M = 3.91$, $SD = 1.87$; ADHD: $M = 1.66$, $SD = 1.46$), and FT7 [$t(19) = 2.96$, $p = 0.008$] (DCD: $M = 3.32$, $SD = 1.19$; ADHD: $M = 1.59$, $SD = 1.49$). During unsuccessful inhibition, the DCD group had heightened peaks in activation for P300 compared to the ADHD group for electrodes AF7 [$t(19) = 2.44$, $p = 0.025$] (DCD: $M = 4.54$, $SD = 2.60$; ADHD: $M = 2.08$, $SD = 1.76$), F7 [$t(19) = 3.05$, $p = 0.008$] (DCD: $M = 5.27$, $SD = 1.82$; ADHD: $M = 2.64$, $SD = 2.13$), and T7 [$t(19) = 2.56$, $p = 0.018$] (DCD: $M = 3.00$, $SD = 1.20$; ADHD: $M = 1.46$, $SD = 1.54$).

In addition, several differences in central and fronto-central electrodes were present between the DCD and ADHD groups for N200, including FC2 [$t(19) = 2.29$, $p = 0.034$] (DCD: $M = -0.96$, $SD = 2.31$; ADHD: $M = 1.10$, $SD = 1.59$), FC6 [$t(19) = 2.60$, $p = 0.022$] (DCD: $M = -2.01$, $SD = 2.14$; ADHD: $M = -0.33$, $SD = 0.57$), C2 [$t(19) = 2.13$, $p = 0.047$] (DCD: $M = -1.14$, $SD = 2.13$; ADHD: $M = 0.71$, $SD = 1.74$), and C6 [$t(19) = 2.32$, $p = 0.032$] (DCD: $M = -2.93$, $SD = 2.31$, ADHD: $M = -1.02$, $SD = 0.94$) during successful inhibition. Furthermore, there were significant differences between groups for N200 peaks in several electrodes during unsuccessful inhibition, including C1 [$t(19) = 2.42$, $p = 0.027$] (DCD: $M = -0.93$, $SD = 2.61$; ADHD:

M = 1.17, SD = 1.30), and C2 [$t(19)$ = 2.31, p = 0.034] (DCD: M = −1.29, SD = 2.48; ADHD: M = 0.59, SD = 1.14). In all aforementioned differences in N200, the DCD group showed a significantly more negative amplitude than the ADHD group.

DISCUSSION

This study lays important groundwork in the DCD literature by examining endophenotypic overlaps and differences in executive functions. This novel design includes a stop-signal task and neurophysiological measurements among adults with DCD, ADHD, both conditions, and typically developing individuals (control group). As expected, the behavioral results showed few differences between all groups. One difference was present in that the go accuracy of individuals with co-occurring DCD + ADHD was lower than for typically developing individuals. Also in line with our expectations, several differences were found as indexed by ERPs between all groups, with additional differences found in direct comparison of the DCD versus ADHD groups. The latter comparison showed that many electrodes had significantly different amplitudes for components P300 and N200 between the DCD and ADHD groups. There were several electrodes, especially in central locations, which indicated differences in amplitude for P300 and N200 components on stop trials. These differences were not present for go trials which signifies that the general presence of the stop-signal may activate a specific neurophysiological response. This is in line with other research comparing those with ADHD to typically developing individuals in various age groups (e.g., Kok et al., 2004; Ramautar et al., 2006; Johnstone et al., 2007; Senderecka et al., 2012). While N200 is often viewed as an index of cognitive flexibility in typically developing individuals, P300 is viewed as an index of attention, and both have implications for attention and response inhibition in the SST (Chikara et al., 2018; Huster et al., 2020). Given that there were several key differences in these components in the present study, mechanisms of attention, response inhibition, and/or cognitive flexibility seem to differ for DCD and ADHD.

Aside from go accuracy, those with combined DCD + ADHD did not perform significantly different from all other groups at the behavioral level. This result aligns with previous studies of adults with DCD or ADHD compared to typically developing individuals where no differences in behavioral results were present between groups (Bekker et al., 2005; MacLaren et al., 2007; He et al., 2018a). Due to high demands on inhibitory performance in the SST (see Rubia et al., 2001), these results are unlikely to be accounted for by a floor effect in the clinical group, as they perform similarly to the control groups. Should our result hold in larger samples, it would indicate adults with DCD and/or ADHD can compensate in order to perform as well as typically developed adults in overt responses on the stop-signal task (i.e., inhibitory control and related engagement of attention).

The general absence of differences in overt behavior emphasizes the lack of diagnostic power of typical measures of accuracy and reaction times regarding differential diagnosis of DCD or ADHD in adults. Differences in task performance on other inhibition tasks (e.g., Stroop task) have previously been considered as indicators of possible neurological differences among those with ADHD in particular, but similarly are not sufficient for a diagnosis (Homack and Riccio, 2004). This could be due to effortful compensation in adults, or more broadly because inhibitory control is extremely complex and can be gaged differently by various inhibition tasks (Mirabella, 2021).

Evidence at the neural level indicates there may be unique neural signatures in evoked potentials between the DCD and ADHD groups, supporting findings of other studies (Langevin et al., 2014; McLeod et al., 2014). In the current study, several differences in activation were present in the N200 component for DCD versus ADHD groups on stop trials, especially in central regions during successful inhibition (i.e., correct performance on a stop trial). Interestingly, the amplitude for the DCD group was consistently larger than in the ADHD group regardless of successful versus unsuccessful inhibition. This provides evidence of separate ways of engaging attentional and inhibitory resources between these groups to achieve the same overt response.

Importantly, while behavioral performance did not differ between groups in the majority of parameters, inhibition and task engagement are not necessarily employed with the same underlying neural mechanisms across groups. It is unclear if these underlying mechanisms also translate to increased effort and/or fatigue, but this should be explored in more detail in future research. Given that compensation is more readily achieved when a task is less complex for DCD groups in particular (e.g., Pratt et al., 2014), it may also be the case that the task was too complex for the DCD + ADHD group with a higher symptom load.

Overall, the present study provides several novel contributions to the DCD and ADHD literature. First, to our knowledge it is the first study to compare inhibition performance between adult participants with DCD versus ADHD using a SST, as well as group comparisons between those with DCD, ADHD, DCD + ADHD, and typically developing adults. It is also the first study to incorporate an additional layer of EEG measurement to examine such group differences during the inhibition of a motor response to a visual cue. While most research on symptom differentiation relies on self-report questionnaires (Eisenbarth et al., 2008), some studies have also investigated endophenotypes in DCD or ADHD via motor performance or attentional performance exclusively with single-occurring DCD or ADHD compared to typically developing individuals, but rarely both. Third, investigations on adults with both ADHD and DCD are extremely rare. Therefore, examining this population can provide researchers with important insights into the endophenotypes and clinical picture of ADHD and DCD in adults.

Limitations and Future Directions

There are several limitations of the present study which should be considered. First, the sample sizes were small, in particular those of the clinical sub-groups. A normality check performed due to unequal group sizes indicated SSRTs of the typically developing (control) group were not normally distributed, even though the typically developing adults comprised of the largest group of the four in this study. Nonetheless, normality checks

for other groups passed. Small and unequal group sizes are not a new problem in DCD and ADHD research, especially when involving neuroscientific measurements (e.g., Querne et al., 2008; McLeod et al., 2014). In that respect, our sample sizes are in the range of previous studies. Future research should replicate this initial study with larger groups. While some shortcomings are expected in pilot testing, the present study is nonetheless important and novel by including diverse groups of adults with DCD, ADHD, DCD + ADHD, and typically developing individuals.

A second limitation is the duration of the study sessions. Participants completed 768 trials of the stop-signal task, and this took most participants around 40 min. While the participants all took breaks between blocks, this can still be straining, especially to participants with difficulties in sustaining attention. There is evidence of this for the DCD + ADHD group in particular, who performed significantly worse than typically developing individuals in go accuracy, which are the least complex trials in the task. This could be explained by a difficulty managing competing cognitive resources of the task (e.g., executive versus inhibitory performance) and could be related to a higher symptom load in the DCD + ADHD group.

Another limitation is that the mean performance measures (i.e., accuracy and reaction-time) did not reveal many substantial differences but they do not confirm a null hypothesis for behavioral results in the present study. Our main focus was to examine if neurophysiological differences would differ between groups for components linked to attention and motor performance with the expectation that behavioral data would reveal no differences, as observed in other studies with related samples (e.g., Bekker et al., 2005; MacLaren et al., 2007; He et al., 2018a). More robust cognitive models could potentially reveal more subtle differences between groups not detectable in mean RTs or accuracy. Future research should consider the use of cognitive models (e.g., the diffusion model, Ratcliff and McKoon, 2008; White et al., 2010) in behavioral data for those with DCD, ADHD, and both conditions. In addition, these models might better differentiate symptoms present in both DCD and ADHD related to visuo-motor integration deficits in particular (Kurdziel et al., 2015; Nobusako et al., 2018). Future research should also consider the utilization of a task with visuo-motor components in order to better understand the potential differences between DCD and ADHD in visuo-motor integration and inhibition, and how it is cognitively employed in relation to profiles of each disorder.

Additional limitations include the participant demographics (e.g., majority female, differing equipment at testing locations). Also, participants were recruited in two different sites. While the laboratory equipment and practices of the researchers remained as consistent as possible, some differences between the testing locations existed, such as the visual angles for the task. On the other hand, it may be an advantage that multinational groups of individuals may help to generalize the findings and is a method that has been used before with DCD (e.g., United Kingdom and Israel, Kirby et al., 2010). We argue that the benefit of increasing the sample size outweighs the possibly subtle differences in laboratories. Nonetheless, future research should replicate these findings with a larger population and balanced groups, but also across additional test sites and cultures.

Another limitation is the lack of specificity in N200 and P300 in general. ERP components are broad constructs, but are useful in the present study to indicate several processes in executive functioning (e.g., attention, action, inhibition; Huster et al., 2020). A source analysis for P300 and N200 components (e.g., Moores et al., 2003; Huster et al., 2010; Hong et al., 2017) should be considered in dedicated future research with larger samples to understand the localization of processes involved in executive functioning during a SST. Given that the present study aimed to identify any potential differences in ERPs as foundational evidence for future research, we reported all relevant electrode sites. However, it also is essential to replicate amplitude differences with *a priori* hypotheses in larger samples with respect to specific electrodes or sensor sites to account for potential false positives by testing many sensors.

Finally, future research should consider approaches with multiple levels of neurophysiological assessment, including neuroimaging and neurostimulation. Recent pilot work using TMS in adults with DCD versus typical adults has shown to be promising in identifying correlates of motor symptoms of DCD at the neural level (He et al., 2018b; Hyde et al., 2019). Combined approaches between some of these methods, such as EEG and TMS, could be particularly effective in disentangling DCD and ADHD symptoms at the neural level.

The present study provides an important initial step in identifying underlying neural processes which may not be reflected in behavioral performance of adults with DCD and ADHD. In addition to further work needed to confirm our findings in behavioral and electrophysiological differences in adults with DCD and/or ADHD during the SST, there is still a need for more research on other relevant endophenotypes in DCD and ADHD which can be compared in other paradigms (e.g., attention; Conzelmann et al., 2010). Also, we have not identified the specific compensatory mechanisms that were used (e.g., adaptive versus maladaptive), or if they were based on motor or executive functioning processes. So far we can only assert that there is evidence in the present study that compensation is being employed by participants in clinical groups. Future research should consider identifying more specific features of compensation mechanisms for those with DCD or ADHD, for example, in different age groups with longitudinal designs.

CONCLUSION

This pilot study innovatively demonstrated that inhibitory control may be a relevant endophenotype at behavioral and neurophysiological levels for the differentiation of DCD, ADHD, and co-occurring DCD and ADHD. Crucially, we identified patterns of varying P300 and N200 amplitudes, suggesting there are unique executive mechanisms utilized to inhibit a motor response between groups. At the same time, our results (i.e., group differences in ERPs but largely similar behavioral performance between groups), may reflect the potential strength

and success of compensatory mechanisms in individuals with DCD and/or ADHD. This study serves as an important foundation for future explorations into the overlapping executive functioning processes in DCD and ADHD.

DATA AVAILABILITY STATEMENT

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

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by University of Mannheim and Oxford Brookes University Ethics Committees. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

EM, MM, KW, MZ, and GA were involved in the conception or design of the work, data interpretation, critical revision of article, and approval of the version submitted for publication. EM tested all participants, conducted data analysis of measures, behavioral data, electrophysiological data, conducted the literature review, and formulated the initial draft of the manuscript. MM developed the stop-signal task used in this study in Matlab and wrote R scripts for behavioral analysis and trigger recoding. MZ provided funding for participants recruited in Germany. KW provided funding for participants recruited the United Kingdom. All authors contributed to the article and approved the submitted version.

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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.2021.629479/full#supplementary-material

Supplementary Table 1 | Descriptive measures of participants removed from behavioral analysis (n = 5).

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The Differential Effects of Auditory and Visual Stimuli on Learning, Retention and Reactivation of a Perceptual-Motor Temporal Sequence in Children With Developmental Coordination Disorder

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This study investigates the procedural learning, retention, and reactivation of temporal sensorimotor sequences in children with and without developmental coordination disorder (DCD). Twenty typically-developing (TD) children and 12 children with DCD took part in this study. The children were required to tap on a keyboard, synchronizing with auditory or visual stimuli presented as an isochronous temporal sequence, and practice non-isochronous temporal sequences to memorize them. Immediate and delayed retention of the audio-motor and visuo-motor non-isochronous sequences were tested by removing auditory or visual stimuli immediately after practice and after a delay of 2 h. A reactivation test involved reintroducing the auditory and visual stimuli after the delayed recall. Data were computed via circular analyses to obtain asynchrony, the stability of synchronization and errors (i.e., the number of supplementary taps). Firstly, an overall deficit in synchronization with both auditory and visual isochronous stimuli was observed in DCD children compared to TD children. During practice, further improvements (decrease in asynchrony and increase in stability) were found for the audio-motor non-isochronous sequence compared to the visuo-motor non-isochronous sequence in both TD children and children with DCD. However, a drastic increase in errors occurred in children with DCD during immediate retention as soon as the auditory stimuli were removed. Reintroducing auditory stimuli decreased errors in the audio-motor sequence for children with DCD. Such changes were not seen for the visuo-motor non-isochronous sequence, which was equally learned, retained and reactivated in DCD and TD children. All these results suggest that TD children benefit from both auditory and visual stimuli to memorize the sequence, whereas children with DCD seem to present a deficit in integrating an audio-motor sequence in their memory. The immediate effect of reactivation suggests a specific dependency on auditory information in DCD. Contrary

to the audio-motor sequence, the visuo-motor sequence was both learned and retained in children with DCD. This suggests that visual stimuli could be the best information for memorizing a temporal sequence in DCD. All these results are discussed in terms of a specific audio-motor coupling deficit in DCD.

Keywords: procedural memory, rhythm, sensory modality, circular analyses, non-regular sequence, tapping

HIGHLIGHTS

- General deficit in audio and visual motor synchronization with rhythmic stimuli in DCD.
- Auditory cueing improves learning and reactivation but not retention in DCD.
- Learning and retention of a visual sequence are preserved in DCD.

INTRODUCTION

Perceptual-motor procedural leaning is the acquisition of new perceptual-motor skills (a series of simple or complex movement elements) with practice, and procedural learning tasks are numerous (Doyon and Benali, 2005; Doyon, 2008). Even if procedural learning of temporal sequences (with no spatial component) has been subject to fewer studies than spatio-temporal sequences (with a low temporal component, as in the traditional Serial Reaction Time Task), both kinds of learning involve learning the order of a repeated sequence (of spatial and/or temporal parameters, see Shin and Ivry, 2002). During practice, participants learn to predict the location and/or time of the subsequent stimulus, thus becoming faster to respond or synchronize with the stimuli. Temporal sequence learning is typically found in music training. For example, at the very beginning of training for drumming and the basis of rhythm in music, temporal sequences are practiced with low spatial parameters (only one drum and one drumstick). In this case, learning requires perceiving sensory input items, memorizing them in a structured temporal sequence through repetitive practice, retaining the sequence for a certain period and then retrieving this temporal sequence so as to recall it (Patel, 2003; Konoike et al., 2012, 2015).

Experimentally, the production of time intervals can be assessed via sensori-motor synchronization (SMS), which is synchronization of a motor output with a sensory stimulus (Fraisse, 1948; Fraisse et al., 1958; Repp et al., 2011; Repp, 2005; Repp and Su, 2013). Several studies have investigated SMS with isochronous stimuli, i.e., with identical time intervals between two stimuli (Jäncke et al., 2000; Chen et al., 2002; Pollok et al., 2009; Blais et al., 2014, 2015). Studies using an SMS paradigm in healthy adults highlight that synchronization with auditory stimuli is stable (Sowiński and Dalla Bella, 2013). Moreover, the literature shows that SMS depends on the sensory modality of the stimuli. When participants are required to tap with their index finger in synchronization with tones (auditory sequence) or flashes (visual sequence), SMS with an auditory stimulus is more accurate and stable than SMS with a visual stimulus (Fraisse, 1948; Semjen and Ivry, 2001; Chen et al., 2002; Repp and Penel, 2002; Patel et al., 2005; Tierney and Kraus, 2013; Blais et al., 2014, 2015). This suggests that rhythmic movements tend to synchronize with auditory more than visual rhythms (Repp and Penel, 2004; Kato and Konishi, 2006). Moreover, SMS is less accurate and stable with non-isochronous stimuli, i.e., a sequence with different time intervals between two stimuli (Patel et al., 2005; Andreou et al., 2015). Therefore, learning is required to achieve synchronization with non-isochronous (auditory or visual) stimuli, which involves alternating short and long delays between consecutive stimuli.

Regarding developmental coordination disorder (DCD), many studies have found evidence of impaired sensorimotor timing, and especially SMS, irrespective of the modality of the stimuli (auditory or visual stimuli) and the type of response (unimanual, bimanual, or verbal) (Volman and Geuze, 1998; Volman et al., 2006; de Castelnau et al., 2007, 2008; Whitall et al., 2008; Debrabant et al., 2013; Blais et al., 2017; Puyjarinet et al., 2017; Blais et al., 2018; Trainor et al., 2018; Lê et al., 2020). However, a recent hypothesis was postulated by Trainor et al. (2018) that one core deficit of DCD could be a specific auditory timing deficit. This deficit would lead to specific impairment of audio-motor synchronization in DCD compared to typical development.

Regarding learning, the model by Nicolson and Fawcett (2007) predicts that procedural learning would be altered in children with DCD because of a dysfunction of the cortico-striato-thalamo-cortical network. However, studies investigating this issue in DCD have reported inconsistent results (Wilson et al., 2003; Gheysen et al., 2011; Lejeune et al., 2013; Blais et al., 2018; Lê et al., 2020). Very recently, a specific deficit in learning and the retention of an auditory temporal non-isochronous sequence using verbal responses were found in DCD (Lê et al., 2020). The deficit was less apparent for learning and the retention of a visual temporal non-isochronous sequence. On the contrary, controlling temporal parameters with visual stimuli seems to be less affected and repeated practice allows learning and retention of the visual temporal non-isochronous sequence in DCD and typically-developing (TD) children equally. These results highlight that DCD children seem to present with an alteration in audio-verbal coupling that is not reduced despite repeated practice. This is in line with the hypothesis of Trainor et al. (2018).

Special emphasis is placed on dynamic changes in memory after learning. The memory dynamic corresponds to the retention and reactivation processes (Tallet, 2012; de Beukelaar et al., 2016; Fogel et al., 2017). The retention process is evaluated by recall tests without stimuli (immediately after practice and after a time delay) and reactivation is evaluated via reintroduction of the stimuli further to retention. Withdrawal of the stimuli

during recall tests may reveal a persistence or forgetting of the memory trace and reintroduction of the stimuli may reactivate the memory trace having forgotten it. Therefore, in the present study, participants were required to practice temporal non-isochronous sequences by tapping on a keyboard in synchrony with auditory or visual stimuli. Afterward, they had to recall the sequences immediately after practice and recall again after a delay of 2 h. During the reactivation test, after the delayed recall (DEL), they were required to reproduce the sequence with the auditory or visual stimuli.

On this basis, this study aims to test the hypothesis for a specific audio-motor coupling impairment using manual responses in DCD when learning, retaining, and reactivating a new temporal sequence presented with either auditory or visual stimuli. In accordance with the hypothesis of an auditory timing deficit (Trainor et al., 2018), we expected that, compared to TD children, DCD children would have a deficit in SMS, learning, retention, and reactivation for a new audio-motor temporal sequence compared to a new visuo-motor temporal sequence. More operationally, we expected children with DCD to demonstrate a lesser decrease in mean asynchrony (and a lesser increase in stability) when practicing the audio-motor sequence compared to the visuo-motor temporal sequence. Moreover, for retention and reactivation, we expected that children with DCD would have a higher increase in asynchrony (and a lower increase in stability) for the audio-motor sequence compared to the visuo-motor sequence. In contrast, TD children were expected to have a higher increase in asynchrony (and a lower increase in stability) for the visuo-motor sequence compared to the audio-motor sequence.

MATERIALS AND METHODS

Participants

Twelve children with DCD and 20 TD children aged 8–12 years took part in this study. They were all right-handed, as assessed by the 10-question version of the Edinburgh Handedness Inventory (Oldfield, 1971; mean laterality quotient: 88.77 ± 20.33; range: 20–100). Seven more children were examined for this study, but their MABC score did not meet the inclusion criteria of Movement Assessment Battery for Children (M-ABC) <5th percentile, so they were not included in the study. We did not include children with musical skills (more than 4 h a week of formal practice for over 1 year). Participants had corrected-to-normal vision and hearing, as verified by a pre-experimental questionnaire. The children were enrolled in the DYSTAC-MAP study (ANR-13-APPR-0010). Eleven DCD and 18 TD children who passed an MRI participated in the study by Lê et al. (2020).

The inclusion criteria for the DCD group were: (1) no attention deficit/hyperactivity disorder according to DSM-5 (American Psychiatric Association, 2013); (2) diagnosis of DCD by a pediatrician; and (3) a total impairment score in the M-ABC (Henderson and Sugden, 1992; Soppelsa and Albaret, 2004) lower than the 5th percentile. The TD group was included with a total score higher than or equal to the 15th percentile. All children were clinically screened for neurodevelopmental disorder according to the DSM5 criteria. Children with comorbidities including ADHD, specific language impairment and developmental dyslexia were excluded from both groups. Moreover, the children did not have any clinical signs of verbal dyspraxia. None of the children had an intellectual disability, which was assessed via two sub-tests of the Wechsler Intelligence Scale for Children, 4th version (Similarities and Picture Concepts; Wechsler, 2005). The protocol was promoted by the French Ethical Committee of the Institute for Medical Research (Inserm, 2014-AO1239-38).

The participant characteristics are given in **Table 1**.

Materials

In the experiment, a computer with Presentation software (Version 18.0, Neurobehavioral Systems, Inc., Berkeley, CA, United States[1]) was placed in front of the experimenter. This computer gave visual instructions and visual stimuli to a connected 24″ screen, located 80 cm from the participants. Auditory stimuli were sent through headphones.

The participant's responses were collected via the same software using the key of the computer keyboard in front of him/her. The keyboard was connected to the computer via a USB port. The Presentation software recorded every time a key was pressed, which allowed recording with time precision in the tenths-of-milliseconds range. We ensured that the mean and stability for the uncertainties were very low and stable.

Task

Control Task: Synchronization With Isochronous Sequence

All the children had to synchronize with a sequence of 10 stimuli appearing at an isochronous interval of 1 s, by tapping the key of the keyboard with the right index finger.

Two modalities were tested: auditory stimuli were given via short tones (100-ms duration, 500 Hz) through headphones and visual stimuli were given via yellow squares (100-ms duration), which appeared in the center of the computer screen.

[1]www.neurobs.com

TABLE 1 | Motor and IQ assessment in both groups.

	TD (*n* = 20; 10 girls)		DCD (*n* = 12; 4 girls)		t(30)	p
	M	SD	*M*	SD		
Age (years)	10.17	1.30	9.63	1.18	1.59	0.12
M-ABC percentile	50.57	25.84	1.36	1.70	7.39	$3.09.10^{-8}$
WISC-IV – SIM	12.7	2.93	12.25	3.81	0.92	0.36
WISC IV – PC	10.15	2.05	9.41	1.92	1.44	0.15

M, mean; SD, standard deviation; M-ABC, Movement Assessment Battery for Children; WISC-IV, Wechsler Intelligence Scale for Children; SIM, Similitaries; PC, Picture Concepts.

Experimental Task: Learning to Synchronize With Non-isochronous Sequences

Practice (with stimuli)

The participants were asked to learn two non-isochronous sequences by tapping the right index finger on the key in synchrony with auditory stimuli (one sequence) and visual stimuli (another sequence). The two sequences were a series of 11 stimuli, which appeared at non-isochronous intervals. The auditory sequence included 11 brief sounds (100-ms duration, 500 Hz) and came through headphones, and the visual modality was in the form of 11 yellow squares (100-ms duration), which appeared in the center of the computer screen. The inter-stimulus interval varied between 500, 900, and 1,650 ms within each sequence in a pre-established pseudo-randomized order (**Figure 1**). Please note that the sequences were presented in a counterbalanced way, so that the results could be interpreted with respect to the duration of the sequences.

The children had to learn two non-isochronous sequences (auditory and visual) (**Figure 1**). The participants were warned that they had to reproduce the sequence without stimuli (without a metronome) at the end of the practice [immediate recall (IMM)] and 2 h after the practice without stimuli (DEL) and then with stimuli (reactivation).

Immediate and delayed recall (without stimuli)

In these tasks, the children had to recall sequences by tapping the key without the stimuli as accurately as possible.

Reactivation (with stimuli)

The children had to recall sequences by tapping the key with the stimuli as accurately as possible. This task assessed reactivation resulting from reintroducing the stimuli (environmental model).

Procedure

The experiment included several tasks, performed as follows:

Control Task: Synchronization With an Isochronous Sequence

The order of the auditory and visual modalities was counterbalanced between the participants for the isochronous synchronization tasks. Two trials were performed per modality.

Experimental Tasks

The children were required to perform the practice session using one of two modalities, followed by IMM, during which the metronome (i.e., visual or auditory sequence) was removed. The second modality was then practiced, followed by IMM. Two hours after the practice, both modalities were re-tested during DEL without the stimuli and in reactivation, during which the stimuli were reintroduced. During these 2 h, the children and their parents left for lunch. The order of the modalities (auditory or visual) and sequences (Sequence 1 or Sequence 2) was counterbalanced between the participants (**Figure 2**). Therefore, one participant learned Sequence 1 with auditory stimuli and Sequence 2 with visual stimuli whereas another participant learned Sequence 1 with visual stimuli and Sequence 2 with auditory stimuli.

Practice

For each sequence, one per modality, each participant had 30 practice trials to learn the sequence. At the end of each trial a visual feedback was presented to the participants as a smiley face, indicating performance (**Figure 3**). This task corresponded to the learning phase and was used to test the effect of the practice.

Immediate recall

Immediately after the practice phase, the participants had three trials to recall the sequence without the stimuli. They performed the visual sequence without any stimuli immediately after practicing the visual sequence, and performed the immediate auditory recall without any stimuli immediately after practicing the auditory sequence. No feedback was given. They started when they wanted and stopped when they thought they had finished the sequence. After a 10-min break, the participants practiced the

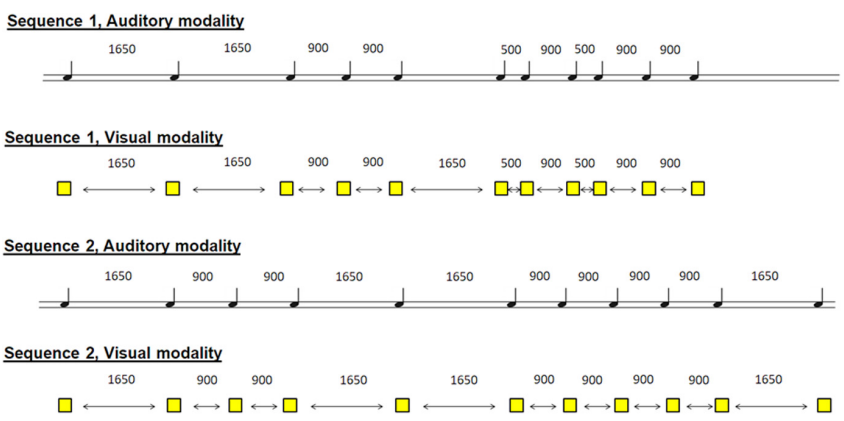

FIGURE 1 | Schematic representation of the 4 sequences. 2 sequences in the auditory modality and 2 sequences in the visual modality. The participant had to learn one sequence in one modality and the other sequence in the other modality. Sequences and modalities were presented randomly among participants.

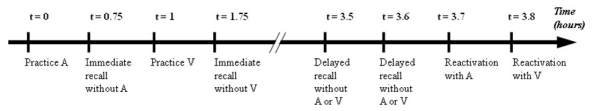

FIGURE 2 | Tests of the experimental protocol : practice, immediate recall, differed recall and reactivation for auditory sequence (A) and visual sequence (V). Immediate retention corresponds to process between practice and immediate recall. Differed retention corresponds to process between immediate recall and differed recall (without stimuli). Reactivation corresponds to process between differed recall (without stimuli) and reactivation (with the reintroduction of the stimuli).

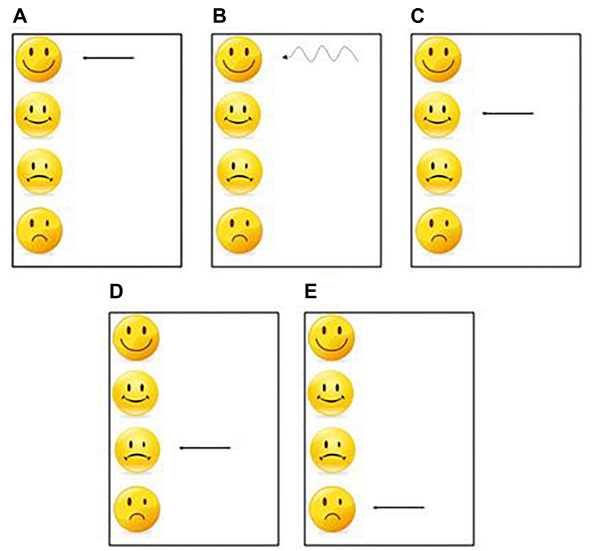

FIGURE 3 | One of these 5 feedbacks was presented at the end of each practice trial. **(A)** The first feedback was displayed when the participant had an average synchronization between −20 ms and +20 ms and a standard deviation less than 20 ms. **(B)** The second feedback was displayed when the participant had an average synchronization between −20 ms and +20 ms and a standard deviation greater than 20 ms. **(C)** The 3rd feedback was displayed when the participant had an average synchronization between −40 ms and −20 ms or between +20 and +40 ms. **(D)** The 4th feedback was displayed when the participant had an average synchronization between −60 ms and −40 ms or between +40 ms and +60 ms. **(E)** The 5th feedback was displayed when the participant had an average synchronization less than −60 ms or greater than 60 ms.

other sequence using the other modality [30 trials in the practice session + three IMM trials].

Delayed recall

Two hours after the practice session, the participants had to recall both sequences (three trials per sequence) without the stimuli. The order of the sequences was free. Children did not verbalize the number of the sequence but labeled the sequence "visual/square" or "auditory/tones" before starting. They started when they wanted and stopped when they thought they had finished the sequence. No feedback was given. Please note that seven TD children (35%) and five children with DCD (41.6%) were not able to reproduce the DEL sequences.

Reactivation

Participants had to produce the two sequences (three trials per sequence) with the stimuli in the same order as the practice. No feedback was given. Unlike DEL without a stimulus, this task was used to test reactivation by reintroducing the metronome.

Data Analysis

Practice and Reactivation

Asynchrony and stability were calculated via a circular data processing method (Fisher, 1995) using CircStat. CircStat is a MATLAB Toolbox (MATLAB version 2015a) for circular statistics (Berens, 2009), recommended for cyclical data, particularly suited to synchronization data and sensitive to individual differences (Dalla Bella and Sowiński, 2015). Circular data processing has been used in the literature during manual tapping on a synchronization task for an isochronous rhythmic sequence in healthy adults (Sowiński and Dalla Bella, 2013), children with or without a neuro-developmental disorder (Puyjarinet et al., 2017) and patients with neurodegenerative diseases (Martin et al., 2017).

Processing involves representing each finger tap with an angle (unitary vector) on a 360° polar scale, where the circle represents the inter-beat interval of the stimuli. The resultant angle of vector *R* represents synchronization accuracy (Sowiński and Dalla Bella, 2013; Dalla Bella et al., 2017). For each subject and each trial, we obtained a resultant vector angle that we transformed into an absolute value (in order to average the data across the trials). Subsequently, the vector angles were converted into a percentage of asynchrony to obtain data on a linear scale and for better understanding. To give an example of conversion: an angle of 0° was converted to 0% asynchrony and an angle of 180° was converted to 50% asynchrony. The higher the percentage, the lower the synchronization. The length of resultant vector *R* (Sowiński and Dalla Bella, 2013; Dalla Bella et al., 2017) is referred to as synchronization stability and varies from 0 to 1: 0 corresponds to a uniform and random distribution of responses around the circle while 1 corresponds to a uniform distribution of responses in one direction. In other words, the longer the vector length is close to 1, the greater the stability for the synchronization of responses within the trials. The vector angle and the vector length were obtained via circular statistics using CircStat (Berens, 2009) in MATLAB, based on temporal tapping data.

For each subject and each trial, we obtained a percentage of absolute asynchrony representing accuracy and a vector length representing the stability of sensorimotor coordination synchronization. Please note that the first response was never taken into account in data processing because it was considered a "warm-up" step. Every three consecutive trials of the 30 practice trials were averaged to obtain 10 blocks of three practice trials for absolute asynchrony and vector length.

Immediate Recall and Delayed Recall (Without Stimuli)

These recalls without stimuli led to other analyses because (1) we recorded tap time only (with no stimulus) and (2) the participants

started when they wanted, so the first interval was extremely variable from one individual to another.

The first tap had to align with 0° (as the first response was synchronized with the first imaginative stimulus). The first tap time was subtracted from all other times further to the responses so the first tap was well aligned with 0° and the following taps were in tempo with what the participant had done.

Between the responses, we added an accumulatively increasing stimuli time by putting the first 0° stimulus aligned with the first dummy response. From there, we performed the same data processing as the practice and reactivation sessions in order to obtain the vector angle and vector length of the three trials. The angle values of the three tests were highlighted as an absolute value so as to be able to average them. Finally, the angle was converted into a percentage.

Control Task: Synchronization With an Isochronous Sequence

Asynchrony (accuracy) and vector length (stability) were calculated using a fixed inter-stimuli interval of 1,000 ms. The number of errors was computed because this appeared to be a potential learning deficit marker in DCD (Blais et al., 2017). Errors corresponded to the additional taps compared to what the rhythmic stimuli proposed.

Statistics

Data normality was verified using the Kolmogorov–Smirnov test ($p > .05$). The homogeneity of variance was verified for each analysis of variance (ANOVA); *df* and *p*-values underwent Greenhouse–Geisser correction, if necessary.

For the control synchronization task using an isochronous metronome, statistical Group (2) × Modality (2) analyses of variance (ANOVAs) were carried out with repeated measures on Modality (Auditory; Visual), to compare the children with DCD with the TD children (controls) on asynchrony, vector length and number of errors ($p < 0.05$).

For the practice session, statistical Group (2) × Modality (2) × Block (10) ANOVAs were carried out with repeated measures on Modality (Auditory; Visual) and Block (B1–B10) on asynchrony, vector length and number of errors.

For immediate retention, statistical Group (2) × Modality (2) × Immediate Recall (2) ANOVAs were carried out with repeated measures on Modality (Auditory; Visual) and Immediate Recall (B10; IMM) for asynchrony, vector length and number of errors. Please note that we compared the last practice block (B10: mean of the last three practice trials) with the immediate retention block (IMM: mean of three retention trials).

For delayed retention, statistical Group (2) × Modality (2) × Recall (2) ANOVAs were carried out with repeated measures on Modality (Auditory; Visual) and Recall (IMM; DEL) for asynchrony, vector length and number of errors.

For reactivation, statistical Group (2) × Modality (2) × Reactivation (2) ANOVAs were carried out with repeated measures on Modality (Auditory; Visual) and Reactivation (DEL; REAC) for asynchrony, vector length and number of errors.

The *p*-value was fixed at $p < 0.05$ for each analysis. η^2 was reported for significant effects on the ANOVA. Separate *post hoc* *t*-tests were computed for independent groups using a Bonferroni correction for multiple comparisons.

RESULTS

Control Task: Synchronization With an Isochronous Sequence

Asynchrony (Accuracy)

The ANOVA revealed a main Group effect on asynchrony [$F(1,30) = 7.496$, $p = 0.01$; $\eta^2 = 0.199$]. Asynchrony was higher in the DCD group (12.4% ± 7.2) than the control group (6.9% ± 4.2) irrespective of Modality, reflecting a lower synchronization accuracy in children with DCD than the control children.

Vector Length (Stability)

The ANOVA revealed a Group effect on vector length [$F(1,30) = 12.881$, $p = 0.001$; $\eta^2 = 0.184$]. Vector length was lower in the DCD group (0.675 ± 0.112) than the control group (0.805 ± 0.090) irrespective of Modality, reflecting lower synchronization stability in children with DCD than the control children.

The ANOVA revealed a Modality effect on vector length [$F(1,30) = 8.508$, $p = 0.006$; $\eta^2 = 0.008$]. Vector length was higher in the auditory Modality (0.801 ± 0.147) than the visual Modality (0.711 ± 0.137) irrespective of the Group, reflecting higher synchronization stability in the auditory modality than the visual modality for both groups.

Number of Errors

The ANOVA revealed a Group effect on the number of errors [$F(1,30) = 5.993$, $p = 0.020$; $\eta^2 = 0.166$]. The number of errors was higher in the DCD group (1.645 ± 0.950) than the control group (1.078 ± 0.334) irrespective of the Modality.

Experimental Task: Learning Non-isochronous Sequences

B1–B10: Practice Effect

Asynchrony (accuracy)

The ANOVA revealed a main Group effect on asynchrony [$F(1,30) = 5.682$, $p = 0.023$; $\eta^2 = 0.008$]. Asynchrony was higher in the DCD group (25.5% ± 10.3) than the control group (19.4% ± 10.8), irrespective of Modality and Block, reflecting lower synchronization accuracy in children with DCD than the control children.

The ANOVA revealed a main Block effect on asynchrony [$F(9,270) = 20.511$, $p < 0.001$; $\eta^2 = 0.116$]. Asynchrony was higher during Block 1 (29.8% ± 8.2) than Block 10 (16.7% ± 10.7) [$t(30) = 8.897$; $p = 6.45\ 10^{-10}$) irrespective of the Group and Modality, suggesting increased accuracy with practice for both groups.

The ANOVA revealed Block × Modality interaction on asynchrony [$F(9,270) = 4.080$, $p < 0.001$; $\eta^2 = 0.022$]. Irrespective of the Group, asynchrony decreased with the Block, most significantly in the auditory Modality (**Figure 4A**), suggesting

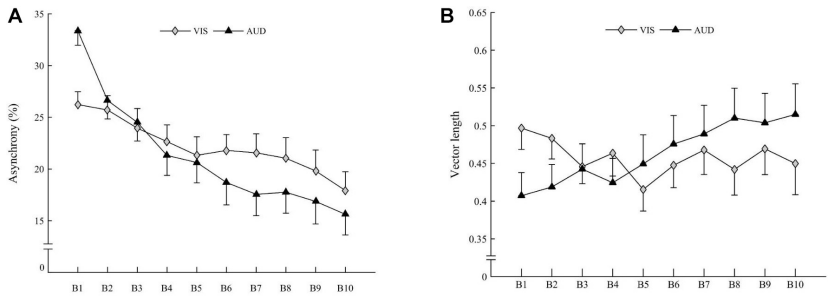

FIGURE 4 | (A) Mean asynchrony of children (both groups averaged) for visual modality (gray diamonds) and auditory modality (black triangles). **(B)** Mean vector length of children (both groups averaged) for visual modality (gray diamonds) and auditory modality (black triangles). Vertical bars represent inter-individual variability (standard error).

that accuracy significantly increased with practice for both groups in the auditory Modality.

Vector length (stability)

The ANOVA revealed a Group effect on vector length [$F(1,30) = 4.534$, $p = 0.041$; $\eta^2 = 0.009$]. The Vector length was lower in the DCD group (0.40 ± 0.18) than the control group (0.49 ± 0.18) irrespective of the Modality, reflecting lower synchronization stability in children with DCD than the control children.

The ANOVA revealed a Block × Modality interaction on vector length [$F(9,270) = 3.002$, $p = 0.001$; $\eta^2 = 0.026$]. Irrespective of Group, the vector length increased with the Block for the auditory Modality [$t(30) = 3.19$; $p = 0.003$] but not for the visual Modality [$t(30) = 1.55$; *ns*], suggesting a stability increase with practice for both groups for the auditory Modality only (**Figure 4B**).

Number of errors

The ANOVA revealed a main Group effect on the Number of errors [$F(1,30) = 6.213$, $p = 0.018$; $\eta^2 = 0.122$]. The Number of errors was higher in the DCD group (1.241 ± 1.217) than the control group (0.631 ± 0.766), irrespective of the Modality and Block.

The ANOVA revealed a Modality × Block interaction on the Number of errors [$F(9,270) = 2.565$, $p = 0.007$; $\eta^2 = 0.022$]. Irrespective of the Group, the Number of errors decreased with the Block for the auditory Modality only [$t(30) = 3.101$; $p = 0.004$].

End of Practice (B10) vs Immediate Recall: Immediate Retention

Asynchrony (accuracy)

The ANOVA revealed a main Group effect on asynchrony [$F(1,30) = 5.230$, $p = 0.029$; $\eta^2 = 0.009$]. The Vector angle was higher in the DCD group (23.6% ± 10.7) than the control group (17.8% ± 10.1) irrespective of the Modality and IMM, reflecting lower synchronization accuracy in children with DCD than the control children.

The ANOVA revealed an IMM effect on asynchrony [$F(1,30) = 16.397$, $p < 0.001$; $\eta^2 = 0.123$]. Asynchrony was higher in IMM (23.2% ± 9.6) than at the end of the practice (B10) (16.7% ± 10.7) irrespective of the Group and Modality suggesting decreased synchronization accuracy for immediate retention, when stimuli were withdrawn.

Vector length (stability)

The ANOVA revealed an IMM effect on vector length [$F(1,30) = 26.712$, $p < 0.001$; $\eta^2 = 0.470$]. The Vector length was lower in IMM (0.31 ± 0.11) than at the end of the practice (B10) (0.48 ± 0.23), suggesting that stability decreased when stimuli were withdrawn, irrespective of the Group and Modality.

Number of errors

The ANOVA revealed a main Group effect on the number of errors [$F(1,30) = 9.651$, $p = 0.004$; $\eta^2 = 0.174$]. Irrespective of the IMM and the Modality, the number of errors was higher in the DCD group (1.43 ± 1.23) than the control group (0.57 ± 0.75).

The ANOVA revealed an IMM effect on the number of errors [$F(1,30) = 17.252$, $p < 0.001$; $\eta^2 = 0.005$]. The Number of errors was higher during IMM (1.08 ± 1.16) than during the final block of the practice (B10) (0.71 ± 0.88) irrespective of the Group and Modality.

The ANOVA revealed a Group × IMM interaction [$F(1,30) = 11.172$; $p = 0.002$; $\eta^2 = 0.003$], Modality × IMM interaction [$F(1,30) = 5.627$; $p = 0.024$; $\eta^2 = 0.014$] and Group × Modality × IMM interaction on the number of errors [$F(1,30) = 5.254$; $p = 0.029$; $\eta^2 = 0.013$]. For the auditory Modality only, the number of errors increased in the DCD group between the end of the practice (B10) (0.92 ± 0.76) and the IMM (2.25 ± 1.49) [$t(30) = 3.844$; $p < 0.001$], whereas for the visual modality, the number of errors did not increase between B10 (1.11 ± 1.06) and the IMM (1.47 ± 1.20) [$t(30) = 1.65$; *ns*] (**Figure 5**).

Immediate Recall vs Delayed Recall: Delayed Retention

Asynchrony (accuracy)

The ANOVA revealed no significant effect or interaction.

Vector length (stability)

The ANOVA revealed no significant effect or interaction.

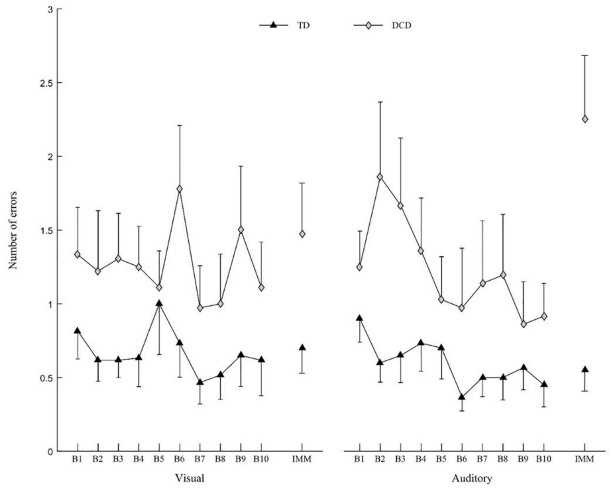

FIGURE 5 | Mean number of errors for DCD group (gray diamonds) and TD group (black triangles) for visual (left) and auditory modality (right). Vertical bars represent inter-individual variability (standard error).

Number of errors

The ANOVA revealed a main Group effect on the number of errors [$F(1,18) = 8.41$, $p = 0.009$; $\eta^2 = 0.318$]. Irrespective of Modality and Recall phase, the number of errors was higher in the DCD group (1.48 ± 1.02) than the control group (0.61 ± 0.76).

Delayed Recall vs Reactivation: Reactivation

Asynchrony (accuracy)

The ANOVA revealed no significant effect or interaction.

Vector length (stability)

The ANOVA revealed a Reactivation of stimulus effect on vector length [$F(1,18) = 50.333$, $p < 0.001$; $\eta^2 = 0.736$]. Irrespective of the Group and Modality, the vector length was higher (more stable) for Reactivation (0.54 ± 0.19) than DEL without a stimulus (0.27 ± 0.12).

Number of errors

The ANOVA revealed a main Group effect on the number of errors [$F(1,18) = 10.994$, $p = 0.003$; $\eta^2 = 0.212$]. Irrespective of Modality and Reactivation, the number of errors was higher in the DCD group (1.23 ± 1.06) than the control group (0.52 ± 0.71).

The ANOVA revealed a Reactivation of stimulus effect on the number of errors [$F(1,30) = 4.654$, $p = 0.044$; $\eta^2 = 0.009$]. Irrespective of the Group and Modality, the number of errors was higher for DEL without a stimulus (0.98 ± 1.01) than Reactivation (0.56 ± 0.75).

DISCUSSION

The purpose of this study was to test SMS and procedural learning for a sensorimotor temporal sequence specified by auditory or visual stimuli in DCD. We predicted that children with DCD would have more difficulties synchronizing, learning, retaining, and reactivating a new temporal sensorimotor sequence than TD children. Moreover, we expected that difficulties would be modulated by the sensory modality of the stimuli, with a greater learning deficit for auditory than visual stimuli, as per the hypothesis of Trainor et al. (2018). Our results were partially consistent with our hypotheses.

Firstly, during the SMS task using isochronous stimuli, children with DCD demonstrated less accurate and stable synchrony than TD children for both auditory and visual stimuli. They also made more errors than their TD peers. Thus, our results indicate that an overarching synchronization deficit is present in DCD, regardless of the visual and auditory modality of the stimuli, as per previous findings on auditory stimuli (Williams et al., 1992; Whitall et al., 2008; Rosenblum and Regev, 2013; Puyjarinet et al., 2017) and auditory and visual stimuli (Whitall and Clark, 2018; Lê et al., 2020). Given that SMS was also impaired for both visual and auditory stimuli when children had to respond with verbal responses (Lê et al., 2020), it is possible that the general – effector-independent and modality-independent – deficit in SMS is possibly due to a deficit in timing perception in DCD, as proposed by Trainor et al. (2018).

Secondly, the DCD group was as able as the TD group in improving accuracy and stability and decreasing the number of errors with practice on the non-isochronous sequence, which challenges the idea that children with DCD do not use sensory information to improve performance (Whitall et al., 2006; Mackenzie et al., 2008; Roche et al., 2011). Therefore, learning a new temporal perceptual-motor sequence seems to be retained in children with DCD. These results are in line with previous results showing that learning is relatively preserved in DCD (Wilson et al., 2003; Blais et al., 2018; Lê et al., 2020) and challenges the procedural learning deficit hypothesis postulated by Nicolson and Fawcett (2007).

Thirdly, regarding the effect of the sensory modality, we found a more significant improvement in temporal accuracy and stability during practice with auditory compared to visual stimuli in TD children and children with DCD. This result suggests that children with or without DCD benefit more from auditory stimuli than visual stimuli when learning a temporal sequence. However, the benefit of auditory stimuli seems to be transient for children with DCD, who demonstrated a significant increase in errors immediately after the removal of the auditory stimuli (IMM) and after a delay (DEL). The new increase in performance with the reintroduction of the auditory stimuli (reactivation test) suggests that children with DCD have a specific deficit in terms of integrating an audio-motor sequence in their memory. Given that the recall and reactivation tests involved withdrawing and reintroducing environmental information specifying the audio-motor sequence, the modulation of performance in children with DCD suggests that the auditory information provides a guidance effect (Salmoni et al., 1984; Walter and Swinnen, 1994). In other words, children with DCD depend on environmental auditory information during practice. This dependency, which is specific to auditory information, suggests that the auditory information results in the establishment of a perception-action coupling in DCD, as already suggested in Lê et al. (2020). Children with DCD fail to properly reproduce the temporal sensorimotor sequence by

themselves once the auditory information is removed. Children with DCD may be able to transiently adapt to environmental stimuli when present, but are not able to really integrate the temporal sequence in their memory. Another view is that auditory stimuli are so attractive that, when withdrawn, children are prone to making more errors than with visual stimuli, for which withdrawal does not result in as much disruption (Repp and Penel, 2004; Repp and Su, 2013; Thaut, 2015). In this case, our results suggest for the first time that auditory stimuli are more attractive than visual stimuli in DCD children when compared with TD children.

In short, TD children benefited from auditory information at each stage of practice, retention, and reactivation, contrary to DCD children, who benefited from auditory information during practice and reactivation (when the stimuli were present) but not for retention (when the stimuli were removed and the sequence had to be produced from memory). For the first time, these results demonstrate the superiority of the auditory modality from SMS to the procedural learning of a new sensorimotor temporal sequence in TD children. As per the literature, in healthy adults (Repp and Penel, 2002; Chauvigné et al., 2014; Merchant et al., 2015; Iversen and Balasubramaniam, 2016), it is possible for common cerebral structures to underlie both SMS and temporal sequence learning with auditory stimuli. In DCD, even if the auditory information helps improve performance during practice and reactivation (with stimuli), it does not help retention (without stimuli). Therefore, our results are partially in line with the proposal by Trainor et al. (2018), who hypothesized that "motor control of children with DCD would benefit from the addition of rhythmic auditory cues" (Trainor et al., 2018). Our results actually led us to conclude that visual stimuli are more likely to improve the learning and memorization of temporal motor sequences in children with DCD. This result adds to the previous findings of Lê et al. (2020), showing that visual information could be a more appropriate cue for the long-term retention of temporal sequences.

LIMITS AND PROSPECTS

A few limits and prospects can be mentioned for this study. Firstly, given that each child had to learn and retain two temporal sensorimotor sequences, an interference effect may have taken place. As previously explained by Schmidt and Young (1987), when two similar tasks are practiced sequentially, they may interfere each other. Such a phenomenon may have occurred in our study, but we could not investigate it given that learning of the two tasks was counterbalanced. However, it would be interesting to study the role of interference in DCD in the future.

Another limitation of our study is that we did not test the perceptual discrimination abilities of the participants. Errors in sensorimotor synchronization may have resulted from a deficit in timing perception, in line with the recent assumption of Trainor et al. (2018). Trainor et al. (2018) hypothesized that auditory perceptual timing deficits may be core characteristics of DCD, but no studies have yet demonstrated this assumption (Trainor et al., 2018). On the other hand, please note that errors correspond to additional responses (more taps than required). This result could be a marker of a motor inhibition deficit in DCD, as reported in a previous study about learning in teenagers with DCD (Blais et al., 2017, 2018). Therefore, we cannot be sure that errors identified in this study were due to a deficit in (perceptual) processing of the stimuli or a deficit (inhibition) in motor output.

Moreover, there might be heterogeneity in the way children memorize the sequence, explaining why we cannot observe the effects of DEL for instance. It is also difficult to explain the high incidence of children (both TD and DCD) who were unable to reproduce the DEL sequence. In the future, it may be interesting to study individual strategies that could give information on specific processes at stake in the learning and memorization of temporal sequences.

Another limitation is the sample size of our study, with only 12 participants with DCD. However, the exclusion of comorbidities and the restricted inclusion criteria (with a M-ABC score below the 5th percentile) were a real advantage for this study.

Finally, our results open prospects for studying the cerebral correlates of learning in DCD. The model of Doyon and Benali (2005) suggests that sequence learning is supposed to involve the cortico-striato-cortical loop, whereas the cortico-cerebello-cortical loop is involved in sensorimotor adaptation. On this basis, it seems that the cortico-striato-cortical loop could be altered in DCD (Cignetti et al., 2020; Tallet and Wilson, 2020). In this study, we evaluated learning with only 30 practice trials, which corresponds to the fast-learning stage according to Doyon and Benali (2005). This stage involves a large cerebral network, including not only the striatum but also a set of structures such as the cerebellum, motor cortical regions, parietal, prefrontal, and limbic regions. In the future, neural functional connectivity measurements to and from the striatum may be good way to understand the relationships between observable behavior and cerebral indices (Blecher et al., 2016). Studying learning at the slow learning stage (specifically involving the cortico-striatal network) would certainly show additional results on motor learning and memory in children with DCD.

To date, no intervention studies have specifically tested whether children with DCD need more practice compared to TD children in order to reach a similar performance level in motor learning tasks (Schoemaker and Smits–Engelsman, 2015; Smits–Engelsman et al., 2018). In our study, the DCD children may have needed more practice compared to TD children to retain the audio-motor sequence. In other words, children with DCD may require longer to reach saturated learning for auditory stimuli.

All in all, our results encourage the continuation of research on aspects involving procedural memory and neural correlates in DCD to be considered as necessary.

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 human participants were reviewed and approved by the National Ethical Committee of the Institute for Medical Research (Inserm, 2014-AO1239-38). Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

YC, J-MA, MJ, and JT conceived the project and obtained the financial support for this experimentation. MB and JT conceived and planned the experiment, analyzed the results, and wrote the manuscript. MB and SM carried out the experiment. All authors provided critical feedbacks on the manuscript.

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Young Adults With Developmental Coordination Disorder Adopt a Different Visual Strategy During a Hazard Perception Test for Cyclists

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Cycling in traffic requires a combination of motor and perceptual skills while interacting with a dynamic and fast-changing environment. The inferior perceptual-motor skills in individuals with developmental coordination disorder (DCD) may put them at a higher risk for accidents. A key skill to navigate in traffic is to quickly detect hazardous situations. This perceptual-cognitive skill was investigated in young adults with DCD using simulated traffic situations in a hazard perception test in cycling. Nine individuals with DCD (age: 23.0 ± 3.8) and nine typically developing (TD) individuals (age: 24.6 ± 3.5) participated in the study and completed the test while their gaze was tracked using a remote eye tracking device. A questionnaire was used to determine cycling experience and the perception of cycling and anticipation skill in traffic. Despite a longer period to master the motor skill of cycling, individuals with DCD reported to be able to safely cycle in traffic around the same age as TD young adults. In the hazard perception test, individuals with DCD fixated the hazards later, less frequently and for a shorter duration than the TD participants, however, the participants with DCD did not wait longer to react to the hazard than the TD participants. Interestingly, individuals with DCD rated the traffic situations in the test as significantly more dangerous than the TD participants. In conclusion, the differences exposed in the hazard perception test may imply an increased risk of accidents in individuals with DCD. In further research and practice it is recommended that both the motor and the perceptual aspects of cycling are addressed.

Keywords: developmental coordination disorder, hazard perception, cycling, traffic safety, gaze behavior, young adults

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INTRODUCTION

Navigating safely through traffic, whether as a car driver, cyclist or pedestrian, depends on cognitive, and perceptual-motor processes. Irrespective of the transport mode, it is important to correctly assess the situation at any time. According to Endsley's (1995) concept of "situational awareness" this entails three levels: the perception of the environment and events with respect to time and space, comprehension of their meaning and projection of their future states. It is clear that what at first glance occurs unconsciously and automatically is actually a very complex task. Hence, it is not surprising that individuals who experience problems with aspects of these cognitive or

perceptual-motor processes (e.g., ADHD or ASD) also have difficulty assessing traffic situations (Clancy et al., 2006; Cowan et al., 2018; Wilmut and Purcell, 2021). A group that deserves the necessary attention in this regard is individuals with developmental coordination disorder (DCD). DCD is an idiopathic neurodevelopmental disorder characterized by significant impairments in motor coordination and learning.

Individuals with DCD have difficulties with perceptual function (for review see Wilson and McKenzie, 1998), oculomotor function (Warlop et al., 2020), executive function (Tsai et al., 2012), and forward modeling (for review see Wilson et al., 2013) all of which are core abilities within the situational awareness model. For example, when it comes to level 1 (perception of the environment and events) Purcell et al. (2012) demonstrated reduced looming sensitivity in children with DCD when observing cars approaching as a pedestrian. This deficit may then lead to choosing inadequate crossing gaps, as found in a follow-up study of Purcell et al. (2017), indicating lack of comprehension of the meaning of the perceptual input (level 2) and/or underlying problems with projection of future states (level 3). It is also worth noting that individuals with DCD are found to have reduced working memory capacity (Alloway, 2011). This may result in lower performances in high cognitively demanding tasks like a hazard perception test, as shown by Wood et al. (2016). Remarkably, recent research shows that both children and adults with DCD perceive road crossing, as a pedestrian, as a more challenging task than typically developing (TD) peers (Wilmut and Purcell, 2020), which indicates that the individuals are to some extent aware of the risk associated with their perceptual-motor problems. In this respect, in this important to highlight that individuals with DCD also perceive themselves as less competent car drivers and avoid active participation to traffic (Kirby et al., 2011a). Ultimately, individuals with DCD may end up in a negative spiral, given that experiential learning is essential in the education of situational awareness.

In the current study, we build upon this line of research and examine individual's with DCD ability to perceive hazards in traffic while cycling. Hazards in traffic, especially when cycling, are ubiquitous and can be both static (e.g., curbs and potholes) and dynamic (approaching cars or pedestrians crossing the road). By definition, hazard perception involves the three levels of the situational awareness model (Wetton et al., 2011; Vansteenkiste et al., 2016), and therefore requires adequate perception, recognition, and projection of the environment and events. Previous research has shown that gaze behavior (i.e., visual search) is a crucial factor in this process. For example, Zeuwts et al. (2016) showed that young learner cyclists fixate the hazards later, and have slower reaction times than experienced adult cyclists. Also in car driving, effective hazard perception performance appears to depend on how quickly the hazards were fixated (Crundall et al., 2012). Furthermore, when cognitive load is increased, individuals with low working memory capacity fixate less on the hazards, resulting in slower reactions to hazards (Wood et al., 2016).

Perception of hazards is quintessential to ensure safety to the individual, and hence, it is important to have insight into the performance of individuals with DCD in this matter, whom we know have underlying deficits that may put them at risk. This will be investigated with a standardized hazard perception test, in which both gaze behavior and reaction (time) are examined. Based on previous reports, e.g., on reduced looming sensitivity or forward modeling, and consistent with findings of immature gaze behavior in children, we expect less efficient visual search with later fixation and longer reaction times in individuals with DCD. Given the critical role of the perception of risk and the perceived competence of cycling skill in the individuals' decision to actively engage in traffic these factors were also documented.

MATERIALS AND METHODS

Participants

Eighteen adults aged between 19 and 30 years old participated in the current study. Nine of these participants were clinically diagnosed with DCD as a child by a pediatrician and recruited via social media and a database of participants that were involved in previous studies (Deconinck et al., 2006a,b). One participant with DCD was excluded after testing due to insufficient tracking accuracy in the HP-test (details of the included participants shown in **Table 1**). The control group, recruited via convenience sampling, consisted of nine TD individuals who have never been diagnosed with a neurodevelopmental disorder or medical condition that could affect motor behavior. All participants with DCD complied to the diagnostic criteria as described in the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association, 2013). For example, their motor skills were below that expected according to their age (criterion A). This was assessed, as part of this study, with the Movement Assessment Battery for Children-2 (MABC-2; Henderson et al., 2010), which is designed and norm-referenced up to the age of 16. This test battery discriminated between poor and normal motor competence in previous studies in young adults with DCD (Wilmut et al., 2013; Du et al., 2015), and was therefore considered suitable for this study. Age band 3 and the reference values of the 16 olds were used to determine the participants' percentile scores. Two participants with DCD scored at the 25th percentile, which is above the cut-off value for "at risk for DCD." However, they both scored high on the Adult DCD Checklist (ADC; Kirby and Rosenblum, 2008; Kirby et al., 2011b), which assessed past motor difficulties in childhood (section 1 of the checklist; DSM criterion C) and current daily

TABLE 1 | Summary of the participant's characteristics (mean ± SD).

	DCD (*N* = 8)	TD (*N* = 9)
Gender (number of participants)	3 male 5 female	3 male 6 female
Age (year)	23.0 ± 3.8	24.6 ± 3.5
Dominant hand (number of participants)	4 left 4 right	2 left 7 right
MABC-2 percentile	9.3 ± 11.0	68.9 ± 22.8
ADC score section 1	22.9 ± 3.9	1.4 ± 1.1
ADC score section 2	53.1 ± 16.9	10.1 ± 8.6

motor functioning (section 2 of the checklist; DSM criterion B). A score of 17 or higher on the first section of this checklist is indicative for "probable DCD." In the TD group, all participants scored at or above the 25th percentile of the MABC-2 and had a maximum score of 3 on the first section of the ADC. None of the participants reported to have neurological conditions affecting movement (other than DCD in the DCD group; DSM criterion D), and they all had normal or corrected-to-normal vision. The study was approved by the Ethics Committee of the Ghent University Hospital and conducted in accordance with the Declaration of Helsinki.

Apparatus

Cycling Experience and Perceived Cycling Skills Questionnaire

To get a better understanding of the participant's cycling experience, the following cycling milestones were assessed: the age at which the participants started to learn how to ride a bike, the age at which they mastered the motor skill of independently riding a bike, and the age that they were able to safely cycle in traffic. Also, the participants' total years of cycling experience and how frequently they cycle was surveyed. To get an indication on the participants' perception of their own cycling skills the following three questions were added to the questionnaire: a yes-no question on if they thought they could safely cycle in traffic, and two 5-point Likert scale questions assessing how well they perceived their cycling ability and how well they anticipated hazards in traffic (ranging from "not good" to "very good"). From all questionnaire measures, raw data were used for statistical analysis.

Hazard Perception Test

For the development of the HP-test, videos of real-life traffic situations were recorded by cycling through traffic in two Flemish cities with a GoPro Hero3 (30 Hz, full HD and 170° field of view) mounted on a helmet. Some traffic scenarios were staged using volunteers to safely create hazardous situations in calm streets. The recordings were made only on straight streets while constantly looking forward and not turning the head towards specific objects, side streets or other road users. Recordings with head movements were excluded. All clips were stabilized to reduce vibrations resulting from the state of the bicycle path or small head movements using dedicated video stabilization software "Mercalli V2" (ProDad). For this study, a selection of 14 video fragments with a duration of 10–50 s was made. The videos were filmed from the cyclist's point of view and all contained at least one hazard (nine videos contained one hazard, two contained two hazards, and one contained four hazards). A hazard was defined as a traffic situation which exposes the cyclist to an increased possibility of an accident and makes the cyclist brake or change direction in order to avert this accident. Both behavioral predication (BP) hazards (i.e. objects or road users that are already visible prior to developing as a hazard), and environmental prediction (EP) hazards (i.e. potentially hazardous situations that are not visible before the actual hazard occurs, yet, are inferable from other objects than the one causing the hazard) were included (Crundall et al., 2012). See the **Supplementary Material** for an example fragment and **Supplementary Table 1** for a description and details of the included video clips and hazards. This type of test was used by Vansteenkiste et al. (2016) and Zeuwts et al. (2016) and proved useful in testing differences between adults and children. The HP-test was carried out with the Remote Eye Tracking Device (RED) of SensoMotoric Instruments (Teltow, Germany), which registered the participant's gaze during the test. The video fragments were shown on a 22-inch computer screen underneath which the eye tracking device was mounted. Two beams of infrared light illuminated the eyes and the reflections on the cornea were captured by an infrared camera to determine the position of the pupils and hence the direction of the gaze. The system has a manufacturer-reported accuracy of 0.4°. A laptop, running the Experiment Center 3.4 software, was connected to the device and recorded the gaze data at a binocular sampling rate of 120 Hz.

Procedure

Prior to the HP-test, the participants filled in a short questionnaire on their cycling experience and how they perceived themselves as a cyclist. Then, they were asked to take place in front of the screen equipped with the RED. Their position was adapted so that the distance between their eyes and the screen was between 60 and 80 cm, resulting in a visual angle ranging between 24 and 32 degrees (vertical), 31–41 degrees (horizontal). Once the participant was seated comfortably and the device was capturing their eye well, a 5-point calibration was done. When the calibration did not result in an accuracy below 0.6° it was repeated. If this accuracy was not achieved after five trials of calibration, the test was continued with the best possible calibration. Although the RED is quite resistant for small movements, the participants were asked to stay in the same position throughout the experiment to assure good recording of the gaze behavior. At the end of the test, a calibration check was done. The participants were instructed to observe the videos and imagine that they were cycling themselves. They were asked to click the mouse when they would use the brakes, change direction or stop for a hazard. After each fragment the participants were asked how safe the traffic scenario was from the perspective of the cyclist on a 5-point Likert scale ranging from "not hazardous" to "very hazardous." A total of 14 videos with a duration of 20–30 s were displayed, resulting in a total test duration of 10–15 min for the HP-test.

Analysis

Gaze Behavior

Prior to quantitative analysis, the quality of the gaze data was assessed. Data of one participant with DCD was deleted, due to insufficient tracking accuracy (8.61°) throughout the test. The averaged accuracy of the included data was $0.74 \pm 0.33°$. Second, the tracking ratio, which is the percentage of time that eye movements were effectively measured, was evaluated. Trials were excluded when tracking ratio was lower than 80% (Vansteenkiste et al., 2016). For this reason, nine trials (over three participants) were excluded from further analysis. In addition, one video clip was excluded in all participants as none of the participants

reacted to the hazard, so it did not seem to be perceived as hazardous by any of the participants. Finally, in one video the hazard was detectable from the very start of the fragment, which resulted in very different reaction times across the subjects. It was unclear what information or which cues led to the responses, so it was decided to exclude this fragment as well, resulting in 12 video clips included in the statistical analysis. In BeGaze 3.7 (SensoMotoric Instruments, Teltow, Germany), fixations were determined using the SMI fixation detection algorithm. In each video clip the hazards (specified in **Supplementary Table 1**) were determined and indicated as Areas Of Interest (AOI) using the dynamic AOI editor. The AOI's were polygons around the hazards that changed shape and size dynamically along with the movement and looming of the hazard in the video clip. Then, the number of fixations, the duration of the fixations, the duration of the first fixation on the AOI, dwell time (i.e., the total time spent fixating on an AOI), and the timing of the first fixation on the hazard relative to the appearance of the hazard, were calculated per AOI. As the nature and the duration of the traffic situations and the hazards varied between the fragments, *z*-scores were calculated of all gaze behavior measures using the means (M) and standard deviation (SD) of the TD control group per AOI: $z = \frac{Raw\ score - M_{TD}}{SD_{TD}}$. Finally, for each gaze behavior variable, the average of the *z*-scores of all AOI's was calculated.

Response Rate and Reaction Time

Response rate, which referred to the number of hazards that the participants clicked for within the time interval that a hazard was visible on the video clip, was counted and expressed in relation to the total number of hazards. In addition, extra clicks, i.e., clicks before or after the time interval related to the hazard, or additional clicks within this time interval, were summed across all trials. Reaction time was measured in ms from the first appearance of the hazard. As different hazards had different lengths of intervals during which the hazard was visible, reaction time was strongly dependent on the nature of the video. Therefore, reaction times per AOI were also converted into *z*-scores in a similar way to the gaze behavior metrics and the average of the *z*-scores of all AOI's was calculated.

Statistics

To assess the criteria for parametric testing, Kolmogorov–Smirnov tests were conducted for normality and Levene's tests were performed to assess the homogeneity of variance. For normally distributed data with equal variances, independent samples *T*-tests were carried out to investigate differences between TD and DCD on all variables. In the instance of not normally distributed data, non-parametric Mann–Whitney U tests were conducted. In the instance of unequal variances, Welch's corrections were applied. The alpha level was set at 0.05 and effect sizes were reported as Cohen's *d*, which was calculated as: $d = \frac{M_{DCD} - M_{TD}}{SD_{TD}}$. Indicative thresholds for Cohen's *d* are small (0.2), medium (0.5), and large (0.8; Field, 2018). No distinction was made between the BP and the EP hazards as it was no primary aim of this study and due to the small number of BP trials and the small sample size.

RESULTS

A detailed representation of the gaze behavior and response rate data of DCD and TD participants per hazard, can be found in **Supplementary Table 2**.

Cycling Experience and Perceived Cycling Abilities

Results from the questionnaire indicated no difference in the age that children started to learn how to cycle between the DCD group (5.13 ± 1.46) and the TD group [4.44 ± 1.01; $t(15) = -1.129$, $p = 0.277$, $d = 0.671$]. However, the participants with DCD reported to have mastered to motor skill of biking significantly later (6.63 ± 2.07) than the TD participants [4.83 ± 1.00; $t(15) = -2.320$, $p = 0.035$, $d = 1.792$]. One participant with DCD reported that she was, at the time of the test, still not able to safely cycle in traffic as an adult. The remaining participants with DCD indicated they were able to safely cycle in traffic since the age of 9.86 ± 3.29, which did not significantly differ from that of the TD group [10.44 ± 1.74; $t(8.593) = 0.428$, $p = 0.679$, $d = -0.338$]. The TD individuals reported to have, on average, more years of cycling experience (19.67 ± 4.03) than the DCD group [15.25 ± 5.73; $t(15) = 1.857$, $p = 0.083$, $d = -1.096$]. Furthermore, TD participants appeared to cycle more often (4.48 ± 2.56 times per week) compared to their DCD counterparts (2.93 ± 2.75), but no significant effect was detected [$t(15) = 1.199$, $p = 0.249$, $d = -0.603$]. As to the perception of cycling ability, the participants with DCD (3.25 ± 1.04) perceived themselves as significantly less proficient cyclists on the 5-point Likert scale than the TD group (4.56 ± 0.53; Mann–Whitney U = 10.500, $p = 0.010$, $d = -2.477$). Finally, their perception of their anticipation skills in traffic when cycling is significantly below (3.38 ± 0.92) that of the TD participants (4.56 ± 0.53; Mann–Whitney U = 10.500, $p = 0.010$, $d = -2.240$).

Hazard Perception Test

Gaze Behavior

Descriptive statistics for of the gaze behavior variables are presented in **Table 2**. Interestingly, in 21.6% of all hazards presented to the DCD participants, the hazard was not fixated

TABLE 2 | Average *z*-score values for the gaze behavior variables (mean ± SD).

	DCD	TD	*t*	df	*p*	Cohen's *d*
Number of fixations	−0.57 ± 0.73	0.00 ± 0.46	3.211	15	0.006*	−2.235**
Timing first fixation	0.65 ± 0.58	0.03 ± 0.41	−2.606	15	0.020*	1.538**
Average fixation duration	0.18 ± 0.58	−0.01 ± 0.58	−0.665	15	0.516	0.325
First fixation duration	0.22 ± 0.50	−0.03 ± 0.39	−1.185	15	0.254	0.656
Dwell time	−0.63 ± 0.49	0.00 ± 0.46	2.727	15	0.016*	−1.367**

*Significant: $p < 0.05$.
**Large effect size: $|d| > 0.8$.

at all, whereas this was only the case in 6.5% of the hazards in the TD group. The DCD group used significantly less fixations compared to the TD group and the participants with DCD appeared to fixate the hazard significantly later than the TD group. No differences were found between the groups for the average duration of all fixations or the duration of the first fixation on the hazard. However, dwell time on the hazards did differ between the groups, with the individuals with DCD spending less time fixating the hazards than the TD participants.

Response Rate and Reaction Time

Individuals with DCD clicked for 85.29 ± 16.03 percent of the hazards, which did not differ from the response rate of the TD participants [76.47 ± 16.38; $t(15) = -1.120$, $p = 0.280$, $d = 0.539$]. In addition, the participants with DCD tended to make more extra clicks (5.00 ± 4.66) than the TD participants (2.11 ± 2.80), however, this difference was not significant [$t(15) = -1.571$, $p = 0.137$, $d = 1.030$]. On most of the hazards, individuals with DCD seem to respond later (z-score: 0.64 ± 1.32) compared to the TD group (0.11 ± 0.66). However, despite a large effect size, there was no significant difference on this variable [$t(15) = -1.071$, $p = 0.301$, $d = 0.807$].

Perception of Safety

The participants with DCD rated the traffic situations in the videos as significantly more dangerous (3.19 ± 0.44) compared to the TD individuals [2.20 ± 0.77; $t(15) = -3.191$, $p = 0.006$, $d = 1.284$].

DISCUSSION

The current study explored if young adults with DCD perceive and react to traffic hazards differently than TD participants. Individuals with DCD fixated the hazards later than the TD participants, made fewer fixations on the hazards, and spent less time fixating them. However, no significant differences in response rate or reaction time were found.

The questionnaire on cycling experience and perceived cycling abilities revealed that individuals with DCD took more time to learn to ride a bike. However, they also indicated to be able to safely cycle in traffic at around the same age as the TD individuals. Furthermore, the participants with DCD had less experience in cycling and they rated themselves as significantly less proficient cyclists than their TD counterparts. Also, the DCD group rated its anticipation skills to be worse than the TD group. This corresponds with the findings on road crossing, where, over half of the adult respondents with DCD indicated to be not or only somewhat confident in road crossing (Wilmut and Purcell, 2020). It therefore seems reasonable to assume that for individuals with DCD, the issues that have been reported in road crossing will also persist in cycling and other modes of transportation.

It is interesting to note that while gaze behavior was different, i.e., later fixation and shorter dwell times on the hazards in the adults with DCD, no difference was found in the reaction to the hazard. The implication is that the time between the first fixation and reaction to the hazard was longer in TD individuals than in their counterparts with DCD. Judging traffic requires a continuous cycle of perception, appraisal, and prediction of a multitude of visual cues. The advanced first fixation and longer dwell times of the TD adults, therefore, suggest a better "situational awareness" in this group, with a more goal-directed visual search strategy (i.e., toward potential hazards) and more revisits of regions that may be or become potentially hazardous. The advantage of early recognition of an object or event that will become a hazard, is that one has time to anticipate. However, the lack of differences in reaction time between DCD and TD indicates that individuals with DCD seem to not have problems with recognizing an object or event as dangerous and reacting to it. The finding that they pick up hazards later and are less attentive to what may develop as hazardous later on, may suggest poor predictive abilities in hazard perception in DCD. The implication is that these individuals would need to react to dangers, rather than being in a position where they can anticipate. Although no differences were found in the number of extra clicks between the groups, the higher total number of clicks in the DCD group may suggest that they react more to anything that may be hazardous, rather than anticipating actual hazards. It is likely that this will contribute to an increased perception of risk in DCD, as found in this study and consistent with Wilmut and Purcell (2020).The differences in gaze behavior may also be influenced by other factors. First, individuals with DCD are known to demonstrate oculomotor problems which might have impacted on the saccadic behavior in this task (Sumner et al., 2018). These oculomotor deficits have been found to surface in other daily tasks. For example, Wilmut et al. (2006) found a delayed initiation of eye movements in a sequential pointing task, and, in catching, children with DCD require more time to fixate and track the ball (Licari et al., 2018). Secondly, the gaze behavior in the hazard perception task may also be reflective of an increased need to focus on the path, as found in adults with DCD while walking (Warlop et al., 2020). If this would also be the visual strategy used in cycling in individuals with DCD, it may have distracted them from detecting hazards, as these usually occur further down the road. This suggestion should be subject to further research. Thirdly, it should be noted that the videos used in the current test were recorded on TD adults. As individuals with DCD are known to adopt compensatory strategies, they might also cycle slower than TD individuals. As a consequence, the videos shown in the current test could reflect a "normal" optical flow for a TD individual, whereas it displayed a faster flow than what individuals with DCD are used to, which might have led to a less appropriate gaze behavior in the DCD group. Fourthly, the difference in gaze behavior might also be caused by a lack of cycling experience in the DCD group. Young inexperienced cyclists were found to have delayed first fixations and slower reaction times to hazards than experienced adults (Vansteenkiste et al., 2016; Zeuwts et al., 2016), so the "immature" gaze behavior of individuals with DCD might be a reflection of a lack of experience with traffic too. Finally, the altered gaze behavior may be related to the cognitive requirements of the task. The hazard perception test may be more demanding for individuals with DCD, who are known to have poorer working memory capacity

(Alloway, 2011). Similar results, with later fixations and reduced fixation times on the hazards, were found in hazard perception tests under increased cognitive load (Wood et al., 2016). In contrast to our study, this resulted in reduced hazard perception performance. It is possible that with higher cognitive load, with for example addition of a motor component, the differences in gaze behavior found in this task may be accompanied by reduced hazard perception performances in individuals with DCD.

In this study, we investigated the hazard perception skills of DCD, while neutralizing the motor challenges related to cycling. The benefit of this approach is that it enabled us to demonstrate that in terms of perception, comprehension, and projection of visual cues and events alone, individuals with DCD already experience problems. In combination with their motor problems, this may lead to a higher risk of accidents during cycling in traffic. The disadvantage of the current paradigm is of course that the motor response was limited to a mouse click. In cycling, leg movements need to be coordinated with accurate arm and hand movements, while balancing on the bike and responding to dynamic traffic situations. This does not only add to the motor difficulty of the task, but also increases the cognitive load, which may negatively impact both the gaze behavior and the reactions toward hazards in traffic (Wood et al., 2016). To get a better understanding of the problems in cycling with DCD, future research should consider more complex tasks with a combination of both the perceptual and motor aspect of the task. Also, while our findings are supported with large effect sizes, it must be acknowledged that the sample size is relatively small. As DCD is a heterogeneous disorder, we recommend future studies to include more participants and to broaden the age range of the sample to children as well.

CONCLUSION

In conclusion, the gaze behavior of young adults with DCD differs from that of TD individuals in a hazard perception task, characterized by a delayed fixation on hazards, fewer fixations and less time spent fixating the hazards. It is unclear whether this altered gaze behavior causes an increased risk for accidents, as no differences were found in the reactions to the hazards. However, it does indicate that not only the motor difficulties should be taken into consideration in therapy for cycling. This is all the more important as the perceived risk experienced by the adults with DCD may lead to withdrawal of active participation in traffic. As cycling is part of a healthy lifestyle and an increasingly important means of transport, future studies should investigate interventions targeting the specific problems highlighted in this study.

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 human participants were reviewed and approved by the Ethics Committee of Ghent University Hospital. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

PV, FD, and ML contributed to the conception of the study and critically reviewed the manuscript. PV designed the study protocol. GW collected and analyzed the data and wrote the first draft of the manuscript. All authors approved the final version of the manuscript for submission.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2021.665189/full#supplementary-material

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Effects of Graded Exergames on Fitness Performance in Elementary School Children With Developmental Coordination Disorder

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Developmental Coordination Disorder (DCD) is a common childhood disorder affecting movement and coordination skills, fitness, and academic performance. Increased physical fitness may have a positive influence on physical and mental health outcomes in children with DCD. Yet, little has been done to develop interventions to improve fitness performance in this group. The purpose of this study was to determine the effects of graded exergames in 7 to 12-year-old children with DCD and typically developing (TD) peers. Participants (32 DCD and 28 TD children) received a 30-min training session twice weekly for 10 weeks. Performance on motor coordination (MABC-2 test), balance, aerobic, and anaerobic fitness tests were assessed at the beginning and end of training. In addition, enjoyment and perception of exertion were measured for each participant during the training period. Both children with DCD and TD children significantly improved on motor coordination, balance, aerobic, and anaerobic fitness at the end of the training. A significant Group by Time interaction was observed on the MABC-2 total [$F_{(1,55)} = 13.19$; $p < 0.001$] and balance scores [$F_{(1,55)} = 26.83$; $p < 0.0001$], with the DCD group demonstrating larger improvements than the TD children. Both groups enjoyed the program throughout the training period even though they rated the training to be of high intensity. These findings indicate that graded exergames may serve as potential treatment for impaired fitness in children with DCD. Regular participation in graded exergames in school settings may be needed to enhance and maintain fitness performance in young children with and without DCD.

Keywords: fitness performance, children, active video game, developmental coordination disorder, training intensity, balance, agility

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INTRODUCTION

Children with developmental coordination disorder (DCD) exhibit severe motor clumsiness that interferes with academic achievement and the activities of daily living (American Psychiatric Association, 2013). DCD occurs in ∼5–6% of children worldwide and is not explained by medical conditions or low IQ (American Psychiatric Association, 2013). Poor fitness performance is well-documented in children with DCD in both high- and low-income countries (Faught et al., 2005; Tsiotra et al., 2009; Rivilis et al., 2011; Ferguson et al., 2014; Lifshitz et al., 2014; Farhat et al., 2015).

In general, children with DCD are more likely to have decreased physical fitness compared to peers with typical development (TD), perhaps due to an activity deficit resulting from poor motor proficiency and withdrawal from physical activity (Hay and Missiuna, 1998; Tsiotra et al., 2009; Cairney et al., 2017). Despite the enormous number of interventions developed to address DCD symptomatology, little research has been done to address DCD-related fitness impairments. Searching for effective methods to increase fitness performance is critical if we are to improve health and wellness in children with DCD.

Physical fitness consists of a set of measurable characteristics that people gain through various physical efforts (Deuster, 1997; Corbin et al., 2000). These include components such as cardiovascular endurance, body composition, muscle strength, endurance, flexibility, balance, coordination, agility, and reaction time (Deuster, 1997; Corbin et al., 2000). Among children, higher levels of physical fitness have been associated with healthy body weight, optimal psychological and bone health, and lower risk for obesity and cardiovascular diseases (Boreham et al., 2001; Biddle et al., 2004; Ortega et al., 2008; Janssen and LeBlanc, 2010). Additionally, adequate physical fitness is positively associated with high academic achievement (Castelli et al., 2007; Wittberg et al., 2012). Despite these benefits, recent studies have shown a decline in physical fitness among children worldwide (Lang et al., 2018; Tomkinson et al., 2019). It is therefore necessary to maximize efforts that will ensure that children of all abilities and socioeconomic backgrounds are provided with an opportunity to increase physical fitness and to participate in meaningful daily activities (Faigenbaum et al., 2011).

Exercise is the most frequently used treatment for children with DCD. Previous exercises that have been tested in individuals with DCD vary in type, intensity, frequency, and duration (Smits-Engelsman et al., 2018). Two very different popular approaches to exercise in the DCD literature are task-oriented functional exercises and active video games or exergames (Blank et al., 2019). While task-oriented exercises have been reported to produce greater improvements in motor coordination compared to exergames, evidence shows that exergames can serve as a useful adjunct to therapy (Blank et al., 2019). Unfortunately, there is currently little or no evidence to guide caregivers (health professionals, educators, coaches/physical trainers) to select the most effective type of exercise for the treatment of impaired physical fitness in young children with DCD. This lack of evidence suggests that treatment for poor fitness performance may be sub-optimal.

Active video games have been proven to be safe, engaging, enjoyable, and beneficial for improving motor coordination in children and adolescents with DCD (Ferguson et al., 2013; Hammond et al., 2014; Bonney et al., 2017a; Mentiplay et al., 2019). More recently, graded active video exercises have been demonstrated as a feasible approach to promoting physical fitness in youth with DCD (Bonney et al., 2018). However, we are not aware of any published research that has tested graded active video exercises in elementary school-aged children with or without DCD. Therefore, the purpose of this study was to determine the effects of graded active video exercises on health- and skill-related fitness measures in children with DCD and typically developing peers. Specifically, the following objectives were pursued: (1) to determine whether elementary school-aged children would tolerate graded active video games, (2) to quantify exercise intensity during the training period, and (3) to examine changes in physical fitness and motor performance at pre- and post-intervention.

MATERIALS AND METHODS

Study Design

This study was conducted as a pre-post design.

Participants

Thirty-two ($n = 32$) elementary school children (aged 7–12 years) with DCD (15 boys, 17 girls) and twenty-eight ($n = 28$) age- and gender-matched TD children (13 boys, 15 girls) attending school in an economically-deprived area of Cape Town, South Africa, participated in this study. The study was approved by the Human Research Ethics Committee (HREC) of the University of Cape Town, South Africa (HREC: 209/2018). Parents provided written consent, and each child gave assent before participation. Sample size was established with G*Power 3.1. (Faul et al., 2007) based on the assumption of an effect size of 0.7 and at least 90% power. Thus, a sample size of 24 children per group was deemed adequate to examine the hypothesis. All 32 children meeting the DCD criteria were offered the intervention.

Procedure

Identification of Participants

Participants were identified using a three-step process (see **Figure 1**). First, children were tested on the 20 m shuttle run (SR) test after consent procedures had been completed. Second, children who performed below the 20th percentile on the SR test (fitness performance was evaluated based on VO_2 max percentile scores published by Kolimechkov et al., 2019) and whose parents had completed a demographic questionnaire were assessed on the Movement Assessment Battery for Children, second edition (MABC-2 test) (Hendersen et al., 2007). The parental demographic questionnaire (Ferguson et al., 2013; Bonney et al., 2017b) asked about the pregnancy history of the child's mother, presence of visual, auditory, intellectual, or motor difficulties, established medical diagnoses (e.g., cerebral palsy), and whether the child experienced movement difficulties. Last, children whose parents gave informed consent to undergo additional testing were further evaluated for DCD using the DSM-5 diagnostic criteria (American Psychiatric Association, 2013) and were asked to take part in the training. Briefly, children with movement difficulties reported by their parents and who scored below the 16th percentile on the MABC-2 test; whose parents reported no diagnosis of a significant medical condition known to affect motor performance, visual, auditory, or attentional problems; and whose teacher confirmed the absence of cognitive impairment were identified as having DCD. The children with DCD were then matched with typically developing (TD) peers whose motor performance was ≥25th percentile on the MABC-2 test. The TD children had no problems with academic work (as confirmed by their class teacher), and their parents reported

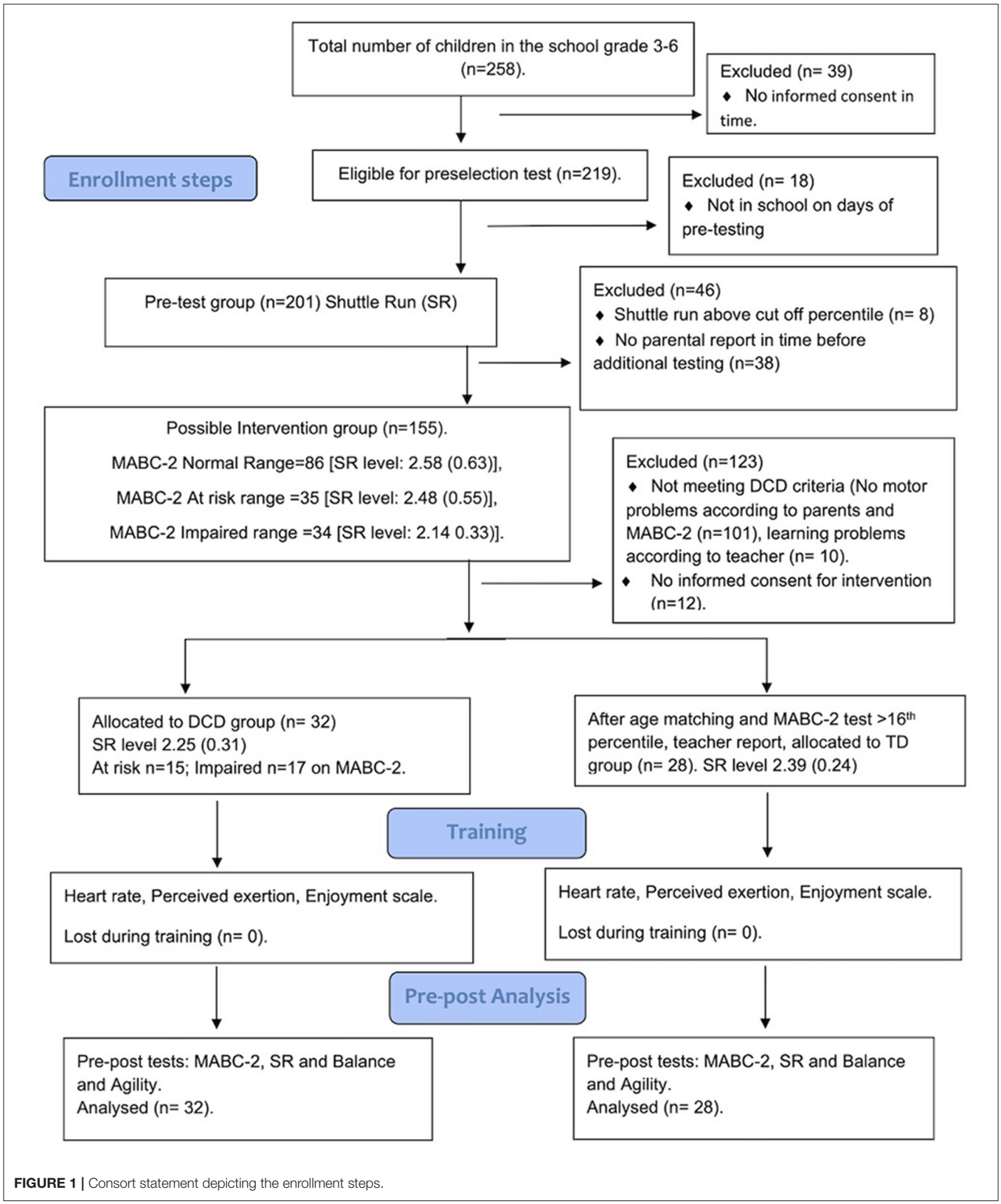

FIGURE 1 | Consort statement depicting the enrollment steps.

no diagnosis of a significant medical condition known to affect motor performance. For ethical reasons, we invited all children who met the DSM-5 diagnostic criteria for DCD to participate in the study. However, children were excluded if they had any documented medical condition that hindered their participation in the training. Selected children were also assessed on the balance and agility tests described below.

Assessments

All pre- and post-tests were administered by a team of trained physical therapists not involved in the training. Testing was spread over several days (14 days) to avoid fatigue. The MABC-2 and Foam Balance tests were administered individually. Sprints and agility tests were performed in pairs at the school's playground. For these tests, children were tested in alternating. Thus, while one child was being tested, the other was allowed to rest. The 20 m shuttle run test was done in groups of four or five children supervised by three or four testers.

Post-tests were planned to take place in the last 3 weeks before the school holidays. However, several unanticipated events occurred in this period, including water shortages, poor sanitation, interruption of electricity supply, and unanticipated community protest action regarding poor municipal service delivery resulting in the temporary closure of the school over safety concerns. Consequently, post-testing was stopped after the first week of testing. Furthermore, no testing was allowed after the children returned to school to complete the last quarter of the academic year. This is because research activities are usually not permitted in schools at this time of the year in the Western Cape Province of South Africa. In view of this, many of the children had incomplete data. We did not exclude children with incomplete test results because at least one post-test result was available for each child (see **Table 1** for details of the study design).

TABLE 1 | Study design.

Activity	Description	Duration
Pre-selection assessments	Children were assessed on SR and MABC-2 tests	2 weeks
Pre-training assessments	Children were assessed on balance and agility performance	2 weeks
Training	Week 1: Gaming with no extra challenge	
	Week 2: Gaming with no extra challenge	
	Week 3: Gaming on Airex Mat (L 50 × W 40 × H 1.5 cm)	
	Week 4: Gaming on Airex Mat (L 50 × W 40 × H 1.5 cm)	
Holidays	Students went on holidays; training was halted during this period	2 weeks
	Week 5: Gaming on Foam pad (L 47 × W 39 × H 6 cm)	
	Week 6: Gaming on Foam pad (L 47 × W 39 × H 6 cm)	
	Week 7: Gaming on Foam pad (L 47 × W 39 × H 6 cm) plus 1 kg vest	
	Week 8: Gaming on Foam pad (L 47 × W 39 × H 6 cm) plus 1 kg vest	
	Week 9: Gaming on Foam pad (L 47 × W 39 × H 6 cm) plus 2 kg vest	
	Week 10: Gaming on Foam pad (L 47 × W 39 × H 6 cm) plus 2 kg vest	
Post-training assessment	Children were assessed on MABC-2, SR, balance, and agility tests	3 weeks

Measures Taken at Pre- and Post-Training

Shuttle Run Test

The Shuttle Run (SR) test was used to evaluate aerobic fitness performance. The test was performed in accordance with recommendations proposed by Leger and Lambert (1982). The highest level achieved by participants was recorded and used in the analysis. The SR test has been demonstrated to have good test-retest reliability (ICC = 0.93) and validity ($r = 0.72$) and had been used in an earlier study involving South African children (Ferguson et al., 2013).

Movement Assessment Battery for Children Test, Second Edition (MABC-2 Test)

The MABC-2 test measures motor performance in children aged 3–16 years (Hendersen et al., 2007). The test involves eight motor tasks divided into three categories: manual dexterity, aiming and catching, and balance skills. The raw scores for each category were converted to standard scores and percentiles, and these values were summed to generate overall percentile scores. Percentiles can be interpreted as follows: normal motor development (≥25th percentile), being at risk for motor difficulties (5th percentile < x ≤16th percentile), or having significant motor difficulty (≤5th percentile) (Hendersen et al., 2007). The MABC-2 test has demonstrated good validity and test-retest reliability with ICC values ranging from 0.92 to 0.98 (Hendersen et al., 2007).

Ladder Agility Test (LAT)

The LAT was used to measure children's agility performance (Smits-Engelsman et al., 2019). The LAT consists of two off-the-shelf 4 m agility ladders. These ladders were adapted by moving the 10 crossbars and fixating them at different distances to create two different ladders, "normal" and "accuracy." For the "normal ladder" all the squares had equal sizes; the 10 3-cm broad yellow bars were separated by 36 cm. The "accuracy ladder" had unequal spaces between the bars to increase the spatial demands of the task. The first square was 44 cm in length, and the distance between the bars in successive squares decreased by 2 cm (44, 42, 40, 38, 36, 34, 32, 30, and 28 cm). Hence, both ladders had equal length (354 cm).

Both ladders with rungs of nine squares were positioned on the floor, and a designated turning point was marked 50 cm at one end of the ladders. From one end of the ladder, participants were instructed to step into each square as quickly as possible, make a 180° turn at the other end, and run back to the starting position. Participants began with both feet behind the crossbar of the first square and completed the test when they got back to the starting point. Upon hearing the "go signal," the child was required to run forward using the required stepping pattern

(single run: one foot in each square, or double run: two feet per square), turn at the turning point demarcated with a colored cone, and return to the starting point by following the same running pattern (Smits-Engelsman et al., 2019). Each participant performed two repetitions on each of the four items: type of ladder (normal and accuracy) with each running pattern (single or double run). Children had a 30 s rest between the two ladder types to ensure adequate recovery. The time taken to complete a full lap on each trial was recorded. Also, the number of mistakes produced during testing was recorded. A mistake was defined as either missing a square, placing the wrong foot or feet in the squares, and/or stepping on the bar separating two consecutive squares. The maximum number of mistakes allowed was three. If three or more mistakes in one run were made, an extra trial was given.

Foam Balance Test

Participants' balance was assessed using the foam balance test. For this test, a child was required to assume a one-legged stance on an Airex balance foam [i.e., a high-density closed-cell foam pad (47 × 39 × 6 cm, 0.7 kg)]. Participants' performance was timed using a stopwatch. Each subject completed two trials per leg in a maximum of 20 s. A 10-s rest was allowed between trials. A maximum of 5 s practice session was allowed prior to the start of each test so that subjects could gain some familiarity with the support surface. For all trials, the subjects placed their hands on their hips, and timing started when the opposite foot was lifted from the floor and stopped when the child could no longer control his or her posture or dropped the elevated foot. The best time recorded (in s) was used for the analysis.

10 × 5 m Sprint Test-Straight

For this test, each participant was required to perform 10 quick runs over a distance of five m without stopping (Bonney et al., 2019). Colored cones were used to demarcate a 5-m running course in a designated hall. The participant begins at the starting point and runs toward the opposite end as quickly as possible. After every 5 m, the participant turns around and continues to run until 10 laps are completed. The time used to complete 10 laps was recorded (measured in s). This test was conducted individually under the supervision of a trained assessor. The test has good reliability in typically developing children (Bonney et al., 2019).

The Functional Strength Measure (FSM)

One upper (overarm throwing of a sandbag) extremity item and one lower extremity (standing-long-jump) item of the FSM (Aertssen et al., 2016) were used to assess functional muscle strength. These test items assess maximal explosive power (distance in cm). The FSM test has excellent psychometric properties in this population (Aertssen et al., 2016).

Körperkoordinationstest Für Kinder (KTK)

Two items of the Kiphard and Schilling (2007) were used to measure dynamic coordination and body control. The KTK consists of four test items: (a) walking backwards on a balance beam (3, 4.5, and 6 cm), (b) hopping over an obstacle, (c) jumping sideways over a wooden board, and (d) moving sideways using two wooden platforms. Specifically, two tests—jumping sideways, i.e., jumping from side to side, two-legged, for 15 s, and shifting platforms, i.e., moving sideways on two wooden boards for 20 s,—were used in this study. Two trials were given, and a summed score was used for the analysis. Test-retest reliability coefficients for the raw score are reported to be $r = 0.95$ (Kiphard and Schilling, 2007).

Measures Taken During the Training Period

Heart Rate

Heart rate was measured for each participant during the training with a Polar heart rate monitor (Polar S810). The device was strapped across the participant's chest, and heart rate readings were recorded with an accompanying wristwatch. Resting heart rate (HR) and peak heart rate were recorded, and the estimated maximum heart rate was computed using the formula: estimated maximum heart rate ($EMHR_{max}$) = 206 – (0.88 × age) (Robergs and Landwehr, 2002). Also, the percentage of the estimated HR reached during the training was calculated to ascertain whether an individual child's maximum HR was above the recommended level.

Perceived Exertion

The Borg Rating of Perceived Exertion (RPE) scale was used to monitor the intensity of the exercises during the training (Borg, 1998). The Borg RPE scale consists of numerical values 6–20, where 6 means "no exertion at all" and 20 means "maximal exertion." The Borg RPE scale is reported to be valid and reliable (Day et al., 2004).

Enjoyment Rating Scale

To quantify participants' enjoyment level during the training session, the Enjoyment scale was used (Jelsma et al., 2014). The Enjoyment scale contains five smiley faces with numeric scores (0–4, 0 meaning "Not fun at all," and 4 "Super fun"). Participants were required to choose a smiley face to indicate their enjoyment level.

Training

The graded exergames training used in this study was designed using the Nintendo Wii games and was based on a published protocol (Bonney et al., 2018). In developing the intervention, two main criteria were used to select appropriate games: (1) games should require whole body movement to control the avatar, and (2) games should be responsive to external modifications without limiting playability. Based on these criteria, games such as the "Hula Hoop," "Perfect 10," "Jogging," "Soccer Heading," "Obstacle Course," and "Torso Twists" were included. Foam pads (1.5 and 6 cm-thick closed cell foam pads) and vests with sandbags (with weights of 1 and 2 kg) were used to increase the physical demands of the selected games and to progressively increase the level of postural challenge required. A detailed description of the training is provided in Appendix 1 (**Supplementary Material**). Each participant was required to play 5–6 games twice for 30 min per session, twice weekly for 10 weeks. Half the children trained on Mondays and Wednesdays, and the other half on Tuesdays and Thursdays. Each 30-min

TABLE 2 | Characteristics of TD and DCD groups at the start of the training.

Variable	TD ($n = 28$)		DCD ($n = 32$)	
	Mean	***SD***	**Mean**	***SD***
Age	9.82	1.42	9.31	1.12
BMI	16.87	2.94	17.46	3.81
Shuttle run level*	2.39	0.24	2.25	0.31
MABC-2 total standard score**	9.25	1.48	5.13	1.70

*Significantly different between TD and DCD at $p < 0.05$.
**Significantly different between TD and DCD at $p < 0.001$.

training session started with warm-up games (e.g., "Basic Steps" or Jogging") and included the performance of games from the three available game categories (anaerobic fitness, balance, and yoga). During the first 2 weeks, the participants were instructed to familiarize themselves with the selected Wii games; hence, no alterations were introduced throughout this period. From Week 3 to Week 10, games were adjusted to increase the postural demands and physiological load. This was done through the use of a foam pad and vest filled with two weights. The training was given to a maximum of six participants simultaneously in an enclosed room. Six Wii consoles and TVs were arranged and partitioned so that participants were not distracted by other players. Each session was supervised by one physical therapist and one fitness trainer. One person was responsible for fetching the children from class and fixing the heart rate monitors and weight vests. The other was solely responsible for supervising the training and ensuring that each child had enough time on task. If a child missed a training session, catch-up sessions were held on Fridays.

Data Analysis

Data were checked for normality and equality of variances, and appropriate parametric or non-parametric analyses were performed. Baseline differences in demographic characteristics, and motor performance between the training groups were calculated using chi-square tests (sex) or *t*-tests (age, BMI, MABC-2 total, and SR). To estimate the intensity of the training, averages of the RPE, and HR over 10 weeks were analyzed. The main effect of Time and the interaction of Time with Group on the HR, RPE, and Enjoyment scale were analyzed using repeated measures ANOVA with Time (10 weeks) as the within-group factor and Group (TD/DCD) as the between-group factor.

Changes in fitness and motor outcomes before and after training were analyzed using repeated measures ANOVA with time (pre-post) as within-group factors and group (TD/DCD) as the between-group factor. If interactions emerged, *post-hoc* tests were performed. The standardized mean difference was calculated by subtracting the mean of the scores at the post-test from the mean at the pretest and dividing this difference by the pooled standard deviation. The magnitude of the effect size was interpreted using Cohen's Conventions: small = 0.2, medium = 0.5, large = 0.8 (Cohen, 1988).

The individual peak HR was compared with the percentage of the estimated maximum HR needed for moderate intensity training. Next, the correlation between peak HR and RPE and between peak HR and Enjoyment scores was determined to ascertain whether greater exertion made playing the games less fun. To compensate for test-retest bias and determine change at an individual level, we calculated the number of children who improved more by estimating the measurement error (SEM) and the smallest detectable difference (SDD = 1.96 × SEM) on motor, sprint, and agility tests. All statistical analyses were performed using the Statistical Package for the Social Sciences (SPSS Inc., version 26), and the level of significance was set at $p < 0.05$.

RESULTS

Group Differences Before Training

No group differences were found between DCD and TD groups with regard to age, gender, weight, height, or BMI. Significant differences were found between DCD and TD children on the MABC-2 and Shuttle Run tests (**Table 2**).

Comparison of Motor and Fitness Performance Between DCD and TD Groups

Pre-Post-Comparison

The means for pre- and post-test outcomes with statistics (main effect of Time) are shown in **Table 3**. After training, balance performance was better (MABC-2 Balance and Foam task in left leg). Large differences were found on the anaerobic and aerobic items (10 × 5 m and SR) as well as on the Ladder Agility Tasks (**Table 3**). On only two variables was an interaction effect for Time by Group found; for the MABC-2 total score [$F_{(1, 55)} = 13.19$; $p < 0.001$] and for the MABC-2 balance sub score [$F_{(1, 55)} = 26.83$; $p < 0.0001$], indicating larger improvement in the children with DCD compared to TD on this measure (**Table 4** shows pre- and post-test means for the TD and DCD groups, separately).

Table 5 shows that the training effect was rather specific; aerobic and anaerobic capacity and agility showed large effect sizes. Balance improved moderately, whereas skills not trained (aiming and catching and manual dexterity) and explosive power (FSM) showed no improvement.

Individual Change

For some of the measures used in this study, SEM and SDD were known from psychometric studies (Holm et al., 2013; Bonney et al., 2019). In **Table 6**, the percentage of children who improved more than SEM and SDD is shown and indicates that at least half the children benefitted from the training more than the SDD on the test. Among children with DCD, 52% significantly improved on the LAT while this was 36% on the Balance sub score.

Participants' Characteristics During the Training

The mean Max HR during the training was 139.3 ± 7.2 beats per minute (bpm) and was not different for the two groups [$F_{(1, 58)} = 2.47$; $p = 0.12$; TD 137.5 ± 7.2, DCD 140.26 ± 6.9]. The estimated max HR was 195.4 ± 1.1; 60% was 118.6 ± 0.67. In most of the sessions (7/10 of the weeks) the maximum heart rate measured reached at least 60% of the estimated max HR for all the children (**Figure 2**). Of all the HR readings, 30%

TABLE 3 | Comparison of motor and fitness performance before and after training (mean, standard deviation, *F*-value for the main effect of Time, *p*-values, and degrees of freedom).

Variables	Time				Statistics		
	Pre		Post				
	Mean	*SD*	Mean	*SD*	*F*	*p*-value	*df*
MABC-2 total score (SS)[$]	6.88	2.57	8.05	2.40	15.44	**0.0001**	57
MABC-2 manual dexterity, (SS)	7.98	2.95	8.09	2.31	0.34	0.61	57
MABC-2 Aiming Catching, (SS)	7.43	2.83	8.09	3.54	2.07	0.16	57
MABC-2 balance, (SS)[$]	7.96	3.19	9.57	2.84	14.80	**0.0001**	57
Foam left (s)	14.07	6.02	16.04	4.86	5.44	**0.024**	54
Foam right (s)	15.17	6.00	16.04	5.56	1.07	0.31	53
Agility ladder: Normal (s)	10.75	2.25	9.22	1.59	41.13	**0.0001**	53
Agility ladder: Accuracy (s)	11.07	2.33	9.42	1.61	43.96	**0.0001**	53
KTK platform (number)	34.26	6.84	39.81	7.35	27.80	**0.0001**	50
KTK side jumps (number)	49.30	12.82	55.20	14.37	8.09	**0.007**	40
10 × 5 m: sprint (s)	24.20	2.99	22.75	2.267	12.37	**0.001**	40
Long jump (cm)	124.60	34.40	124.70	29.73	0.73	0.38	30
Overhand throw (cm)	222.77	51.68	226.20	59.89	0.21	0.65	30
Shuttle run level	2.26	0.239	2.90	1.23	8.38	**0.008**	27

[$] *Significant Time by Group interaction. Significant values are printed bold.*

TABLE 4 | Means and standard deviations of motor and fitness performance variables before and after training for the TD and DCD groups for the children who have participated in the post-test.

Variables	TD					DCD				
	Pre		Post			Pre		Post		
	Mean	*SD*	Mean	*SD*	*n*	Mean	*SD*	Mean	*SD*	*n*
MABC-2 total score (SS),	9.12	1.5	9.16	2.1	25	5.13	1.7	7.16	2.2	32
MABC-2 manual dexterity, (SS)	9.40	2.8	9.08	2.2	25	6.78	2.5	7.25	2.2	32
MABC-2 aiming catching, (SS)	8.72	2.7	9.20	3.1	25	6.39	2.5	6.97	3.3	32
MABC-2 balance, (SS)	9.96	1.9	9.68	2.7	25	6.31	3.1	9.67	3.0	32
Foam left (s)	14.56	6.7	16.12	6.5	25	13.65	5.5	12.58	6.2	29
Foam right (s)	17.13	4.8	17.5	4.5	25	13.55	6.5	14.82	6.1	29
Agility ladder: Normal (s)	10.60	2.1	8.97	1.5	24	12.11	2.7	10.08	1.6	29
Agility ladder: Accuracy (s)	10.66	2.0	9.17	1.5	24	12.07	2.6	10.45	1.7	29
KTK platform (number)	37.70	5.4	41.87	5.9	23	30.62	6.7	36.19	7.8	27
KTK side jumps (number)	56.71	10.2	61.06	13.2	17	43.82	11.9	50.86	13.9	23
10 × 5m – sprint (s)	23.41	3.02	21.86	2.3	20	24.98	2.8	23.65	1.9	20
Long jump (cm)	133.67	28.29	128.07	30.6	15	115.53	38.4	121.33	29.5	15
Overhand throw (cm)	249.47	53.5	242.47	59.7	15	196.06	33.8	209.93	57.4	15
Shuttle run level	2.35	0.14	3.22	1.6	12	2.19	0.28	2.64	0.79	15

were above the 70% level and 80% above the 60% estimated max HR level. This confirms that in most cases an adequate maximum level of intensity was reached. As shown in **Figure 2**, HR fluctuated over the 10 weeks [$F_{(9, 50)} = 12.24$; $p < 0.001$]. The changes reflect a gradual decrease in the first 4 weeks before the break and a small increase after the children returned to the study. Only four children did not reach max heart rate above 60% of estimated max HR in at least 7 of the 10 training weeks.

The mean perceived exertion was rated 12.1 ($SD = 0.93$) indicating "somewhat hard" exercise. The perception of the two groups regarding the intensity of the training was similar

[$F_{(1,58)} = 0.008$; $p = 0.93$)]. Variation over the 10 weeks is depicted in **Figure 3** [$F_{(9, 50)} = 13.27$; $p < 0.001$]. The RPE went down gradually but increased for many children after the 2 kg vest for children was added.

The enjoyment in playing the games fluctuated between Super fun (score 4) and Fun (score 3). No differences between the ratings were found between the TD and DCD group [$F_{(1,58)} = 0.393$; $p = 0.53$]. Only one child (DCD) in week 3 rated the training as "A bit fun." "Boring" or "Not fun at all" was never scored. A main effect of time was found [$F_{(9,50)} = 3.63$; $p = 0.002$]. More detailed analysis showed a cubic trend [$F_{(1,58)} = 4.43$; $p = 0.04$]. There was a drop in enjoyment after week 1, the lowest value was found at training week 3, and then rating went up in the rest of the sessions. From week 7, the loads on the training were increased (week 7 and 8 with 1 kg extra; week 9 and 10 with 2 kg extra), and interestingly, this did not lead to a decrease in the number of children rating the training as "Super fun" (**Figure 4**).

A significant correlation ($r_p = 0.47$, $p = 0.001$) was found between the mean max HR during training and the mean RPE score the children gave after the training. No significant correlations were found between Enjoyment and mean RPE or Enjoyment and mean max HR. Children did not like the training less if it was perceived as harder or lighter.

TABLE 5 | Effect sizes for the changes in motor and fitness performance variables.

Variables	Cohen's *d*
Shuttle run level	0.87
Agility ladder: Accuracy (s)	−0.84
Agility ladder: Normal (s)	−0.80
KTK platform (number)	0.78
10 × 5 m sprint (s)	−0.55
MABC-2 balance (SS)	0.53
MABC-2 total score (ISS) (SS)	0.47
KTK side jumps (number)	0.43
Foam left (s)	0.36
MABC-2 aiming catching (SS)	Ns
Foam right (s)	Ns
MABC-2 manual dexterity (SS)	Ns
Long jump (cm)	Ns
Overhand throw (cm)	Ns

DISCUSSION

This study was performed to examine the effects of graded exergames on motor and fitness performance in a sample of elementary school children with and without DCD. The main finding of this study was that children with DCD and TD children experienced gains in motor coordination (i.e., static and dynamic balance and total body coordination) and fitness performance (i.e., aerobic, anaerobic, and agility performance) after 10 weeks of training, which took place in a school setting. A significant interaction of Group by Time after training was observed and the DCD group was found to have larger improvements than the TD children in balance and total body coordination as measured on the MABC-2 test. Also, our study found that both groups of participants enjoyed the training and perceived its intensity to be high (an average of 12 points on the Borg scale). These findings provide new insights into the effects of graded exergames in elementary school children. Since this paper adapted and tested our earlier protocol in young children with and without DCD (Bonney et al., 2018), we can speculate that the current data seem to validate the graded exergames protocol in individuals with DCD.

There is very limited field research on skills training and fitness training in an elementary school setting. The current study provides additional data to physical education teachers, as well as pediatric, physical, and occupational therapists regarding the importance of graded exergames to enhance physical fitness in children with movement difficulties in school settings. However, the adoption of AVGs within the school environment will ultimately be determined by school staff and by practical constraints (e.g., availability and storage options for the equipment; Norris et al., 2016). The fact that we needed to break down our setup every afternoon and pack it into a secure storage room may be a real barrier for its use outside the experimental setting. Importantly, children with DCD in our study have no other options for intervention. Therefore, using AVGs during breaktimes could help them improve their skills and physical fitness, building confidence to take part in regular playground activities.

On this topic, there are currently limited data to which we can relate our findings. We found only one study that examined the feasibility of the graded Wii protocol in youth (females) with probable DCD (Bonney et al., 2018). In that study, it was reported that older school children demonstrated improvements in aerobic and anaerobic fitness after 14 weeks of training.

TABLE 6 | Percentage of individuals with progress beyond the SEM and SDD per group.

Variable	Total (%)			TD (%)		DCD (%)	
	≥ SEM	≥SDD	Improvement	≥ SEM	≥ SDD	≥SEM	≥SDD
Total MABC-2 ($n = 57$)	34	7	41	14	0	53	13
Cluster balance ($n = 57$)	19	23	42	7	4	27	36
Total agility ladder ($n = 53$)	28	40	68	50	25	10	52
10 × 5 sprint ($n = 40$)	28	23	51	20	30	35	15

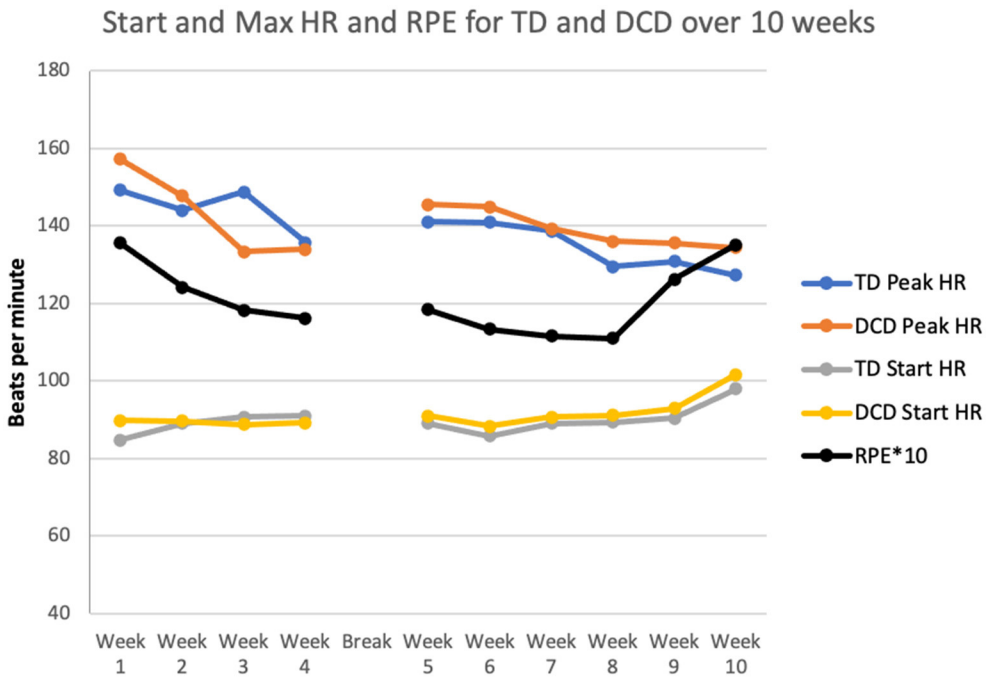

FIGURE 2 | Comparison of heart rate (HR) and Rating of Perceived Exertion (RPE) scale for TD children and children with DCD during the 10 weeks of training.

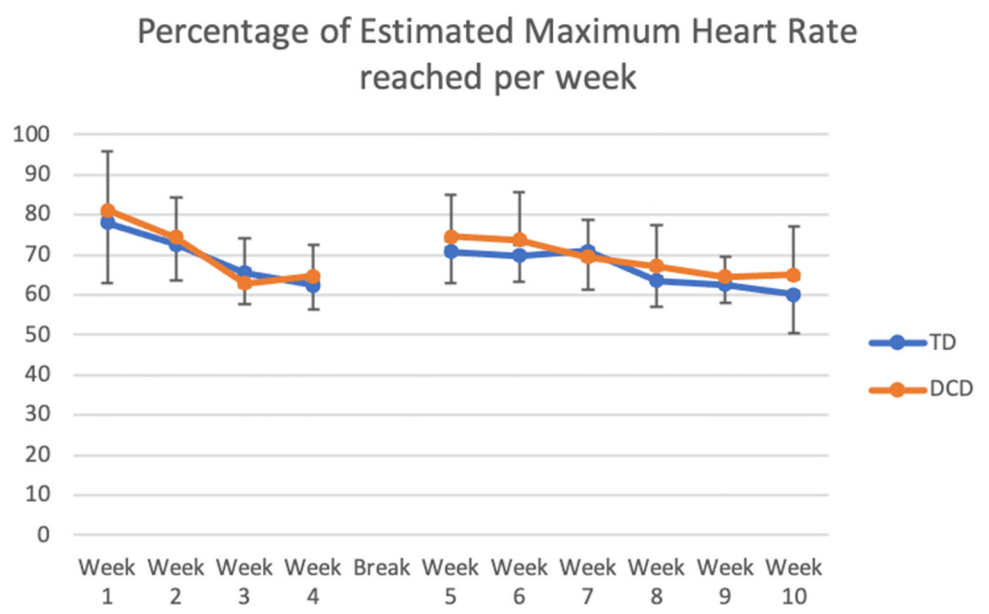

FIGURE 3 | Comparison of the percentage of Estimated Maximum Heart Rate that TD children and children with DCD reached during the 10 weeks of training.

Important points made for the children with DCD in that older age group may be even more relevant for this study. It could be that children with DCD felt more at ease without their peers watching them and the feedback provided by the games may have helped focus their attention inward instead of externally. Another benefit of this type of exercise is the ability to run the program in small spaces within the safety of the school premises. Last, the fact that exercising this way is a fun alternative to the traditional class-based exercise creates an opportunity to expose children with movement difficulties to a variety of options to develop a positive attitude toward exercise (Sheehan and Katz, 2013). This may help reverse withdrawal from physical activity, as motor

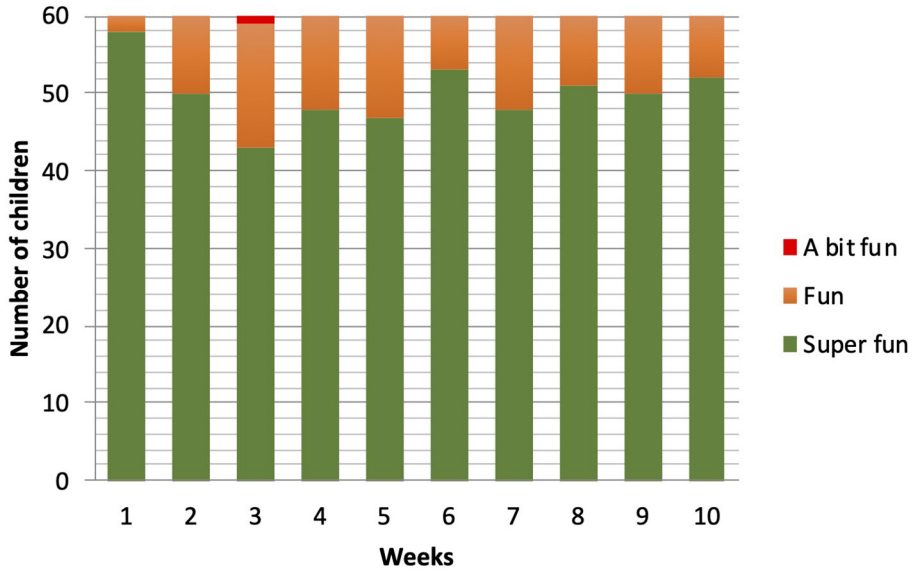

FIGURE 4 | Enjoyment levels reported by the children during the 10 weeks of training.

competence and confidence in one's own motor skills are essential elements in the intrinsic desire to participate in physical activity (Higgs et al., 2008).

In the present investigation, both children with DCD and TD peers made significant gains in aerobic fitness, anaerobic fitness, agility performance, and balance (both static and dynamic) even though the training period was less than that of the previous study (10 vs. 14 weeks 30 vs. 60 min). Participants in our study reported high enjoyment and perceived exertion, which was similar in both groups. The collective experience of the children during the 10 weeks of training was positive (fun to awesome), and they reached the required 60% estimated peak HR. Remarkably, the effect sizes recorded for these gains after training were higher than what was reported in the previous study (Bonney et al., 2018). This is probably a consequence of the low start values, younger age (7–12 vs. 13–16 years) with lower BMI (17.1 vs. 27.5 kg/m^2), and high enjoyment. It is also possible that due to the lack of organized physical activity in the school and neighboring communities, our 2 × 30 min fitness-enhancing graded program was adequate to stimulate short-term gains (after 10 weeks) in motor skills and fitness in these children.

A strength of our study rests on the fact that this study was conducted among 7- to 12-year-old children with DCD and age matched TD peers and adherence with our training was 100%. On the other hand, there are limitations. Our design had no control group, which makes it vulnerable to threats to internal validity. There is the possibility that other events (e.g., history, maturation, testing effects, and statistical regression) than the intervention administered might have caused the observed changes. Unfortunately, it was not possible to include a no-treatment control group because of ethical concerns associated with such a group. Furthermore, our results do not provide insight into the long-term effects of graded exergames on fitness performance in children with DCD. Despite these shortfalls and partly missing data, our study demonstrated the potential value of gradually grading exercises in children with DCD and provides data that serve to validate previous findings on the fitness-promoting benefits of graded exergames for individuals with 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 human participants were reviewed and approved by the study was approved by the Human Research Ethics Committee (HREC) of the University of Cape Town, South Africa (HREC: 209/2018). Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

BS-E, EB, and GF: conceptualization, design, funding, and data acquisition. BS-E and EB: data analysis and interpretation as well as manuscript preparation. All authors critically reviewed and approved the final manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fspor.2021.653851/full#supplementary-material

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White Matter Changes With Rehabilitation in Children With Developmental Coordination Disorder: A Randomized Controlled Trial

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Background and Objectives: Children with developmental coordination disorder (DCD) have difficulty learning motor skills, which can affect their participation in activities of daily living and psychosocial well-being. Over 50% of children with DCD also have attention deficit hyperactivity disorder (ADHD), which further exacerbates their motor problems and impact on quality of life. A rehabilitation approach known as Cognitive Orientation to Occupational Performance uses problem-solving strategies to help children learn motor skills they wish to achieve. While this cognitive approach has been effective for children with DCD, few studies have examined the effectiveness of this approach for children with co-occurring ADHD. Further, the underlying mechanism and neural basis of this intervention are largely unknown.

Methods: In this randomized waitlist-controlled trial, we used MRI to examine white matter microstructure after intervention in 8–12-year-old children with DCD (n = 28) and with DCD and co-occurring ADHD (n = 25). Children in both groups were randomized to either a treatment group or waitlist group at their first MRI. The treatment group began the intervention after their MRI scan and returned for a post-treatment scan at 3 months, and follow-up scan at 6 months; the waitlist group waited 3 months before their second MRI, received the intervention, and then had a post-treatment scan. Each child received intervention once weekly for 10 weeks. Diffusion tensor imaging was used to acquire white matter diffusion parameters and was analyzed using tract-based spatial statistics (TBSS).

Results and Conclusion: Children with DCD showed significant improvement in white matter microstructure in the bilateral anterior thalamic radiation, bilateral sensorimotor tract, bilateral cingulum, fornix, splenium and body of corpus callosum, right inferior fronto-occipital fasciculus, and white matter pathways to bilateral inferior gyri, right middle frontal gyrus, frontal medial cortex, and left cuneus. We suggest that these rehabilitation-induced neural changes in children with DCD occurred in regions

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associated with attention, self-regulation, motor planning, and inter-hemispheric communication, which positively affected brain connectivity and motor function. In contrast, children with DCD and co-occurring ADHD did not show any brain changes following the intervention. Modifications to the treatment protocol might help address the attentional and self-regulatory needs of children with a dual diagnosis.

Clinical Trial Registration: ClinicalTrials.gov ID: NCT02597751.

Keywords: developmental coordination disorder, motor skills disorder, rehabilitation, diffusion tensor imaging, neuroplasticity, CO-OP

INTRODUCTION

Up to 5 to 6% of all school-age children may be affected by developmental coordination disorder (DCD), a neurodevelopmental disorder characterized by difficulty performing and learning motor skills that significantly interferes with daily activities and academic achievement (American Psychiatric Association, 2013). These motor difficulties can lead to higher risks of anxiety, emotional and behavioral problems, low self-esteem (Hellgren et al., 1994; Green et al., 2006; Pratt and Hill, 2011; Lingam et al., 2012; Hill and Brown, 2013; Zwicker et al., 2013; Crane et al., 2017; Harrowell et al., 2017; Li et al., 2018), as well as physical consequences such as obesity and poorer physical fitness (Cairney et al., 2005, 2010; Rivilis et al., 2011; Cairney and Veldhuizen, 2013). A well-known and common co-occurrence of DCD is attention deficit hyperactivity disorder (ADHD), exacerbating motor and functional difficulties in children (Kadesjo and Gillberg, 1999; Piek et al., 1999; Dewey et al., 2000, 2002; Rasmussen and Gillberg, 2000; Pitcher et al., 2003; Martin et al., 2006; Watemberg et al., 2007; Fliers et al., 2009; Barkley, 2014; Blank et al., 2019) and increasing the risk of psychological distress (Piek et al., 2007; Missiuna et al., 2014), antisocial behavior (Rasmussen and Gillberg, 2000), and peer victimization (Dewey and Volkovinskaia, 2018). Motor problems of children with DCD with or without ADHD have been attributed to attention deficits and lack of inhibition (Kaiser et al., 2015; Fong et al., 2016; Thornton et al., 2018). An electroencephalographic (EEG) study suggests that the contribution of attention to motor performance is greater in children with co-occurring DCD and ADHD than children with DCD only (Fong et al., 2016); it, accordingly assumes that improving attention in children with DCD with or without ADHD leads to motor performance improvement (Fong et al., 2016). However, only 30% to 50% of children with DCD and ADHD show improved motor performance following attention-related medications (Bart et al., 2010; Brossard-Racine et al., 2012), and only 50% of children with ADHD receive non-pharmaceutical treatment for their motor difficulties (Fliers et al., 2010, 2011). This controversy in the literature and limited attention to motor problems add complexities to the treatment approaches for children with a dual diagnosis of DCD and ADHD.

Current neuroimaging studies reveal that DCD is associated with involvement of the cerebellum, the parietal lobe, the frontal lobe, the basal ganglia, and the limbic system (Brown-Lum and Zwicker, 2015; Biotteau et al., 2016); each of these regions play a specific role in generating internal models of motor actions (Kawato and Gomi, 1992; Blakemore et al., 2001), updating the internal model (Blakemore and Sirigu, 2003), providing optimal control during motor execution (Shadmehr and Krakauer, 2008), executing motor actions (Shadmehr and Krakauer, 2008), and managing the movement motivation (Merel et al., 2019), respectively.

However, co-occurring DCD and ADHD are associated with unique structural (Langevin et al., 2014, 2015), functional (McLeod et al., 2014, 2016; Thornton et al., 2018), and physiological (Yeh et al., 2012; Fong et al., 2016) properties of the sensorimotor and attentional networks, including the parietal and frontal lobes (Yeh et al., 2012; Langevin et al., 2014, 2015; McLeod et al., 2014, 2016; Thornton et al., 2018) as well as interhemispheric connections and asymmetry (Langevin et al., 2014, 2015). Individuals with co-occurring DCD and ADHD may use compensatory attentional control of motor coordination through increasing cerebral blood flow in the posterior cingulate cortex and the cerebellum (Yeh et al., 2012).

A treatment approach called Cognitive Orientation to daily Occupational Performance (CO-OP) is one of the recommended treatments for DCD as per international clinical guidelines (Blank et al., 2019). CO-OP is a client-centered, task-oriented approach developed for children with DCD to successfully solve motor problems (Polatajko et al., 2001). Previous studies have shown positive results in children with DCD (Ward and Rodger, 2004; Taylor et al., 2007; Zwicker et al., 2015; Capistran and Martini, 2016; Thornton et al., 2016), but given that at least 50% of children with DCD have co-occurring ADHD (Dewey et al., 2002), we wondered if this cognitive approach was effective for children with a dual diagnosis of DCD and ADHD. Results from a single case study of six children with ADHD show promise for the CO-OP approach (Gharebaghy et al., 2015), but studies with larger sample sizes and more rigorous research designs are required to determine CO-OP's effectiveness in children with DCD, with and without ADHD. To better understand if and how CO-OP affects children with DCD with or without ADHD differently, we examined brain changes after 10 sessions of CO-OP intervention using various MRI modalities (resting state, T1-weighted images, and diffusion tensor imaging; DTI). In the current study, we focus on structural neuroplastic changes captured by DTI after CO-OP intervention in children with DCD, with and without ADHD. Understanding the neural mechanisms of CO-OP could further guide the modification and optimization of CO-OP based on specific needs of children with DCD with or without ADHD.

Diffusion MRI is a non-invasive tool measuring both structural connectivity and white matter microstructure by obtaining information about connections between brain regions and their tissue architecture (Jones et al., 2013). It measures water diffusivity in brain tissue and the amount of restriction experienced by water molecules moving in the brain. Water molecules are considerably impeded in white matter, owing to factors such as myelination, fiber diameter or density, as well as membrane permeability (Beaulieu, 2002). This impedance causes directional and anisotropic water diffusivity.

Thus far, diffusion MRI studies have used various analysis methods, including tract-based spatial statistics (TBSS; Williams et al., 2017; Brown-Lum et al., 2020), constrained spherical deconvolution (Hyde et al., 2019), tractography (Zwicker et al., 2012; Debrabant et al., 2016) and graph theory (Debrabant et al., 2016) to understand white matter microstructure in children with DCD. Results have shown that children with DCD have altered white matter microstructure in the corpus callosum (Langevin et al., 2014; Brown-Lum et al., 2020) and sensorimotor, corticospinal, cortico-cerebellar (Zwicker et al., 2012; Debrabant et al., 2016; Williams et al., 2017; Brown-Lum et al., 2020), and frontoparietal pathways (Langevin et al., 2014; Williams et al., 2017; Hyde et al., 2019; Brown-Lum et al., 2020). Structural connectivity between brain regions (e.g., cerebellar lobule VI and right superior parietal gyrus; Debrabant et al., 2016) is also implicated in children with DCD. Children with DCD and co-occurring ADHD have altered white matter in the corpus callosum (Langevin et al., 2014). However, no study investigated longitudinal changes following intervention in children with DCD. In this study, we will compare white matter microstructural properties of children with DCD, with or without ADHD, before and after CO-OP intervention.

MATERIALS AND METHODS

Study Design

In this randomized waitlist-controlled trial (ClinicalTrials.gov ID: NCT02597751), we used computer-generated sequential blocks of four to six, prepared by a statistician to randomize children with DCD, with or without ADHD, into treatment and waitlist groups. To ensure a power of 90% to detect a 3% difference in axial diffusivity (AD) with a type-1 error of 0.01, we used our pilot study on DTI in this population (effect size = 1.1; Zwicker et al., 2012) and estimated a sample size of 27 per group.

Participants

From September 2014 to July 2019, children with DCD and DCD+ADHD were recruited from the Sunny Hill Health Centre for Children, BC Children's Hospital ADHD Clinic, the Vancouver Regional Pediatric Team, and from advertisements in the community (Vancouver, BC, Canada). Children aged 8–12 years were eligible to participate in the study if they were diagnosed with DCD as per the Diagnostic and Statistical Manual–5th edition (American Psychiatric Association, 2013) and the international clinical practice recommendations (Blank et al., 2019) as follows: (1): scored ≤16th percentile on the Movement Assessment Battery for Children—2nd ed. (MABC-2; Henderson et al., 2007); (2) scored in the suspected or indicative DCD range on the DCD Questionnaire (Wilson et al., 2009); (3) parents reported motor difficulties from a young age; and (4) there was no other medical condition that could explain motor difficulties based on parent-report, clinical observations, and/or a medical examination. Children were assigned to the DCD+ADHD group if they had been diagnosed ADHD based on parent report. ADHD symptomatology was quantified for all children using the Conners ADHD Index—parent report form (Conners, 2009). Exclusion criteria included premature birth (gestational age <37 weeks), other neurodevelopmental disorders (e.g., autism spectrum disorder), claustrophobia, and MRI contraindications (e.g., metal braces). After parental consent and child assent as per ethics approval from the University of British Columbia/BC Children's and Women's Research Ethics Board, children were enrolled in the study.

Procedure

Participants started with MRI safety screening and an MRI simulation session. A research nurse accompanied by a graduate student scanned all the participants using MRI at baseline, after 3 months, and after 6 months. After the first MRI scan, children were randomized into treatment and waitlist groups; group assignments were concealed to the research team in an opaque, sealed envelope. Children assigned to the treatment group went through pre-intervention assessment, then received 10 sessions of CO-OP intervention (once a week), followed by post-intervention assessment, a second (post-treatment) MRI scan, and a third follow-up MRI scan 3 months later to determine if brain changes were maintained after the intervention was discontinued. In contrast, children in the waitlist group were scanned 3 months after their first MRI, received their pre-assessments and intervention, followed by a third (post-treatment) MRI. A study design schematic (Izadi-Najafabadi et al., 2020) can be found elsewhere.

Motor assessments were administered by an occupational therapist not involved in the intervention. Assessments included the following: (1) Canadian Occupational Performance Measure (COPM; Law et al., 2014), which measured the child's perceived motor performance and satisfaction on their three chosen motor goals; (2) Performance Quality Rating Scale (PQRS; Martini et al., 2015), which objectively measured the child's quality of motor performance on their motor goals based on a blinded occupational therapist scoring videos of the child performing their motor goals before and after the intervention; and (3) Bruininks-Oseretsky Test of Motor Proficiency-2 (BOT-2) short form (Bruininks and Bruininks, 2005), which measured general motor skills. A more detailed description of behavioral outcome measures and their results can be found elsewhere (Izadi-Najafabadi et al., under review).

Intervention

Using the Pediatric Activity Card Sort (Mandich et al., 2004), an assessment tool for developing activity profiles for children, each child with DCD (with or without ADHD)

identified three functional motor goals (e.g., handwriting, playing basketball, tying shoelaces) on which to work during the CO-OP intervention. An occupational therapist administered 10 one-hour sessions of the CO-OP intervention over 10 weeks as per published protocol (Polatajko et al., 2001) at the Sunny Hill Health Centre for Children or BC Children's Hospital. CO-OP intervention is a problem-solving rehabilitation approach that focuses on the use of cognitive strategies to solve motor problems. Children learned the global strategy of CO-OP called Goal-Plan-Do-Check in the first session and were guided to discover specific cognitive strategies (e.g., supplementing task knowledge, changing body position) to solve motor problems to achieve their chosen goals (Miller et al., 2001; Polatajko and Mandich, 2004). Parents were instructed and encouraged to use CO-OP at home and keep a record of practice time for each goal on each day, per week.

Diffusion Tensor Imaging

Acquisition

A 3-Tesla General-Electric Discovery MR750 MRI scanner with a 32-channel head coil was used to acquire DTI data. Participants were asked to lie very still during the DTI sequence while watching a movie. A minimum of two 32-direction DTI sequences were acquired so that the best sequence could be used for data analysis. DTI acquisition parameters for each DTI sequence with 32 diffusion encoded directions dispersed around a full-sphere were as follows: TR: 7,000 ms; TE: 60 ms; FOV: 220 mm; acquisition matrix: 100 × 100; slice thickness: 2.2 mm; b = 1,000. Three b0 volumes were also acquired at the beginning of each DTI sequence. A graduate student monitored movement during the scan, encouraged the child to stay still, and asked the sequence to be repeated, if needed.

Preprocessing

DTI data preprocessing and analysis were completed using FSL 6.0.1. (Smith et al., 2004). Preprocessing steps included: (1) executing *eddy_cuda* for distortion correction through signal loss (drop-out) detection and replacement (Andersson et al., 2016) as well as correction for susceptibility-induced distortions, eddy currents, and subject motions (within and between volumes; Andersson and Sotiropoulos, 2016; Andersson et al., 2017); (2) implementing automated quality control via *QUality Assessment for DMRI (QUAD)* to extract quality metrices of within-and between-volume average relative motion, absolute relative motion, signal-to-noise ratio (SNR), and contrast-to-noise ratio (CNR; Bastiani et al., 2019); for each participant, the sequence with higher CNR was carried forward for further analysis; (3) visually checking every image to identify any residual distortion and removing any motion-contaminated volumes; any image with less than 20 good quality volumes were excluded from the analysis; and (4) reconstructing diffusion tensors to estimate DTI parameters and create corresponding maps: *fractional anisotropy (FA)*, the degree of anisotropy/directionality of water diffusion in each voxel; *mean diffusivity (MD)*, average amount of water diffusion independent of directionality in each voxel; *axial diffusivity (AD)*, water diffusivity along the tract; *radial diffusivity (RD)*, water diffusivity perpendicular to the tract (Jones et al., 2013).

Statistical Analysis

We performed tract-based spatial statistics (TBSS) to analyze whole-brain white matter microstructural properties without pre-specification of tracts of interest (Smith et al., 2006). TBSS carries out a voxel-wise statistical analysis while controlling for inaccurate alignment and arbitrary smoothing in traditional voxel-based analysis (Smith et al., 2006). TBSS aligns each participant's FA map to the FA map of the most representative participant and then to the standard template (MNI 152 1 mm). The use of the most representative participant as the first step in registration is critical to TBSS reliability (Madhyastha et al., 2014) since it increases the alignment accuracy and reduces inter-subject variability (Smith et al., 2006). A mean FA skeleton was then generated using all participants' aligned FA maps and thresholded at 0.2 to exclude any gray matter residuals and peripheral tracts with high inter-subject variability. Each participant's FA, MD, AD, and RD data were then projected onto the mean FA skeleton to be ready for statistical analysis (Smith et al., 2006). TBSS has shown a high test-retest reliability in longitudinal studies (Madhyastha et al., 2014).

TBSS results were fed into Permutation Analysis of Linear Models (PALM; Winkler et al., 2014) with 5,000 permutations to investigate 3-month maturation effect (scan 1 and 2 of waitlist groups), pre-post intervention effect (scan 1 and 2 of treatment group combined with scan 2 and 3 of waitlist group), and 3-month follow-up effect (scan 2 and 3 of treatment group) for children with DCD and DCD+ADHD. We used a paired t-test design controlling for ADHD-related medications. Family-wise error correction (FWE) with an alpha level of 0.05 was applied to correct for multiple testing errors. We also used PALM to run a generalized linear model and investigate the effect of motor outcomes (PQRS; Martini et al., 2015) and BOT-2 (Bruininks and Bruininks, 2005) on DTI parameters in the two groups. To report the results, we used a sensitive thresholding approach called threshold-free cluster enhancement (TFCE), in which voxel-wise values receive local spatial supports from extended areas of signal (Smith and Nichols, 2009). To label anatomic locations of white matter structures, the Johns Hopkins University ICBM-81 White-Matter Labels (Mori et al., 2005), the White Matter Tractography Atlas (Hua et al., 2008), and the Sensorimotor Tracts Atlas (Archer et al., 2018) were used.

RESULTS

Participants

Eighty children were recruited for this study; two children with DCD + ADHD declined to participate. Of the 78 children enrolled and randomized into treatment and waitlist groups, 37 children were diagnosed with DCD [25 male, 12 female; mean (SD) age: 9.7 (1.5) years] and 41 children were diagnosed with both DCD and ADHD [38 male, 3 female; mean (SD) age: 10.2 (1.4) years]. Nine children with DCD and 16 children with DCD + ADHD were excluded from the analysis due to false inclusion

(DCD: n = 3; DCD + ADHD: n = 12), low MRI quality (DCD: n = 2; DCD + ADHD: n = 1), and child disliked MRI (DCD: n = 4; DCD + ADHD: n = 3). See **Figure 1** for more details.

Accordingly, we compared pre-and post-intervention scans from 28 children with DCD [20 male, 8 female; mean (SD) age: 9.6 (1.4) years] and 25 children with DCD + ADHD [22 male, 3 female; mean (SD) age: 9.9 (1.1) years]. Due to the quality of baseline and follow-up scans, maturation effect (DCD: n = 18; DCD + ADHD: n = 10) and follow-up analyses (DCD: n = 10; DCD + ADHD: n = 13) were performed with fewer participants. At the time of intervention, three children with DCD and 13 children with DCD + ADHD took ADHD-related medications (e.g., Adderall, Biphentin, Concerta). **Table 1** summarizes participant characteristics and **Table 2** highlights DTI quality and head motion parameters per group.

DCD-Only Group

Maturation

Comparison of scan 1 and scan 2 of children in the waitlist group showed a significant decrease in FA and a significant increase in MD and RD of the left anterior thalamic radiation passing through the anterior corona radiata (FA: FWE-$p < 0.05$; MD and RD: FWE-$p < 0.01$) and anterior limb of internal capsule (FA and MD: FWE-$p < 0.05$; RD: FWE-$p < 0.01$) as well as a significant increase in MD and RD (FWE-$p < 0.01$) of the right anterior thalamic radiation, bilateral corticospinal tract, bilateral inferior longitudinal fasciculus, bilateral superior longitudinal fasciculus, bilateral inferior fronto-occipital fasciculus, bilateral cingulum, corpus callosum, and right anterior corona radiata, posterior limb of internal capsule, and retrolenticular part of internal capsule (**Table 3** and **Figure 2**).

Intervention Effect

Comparing pre-and post-intervention DTI parameters of 28 children with DCD showed a significant increase (FWE-$p < 0.05$) in FA of the bilateral anterior thalamic radiation, bilateral sensorimotor tract, bilateral cingulum, fornix, splenium and body of corpus callosum, right inferior fronto-occipital fasciculus, and white matter pathways to the bilateral inferior gyri, right middle frontal gyrus, frontal medial cortex, and left cuneus (**Table 4** and **Figure 3**).

Follow-up

Follow-up analysis of 10 children with DCD in the treatment group did not show any significant changes between post-intervention and follow-up MRI scans. We also did not find any significant changes from pre-intervention to follow-up scans in this small sub-sample.

Motor Outcomes

Regression analysis did not show any relationship between behavioral outcome measures (e.g., PQRS and BOT-2) and changes in white matter microstructure in children with DCD.

DCD + ADHD Group

Children with DCD + ADHD did not show any significant change (FWE-$p > 0.05$) in any of the DTI parameters in the 3-month period before CO-OP intervention (n = 10), immediately after the intervention (n = 25), or in the follow-up scan (n = 13). We did not find any relationship between motor performance and white matter parameters.

DISCUSSION

Our results showed an increase in FA in white matter tracts, not associated with maturation, after CO-OP intervention in children with DCD, including the bilateral anterior thalamic radiation, bilateral sensorimotor tracts, right inferior fronto-occipital fasciculus, bilateral cingulum, fornix, and corpus callosum. During the first 3 months of the study prior to the CO-OP intervention, we observed an overall increase in RD and MD of the white matter microstructure as well as a reduction in FA of left anterior thalamic radiation in children with DCD. We showed that the CO-OP intervention reverted this pattern in children with DCD. Similar to our resting state results (Rinat et al., 2020), children with DCD + ADHD did not show any brain changes after the CO-OP intervention.

Rehabilitation-Induced Neuroplasticity in Children With DCD

Improved FA in white matter tracts is generally thought to reflect improved microstructural properties, such as axonal density and diameter, myelin integrity, or fiber coherence in these tracts (Concha, 2014); however, the specific change in microstructure cannot be identified. It is also important to note that FA values can be imprecise in regions where there are crossing fibers (O'Donnell and Westin, 2011). Notwithstanding these limitations, we showed that FA increased in the bilateral anterior thalamic radiations, bilateral sensorimotor tracts, right inferior fronto-occipital fasciculus, bilateral cingulum, fornix, corpus callosum, and white matter structures to anterior cingulate cortex and frontal lobe after CO-OP intervention in children with DCD. Our results align with preliminary results from a non-parametric combination of voxel-based morphometry and fractional anisotropy in the anterior thalamic radiation and the body and splenium of the corpus callosum after the CO-OP intervention (Izadi-Najafabadi et al., 2020). These regions play multiple roles in human behavior (e.g., emotion regulation, motor control, executive function). In what follows, we will explain how improved FA in each of these white matter tracts is related to the CO-OP intervention and its potential role in self-regulation, attention, and emotion regulation, as well as motor performance.

Attention and Self-regulation

Consistent with behavioral evidence of self-regulation mediating CO-OP's effect (Jokić et al., 2013), our results suggest that CO-OP intervention might play a role in self- and attention-regulation. Most of our findings underlie the default mode network (DMN); *anterior thalamic radiation, cingulum, corpus callosum, inferior fronto-occipital fasciculus*, and *white matter tracts to the cuneus and anterior cingulate cortex* connect DMN regions in the posterior and anterior cingulate cortices, thalamus, cuneus, as well as across the two hemispheres (Teipel et al., 2010;

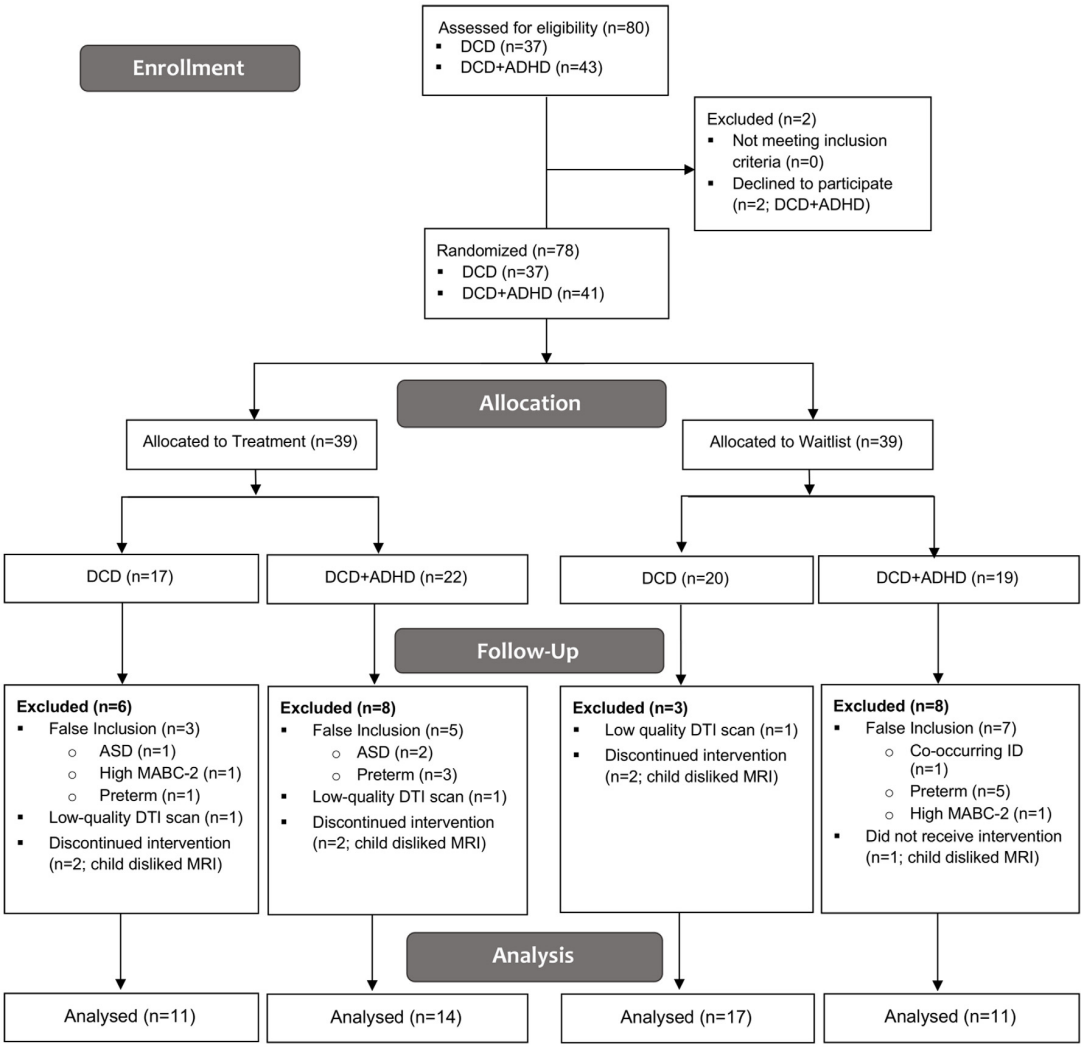

FIGURE 1 | CONSORT Flow Diagram. ADHD, attention deficit hyperactivity disorder; ASD, autism spectrum disorder; DCD, developmental coordination disorder; DTI, diffusion tensor imaging; ID, intellectual disability; MABC-2, Movement Assessment Battery for Children—2nd ed.

TABLE 1 | Participant characteristics.

Variable	DCD (*n* = 28)		DCD + ADHD (*n* = 25)	
	Treatment (*n* = 11)	Waitlist (*n* = 17)	Treatment (*n* = 14)	Waitlist (*n* = 11)
Male sex; N (%)	7 (64)	13 (77)	12 (86)	10 (91)
Age (years); Mean (SD)	10.1 (1.6)	9.3 (1.2)	9.7 (1.2)	10.2 (1.1)
DCDQ (total); Mean (SD)	28.1 (8.6)	32.8 (9.8)	28.1 (6.7)	32.0 (6.8)
MABC-2 (percentile); Median (IQR)	2 (6.2)	2 (4.0)	1 (1.5)	9 (6.0)
Conner's ADHD Index (T-score); Median (IQR)	90 (17)	86 (31)	90 (0)	90 (0)

ADHD, attention deficit hyperactivity disorder; DCD, developmental coordination disorder; DCDQ, Developmental Coordination Disorder Questionnaire; IQR, inter-quartile range; MABC-2, Movement Assessment Battery for Children—2nd ed.; SD, standard deviation.

Luo et al., 2012; Alves et al., 2019). DMN is involved in internally-directed attention, regulating attentional resources, and guiding self-regulatory processes (Bush et al., 2000; Grimm et al., 2009; Kelly et al., 2009; Wiebking et al., 2011; Dixon et al., 2014). The CO-OP intervention uses a global strategy of *Goal-Plan-Do-Check* where children set goals and motivate themselves, perform the task, observe their own performance, judge it to improve it, and then achieve their goal and express their self-satisfaction (Polatajko and Mandich, 2004), which is similar to the three phases of self-regulation (i.e., forethought, performance, and self-reflection) based on Triadic Model of Self-regulation (Zimmerman, 2000).

TABLE 2 | DTI quality and head motion parameters.

Variable		DCD (*n* = 28)		DCD + ADHD (*n* = 25)	
		Treatment (*n* = 11)	Waitlist (*n* = 17)	Treatment (*n* = 14)	Waitlist (*n* = 11)
DTI Quality					
Contrast-to-Noise Ratio (CNR); Mean (SD)	Scan 1	5.7 (1.0)	4.9 (1.7)	5.8 (1.8)	*10.2 (8.5)
	Scan 2	5.9 (1.2)	5.3 (2.0)	6.8 (4.1)	7.0 (3.6)
	Scan 3	5.6 (0.9)	5.8 (2.5)	5.5 (1.9)	6.6 (2.6)
Signal-to-Noise Ratio (SNR); Mean (SD)	Scan 1	65.6 (10.0)	61.0 (11.8)	65.1 (10.7)	61.4 (19.9)
	Scan 2	68.6 (8.0)	67.0 (13.2)	63.9 (10.0)	63.9 (14.8)
	Scan 3	71.7 (6.6)	65.8 (10.8)	65.8 (11.9)	72.4 (7.2)
Head Motion Parameters					
Average Relative Motion (mm); Mean (SD)	Scan 1	0.30 (0.2)	0.42 (0.2)	0.44 (0.2)	0.40 (0.3)
	Scan 2	0.20 (0.1)	0.45 (0.2)	0.41 (0.3)	0.40 (0.3)
	Scan 3	0.30 (0.2)	0.30 (0.2)	0.45 (0.3)	0.32 (0.13)
Average Absolute Motion (mm); Mean (SD)	Scan 1	0.81 (0.5)	1.20 (1.0)	1.20 (1.0)	1.10 (1.4)
	Scan 2	0.83 (0.7)	1.00 (0.5)	0.86 (0.6)	0.80 (0.4)
	Scan 3	0.73 (0.5)	0.80 (0.3)	1.10 (0.9)	0.90 (0.5)

*ADHD, attention deficit hyperactivity disorder; DCD, developmental coordination disorder; DTI, diffusion tensor imaging; SD, standard deviation. *$p < 0.01$: not significant after Bonferroni correction ($p < 0.004$).*

We have also shown that CO-OP intervention induces both increased FA in white matter structures underlying the DMN and increased functional connectivity between the DMN and the anterior cingulate cortex in this population (Rinat et al., 2020). High functional connectivity between regions of the DMN is related to their increased structural connectivity (van den Heuvel et al., 2008; Damoiseaux and Greicius, 2009), in particular, increased FA in the right and left cingulum (Bathelt et al., 2019).

Moreover, CO-OP intervention induces structural changes in other DMN white matter tracts, including the corona radiata (Izadi-Najafabadi et al., 2020). Based on our results from DTI analysis and other analysis (Izadi-Najafabadi et al., 2020; Rinat et al., 2020), we infer that the CO-OP intervention improves both functional and structural connectivity of the DMN, which is responsible for attention-and self-regulation (Shamloo and Helie, 2016; Reddy et al., 2018; Izadi-Najafabadi et al., 2020). We have previously suggested that these functions are implicated in children with DCD due to altered connectivity between the posterior cingulate cortex and DMN (Rinat et al., 2020).

CO-OP intervention improved white matter organization of the *right inferior fronto-occipital fasciculus*, *right anterior thalamic radiation, and bilateral sensorimotor tracts*. These regions are involved in various forms of attention-and self-regulation. The *inferior fronto-occipital fasciculus* guides sustained attention and other executive control functions (Leng et al., 2016). It is the only tract with a rightward asymmetry in our result, which is consistent with the right asymmetry for sustained attention (Pardo et al., 1991). Brown-Lum (2017) reported reduced FA in the *right inferior fronto-occipital fasciculus* in children with DCD. The white matter organization of the *right inferior fronto-occipital fasciculus* and *the right anterior thalamic radiation* are also associated with delayed discounting—the ability to tolerate delays while waiting for future rewards (Olson et al., 2009). Delayed discounting is a critical aspect of motivated behavior and self-regulatory skills, which are known to mediate the CO-OP intervention effects in children with DCD (Jokić et al., 2013).

The *bilateral sensorimotor tracts* immediately beneath the superior corona radiata transfer motor information from the primary motor cortex (M1) and supplementary motor area (SMA) through the corticospinal tract (Klöppel et al., 2008; Vergani et al., 2014). High FA in the superior corona radiata is also associated with improved focused and sustained attention (Stave et al., 2017) and has previously been reported in a different analysis of CO-OP-induced structural changes (Izadi-Najafabadi et al., 2020). The current analysis provides further evidence of the potential role of the CO-OP in attention regulation and motor execution.

Emotion Regulation

We found improved microstructure in regions involved in emotion regulation including *the anterior thalamic radiation, SMA white matter, and inferior frontal gyrus white matter*. Emotion regulation is a process of modifying emotion to increase or decrease emotional experience before, during, and after the motor performance (Beatty and Janelle, 2020). Accordingly, we suggest that the CO-OP intervention might help to regulate emotions required before, during, and after motor learning; the child-chosen nature of motor goals in CO-OP might increase emotional motivation prior to the motor performance, which is known to be the action starter (Beatty and Janelle, 2020). To achieve motor goals, CO-OP enables children to use strategies that distract them from negative emotions (e.g., frustration or embarrassment of their inability to perform a task) during motor skill acquisition. For example, when children use the domain-specific strategy of "attention to doing" during CO-OP, they focus more on their action rather than their feeling. Lastly, when children achieve their motor goals, they experience satisfaction; all these together enhance the likelihood of success in future motor performance and helps with emotion regulation (Beatty and Janelle, 2020). Moreover, similar to a key feature of CO-OP (Missiuna et al., 2001), emotions could be regulated when guided by a significant adult (e.g., therapist or parents; Williams et al., 2020).

TABLE 3 | Effect of maturation on DTI parameters in children with DCD[a].

White matter structure		MNI-space			DTI	Direction	*t*	FWE	Cohen's *d*
		X	*Y*	*Z*	parameter	of change		Sig	
Anterior thalamic radiation L	At anterior corona radiata	−21	31	8	FA	Decreased	3.9	*	0.72
		−24	32	9	MD	Increased	2.5	**	0.46
					RD		4.1	**	0.76
	At anterior limb of internal capsule	−20	17	4	FA	Decreased	3.0	*	0.55
		−12	2	4	MD	Increased	1.3	*	0.24
					RD		3.0	**	0.55
	At posterior limb of internal capsule	−12	−2	6	MD	Increased	2.8	**	0.52
					RD		2.2	**	0.41
	At thalamus	−10	−10	10	MD	Increased	1.5	*	0.27
Anterior thalamic radiation R		25	−35	4	MD	Increased	2.4	**	0.44
					RD		2.2	*	0.40
	At anterior corona radiata	22	31	10	MD	Increased	1.6	*	0.29
					RD		1.9	**	0.36
	At anterior limb of internal capsule	13	1	5	MD	Increased	4.9	**	0.88
					RD		4.6	**	0.66
Sensorimotor tract L	At posterior corona radiata	−25	−25	29	MD	Increased	2.5	*	0.46
					RD		3.8	**	0.70
Sensorimotor tract R	At posterior limb of internal capsule	18	−12	1	MD	Increased	2.9	**	0.54
					RD		2.2	*	0.40
	At thalamus	16	−20	0	MD	Increased	2.9	**	0.48
					RD		2.1	**	0.49
	At pontine crossing	6	−30	−35	MD	Increased	3.9	*	0.71
Inferior fronto-occipital fasciculus L		−23	23	6	MD	Increased	1.9	**	0.35
					RD		2.9	**	0.53
Inferior fronto-occipital fasciculus R		25	23	−7	MD	Increased	1.9	**	0.34
					RD		2.1	**	0.38
		33	−30	5	MD	Increased	3.2	**	0.59
					RD		1.1	*	0.20
Superior longitudinal fasciculus L		−36	−22	29	MD	Increased	1.4	*	0.25
					RD		4.1	**	0.75
Superior longitudinal fasciculus R		39	−36	34	MD	Increased	2.7	*	0.48
					RD		2.2	*	0.40
		31	−53	24	MD	Increased	1.2	*	0.23
					RD		3.4	*	0.62
		43	−46	3	MD	Increased	0.9	*	0.20
					RD		2.2	*	040
		58	−30	−14	MD	Increased	2.8	**	0.51
					RD		3.5	**	0.65
Inferior longitudinal fasciculus L		−29	−30	3	MD	Increased	1.4	*	0.25
					RD		2.4	**	0.43
		−43	−30	−7	MD	Increased	2.2	*	0.40
		−48	−21	−15			3.2	*	0.60
		−43	0	−30			2.8	*	0.50
		−45	−3	−34			1.9	*	0.30
Inferior longitudinal fasciculus R		43	−7	−20	MD	Increased	2.4	**	0.44
					RD		1.9	**	0.36
		48	116	60	MD	Increased	2.4	**	0.43
					RD		3.2	**	0.58
		42	−3	−24	MD	Increased	3.5	**	0.64
					RD		1.9	**	0.34
		45	−15	−10	MD	Increased	2.7	**	0.49
					RD		3.1	**	0.57
		43	−41	−3	MD	Increased	1.9	*	0.34
					RD		2.4	**	0.43
		40	−30	4	MD	Increased	1.9	**	0.34
					RD		4.4	**	0.81
Cingulum L		−8	−25	38	MD	Increased	1.7	*	0.31
					RD		2.0	*	0.37
		−18	−37	38	MD	Increased	2.9	*	0.52
					RD		3.6	*	0.65
Cingulum R		8	−25	38	MD	Increased	2.2	*	0.40
					RD		4.0	*	0.74

(Continued)

TABLE 3 | Continued

White matter structure		MNI-space			DTI	Direction	*t*	FWE	Cohen's *d*
		X	*Y*	*Z*	parameter	of change		Sig	
		10	−36	37	MD	Increased	2.3	*	0.40
					RD		1.8	*	0.33
Anterior corona radiata R		18	28	−10	MD	Increased	2.1	**	0.40
					RD		1.0	*	0.20
		27	17	18	MD	Increased	1.6	**	0.30
					RD		1.2	*	0.20
Posterior limb of internal capsule R		21	−5	13	MD	Increased	2.5	**	0.46
					RD		5.7	*	0.99
Retrolenticular of internal capsule R		28	−21	−3	MD	Increased	4.2	**	0.76
					RD		4.3	**	0.79
Corpus callosum	Genu	13	32	0	MD	Increased	1.1	**	0.30
Forceps minor		−18	54	6	RD	Increased	2.1	*	0.39
Body		4	12	21	MD	Increased	2.2	**	0.41
					RD		1.9	*	0.35
		0	3	24	MD	Increased	1.5	**	0.27
					RD		1.5	*	0.28
	Splenium	−2	−32	18	MD	Increased	2.3	**	0.43
					RD		1.6	*	0.30
		13	−43	13	MD	Increased	1.8	**	0.32

DCD, developmental coordination disorder; DTI, diffusion tensor imaging; FA, fractional anisotropy; FWE, family-wise error corrected; L, left; MD, mean diffusivity; R, right; RD, radial diffusivity; Sig, significant level. [a]*Effects are shown with threshold-free cluster enhancement (TFCE) and a minimum cluster size of 100 voxels. *FWE-p < 0.05; confidence interval: 0.044–0.056. **FWE-p < 0.01; confidence interval: 0.008–0.013.*

The *anterior thalamic radiation* is known for its role in emotional processes and low FA in this structure is related to sadness (Coenen et al., 2012; Jia et al., 2014; Niida et al., 2018). Anterior parts of the *cingulum*, terminating in the frontal lobe, are also involved in attention and executive function (Takahashi et al., 2010; Chiang et al., 2016). The *anterior thalamic radiation* along with the *cingulum* are considered parts of the Papez circuit, involved in emotional processing, semantic memory, and learning (Papez, 1937; Jang and Yeo, 2013; Bubb et al., 2017; Weininger et al., 2019). Extremely high or low levels of inputs from the anterior thalamic nuclei to the anterior cingulate cortex through the *anterior thalamic radiation* and the *cingulum* could increase emotional distraction of behavior (Hartikainen et al., 2014; Sun et al., 2015) and overwhelm attentional resources required for learning (Öhman et al., 2001; Hartikainen et al., 2014). As such, we infer that CO-OP intervention might regulate emotions by allocating more attentional resources to motor performance, which is reflected in neuroplastic changes in white matter microstructure. This interpretation should be considered with caution considering that we did not assess emotion and attention regulation before and after the intervention.

FA in the *white matter tract to the right inferior frontal gyrus* is involved in executive functions, detection of cues (Hampshire et al., 2010), and fine movement control (Liakakis et al., 2011). The *inferior frontal gyrus* has structural connectivity with the *SMA* (Kohn et al., 2014; Vergani et al., 2014), which mediates overt and covert speech initiation (Winsler et al., 2009), as a means of emotion regulation (Beatty and Janelle, 2020). CO-OP uses self-verbalization in the form of overt and covert/inner speech production as a global strategy to solve motor problems (Missiuna et al., 2001) and children's verbal ability predicts their motor outcomes after the CO-OP (Green et al., 2008). Self-verbalization strategies of CO-OP (e.g., global strategy of Goal-Plan-Do-Check and domain-specific strategies of verbal motor mnemonic and verbal rote script; Polatajko and Mandich, 2004) may motivate children by providing effective instructions and selectively attending to the performance-relevant information rather than negative emotions (Anderson, 1997; Beatty and Janelle, 2020). Self-verbalization helps to internalize instructions and use them to acquire the skills (Anderson, 1997). Accordingly, the observed increase in FA of white matter structures underlying the SMA and inferior frontal gyrus might be associated with CO-OP's extensive use of self-verbalization strategy as a regulatory technique.

The SMA also plays a role in planning and executing a motor function. It has structural connectivity with the *cingulate gyrus* (Vergani et al., 2014), transforming emotion into motor experience in situations of reward or punishments (Northoff et al., 2000; Oliveri et al., 2003; Kohn et al., 2014). Taken together, our findings suggest that the CO-OP intervention may play a role in emotion regulation as well. More research is required to better understand CO-OP's role in emotion regulation at behavioral and neural levels.

Motor Performance

We also found increased FA in the *bilateral sensorimotor tracts* immediately beneath the right and left M1, as well as left SMA and parts of the right and left superior corona radiata. Since we did not perform tractography, we are not able to distinguish between corticospinal tracts and corticobulbar tracts originating from sensorimotor cortical areas. In looking at regional microstructure, increased regional FA in the subcortical white matter areas could be explained in an intra-hemispheric or inter-hemispheric context. As for intra-hemispheric improvement, high regional FA underneath M1 might indicate high functional connectivity between M1 and secondary motor areas in each hemisphere (Klöppel et al., 2008; Vergani et al., 2014),

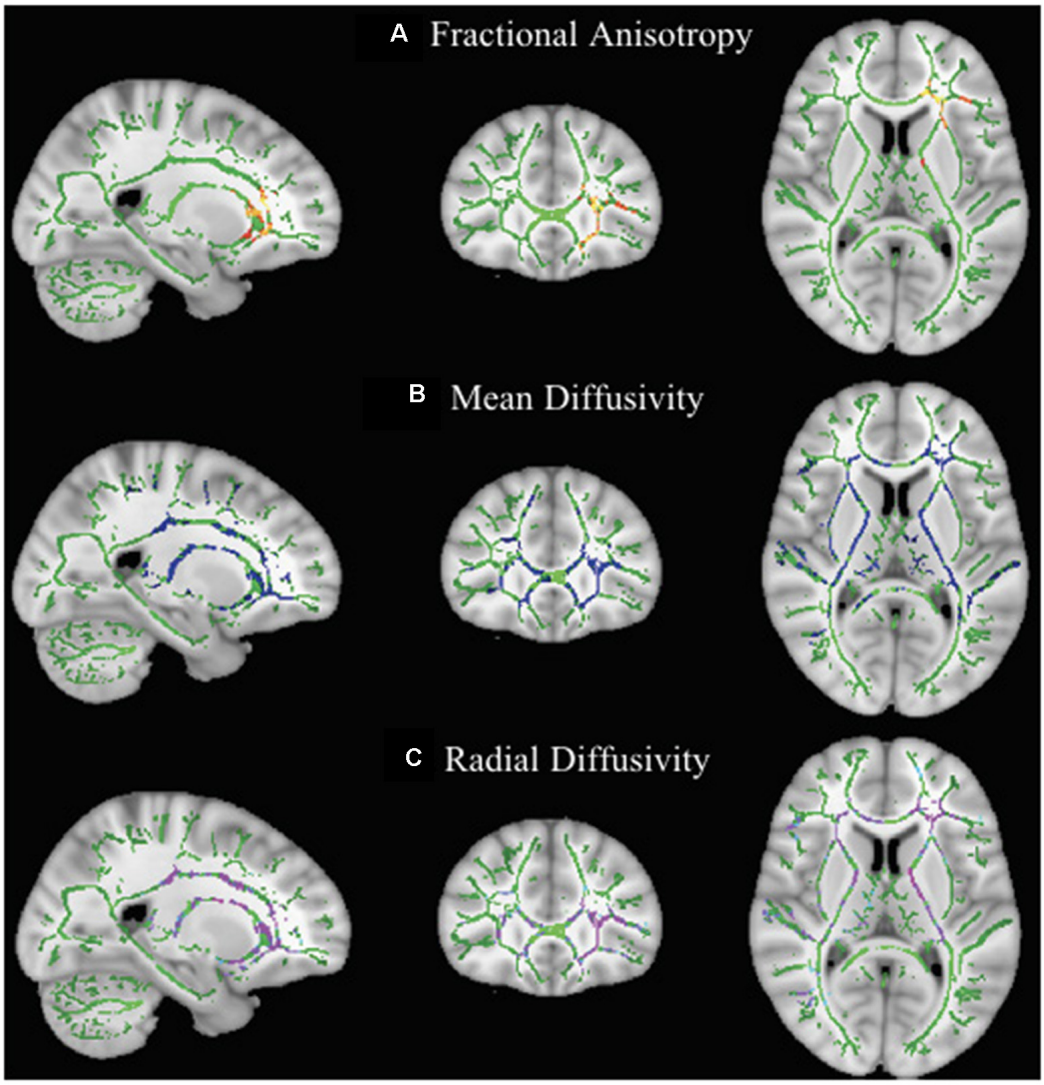

FIGURE 2 | Effect of maturation on DTI parameters in children with DCD. In these images, the white matter skeleton is shown in green and brain structures with altered fractional anisotropy (FA; **A**), mean diffusivity (MD; **B**), and radial diffusivity (RD; **C**) are shown in red, blue, and purple, respectively.

especially in the left hemisphere lateralized for motor control (Guye et al., 2003). However, our resting state results did not show such increased functional connectivity (Izadi-Najafabadi et al., 2020). This discrepancy between our structural and functional results might have different explanations. First, it could be due to a smaller sample size in our resting state analysis or a false positive result in our DTI results. Also, considering the small effect size in the sensorimotor tracts (Cohen's *D* range: 0.18–0.35), these results should be interpreted with caution. Second, it could be related to the unique role of brain function and structure in supporting a specific behavior (Zimmermann et al., 2018). Although structural and functional connectivity are equally important in understanding behavior, they capture independent and complementary features of the brain and do not necessarily show spatial overlap (Zimmermann et al., 2018). Further studies are required to better understand the relationship between brain function/ structure and motor behavior in children with DCD.

Increased FA in the *bilateral sensorimotor tracts* immediately beneath M1 and SMA may be indicative of improved structural connectivity between motor-related brain regions. A transcranial magnetic stimulation study showed that water diffuses from M1 to the subcortical white matter below the prefrontal areas, as well as parts of the corona radiata, internal capsule, cerebral peduncles, and corpus callosum (Klöppel et al., 2008). Also, the SMA sends fibers through the corpus callosum and through the corona radiata to join the corticospinal tract (Vergani et al., 2014). Similarly, we found increased FA in the subcortical white matter immediately underneath M1, SMA, middle and inferior frontal gyrus, frontal medial cortex, as well as parts of the superior corona radiata and corpus callosum. Therefore, we suggest that CO-OP intervention

TABLE 4 | Increased FA after the CO-OP intervention in children with DCD[a].

White matter structure		MNI-space			*t*	FWE Sig	Cohen's *d*
		X	*Y*	*Z*			
Fornix		0	−11	7	2.4	*	0.34
Anterior thalamic radiation L	At precuneus	−19	−55	38	1.3	*	0.18
	At posterior limb of internal capsule	−10	−2	6	4.8	*	0.68
	At red nucleus	−2	−21	−10	3.1	*	0.43
	At parahippocampal gyrus	−21	−34	1	2.9	*	0.41
	At thalamus	−8	−20	3	1.3	*	0.18
Anterior thalamic radiation R	At anterior nucleus	11	−4	10	2.3	*	0.33
	At precuneus	19	−55	38	3.9	*	0.56
	At parahippocampal gyrus	22	−36	3	2.0	*	0.28
	At thalamus	8	−20	3	1.0	*	0.14
Sensorimotor tract L	At M1	−24	−19	46	1.6	*	0.22
	At superior corona radiata	−24	−21	33	1.5	*	0.21
	At SMA	−16	−11	53	1.3	*	0.18
Sensorimotor tract R	At M1	24	−28	47	31.4	*	0.20
	At superior corona radiata	26	−18	34	2.5	*	0.35
	At SMA	12	−12	65	2.2	*	0.31
Inferior fronto-occipital fasciculus R		31	−66	1	5.1	*	0.72
	At cuneus	18	−84	22	1.2	*	0.17
	At external capsule	36	−10	−5	1.8	*	0.27
	At occipital pole	20	−93	2	3.1	*	0.43
	At tapetum	30	−44	17	3.1	*	0.43
Cingulum L	At posterior cingulate cortex	−13	−34	37	3.2	*	0.45
	At posterior corona radiata	−20	−30	35	3.2	*	0.45
	At subgenual anterior cingulate cortex	−6	15	−12	2.6	*	0.37
Cingulum R	At posterior cingulate cortex	13	−34	37	1.2	*	0.17
	At posterior corona radiata	20	−30	34	1.6	*	0.22
	At sub-genual anterior cingulate cortex	6	15	−12	1.5	*	0.22
Corpus callosum	Body	9	−7	28	1.2	*	0.17
	Body	−9	−7	28	2.4	*	0.34
	Body to cingulate cortex	−12	12	25	3.4	*	0.48
	Splenium	−25	−57	16	3.4	*	0.48
	Splenium to cingulate cortex	−12	−35	24	3.7	*	0.52
White matter	To subcallosal area of anterior cingulate cortex	7	21	−19	2.6	*	0.37
	To inferior frontal gyrus L	−32	18	−21	3.5	*	0.49
	To frontal medial cortex R	6	36	−23	1.9	*	0.27
	To middle frontal cortex R	32	23	32	1.4	*	0.21
	To cuneus L	−8	−86	29	3.4	*	0.48

*DCD, developmental coordination disorder; FA, fractional anisotropy; FWE, family-wise error corrected; L, left; M1: primary motor cortex; R, right; Sig, significant level; SMA: supplementary motor area. [a]Effects are shown with threshold-free cluster enhancement (TFCE) and a minimum cluster size of 100 voxels. *FWE-p < 0.05; confidence interval: 0.044–0.056.*

increases FA in the subcortical white matter below motor cortices, facilitates cortico-cortical connectivity between M1 and SMA and/or corticospinal connectivity; this can subsequently improve planning, initiating, and executing a motor task. This interpretation should be considered with caution and further analysis using tractography and graph theory is required to better understand the nature of increased FA in these subcortical white matter structures.

As for inter-hemispheric improvement in FA, right and left M1 and SMA are connected through transcallosal sensorimotor fibers that facilitate interhemispheric inhibition (Fling et al., 2013; Vergani et al., 2014). FA of these transcallosal sensorimotor fibers at the superior portion of the corticospinal tracts are positively correlated to motor ability of children as assessed by MABC-2 (Grohs et al., 2018). Motor learning, especially during bimanual movements, is known to enhance inter-hemispheric interactions (Takeuchi et al., 2012); in our study, all children had chosen at least one bimanual motor goal, such as tying shoelaces or cutting a fruit with a knife, which might explain the improved microstructure of transcallosal sensorimotor fibers.

We did not find any relationship between motor performance and microstructural changes while we had previously reported a relationship between improved motor performance and functional connectivity of the cerebellar lobule I-IV and the DMN related to automatization (Izadi-Najafabadi et al., 2020). Evidence suggests that the brain-behavior relationship is explained differently when considering brain function vs. brain structure. In a large study investigating the whole-brain structural connectivity-behavior relationship vs. functional connectivity-behavior, there were fewer structural connections that were linked to specific behaviors than functional connectivity. And, unlike functional connectivity, a general pattern of structural connectivity is linked to overall cognitive abilities (Zimmermann et al., 2018). These findings may explain why we found a functional connectivity-motor relationship in our results while we did not find such a specific relationship

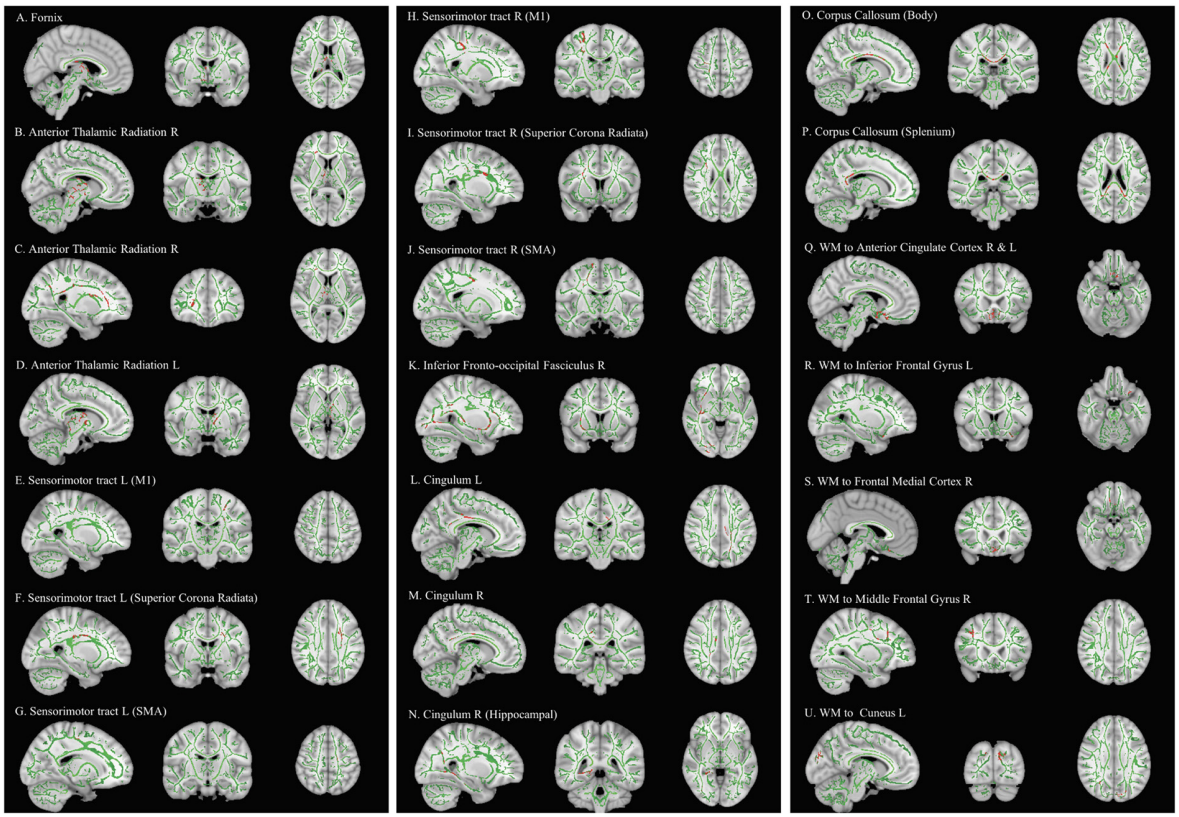

FIGURE 3 | Effect of intervention on fractional anisotropy in children with DCD. In these images, the white matter skeleton is shown in green and brain structures with altered fractional anisotropy are shown in red. L, left; M1, primary motor cortex; R, right; SMA, supplementary motor area; WM, white matter.

using our microstructural measures. Moreover, we suggested that CO-OP was associated with improved microstructural properties of regions involved in self-regulation, attention regulation, and emotion regulation; accordingly, we may not have found a brain-behavior relationship because we did not assess these mediatory behaviors.

Theories to Support Interpretation of Findings

Our results and interpretation of the role of self-regulation, attention, and emotion regulation on motor performance are consistent with the "sensorimotor control framework for emotion regulation" by Williams et al. (2020) and the "Optimizing Performance through Intrinsic Motivation and Attention for Learning (OPTIMAL) theory of motor learning" by Wulf and Lewthwaite (2016).

The sensorimotor control framework for emotion regulation is similar to the internal modeling hypothesis (Adams et al., 2014), with an added component of emotion. Accordingly, all actions start with a motivation or emotional value. To achieve this desired goal, children have to form internal sensorimotor models over time and to learn and plan their movements based on feedback-feedforward processes (Williams et al., 2020). Children may achieve their goal or fail, which engages their emotional response again and modulates their actions; strategies such as avoiding, selective attention, and reappraisal are used to regulate emotions until the child achieves the goal (Braunstein et al., 2017). The OPTIMAL theory of motor learning emphasizes the role of emotions and motivation in motor performance (Wulf and Lewthwaite, 2016). This theory predicts a virtuous vs. a vicious cycle in motor learning; in the virtuous cycle, motivation leads to better motor performance, a sense of autonomy, and self-efficacy, which in turn, motivates the individual to further accomplish motor goals. The vicious cycle, on the other hand, predicts that low motivation results in low motor performance and *vice versa* (Wulf and Lewthwaite, 2016).

Atypical Maturation in Children With DCD

The pattern of brain maturation prior to the intervention in children with DCD contradicts existing reports of longitudinal development in typically developing children and adolescents; typically, white matter microstructure develops with increased FA and decreased MD and RD (Chen et al., 2016; Krogsrud et al., 2016). The observed reduction in FA and increase in MA and RD in this study suggests an overall decline in brain microstructure in children with DCD over a 3-month period. Reduced FA in the left anterior thalamic radiation in children with DCD compared to typically developing children has previously been reported (Brown-Lum, 2017). The current study is the first to report a longitudinal decrease in FA of

this white matter structure in children with DCD, suggesting atypical brain development/maturation might underlie their motor performance difficulty.

In a different analysis, we were not able to find any maturational changes in white matter FA and/or volume in a sample of 12 children with DCD +/- ADHD (Izadi-Najafabadi et al., 2020). The smaller sample size may have reduced the statistical power to detect brain changes, and the inclusion of children with DCD + ADHD could have confounded the results.

In addition, we were not able to find any maturational changes in the white matter microstructure of children with DCD + ADHD prior to the intervention, indicating that their brain development does not follow a typical trajectory. This finding should be interpreted with caution as this might be due to a smaller sample size (DCD: n = 18; DCD + ADHD: n = 10) and reduced statistical power.

No Observed Neuroplasticity in Children With DCD + ADHD

Although CO-OP improved the motor performance of children with DCD + ADHD (Izadi-Najafabadi et al., under review), they did not show any changes in white matter microstructure after the intervention. This is in line with Green et al.'s (2008) study indicating the benefits of CO-OP for children with co-occurring conditions such as ADHD while still experiencing motor problems. Our results are explained considering three evidence-informed statements: (1) the CO-OP intervention relies on attention and self-regulation to mediate its effects on motor performance (Jokić et al., 2013); (2) attention contribution to motor performance is greater in children with DCD + ADHD than children with DCD only (Fong et al., 2016); (3) children with ADHD experience greater difficulty with self-regulation (e.g., attention regulation, emotion regulation; Shiels and Hawk, 2010). In other words, the CO-OP intervention is not able to overcome the attentional and self-regulatory difficulties and its underlying neural mechanism in children with DCD + ADHD. Moreover, children with DCD + ADHD have shown unique brain structure (Langevin et al., 2014, 2015) and function (McLeod et al., 2014, 2016; Thornton et al., 2018) compared to children with DCD. Compensatory attentional control by the posterior cingulate cortex and the cerebellum has been proposed as the neural mechanism of motor coordination in children with DCD + ADHD (Yeh et al., 2012); however, our resting state (Izadi-Najafabadi et al., in press), morphometry (Izadi-Najafabadi et al., 2020), and diffusion imaging analyses did not show similar neural mechanism following the CO-OP.

Additionally, literature suggests that only hyperactivity symptoms of children with ADHD improve over time, and this improvement is highly correlated with reduced FA in the left corticospinal tract (Francx et al., 2015); it is, therefore, hypothesized that development induces a less stimulated/excited motor pathway with decreased FA, which reduces the motor hyperactivity in children with ADHD (Francx et al., 2015). On the other hand, reduced FA in white matter microstructure over time, especially in the frontal and parietal regions, could reflect poorer motor performance as seen in children with DCD (Brown-Lum et al., 2020) and DCD + ADHD (Langevin et al., 2014). Thus, it is unclear whether a reduced FA in the corticospinal tract is beneficial or detrimental in children with ADHD. This highlights how a co-occurring condition such as ADHD complicates our understanding of brain and neuroplasticity following the intervention.

Taken together, some modifications to the CO-OP protocol to better address the attentional needs of children with ADHD (Gharebaghy et al., 2015; Izadi-Najafabadi et al., 2020) or combining CO-OP intervention with medication or other self-regulatory interventions (Reid et al., 2005) might improve its effectiveness and induce permanent brain changes in children with DCD + ADHD.

LIMITATIONS

A smaller than anticipated sample size for the analysis could have biased our results. This study is the first study to use DTI to investigate training-induced brain changes in children with DCD; however, we only performed a voxel-wise analysis of the white matter microstructure, which has spatial inaccuracy (Edden and Jones, 2011). TBSS assumes an accurate registration of the data and it is nearly impossible to detect errors (Jones and Cercignani, 2010); this could add some uncertainty to the results. Further analysis using myelin water fraction analysis, tractography (probabilistic or constrained spherical deconvolution), and graph theory would help to validate our results. In addition, although we conducted the strictest available clean-up pipeline through FSL and excluded any residual motion-contaminated volumes from our data, we did not collect field map data, which would have further cleaned our data and better corrected for the susceptibility-induced distortions. Moreover, we did not find a relationship between behavioral data and white matter microstructure, meaning that behavioral interpretations should be considered with caution.

CONCLUSIONS

Our results indicate that the CO-OP intervention induces microstructural changes in white matter tracts involved in self-, attention-, and emotion-regulation and white matter structures involved in intra-and inter-hemispheric transfer of motor information. These changes were maintained 3 months after the intervention. However, children with a dual diagnosis of DCD and ADHD did not show any microstructural changes following CO-OP intervention, suggesting their different needs in motor-based interventions. These results are consistent with our functional connectivity results of improved motor performance and associated increases in functional connectivity of DMN with the pACC after CO-OP intervention in children with DCD, but not in children with a dual diagnosis of DCD and ADHD (Izadi-Najafabadi et al., 2020).

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 human participants were reviewed and approved by University of British Columbia/BC Children's and Women's Research Ethics Board. Written informed consent to participate in this study was provided by the participants' parent/legal guardian. Child assent was also obtained.

AUTHOR CONTRIBUTIONS

SI was responsible for participant screening, data collection, data processing, statistical data analysis, data interpretation, and drafting the manuscript. JZ conceived and designed the study, coordinated and supervised data collection, and critically reviewed the draft manuscript for intellectual content. All authors contributed to the article and approved the submitted version.

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Genome-Wide Association Study of Motor Coordination

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The ability to finely control our movement is key to achieving many of the educational milestones and life-skills we develop throughout our lives. Despite the centrality of coordination to early development, there is a vast gap in our understanding of the underlying biology. Like most complex traits, both genetics and environment influence motor coordination, however, the specific genes, early environmental risk factors and molecular pathways are unknown. Previous studies have shown that about 5% of school-age children experience unexplained difficulties with motor coordination. These children are said to have Developmental Coordination Disorder (DCD). For children with DCD, these motor coordination difficulties significantly impact their everyday life and learning. DCD is associated with poorer academic achievement, reduced quality of life, it can constrain career opportunities and increase the risk of mental health issues in adulthood. Despite the high prevalence of coordination difficulties, many children remain undiagnosed by healthcare professionals. Compounding under-diagnosis in the clinic, research into the etiology of DCD is severely underrepresented in the literature. Here we present the first genome-wide association study to examine the genetic basis of early motor coordination in the context of motor difficulties. Using data from the Avon Longitudinal Study of Parents and Children we generate a derived measure of motor coordination from four components of the Movement Assessment Battery for Children, providing an overall measure of coordination across the full range of ability. We perform the first genome-wide association analysis focused on motor coordination (N = 4542). No single nucleotide polymorphisms (SNPs) met the threshold for genome-wide significance, however, 59 SNPs showed suggestive associations. Three regions contained multiple suggestively associated SNPs, within five preliminary candidate genes: *IQSEC1*, *LRCC1*, *SYNJ2B2*, *ADAM20*, and *ADAM21*. Association to the gene *IQSEC1* suggests a potential link to axon guidance and dendritic projection processes as a potential underlying mechanism of motor coordination difficulties. This represents an interesting potential mechanism, and whilst further validation is essential, it generates a direct window into the biology of motor coordination difficulties. This research has identified potential biological drivers of DCD, a first step towards understanding this common, yet neglected neurodevelopmental disorder.

Keywords: coordination, development, dyspraxia, neurodevelopment, GWAS, ALSPAC, developmental coordination disorder, motor coordination

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INTRODUCTION

Developmental coordination disorder (DCD) is a neurodevelopmental condition defined by the DSM-5 as a severe impairment of motor skills, usually presenting in early childhood, and in the absence of any other explanatory factor such as a known neurological disorder (e.g., cerebral palsy, acquired brain injury, visual impairment, or intellectual disability) (American Psychiatric Association, 2013).

Developmental coordination disorder is thought to affect approximately 5% of 7–8-year-old children (American Psychiatric Association, 2013), although the estimates vary considerably between study populations and methodological approaches (Wright and Sugden, 1996; Tsiotra et al., 2006; Lingam et al., 2009). In practice, this means there are one or two children with DCD in every school class. Despite this high prevalence and official recognition as a neurodevelopmental disorder in the DSM-IV and -5, DCD remains chronically underdiagnosed by healthcare professionals (Blank et al., 2019). DCD is similarly underrepresented in the research literature, with ten-fold fewer research articles published on DCD than dyslexia between 1985 and 2009 (Bishop, 2010).

Children with DCD have difficulty acquiring fine and/or gross motor skills, making learning age-appropriate activities such as riding a bicycle or catching a ball extremely challenging, even given opportunity to practice. They are more likely to struggle with daily tasks, academically and socially, and are more likely to have overall poorer quality of life in adulthood (Stephenson and Chesson, 2008; Summers et al., 2008; Harrowell et al., 2018; Cleaton et al., 2019). These motor difficulties initially manifest in early childhood and have been shown to persist into adolescence in approximately half of individuals (Losse et al., 1991; Cantell et al., 1994) and a similar proportion continue to be affected into adulthood (Kirby et al., 2008, 2011). There is a current absence of longitudinal data to understand whether the proportion of individuals who improve is a result of access to intervention, developing their own coping strategies, through extensive and deliberate practice or through some other mechanism (Kirby et al., 2011). Therefore, the impact of DCD persisting into adult life is likely to be underestimated.

Many children receive their initial referral for motor function assessment because of their poor handwriting (Barnett and Prunty, 2020). This is when coordination problems are often first noticed in school and begin to affect their academic achievement. Poor academic outcomes are reported in children with DCD. A recent study found that individuals with DCD were less likely to finish school with 5 or more GCSEs (OR 0.27, 95% CI 0.21–0.34), placing them at a considerable disadvantage compared to other students (Harrowell et al., 2018).

Children with DCD require substantial extra support to successfully perform daily activities, as they often find basic tasks such as eating, washing, or cleaning their teeth extremely challenging (Summers et al., 2008). This extra support impacts upon family quality of life and can have a substantial financial and time cost to their carers (Cleaton et al., 2019). Further contributing to this, behavioral problems and difficulties with social interactions can make these relationships more challenging, with these problems often persisting into adulthood (Stephenson and Chesson, 2008).

Co-occurrence with other neurodevelopmental conditions is extremely common, particularly with attention deficit hyperactivity disorder (ADHD) and developmental language disorder (DLD) (Lingam et al., 2010). The reason for these co-occurring conditions is unknown; whether they are due to shared genetic and/or environmental risk factors, or perhaps that one may act as a risk factor for another. Anxiety and depression are commonly associated with DCD, placing individuals at a greater risk of a lifetime of increased vulnerability to mental health issues (Kirby et al., 2008). Individuals with DCD are less likely to participate in physical activities (Cantell et al., 1994) and have a higher chance of becoming overweight (Cairney et al., 2005), increasing their long-term risk of developing obesity related health problems.

The underlying etiology of DCD is multifactorial, and both environmental and genetic factors are thought to play a role. One major risk factor that has been repeatedly associated with DCD risk is premature birth (and therefore low birth weight) (Lingam et al., 2009; Larsen et al., 2013). Similarly, low socio-economic status (SES) has been shown to correlate with increased risk of DCD (Lingam et al., 2009). While these environmental factors have been robustly associated with increased risk of DCD, the mechanism by which they act upon early motor development remains unknown.

It is also widely accepted that genetics play a major role in an individual's risk for developing DCD, although no specific genes or molecular pathways have been reported, to date. Several twin studies have examined the heritability of DCD and revealed genetic contribution estimates from 0.44 (Moruzzi et al., 2010), through to 0.7 (Lichtenstein et al., 2010) and 0.8 (Martin et al., 2006). Although these studies are relatively small scale and vary in their inclusion criteria, they indicate a relatively high potential genetic contribution, particularly for a neurodevelopmental disorder.

Genetic investigations into the underlying cause of other neurodevelopmental disorders such as dyslexia and DLD have identified both Mendelian variants and complex genetic models of susceptibility to contribute to their genetic basis (Becker et al., 2017; Mountford and Newbury, 2017). Mendelian variants are usually single genetic variants that have a negative impact on their resulting protein, preventing it from functioning as it should. This type of Mendelian genetic variation generally results in specific (and often more severe) forms of neurodevelopmental disorders and are extremely rare.

Complex genetic models consider genetic susceptibility or risk conferred by genetic variants that are more commonly found in the general population and interact with environmental factors to increase overall risk of developing a condition. Environmental influences can also act as protective factors, such as participation in sporting activities. So far, other neurodevelopmental disorders have examples from both of these genetic models. It is therefore highly likely that DCD will have a similar genetic etiology combining rare Mendelian variants in some rare cases, but more commonly an overall complex genetic risk which is influenced

by risk and protective environmental factors. It is likely to be genetically and environmentally multi-factorial, with many subtle influences.

There have only been a few studies into the genetic basis of DCD, and so far, no genes have been identified as causative. One study reported a large Canadian family where five of seven children and their mother had a diagnosis of DCD (Gaines et al., 2008). This inheritance pattern is strongly suggestive of a fully penetrant dominant Mendelian genetic variant, which has been inherited from the affected mother, however, no investigation into the underlying genetic cause was reported.

Copy number variations (CNVs), large insertions and deletions of regions of the genome, have also been implicated in DCD and other neurodevelopmental disorders such as autism spectrum disorder (ASD) (Sanders et al., 2015), intellectual disability (Coe et al., 2014) and ADHD (Lionel et al., 2011). These insertions or deletions of genetic material can be inherited or occur sporadically. CNVs occur as a normal part of our individual genomic variation, and we each carry about ten unique or very rare changes. The presence of some specific CNVs results in a clear syndrome, for example certain deletions carried on 16p11.2 (chromosome 16) cause a specific type of language impairment called childhood apraxia of speech (CAS) (Newbury et al., 2013; Fedorenko et al., 2016). More frequently, the effect of an individual CNV is difficult to determine, depending on which genes are contained within the deleted or duplicated region and how much it affects their ability to function. Perhaps surprisingly, the majority of CNVs have no clear role or effect on cell function and appear to be completely tolerated.

The presence of more and/or larger genomic regions, known as enrichment or increased burden of CNVs have also been detected in individuals with DLD when compared to controls with typical language development (Simpson et al., 2015; Kalnak et al., 2018). Individuals with a higher burden, i.e., more CNVs or larger regions of their genomes contained CNVs, tend towards a more severe phenotype. In all these CNV studies, the statistical differences are extremely subtle, and difficult to detect. This is further hindered by individual CNVs resulting in highly heterogeneous phenotypes, and even some of the most common and best understood show a varied phenotype (Mountford et al., 2020).

Enrichment of CNVs has been reported in individuals with DCD (Mosca et al., 2016). This provides a direct link between the overall number and size of copy number changes and DCD, as has been established in other neurodevelopmental disorders.

Also, in cases where individuals carried a pathogenic CNV known to cause other neurodevelopmental disorders (e.g., the 16p11.2 deletion which results in CAS), Cunningham et al. (2019) showed that carriers of these pathogenic CNVs were more likely to have coordination difficulties. While this does not demonstrate a clear association between specific CNVs and coordination, it suggests a link between known causes of neurodevelopmental disorders and motor function. Taken together, these two studies are suggestive of a role of CNVs in motor coordination, and an interesting avenue for further investigation.

For common genetic disorders, the first line of investigation usually comprises of a genome-wide association study (GWAS), in which common genetic variants are compared between unrelated cases and controls. These genetic variants are single base pair changes, called single nucleotide polymorphisms (SNPs), and represent common variation within the general population. Only one GWAS has so far been performed to look for regions of the genome which are commonly shared by individuals with DCD. The study looked at 890 individuals with a diagnosis of DCD co-occurring with ADHD, and did not find any variants that reached genome-wide significance, but did report an enrichment for common variants located within genes with a known neurological function (Fliers et al., 2012). A particularly interesting finding from this study, was that eight of the nine genes that contained variants of suggestive association, played a role in mechanisms of neurite outgrowth and muscle function. Although this is a small and underpowered study, they suggest that motor coordination difficulties with ADHD are associated with genes with both neurological and muscular functions, however, this is yet to be fully elucidated.

In a wider sense, the underlying systems through which DCD and early coordination difficulties manifest are unknown and remains a major unanswered question in the DCD field. The current literature tends towards the theory that subtle neurological changes in the brain, including the cerebellum, may underlie DCD, as opposed to motor or muscular function (Licari et al., 2015; Blank et al., 2019). A recent study used diffusion tensor imaging (DTI) to identify differences in white matter between children with DCD and neurotypical controls (Brown-Lum et al., 2020). They detected white matter differences in three pathways: the corticospinal tract, posterior thalamic radiation, and cerebellar pathways. All three white matter pathways are involved in motor or sensorimotor function, providing compelling evidence that differences in axonal development in these regions may underlie DCD. The identification of genes associated with motor coordination may help to further delineate the underlying etiology and has the potential to link neurological changes to gene function.

One approach to the specific investigation of a disorder in clinical cohorts is the investigation of underlying traits in the general population. This can help to increase sample size and identify underlying molecular mechanisms. Here, we use the Avon Longitudinal Study of Parents and Children (ALSPAC) population data set to perform the first quantitative GWAS of motor coordination in a population cohort. We use four measures from the Movement Assessment Battery for Children (MABC) (Henderson and Sugden, 1992), the gold-standard measure of motor coordination difficulties in the ALSPAC dataset. The ALSPAC cohort provides data on a subset of children across four of the individual MABC tasks: heel-to-toe walking, placing pegs, threading lace and throwing a bean bag into a box (Henderson and Sugden, 1992). Collectively, these test items represent the most robust measure of fine and gross coordination in children with associated genetic data. Lingam et al. (2009) used three of these measures in ALSPAC (heel-to-toe walking, placing pegs, throwing a bean bag) along with additional criteria (see "Discussion") to estimate that 1.8% of children at 7 years old meet the diagnostic criteria for DCD, and an additional 2.23% have probable DCD. Although the full MABC has not been performed

in this cohort, Lingam et al. (2009) showed that the three MABC tasks reported in their study provide a reliable measure of three main domains of motor coordination (balance, manual dexterity, and ball skills). Lingam et al. (2009) further showed that these tasks had concurrent validity with other coordination tests from their own measures and from other studies (Van Waelvelde et al., 2004). In the current study we strengthen the measure of motor coordination by supplementing the three MABC tasks with an additional manual dexterity task ("threading lace" from the MABC) also used by Lingam et al. (2009).

Here we report the first genes to be directly implicated in motor coordination in children, generating the first window into the genetic basis of DCD.

MATERIALS AND METHODS

Ethical Approval

Ethical approval for this study was obtained from Oxford Brookes University Research Ethics Committee (UREC #191311).

Ethical approval for ALSPAC (B2341) was obtained from the ALSPAC Ethics and Law Committee and the Local Research Ethics Committees[1]. Informed consent for the use of data collected via questionnaires and clinics was obtained from participants following the recommendations of the ALSPAC Ethics and Law Committee at the time. Consent for biological samples has been collected in accordance with the Human Tissue Act (2004).

Avon Longitudinal Study of Parents and Children Population Cohort

The study was performed using a large UK population cohort; the Avon Longitudinal Study of Parents and Children (ALSPAC) consisting of 14,541 pregnancies to mothers in the Avon region with anticipated delivery dates of between 1st April, 1991, and 31st December, 1992 (Boyd et al., 2013; Fraser et al., 2013). Of these initial pregnancies, there were a total of 14,676 fetuses, resulting in 14,062 live births and 13,988 children who were alive at 1 year of age.

Avon Longitudinal Study of Parents and Children offers a broad range of developmental phenotype measures spanning the participants' lives, including measures of gross and fine motor skills. Please note that the study website contains details of all the data that are available through a fully searchable data dictionary and variable search tool http://www.bristol.ac.uk/alspac/researchers/our-data/

A subset of children (N = 8,365) was genotyped by ALSPAC using Illumina Human Hap 550-quad arrays which allows the direct characterization of more than half a million common European genetic variants across the Human genome. These data were jointly phased using SHAPEIT2 (Delaneau et al., 2013), which uses relationship information to improve phasing accuracy, and imputed to the Haplotype Reference Consortium (HRC) panel (pre-release 2015). This imputation phase allows the prediction of uncharacterized variation using genetic data from unrelated individuals. The imputation dataset was filtered to include SNPs with an imputation quality score >0.8 (i.e., those with high confidence genotype calls).

Children were excluded from the current study if there were missing data from one of the four measures of coordination from the MABC (Henderson and Sugden, 1992); heel-to-toe walking (F7CR015), placing pegs with preferred hand (F7CR105), threading lace (F7CR211), and throwing a beanbag into a box (F7CR331). In total, data were available for 6500 children across all four measures.

DSM-IV/5 criteria for DCD include a qualification that children with a visual or physical disability which limits movement, or moderate intellectual disability should be excluded from a diagnosis of DCD. Children with a parental report of eyesight problems requiring special arrangements at school (age 7.5–KR566) or visual impairment (age 8.5–SA036A) were excluded. Similarly, parents who reported the child has physical problems requiring special arrangements at school (age 7.5–KR567), the child has ever had a physical disability at (age 8.5–SA037A), or the school reported the child has sensory and/or physical needs (visual impairment, hearing impairment, multi-sensory impairment, physical disability) requiring formalized government special education needs support at (age 11–12–PLASCC65) (N = 159) were excluded. Individuals were excluded if they answered yes to any of the five visual or physical exclusions, if the answers were inconsistent between measures across the time points, or if they were missing data across all three time points.

Finally, children were excluded if they showed evidence of moderate to severe intellectual disability (Weschler Intelligence Score for Children III–Full IQ $\leq$ 50 at age 8.5–F8WS112) (N = 669). Children with missing IQ data were excluded unless they scored above expected (level 5) in the nationally administered key stage 3 (KS3) assessments at age 13–14 in English (ks3_leve), math (ks3_levm), and science (ks3_levs) and were not reported as ever needing provisions for special educational support (age 7.5–KR561, age 8.5–SA030, and age 11–12–PLASCC40). We could confidently conclude that these children did not have moderate intellectual disability. Individuals missing IQ, KS3, and special educational support provision data were excluded.

Individuals were further excluded from this dataset if they did not have imputed genetic data available (N = 1097). Finally, to ensure that all children in the cohort are unrelated, in the case of twins one twin from each pair was removed from analyses (N = 33). The final cohort consisted of 4,542 children (2183M, 2359F).

Phenotype and Derived Measures

Four components of the MABC (Henderson and Sugden, 1992) were identified as representative measures of motor coordination at age 7. The four tasks assess three aspects of coordination: balance (heel-to-toe walking), manual dexterity (placing pegs, threading lace), and ball skills (throwing bean bag into box). Heel-to-toe walking (F7CR015) measures the number of steps the child takes before stepping off a straight line. Throwing a beanbag (F7CR331) is the number of throws out of 10 where the

[1] http://www.bristol.ac.uk/alspac/researchers/research-ethics/

beanbag is successfully thrown into a box using their preferred hand. Finally, the placing pegs task (F7CR105), inserting 12 pegs into a board, and the threading lace task (F7CR211), threading a lace through a series of holes in a board, are both measured as completion time. These raw scores for each individual measure were age-adjusted (Age in months at test), and the distributions of these individual measures are shown in **Figure 1**.

Scores were then expressed as percentile performance against the entire cohort. The placing pegs and threading lace scores were inverted so that for all scores a higher percentile denotes better performance. Each age-adjusted percentile score was transformed into an inverted point scale across the full range (such that individuals scoring between the 0 and 10th percentiles score 9, between the 10 and 20th percentiles score 8, and so on through to individuals between the 90 and 100th percentiles who score 0). Scores were summed across all four measures to generate a summed measure of overall motor coordination (SumQMS4) which was normally distributed (**Figure 2**) and ranged from 0 to 36 (mean 18.02, SD 6.71), where a higher score denotes worse performance. Note that although these measures spread across the full range of performance, the MABC was designed such that there will be a ceiling effect in typically developing children. As such, this test does not sensitively allow us to distinguish between children at the top of the motor skill range (Henderson et al., 2007).

In addition to the quantitative measure of motor ability, a binary measure of motor coordination was also derived. In this binary measure, each of the individual four motor measures were transformed to a point scale, this time focusing upon the lower tail of the distribution–the individuals with the poorest performance. Individuals scoring between the 0 and 2nd percentiles scored 5, between the 2 and 5th percentiles scored 4, between the 5 and 10th percentiles scored 3, between the 10 and 15th percentiles scored 2, between the 15 and 25th percentiles scored 1, and anyone above the 25th percentile scored 0, where again, a higher score denoted worse performance. Cases with motor coordination difficulties were defined as having a total summative score of $\geq$8 across all four tasks (a score that would require an average performance below the 10th percentile across all four tasks, N = 214). Controls were defined as individuals with score = 0 ($\geq$25th percentile on all four tasks, N = 1737). The number of cases falling below this categorical cutoff was N = 214 or 5% of included children.

The available test battery does not allow us to unequivocally say that the children with motor coordination difficulties had DCD, as not all diagnostic criteria could be applied. We thus refer to this group as "probable" DCD (pDCD), allowing us to perform a case-control GWAS. Please note, this differs from the criteria used by Lingam et al. (2009) who also considered impact upon daily life as criteria for DCD and pDCD in accordance with the DSM-IV. All other individuals were excluded from analyses.

Graphs were plotted using the ggplot2 package[2] (Wickham, 2016) within RStudio (v3.5.1).

[2]https://ggplot2.tidyverse.org

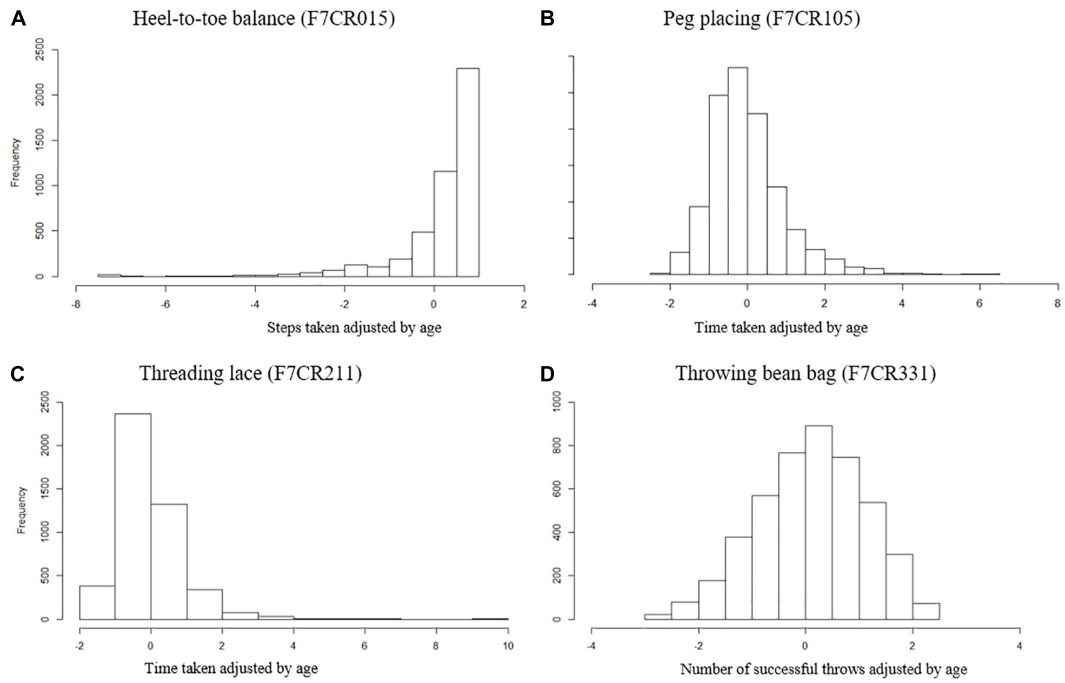

FIGURE 1 | Distribution of MABC test items age adjusted. **(A)** Heel-to-toe walking (F7CR015) showing the number of steps taken (high score denotes good performance). **(B)** Placing pegs (F7CR105) showing the time taken to complete the task (low score denotes good performance). **(C)** Threading lace task (F7CR211) showing the time taken to complete the task (low score denotes good performance). **(D)** Throwing bean bag (F7CR331) showing the number of throws that successfully hit the target (high score denotes good performance).

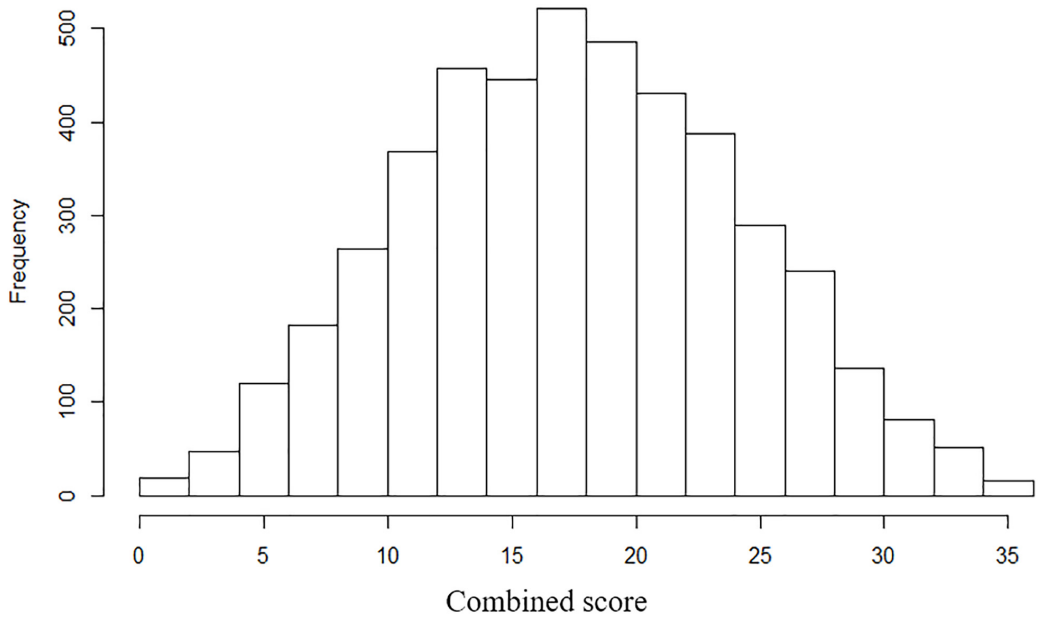

FIGURE 2 | Distribution of summed motor coordination measures (SumQMS4) derived from the MABC test components to reflect an overall motor coordination score, and a maximum possible score of 40.

Genome Wide Association Study

The imputed genotype dataset contained 4,774,020 autosomal and X chromosome SNPs (chrs1-23) for the 4,542 individuals included in the study. A power calculation indicated that this sample provides 90.4% power to detect a variant that explains 1% of trait variance at a genome-wide threshold of significance and 99.0% power at a significance threshold of 1×10^{-5} [assuming a minor allele frequency (MAF) of 0.1, complete linkage disequilibrium (LD) between marker and causal variant]. This means we have sufficient power to detect common contributory variants that account for 1% of the population variation in motor control ability.

Standard quality control measures were performed on genome-wide SNP data prior to analysis (Anderson et al., 2010); SNPs with a minor allele frequency <5%, a per SNP call rate of <5%, a Hardy-Weinberg equilibrium $P < 5 \times 10^{-7}$ ($N = 70$) or a heterozygosity rate more than 3SD from the mean were excluded from analysis. Genotype rates were compared between motor difficulty cases and controls, and SNPs with a differential missing rate ($P < 1 \times 10^{-5}$, $N = 4$) were excluded from further analysis, leaving a total of 4,774,020 high quality SNPs. Individual genotype rate was checked, however, as this was an imputed dataset, all individuals had ≥95% coverage across SNPs.

One factor that can impact genetic association is differences in ancestry between individuals. To avoid this, genetic ethnicity was checked using a Principal Component Analysis (PCA) with the smartpca perl script[3] from the Eigensoft package (Patterson et al., 2006; Price et al., 2006). These analyses compared a pruned set of low LD SNP data (95,225 variants) with the Human Genome Diversity Project (HGDP) dataset[4] which includes seven populations of broad ethnicity (African, American, Central South Asia, East Asia, European, Middle East, and Oceania). The first and second Principal Components were plotted using the ggplot2 package (see footnote text 2) (Wickham, 2016) within RStudio (v3.5.1). All individuals appeared to be of European descent.

A set of 4,774,020 SNPs were analyzed for association using a general linear regression model within PLINK for the quantitative measure of coordination (SumQMS4), and a logistic model for the binary pDCD case/control phenotype. Genome-wide Manhattan and QQ plots were plotted in the qqman package (Turner, 2014) in RStudio (v3.5.1). Zoomed-in plots for suggestively associated loci were generated using Locus Zoom v0.12.0 (locuszoom.org). Genome-wide power calculation was performed using the online Genetic Power Calculator[5].

RESULTS

Quantitative Motor Coordination GWAS

Here we describe the first genome-wide association analysis of overall quantitative motor coordination (SumQMS4) in 4,542 children. This approach examines common genetic variants (SNPs) that are correlated with behavioral outcomes. No SNPs

[3]https://github.com/chrchang/eigensoft/wiki/smartpca

[4]https://www.internationalgenome.org/data-portal/data-collection/hgdp

[5]https://zzz.bwh.harvard.edu/gpc

met genome-wide association ($P \leq 5 \times 10^{-8}$), however, we identified 59 SNPs across seven genomic regions that met the threshold for suggestive significance at $P \leq 1 \times 10^{-5}$ (**Figure 3**). These regions contain several potential genes of interest, that form potential pathways to investigate in future studies.

Figure 3A shows a Manhattan plot indicating suggestively associated regions on chromosomes 3, 4, 5, 6, 14, 15, and 20. **Figure 3B** shows the QQ plot of expected versus observed SNP association *P* values, indicating the absence of population stratification or other confounding variables.

Table 1 lists the seven chromosomal regions associated with SumQMS4, including flanking SNPs and genes. The full association results for all 59 top SNPs with $P \leq 5 \times 10^{-5}$ can be found in the **supplementary materials** (**Supplementary Table 1**).

From the seven associated regions, three chromosome regions (3p25.2, 6p12.1, and 14q24.2) contained more than one suggestively statistically associated SNP, which is considered a marker of "true" association. Region 3p25.2 spans chr3:13076226–13114852 (rs11128630–rs62232913) and contains 16 SNPs with a $P \leq 8.34 \times 10^{-6}$. **Figure 4A** shows a zoomed in view of the locus, showing that the association region lies within the gene *IQSEC1*. Region 6p12.1 spans region chr6:53632969–53654299 (rs9395876–rs4610551) and contains eight SNPs with a minimum *P* value of 9.43×10^{-6}. The zoomed in locus view (**Figure 4B**) shows that this region of high association does not include any coding variants but is directly flanked by *LRRC1*. The most significantly associated SNP was rs8008210 ($P = 1.88 \times 10^{-6}$) which falls within the 14q24.2 region (chr14:70875943–70935875) (**Figure 4C**). This region contains 31 significant SNPs (rs8012142 to rs2293877) and overlaps fully with the entire *ADAM21* gene, the 3' end of *ADAM20* and the 5' of *SYNJ2BP* (**Figure 4C**).

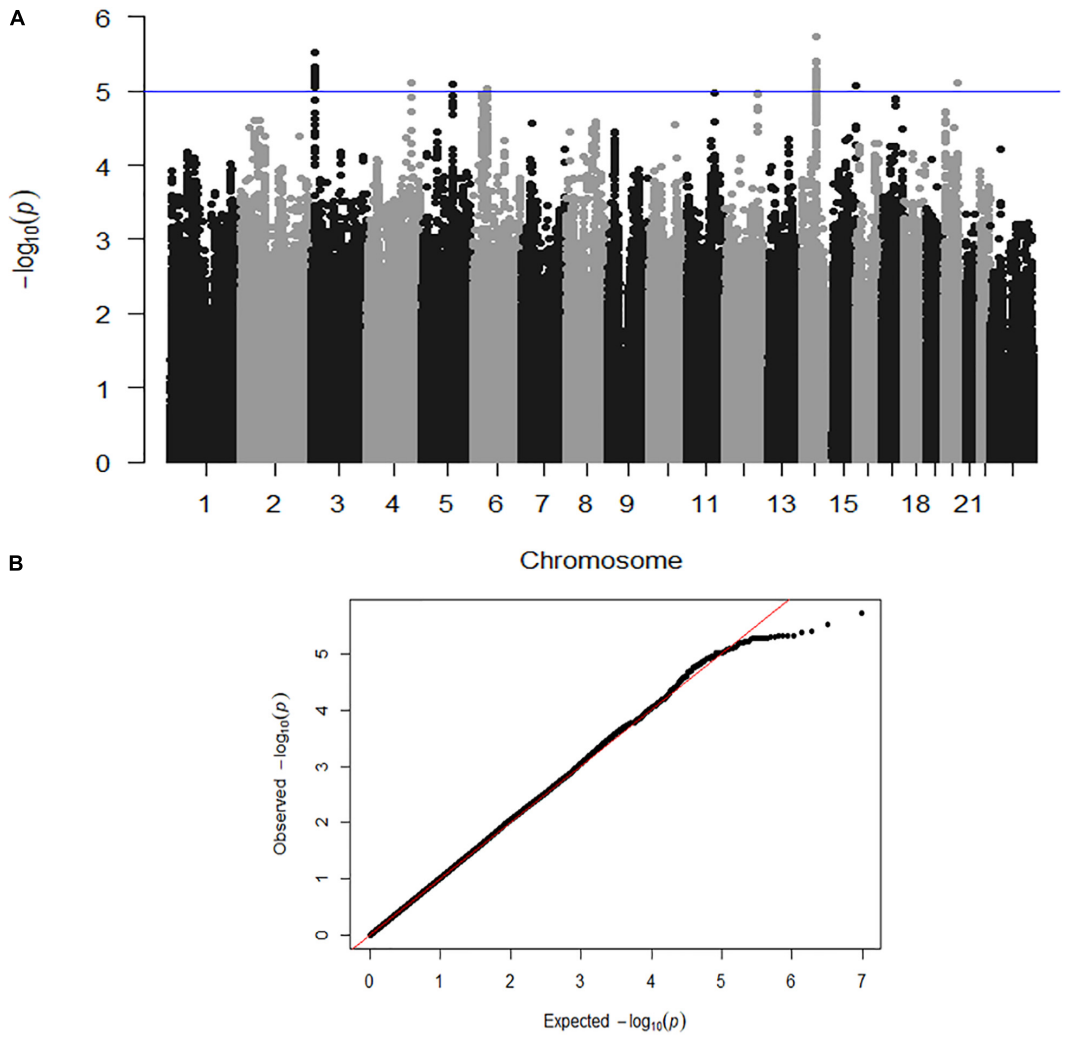

FIGURE 3 | (A) Genome-wide association shown by a Manhattan plot indicating regions of nominal significance. Each point refers to a single genetic variation (SNP). The X-axis shows the position of SNPs along each of the 23 chromosomes. The Y axis shows the log-*P*-value indicating the strength of correlation between the genetic variant and the behavioral outcome (SumQMS4). **(B)** QQ plot showing the observed and expected association values are free from confounding population stratification.

TABLE 1 | Motor coordination (SumQMS4) GWAS top-hits by regions.

Position	Region	Flanking SNPs	No. SNPs	Min *P* value	Gene	Flanking genes	pDCD Case/control min *P* value
chr3:13076226–13114852	3p25.2	rs11128630–rs62232913	16	3.07×10^{-6}	*IQSEC1*	–	0.06095
chr4:157215618	4q32.1	rs17034349	1	7.90×10^{-6}	–	*CTSO-PDGFC*	0.07066
chr5:110555735	5q22.1	rs75575712	1	8.05×10^{-6}	–	*WDR36-CAMK4*	0.006457
chr6:53632969–53654299	6p12.1	rs9395876–rs4610551	8	9.17×10^{-6}	–	*KLHL31-LRRC1*	0.4283
chr14:70875943–70935875	14q24.2	rs8012142–rs2293877	31	1.88×10^{-6}	*SYNJ2BP, SYNJ2BP-COX16, ADAM21,* and *ADAM20P1*	–	0.01161
chr15:100783527	15q26.3	rs12324426	1	8.48×10^{-6}	*ADAMTS17*	–	0.06136
chr20:51577026	20q13.2	rs2904292	1	7.78×10^{-6}	–	*LINC01524-TSH7Z2*	0.03321

The flanking SNPs, minimum P value are reported for each region. Genes contained within the associated region are listed, and in the case of intragenic regions we list flanking genes. The minimum P value from the corresponding region of the pDCD case/control GWAS is also listed (Bonferroni corrected P = 8.5×10^{-4}).

Association With pDCD Case Control

As the quantitative motor coordination score looks for association with motor coordination across the whole population regardless of ability, we then examined the top 59 most significant SNPs for association with the binary coordination difficulties status (pDCD cases = 214, controls = 1,737). The minimum *P* value associated with each region is reported in **Table 1**. None of the top 59 SNPs from the motor coordination GWAS were found to be associated with motor difficulty case-control status (Bonferroni correct *P* threshold is set to 8.5×10^{-4} to account for multiple testing). Individual association results for of the 59 top SNPs for the motor difficulties case/control association each can be found in **Supplementary Table 1**.

DISCUSSION

Here we report the first genetic association study of quantitative motor coordination in a population-based cohort of children. While no SNPs reached the threshold for genome-wide significance ($P \leq 5 \times 10^{-8}$), three chromosome regions (3p25.2, 6p12.1, and 14q24.2) contained more than one suggestively associated SNP ($P \leq 1 \times 10^{-5}$). The identification of a potential association of motor coordination with the genes *IQSEC1*, *LRRC1*, *SYNJ2BP*, *ADAM20*, and *ADAM21* do not offer a single clear potential molecular mechanism for motor coordination.

In a GWAS approach, we observe a statistical correlation between genetic variants and outcomes. This method does not provide information regarding the functional actions of those genetic variants. However, some information can be gained by considering the roles of the genes in which the associated variants fall. The 38Kb region on 3p25.2 ($P \leq 8.34 \times 10^{-6}$) lies across the 5' end of *IQSEC1*, including the 5' untranslated region and coding exon 1. *IQSEC1* (OMIM *610166) also referred to as *BRAG2* or *GEP100*, the IQ motif and SEC7 domain containing-protein are involved in a wide range of cellular processes including cancer metastasis, angiogenesis, myoblast fusion and integrin trafficking (D'Souza and Casanova, 2016). IQSEC1 plays an important role in the structural organization and regulation of neurotransmitters present at the postsynaptic surface and is implicated in axon guidance through the Slit-robo pathway (Onel et al., 2004). Recessive, rare mutations in *IQSEC1* are associated with intellectual developmental disorder with short stature and behavioral abnormalities (OMIM #618687). Ansar et al. (2019) recently reported the first case of bi-allelic pathogenic variants in *IQSEC1*. Functional studies suggested that the pathogenic *IQSEC1* variants resulted in defects in axon guidance and dendritic projection processes. Affected family members presented with severe intellectual disability, short stature, speech aphasia and behavioral problems. Note that the variants described by Ansar differ from the common variations analyzed in the present study. The Ansar variants are rare pathogenic variations that have a clearly detrimental effect upon protein function, while the variants identified in the GWAS are common variants that may not directly impact protein coding, and are therefore expected to have only subtle effects on protein function. Interestingly, Ansar et al. reported that motor milestones were delayed, however, this is a common presentation in severe intellectual disability. Its known role in axon guidance and neurodevelopmental disorders make *IQSEC1* a plausible and interesting candidate for motor coordination, however, follow-up studies are necessary to validate this.

Interestingly, and perhaps relevant to a potential muscular process in DCD, *IQSEC1* is known to play a role in myoblast fusion, through the formation and repair of muscle (D'Souza and Casanova, 2016). Initially identified in fruit flies, and later confirmed in mammalian cell lines, *IQSEC1*/*BRAG2* knockouts showed impaired myoblast fusion (Chen et al., 2003). Although there is insufficient evidence for a direct effect, this highlights the potential for investigating underlying molecular mechanisms for DCD in model organisms.

The 21Kb region located on 6p12.1 ($P \leq 1 \times 10^{-6}$) overlaps with the non-coding region immediately upstream of *LRRC1*, leucine rich repeat containing 1. *LRRC1* (OMIM *608195) is not

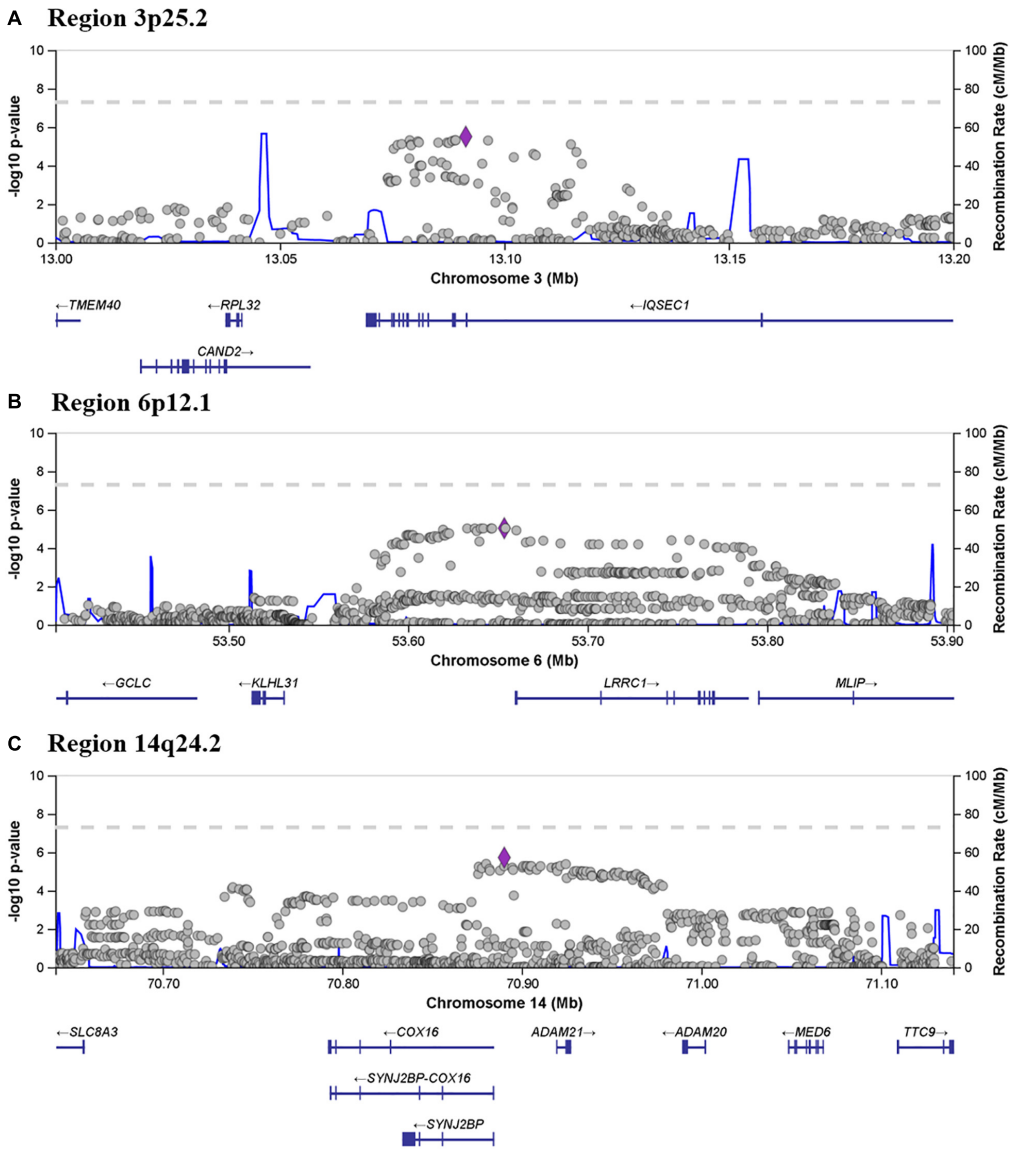

FIGURE 4 | Locus zoom showing associated and flanking regions in **(A)** 3p25.2, **(B)** 6p12.1 and **(C)** 14q24.2, where more than one SNP met the criteria for suggestive association. Genes located within and flanking each of the three regions are indicated below each panel.

reported to be associated with any genetic disorders. Research on the function of this gene is limited, however, it is reported to be involved in the Wnt/β-catenin signaling pathway, which is important in early development, and plays a role in cancer (Daulat et al., 2019).

The most significantly associated SNP fell within a 60Kb region within 14q24.2, overlapping with the 5' region *SYNJ2BP*, the 3' end of *ADAM20*, and the entirety of *ADAM21*. SYNJ2BP, synaptojanin 2 binding protein (OMIM *609411) is located on the outer membrane of the mitochondria (Hartmann et al., 2020) and forms an unusual gene read-through product with neighboring gene *COX16* (OMIM *618064) (**Figure 4**), a known mitochondrial complex IV assembly factor (Cerqua et al., 2018). Mitochondria play a vital role in brain function, and therefore this represents an interesting potential mechanism to further explore. On the other hand, *ADAM20* (OMIM *603712) and *ADAM21* (OMIM *603713), metallopeptidase domain 20 and 21, have not been previously reported to be associated with disease, and both are testes specific proteins (Hooft van Huijsduijnen, 1998).

Fliers et al. (2012) reported an enrichment of genes implicated in neurite outgrowth and muscle function in their GWAS of motor coordination measures in a cohort of children who met the criteria for ADHD. The genes and pathways identified in this study comprise a similar narrative - that genes with a known involvement in neuronal function are enriched. The total number of genes that were robustly associated in our study are too low to undertake a pathway analysis. The results of both studies indicate

that neuronal mechanisms play a role in motor coordination, and that this is a promising area of future research.

Neither Fliers et al. (2012) nor the present study identified any SNPs that met the threshold for genome-wide significance. This is likely due to the relatively small sample sizes in both studies. As we could detect only a moderate effect size, far larger sample sizes are necessary to obtain statistically robust results. For example, a recent GWAS on schizophrenia successfully identified several novel and biologically validated regions of association (Schizophrenia Working Group of the Psychiatric Genomics Consortium., 2014). The success of this study is in part due to the sample size of more than 36,000 cases and 110,000 controls. The present study case/control analysis had only 214 cases and 1,737 controls, which meant that analyzing the derived quantitative measure of coordination (SumQMS4) provided more power than a case/control GWAS. Sample size is a major limitation of this study, and hence the findings should be considered as preliminary. Larger sample sizes, in the tens of thousands, are necessary for future GWAS, making meta-GWAS that combine multiple datasets a viable and potentially fruitful direction, although these approaches can present their own difficulties in terms of robust and consistent phenotype measurement.

The ALSPAC study contained a limited range of measures relating to motor coordination, however, the four components of the MABC are included in the current edition (MABC-2; Henderson et al., 2007) and are widely used for assessing and diagnosing motor coordination difficulties (Henderson and Sugden, 1992; Henderson et al., 2007; Blank et al., 2019). The complete MABC/MABC-2 is intended for identifying children with coordination difficulties and is not designed to accurately measure subtle differences in children within the typical range of ability. By deriving a composite measure of motor coordination (SumQMS4) using the four MABC elements, we were able to produce a general approximation of coordination ability at age 7 using the available data. Using a derived quantitative measure of coordination permitted us a far larger sample size ($N = 4542$) than a case-control analysis (motor difficulties case $N = 214$, controls $N = 1737$).

We are able to resolve the "tail" of children with coordination difficulties more reliably than those within the typical range. By defining the binary motor difficulties case group based on poor performance across multiple tests (<10th percentile), we defined an approximation for pDCD cases. It should be noted that this "case" definition is limited, and a full test battery is required to confidently diagnose DCD cases.

Lingam et al. (2009) used the same ALSPAC cohort to report the prevalence of DCD as 1.8%, and 2.23% met the criteria for probable DCD. The full available cohort ($N = 7399$) was included in their analysis as they were not limited to participants with available genotype data ($N = 6500$), prior to exclusions. Lingam et al. used three of the MABC elements (heel-to-toe walking, placing pegs and throwing a bean bag into a box) to represent each of the realms of coordination. Additional criteria (evidence that coordination difficulties impact their daily life) were applied to robustly define a DCD diagnosis in accordance with the DSM-IV definition. A DCD diagnosis was only applied if children had substantial handwriting difficulties at key stage 1 (age 7) and a parent reported difficulties with daily living. As in our study, Lingam et al. also excluded children with known non-developmental explanatory conditions (i.e., visual impairment, or medical condition), and children with mild intellectual disability (WISC FIQ < 70). In comparison, our inclusion criteria are more relaxed to allow us to capture motor difficulties more broadly, rather than a confident diagnosis of DCD or pDCD. While we recognize that using the additional life impact criteria would greatly improve confidence of diagnosis, it would have further reduced the already limited sample size, highlighting the need for balance between power and specificity.

Our results suggest a potential neuronal etiology to motor coordination difficulties, supported by the current literature (Licari et al., 2015; Blank et al., 2019; Brown-Lum et al., 2020). The identification of suggestive association of motor coordination with the genes *IQSEC1*, *LRRC1*, *SYNJ2BP*, *ADAM20*, and *ADAM21* do not offer a single clear potential molecular mechanism for motor coordination. Instead, it indicates *IQSEC1*, and the potential role of axon guidance and dendritic projection processes in motor coordination. This represents the most interesting candidate gene, although further validation would be necessary to understand the underlying mechanism.

Evidence that axonal development may be disrupted in children with DCD comes from a recent DTI study which compared the white matter structure of 31 children with a diagnosis of DCD to 30 neurotypical children (Brown-Lum et al., 2020). They found that children with DCD showed white matter differences in the corticospinal tract, posterior thalamic radiation, and the cerebellar pathways; all three of these regions have known roles in motor coordination. The low axial diffusivity observed in these areas are strongly suggestive of alterations in axonal structure and/or function within these regions (Brown-Lum et al., 2020). Brown-Lum et al. (2020) observed evidence of axonal changes in these regions, further supporting the potential role for axonal function in DCD.

Other neurodevelopmental disorders such as DLD provide a model for the genetic study of DCD and have uncovered underlying molecular mechanisms in related pathways. For example, Prasad et al. (2012) discovered a rare copy number variant in the gene *ROBO2* that was associated with ASD, while common variants in the same gene have been associated with expressive vocabulary development in infants (St Pourcain et al., 2014). The association of common variants in *ROBO2* with expressive vocabulary directly implicates the Slit-robo pathway and axon guidance as potential mechanisms for language development, and this mechanism is reflected in the association observed here with *IQSEC1*. A second example is the gene *FOXP2* in which some specific rare genetic variants result in a subtype of language disorder called childhood apraxia of speech (CAS OMIM #602081) (Lai et al., 2001). In contrast to this specificity between genotype and phenotype, common variant within the *FOXP2* gene have been associated with both ADHD (Demontis et al., 2019; Soler Artigas et al., 2020) and

intelligence (Lam et al., 2019) in GWAS studies. The role of rare and common variants specific to DLD and language disorders was comprehensively reviewed by Mountford and Newbury (2017).

It is highly likely that DCD will have a similar pattern to that seen in DLD, ASD, and other neurodevelopmental disorders, whereby there are both familial inherited variants that underlie specific phenotypes, and common variants in genes that contribute to motor coordination difficulties. The identification of these genes through a combination of family and large GWAS studies will contribute greatly to the underlying causes of DCD and help to support its recognition as an important neurodevelopmental disorder.

DATA AVAILABILITY STATEMENT

ALSPAC genotype data analyzed in this study are available upon application as outlined at http://www.bristol.ac.uk/alspac/researchers/access/. The ALSPAC website additionally contains details of all the data that is available through a fully searchable data dictionary and variable search tool (http://www.bristol.ac.uk/alspac/researchers/our-data/).

ETHICS STATEMENT

Ethical approval for this study was obtained from Oxford Brookes University Research Ethics Committee (UREC #191311). Ethical approval for ALSPAC (B2341) was obtained from the ALSPAC Ethics and Law Committee and the Local Research Ethics Committees (http://www.bristol.ac.uk/alspac/researchers/research-ethics/). Informed consent for the use of data collected via questionnaires and clinics was obtained from participants following the recommendations of the ALSPAC Ethics and Law Committee at the time. Consent for biological samples has been collected in accordance with the Human Tissue Act (2004).

AUTHOR CONTRIBUTIONS

DN, AB, and HM designed and conceived the experiment. AH was the ALSPAC data buddy for this project and compiled and verified all ALSPAC datasets. DN and HM performed the genetic analyses. DN, HM, and AB wrote the manuscript. All authors contributed to the article and approved the submitted version.

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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.2021.669902/full#supplementary-material

Supplementary Table 1 | Full list of association results for the 59 SNPs that met the threshold for suggestive association ($P \leq 5 \times 10^{-5}$) on the SumQMS4. The statistical significance for each of the 59 SNPs is also reported for the binary DCD case/control association test.

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IMAGINE THAT! MENTAL TRAINING FOR CHILDREN WITH DEVELOPMENTAL COORDINATION DISORDER

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Movement is important for children's health and well-being. Most children find it easy to learn to move but children with developmental coordination disorder (DCD) find it hard. It can be tricky for them to plan and control their movements. DCD affects 1 in every 20 children. It makes important tasks difficult, like getting dressed or playing games and sports. Scientists have found that children with DCD have different activity in some brain areas compared to other children. Mental training can increase activity in these areas of the brain. One type of mental training is motor imagery, which involves imagining doing movements. Another type of mental training is action observation, which involves carefully watching how people make certain movements. These techniques can help children with DCD get better at moving. This means that doing mental training might help make life easier for children with DCD.

WHAT IS DEVELOPMENTAL COORDINATION DISORDER?

DEVELOPMENTAL COORDINATION DISORDER

A medical condition that makes it difficult for people to plan and control movements.

Think about a time when you reached for something, maybe a cup of juice, and knocked it over! Although you had probably made this movement successfully many times before, sometimes movements do not turn out as we planned. This is rare for most of us, but it is a daily problem for children with **developmental coordination disorder** (DCD). DCD is a medical condition that makes it difficult for children to learn to move skillfully. Their movements look clumsy, and they often make mistakes. DCD is usually diagnosed in children between ages 5–8, and it affects 1 in every 20 children. That means, on average, one child in every school class may have DCD—so it might even affect someone you know. DCD causes big problems for these children. They find it hard to do everyday tasks like feeding themselves or getting dressed, which can be very frustrating for them. They also struggle with playground games and sports, as they are not able to move as well as other children their age. This means they often also struggle to make friends or to do well in school. These things make day-to-day life more difficult for children with DCD. The good news is that scientists are starting to understand what causes DCD. They are also finding ways to help children with DCD to move better.

WHAT CAUSES DCD?

Scientists do not yet know the exact cause of DCD. Research using brain scanning techniques is starting to indicate why DCD might occur. Scientists have shown that children with DCD have different brain activity than children without DCD [1]. There are three main brain areas involved in movement, which are less active in children with DCD (Figure 1) [1]. The first area is an area across the center of the brain that helps to plan and prepare movements. The second area is more toward the front of the brain and is involved in copying and imagining movement. The third area is at the back of the brain and helps us to coordinate our movements. The lower activity in these areas might explain why children with DCD struggle to perform everyday movements.

We learn important movements, such as throwing, catching, or kicking, through practice. As we practice, we build up a picture in our minds about how the movement should look and feel when we do it well. We then use the picture to help us plan how to do the movement. We learn by comparing how the movement looks and feels against the mental picture. If the movement matches the picture, we know we did it right. If it does not match, we know we did it wrong and can try to correct it. Scientists call this picture an **internal model**. Scientists think that children with DCD might struggle with movements because they cannot create an internal model as they practice. This makes it hard for them to plan movements because they do not know how the

INTERNAL MODEL

A mental picture of what a movement should look like and feel like when we do it well.

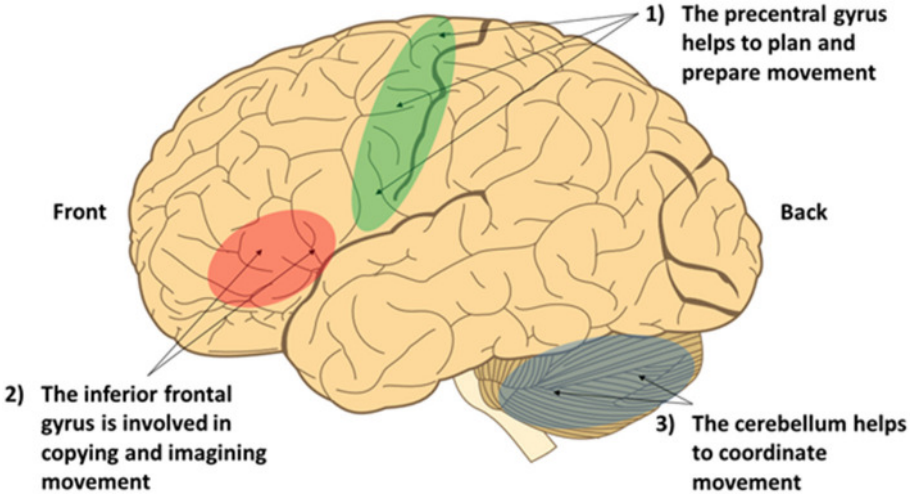

Figure 1

Brain areas that are important for movement but are less active in children with DCD. (1) The precentral gyrus (green) helps to plan and prepare movement. (2) The inferior frontal gyrus (red) is involved in copying and imagining movement. (3) The cerebellum (gray) helps coordinate movements.

movements should look or feel. This means that they do not know if they are doing a movement correctly, and so they struggle to improve. The brain areas shown in Figure 1 are believed to help create internal models as we practice [1]. This could explain why children with DCD have less activity in these brain areas.

CAN DCD BE TREATED?

Children with DCD may face difficulties all their lives, so they must learn to live with them. They might adapt tasks to make life easier. For example, they may use Velcro-strap shoes to avoid tying laces, or they might avoid wearing certain shirts because they struggle to fasten the buttons. They might even completely avoid doing certain activities. For example, they may skip P.E. lessons in school, or avoid taking part in playground games and sports teams. This is a problem, because regular exercise is important for physical and mental health. The good news is that, once it has been diagnosed, children with DCD can be helped to improve their movement skills. Current techniques focus on doing repetitive physical practice. Therapists may ask children with DCD to repeat movements over and over again. To help the children, therapists might make tasks easier or split them up into smaller parts. However, scientists have suggested that just practicing movements is not enough to help children with DCD to improve. Instead, mental training that targets the less-active brain regions could be helpful [1].

CAN MENTAL TRAINING HELP CHILDREN WITH DCD?

MOTOR IMAGERY

Imagining how performing a movement would look and feel.

Scientists believe that mental training techniques can help children with DCD. One mental training technique that can improve movement is called **motor imagery**. Motor imagery involves imagining

movements, encouraging people to imagine both how a movement should look and how a movement should feel. You could try it yourself. Choose an object near you, reach for the object, grasp it, and bring it back toward you. Think about what you see and how the movement feels. Were there any sounds you heard as you moved? Now, without moving, imagine seeing your hand and arm reach and grasp the object, and imagine the feelings and sounds of doing it. That is motor imagery! Scientists in Australia have shown that motor imagery training can help children with DCD (aged 7–12) improve their movements [2]. The scientists asked one group to imagine and then practice doing movements like catching a ball, several times over 5 weeks. Children who did this motor imagery improved more than other groups that just did physical practice, or that did no training. But why does motor imagery work? Well, when we do motor imagery, the brain areas shown in Figure 1 are all more active [3]! Since these brain areas are less active in children with DCD, motor imagery helps to activate them. By doing motor imagery to activate these brain areas on a regular basis, children with DCD might be able to improve their movements.

Although motor imagery helps, it is not easy for children with DCD to imagine how a movement looks and feels. In fact, many children with DCD struggle to imagine themselves doing movements [2]. Scientists are investigating ways to help make motor imagery easier for these children. One way to help is by showing them movements. Watching movement is called **action observation**. Think about when you are in a P.E. lesson. You often watch your teacher do a movement and then copy it. This activates similar areas of the brain to motor imagery. For example, children with DCD could be given a video showing them what the movement should look like and asked to imagine the feelings of doing the movement at the same time (Figure 2). This is a bit like watching your favorite soccer player taking a penalty kick on television, while trying to imagine that *you* are the one kicking the ball and scoring the winning goal! Scientists call this **combined action observation and motor imagery (AOMI)** [3]. Doing AOMI means children with DCD do not have to imagine what the movement looks like because it is shown to them on video. This should make it easier, as they only need to imagine the feeling of the movement whilst they watch the video. Scientists have started to investigate brain activity when people do AOMI, and guess what? It causes more activity in the brain areas involved in movement than just doing motor imagery [3]! For this reason, AOMI might be better than motor imagery alone for improving movement in children with DCD.

ACTION OBSERVATION

Watching people perform movements, either on video or in live demonstrations.

COMBINED ACTION OBSERVATION AND MOTOR IMAGERY (AOMI)

Watching a video of a movement while at the same time imagining the feeling of performing the movement.

Scientists in the UK have started to research whether AOMI can help children with DCD to move better. One study looked at how well children with DCD (aged 7–12) could copy the movements of another person, and they found that AOMI improved their ability to copy [4]. AOMI was even more helpful than motor imagery! This means that AOMI may help these children learn movements more easily when copying demonstrations. Other scientists have shown that AOMI can

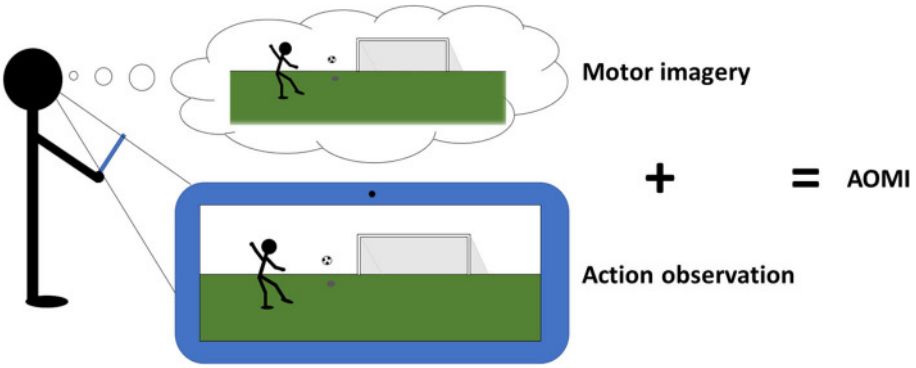

Figure 2

Combined action observation and motor imagery (AOMI) involves watching a video of a movement (such as kicking a ball) and, at the same, time imagining the feeling of doing that movement.

help children with DCD (aged 7–11) to learn quicker and to move their hands and eyes more skillfully [5]. These two experiments are the first to show that AOMI can help children with DCD to get better at planning and controlling their movements. In both experiments, improvements were found after only one session of repeated AOMI practice. As AOMI involves watching how the movement should look whilst imagining how the movement should feel, it might help children with DCD to develop an internal model that they can use to help them improve their movements [4, 5]. Current research is trying to find out exactly how effective AOMI can be in helping children with DCD to get better at everyday movements.

CONCLUSION

DCD is a complex medical disorder that can make everyday movements difficult and frustrating for many children. Although the cause is not fully known, science is helping us to understand the role of brain activity in DCD. Motor imagery can help children with DCD to improve their movements, but more recent research shows us that combining imagery with action observation may be even better. Scientists are now hopeful that AOMI can support children with DCD to move better, helping them to perform their daily activities more easily, and improving their quality of life.

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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.

YOUNG REVIEWERS

THE TI SUMMER INTERNS, AGES: 14–16

We are Belle, Brodi, Emma, Isobel, Leah, Ella, and Grace. We are a group of high school students from different schools across the Sunshine Coast participating in a Summer Internship at the Thompson Institute. We all have an interest in Science and in particular Mental Health and Neuroscience, and enjoy learning more about these topics by reading the articles in Frontiers for Young Minds. We enjoy collaborating with researchers to help them better understand the adolescent experience. We

were helped to review this article by researchers from the Thompson Institute while participating in the Summer Internship.

AUTHORS

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I am a postdoctoral research associate at Manchester Metropolitan University. I completed my Ph.D. at Teesside University in 2020. My previous research investigated the effects of combined action observation and motor imagery (AOMI) on imitation in children with and without developmental coordination disorder (DCD). I am now continuing to research the benefits of AOMI for children with DCD and how best to improve daily activities for this population. *m.scott@mmu.ac.uk

GREG WOOD

I am a senior lecturer in motor control and learning at Manchester Metropolitan University. I completed my Ph.D. in 2011, which looked at anxiety, eye movements, and performance of footballers. My research now focuses on vision in sport and people with movement problems. I am very interested in issues related to developmental coordination disorder and hand amputation.

PAUL S. HOLMES

I am a professor of motor cognition and deputy pro-vice chancellor in the Faculty of Health and Education at Manchester Metropolitan University. My research interests include motor cognition in human performance and movement rehabilitation. I have published widely, focusing on motor imagery and action-observation mechanisms. I have also worked as a sport psychologist in high-performance sport and have traveled with teams from Great Britain and England to major championships across the world.

BEN MARSHALL

I am a lecturer in sport and exercise psychology at Manchester Metropolitan University. I completed a Ph.D. in 2019, which investigated how combined action observation and motor imagery can be used to improve the hand-eye coordination of children with developmental coordination disorder. I am now interested in researching how we can use virtual reality to improve combined action observation and motor imagery interventions. In my spare time, I like cycling in the beautiful countryside near my home (when it is not raining).

JACQUELINE WILLIAMS

I am an associate professor from Victoria University in Melbourne, Australia. In my research, I work to understand the impact of developmental coordination disorder on children and their families. I work to develop strategies to reduce the impact of DCD and I engage with teachers, doctors, and occupational therapists, as well as the wider community, to raise awareness of DCD and its impact. Away from work, I enjoy exercising and spending time with my young children, and I love to sit with a cup of tea and a good book.

DAVID J. WRIGHT

I am a senior lecturer in psychology at Manchester Metropolitan University. My research focuses on two mental training techniques called action observation and motor imagery. I am interested in the brain activity when people do these techniques, as well as how they can be used to help people improve their movement skills. I am currently exploring how combined action observation and motor imagery interventions can help children with developmental coordination disorder learn how to perform everyday movement tasks. When I am not working, I enjoy spending time with my wife and two young children, running, and riding my bike. *d.j.wright@mmu.ac.uk

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