Leonard, H.C. & Hill, E.L. (2015, in press). Executive difficulties in developmental coordination disorder: Methodological issues and future directions). *Current Developmental Disorder Reports*. Doi: 10.1007/s40474-015-0044-8 (Springer)

Executive difficulties in developmental coordination disorder: Methodological issues and future directions

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Keywords: Developmental Coordination Disorder, dyspraxia, motor development, executive function

Abstract

Motor skills and cognition have often been studied separately, but there is increasing understanding of the close relationship between these abilities over development. Motor coordination difficulties are central to the diagnosis of Developmental Coordination Disorder (DCD), and recent evidence suggests that certain cognitive processes, known as ‘executive functions’, may be affected in individuals with this neurodevelopmental disorder. In this article, we review the research concerning executive functions in DCD, considering behavioural, neuroimaging and questionnaire studies of a range of processes. We highlight methodological issues relating to our current understanding of executive functioning difficulties in DCD, including problems with interpretation of results based on the tasks used. We suggest future directions for research in this area, including the relationship of laboratory research to interventions within ‘real-world’ contexts.

Introduction

Motor skills are essential for activities of everyday life, and the ability to move around and to manipulate objects impacts our understanding of the world [1, 2]. This relationship between motor skills and cognition is mirrored in the close interrelation of the neural areas associated with motor function (e.g., the cerebellum) and cognitive control (e.g., the prefrontal cortex) [3, 4]. However, motor and cognitive abilities are most often studied separately and, although motor difficulties are recorded in many neurodevelopmental disorders, the focus of psychological investigations in atypical development is usually cognition [3, 5]. Developmental Coordination Disorder (DCD), on the other hand, provides an ideal opportunity to investigate the relationships between motor and cognitive abilities as it is a disorder diagnosed on the basis of difficulties in acquiring and executing motor skills. These difficulties are not due to a medical condition, and have an impact on activities of daily living and academic achievement [6]. In this article, we review the literature regarding a particular group of top-down cognitive processes, known as ‘executive functions’, in DCD, and consider the reciprocal relationship between these processes and motor impairments. In doing so, we highlight a number of methodological issues raised by these studies and consider future directions for this research, both in order to improve our own understanding of DCD and to increase educational and clinical professionals’ awareness of the disorder and any associated difficulties in cognition.

What are executive functions?

Although there are numerous definitions of executive functions (EFs) throughout the neuropsychological literature, there is general agreement that they are a range of processes or ‘higher-order’ thinking skills, which direct cognition and behaviour toward a particular goal [7] and are under voluntary, conscious control [4, 8]. While these complex cognitive skills have traditionally been related to the functioning of the prefrontal cortex, there is increasing evidence of structural and functional connections between the prefrontal cortex and the cerebellum, which is usually associated with motor skills [3,4]. This close interrelation between neural pathways is likely to drive the relationships between motor and cognitive deficits seen in a range of neurodevelopmental disorders [3], which are usually measured through behavioural performance on motor and EF tasks. Understanding executive functioning therefore requires a multi-level approach, considering the biological and cognitive processes that influence EF behaviour, as well as specific environmental influences, such as the pressures of a classroom environment, which could affect each of these levels of causation [9].

At the cognitive level, three ‘core’ EFs have been suggested [10], namely: *working memory*, which represents the ability to store information in memory while processing another task; *inhibition*, which involves exerting control over one’s natural responses (e.g., suppressing a response even when there are highly rewarding internal and external outcomes of not supressing it), and *cognitive flexibility* (or switching), which allows one to be flexible in approaching a problem by adapting to different rules or demands of the task [8]. Studies from adult neuropsychological patients highlight two further executive processes related to frontal lobe functioning [11]: *planning*, which involves developing goals, monitoring performance and adjusting behaviour in order to achieve these goals, and *fluency*, which is the ability to generate a number of responses around a particular theme, thus testing the efficiency of search processes and creative thinking [12]. It is the broader view of these five executive functions that will be considered in the following review. EFs develop gradually between infancy and early adulthood [13, 14], and each EF may follow a different developmental trajectory [8, 14]. Aspects of EF pervade all areas of our everyday life and are closely related to measures of intelligence [15] and to academic achievement [16, 17]. Given the close relationship between neural pathways related to motor and EF skills [3, 4], investigating EFs in children with DCD has important implications for academic and employment outcomes. With this in mind, we will now review the literature regarding EFs in DCD, first considering the results from behavioural measures and standardised tests.

Behavioural measures of EF in DCD

A recent meta-analysis [18] reviewed DCD research between 1998 and August 2011, and reported clear difficulties in EF across a range of standardised and experimental measures assessing planning [19], inhibition [20-23], working memory [21, 22, 24-26] and cognitive flexibility [21, 22, 27]. This pattern is also evident in more recent research and studies not included in the meta-analysis, including those with children ‘at-risk’ of DCD who have some motor impairments (i.e., those who demonstrate motor difficulties or meet criteria for DCD in screening studies, but who have not received a formal diagnosis) [28-43]. Additional difficulties have been highlighted in fluency in children with DCD and those ‘at-risk’ [42]. However, despite this overall picture of EF impairments across studies, closer inspection of the methodologies used highlights some key issues that are important for our understanding of these results. One problem which has been highlighted in the EF literature is the use of tasks which are highly complex or which tap multiple executive functions [9], and may therefore depend on a range of other cognitive abilities and general IQ to perform them successfully. This may produce different results across studies that use the same task with different samples of children with DCD [29, 41] or DCD compared to an 'at-risk' sample [21, 28]. Another issue concerns the demands of the task in terms of the domain being tested, i.e., whether the task requires visuospatial processing, verbal comprehension and verbal or motor responses. The task demands of the different EF studies are depicted in Figure 1 and will be considered in more detail below.

---Figure 1 about here---

As depicted in Figure 1, the behavioural studies of EF in those with a diagnosis of DCD and 'at-risk' samples (hereafter, 'DCD') have employed tasks that rely on the processing of verbal, motor and visuospatial information, with only a relatively small proportion of these studies comparing performance across domains. This is important because it may be that those with DCD would perform at a typical level if, for example, a verbal task was employed, while an EF impairment might be reported for a task requiring a significant motor response. In the case of goal-directed reaching, for example, studies of *motor* planning in DCD have reported significant difficulties in planning a reaching movement to either end in a comfortable position [29, 38, 41] or to carry out different end actions, such as placing, throwing or lifting an object [36]. However, as cognitive demand increases, studies of 'cognitive' planning (such as planning a sequence of moves in a game in order to reach an end goal) have reported mixed results [19, 29, 41].

Tasks of motor inhibition have also presented different patterns of difficulties depending on the task used and the measure taken: a greater number of errors in inhibition have been reported in some cases [20, 33, 40, 42], whereas other studies reported a similar number of errors but slower and more variable response times in DCD compared to typically-developing peers [21, 23, 28, 30]. Some recent follow-up analysis in our research group suggested that the DCD group made more errors in a motor test of inhibition, but that typical error rates on a verbal inhibition task were related to significantly slower response times in DCD [43].

Thus, understanding both the errors and the task completion time has implications for detection and support of EF difficulties in everyday life: for example, making errors on an EF task in a classroom situation, such as remembering a list of instructions while completing a piece of work (a working memory task), might be more evident to a teacher than taking longer to change strategy when encountering a problem (a cognitive flexibility task). Moreover, if children with DCD can achieve similar performance to their peers if given longer to complete a task, then raising teachers' awareness of this could result in an improvement in classroom functioning and academic achievement.

As well as the motor coordination impairments which are central to the diagnosis, many individuals with DCD are also reported to have difficulties with visuospatial processing across a range of measures [18, 44]. Tasks that rely on visuospatial processing may therefore engender poor performance in individuals with DCD, aside from any problems with executive functioning *per se*, and so it is important to compare performance across task domains (see Figure 1). Research conducted by Alloway and colleagues [24-26, 31] gives some support to this suggestion, demonstrating significantly poorer visuospatial working memory compared to verbal working memory in children with DCD. Our recent study [42] also found significantly poorer visuospatial working memory in children with DCD compared to typically-developing controls, but no difference in verbal working memory. Furthermore, when comparing performance across verbal and nonverbal measures, our study highlighted difficulties for DCD in *nonverbal measures of EF only*. It may be that studies employing only tasks that require a visuospatial or motor element show greatly reduced EF abilities in DCD.

Another important issue to consider when interpreting the results of the EF studies identified above is the sampling procedures used. First, the vast majority of the studies investigating EFs in DCD have focused on children between the ages of 5-11 years. Given that the EF literature highlights the prolonged development of EFs over adolescence and into early adulthood [8, 14], restricting the age range to early childhood limits our understanding of the impact of EF on everyday life in DCD. Two studies of motor planning have recruited both children and adults with DCD [36, 38] and reported that although there was some improvement between the two ages, adults with DCD continued to show atypical movement planning. However, there are no equivalent data available on the more ‘cognitive’ planning tasks used with children, e.g., the Tower of London task [45]. On the basis of these data, and given that the demands of school and then employment increase after early childhood, it is vital that we understand the later development of EF in DCD and the potential age-related changes in the impact of EF difficulties on everyday life.

A second point relating to sampling is that DCD is a heterogeneous condition and can often overlap with other clinical diagnoses, or present subclinical symptoms of other disorders [46, 47]. These disorders, such as Attention Deficit-Hyperactivity Disorder (ADHD) and Autism Spectrum Disorder (ASD), often have different profiles of EF difficulties [48] and it may be that EF impairments in DCD in some of the studies are related to these overlapping symptoms or diagnoses, rather than motor difficulties that are central to the disorder. Some researchers have suggested that only a subgroup of individuals with DCD have executive function problems [49-51] and that these cases may be those with more co-occurring features of other developmental disorders. However, research suggests that even those children with DCD with relatively ‘pure’ motor difficulties (i.e., no other clinical diagnoses) are impaired across a range of EF tasks [21,42]. In our recent study, this was further supported by the fact that these difficulties were evident after ADHD-related symptoms were controlled in the analyses, and because children who we screened for motor difficulties, but had not been referred for clinical diagnosis, demonstrated a highly similar pattern of functioning across the EF tasks [42]. These points will be considered further later in the article, along with the future directions for EF research in DCD. We will now turn to some recent literature that has taken somewhat different approaches to the study of EF in DCD, first by conducting neuroimaging during EF tasks, and second, by investigating EF in ‘real-life’ contexts through questionnaires.

Neuroimaging of EF in DCD

As discussed earlier, while motor and EF abilities are often investigated separately, these skills are highly interconnected on a neural level, and these connections change over development [3, 4, 14]. A limited amount of research has investigated the neural correlates of EF performance in DCD, and these studies have focused on working memory and inhibition using visuospatial or motor tasks [23, 33-35]. All have found behavioural impairments in children with DCD, either in errors or reaction times, as well as atypical neural functioning underlying the tasks. Two studies have used event-related potentials (ERPs) to test the temporal dynamics of brain responses to visuospatial working memory (VSWM) tasks in DCD, using a delayed match-to-sample paradigm in which a stimulus was presented, followed by a delay, and participants were required to make a judgement as to the similarity of a second stimulus to the one presented previously [33, 35]. These studies reported significantly smaller amplitudes of two positive deflections, the P3 and the ‘positive slow wave’, in children with DCD compared to controls. The authors suggested that this reflected a reduced allocation of attentional resources to the stimulus and reduced processing during the retrieval stage, respectively [33]. To our knowledge, no studies of DCD have used functional Magnetic Resonance Imaging (fMRI) during a test of VSWM, but the smaller P3 amplitude cited above might implicate the corpus callosum and suggest atypical hemispheric lateralisation or transfer of information [33]. Another site of interest for fMRI would be the lateral cerebellum, recruited in typically-developing children but not adolescents and adults in an oculomotor VSWM task [52]. This area has been linked to poor performance in motor learning tasks [53] and might therefore be involved in both the motor and VSWM difficulties seen in DCD.

Two studies have investigated the neural underpinnings of motor inhibition in children with DCD, one using ERPs [34] and the other using fMRI [23]. In the ERP study [34] children were required to respond by pressing a pedal with the corresponding foot when a particular stimulus appeared on the left or right side of a screen. Before the stimulus appeared, the children received a cue which was either congruent or incongruent with the side of the screen on which the stimulus would be presented. The amplitude of the P3 component measured from posterior channels was reduced in DCD and there was also a longer latency compared with typically-developing controls, reflecting slower reaction times and poorer inhibitory control. The N2 component, measured at 150-200ms after the stimulus, did not differ between groups, but is not always found to be related to inhibition in children [54].

The fMRI study [23] detected some atypical activation in DCD during a Go-NoGo task. Here a prepotent response was built up to respond to a particular stimulus (in this case, when two sequentially-presented stimuli were the same: Go trials), and this response was then to be inhibited when a different stimulus was presented (when the stimulus was an ‘X’: NoGo trials). Behaviourally, children with DCD were able to inhibit responses on NoGo trials to the same level as their peers, but were slower and more likely to make errors on Go trials than controls. Inspection of the functional neuroimaging data underlying this performance provided some insight into this performance: children with DCD had greater activation of the anterior cingulate cortex, which is thought to reflect conflict monitoring [55, 56]. This pattern has been found in healthy adults when a conflict monitoring decision is difficult [57], and therefore suggests that children with DCD may have demonstrated different functional activation because the task was more challenging for them than controls. Activation was also greater in the left hemisphere than the right in the children with DCD, which was the opposite pattern to controls, suggesting that similar behavioural performance can be achieved through atypical neural functioning. Understanding the functional pathways subserving EF performance in the laboratory will be of great importance in future research, as will utilising structural MRI and diffusion tensor imaging to identify any differences in neural structures and their connecting pathways. However, it is also important to understand EF in everyday life, and it is to this research that we now turn.

EF in ‘real-life’ contexts

While standardised and experimental measures of EF have been conducted with children with DCD, the assessment of ‘everyday’ EF has relied on questionnaire-based measures completed by adults. These measures have assessed skills such as time management, organisation and planning, with individuals with DCD reporting difficulties in all of these areas, using a range of questionnaires [58-61]. Interestingly, when Kirby et al. [58] asked adults with DCD to report their strengths and weaknesses, around half of the individuals described skills such as planning and organisation as a weakness, but 20% described these skills as a strength. This prompts a question regarding the relationship between laboratory and ‘real-life’ measures of EF: would the individuals who describe EF as a strength outside of the laboratory perform better than those who report it as a weakness when completing standardised and experimental EF measures? To our knowledge, no research has investigated this question in DCD, but evidence from other clinical and non-clinical groups has suggested that there is a very weak relationship between performance-based and rating measures of EF in both children and adults [62]. The authors suggest that this may be due to the constrained conditions of the performance-based measures, in which the goals and structure of the task are provided by the experimenter, and the unconstrained conditions in everyday contexts, when individuals must discover and create their own task structures and set their own goals. However, this does not mean that the standardised and experimental measures of EF conducted with children with DCD are not useful or valid; these tasks enable us to assess how well they perform tasks in highly structured conditions [62], which could have implications for the way in which activities are managed or set out in the classroom for these children. Nevertheless, it is important to remember that ‘passing’ one of these tests may not be representative of individuals’ abilities to set their own goals and manage their behaviour in more complex, less constrained situations, and so it will be necessary to combine these different measures, along with more unconstrained performance-based tasks, in future research in order to produce a more rounded understanding of the EF profile in DCD. This and other future directions are considered further in the next section.

How can we build on the current knowledge?

Throughout this review of the literature into EF in DCD, it has become clear that definitional, measurement, diagnostic and contextual issues associated with the study of EF need to be considered fully before we can understand if difficulties in EF are evident across the disorder, or whether these difficulties are confined to a subgroup with a range of other impairments [49-51]. Furthermore, it will be important to understand whether there are different constellations of EF impairments in DCD (e.g., some individuals may have problems with executive-loaded working memory and planning, while others are more impaired in inhibition and switching), and if these varying patterns of impairment have a significant effect on overall functioning. It may be that a continuous model of EF impairments is preferable, with the *number* of affected EFs providing a more useful indicator of the severity of functional impairment than the *types* of EFs affected. Finally, understanding whether relatively good performance in any EF can compensate for difficulties in other areas will be of great value, allowing the identification of potential protective factors in EF development [13].

In order to address these and other points raised in this review when designing a study, it will be necessary in the first instance to consider the component skills required to complete the tasks, i.e., could impairments in these skills be the cause of poor performance in individuals with DCD, rather than difficulties with EF? As depicted in Figure 1, only some studies compare performance on tasks measuring the same EF across different domains, and we suggest that this practice should be adopted more widely in EF research. The breadth of EFs tested is also important; the pattern in Figure 1 demonstrates that some EFs, such as working memory, are much more widely studied than cognitive flexibility and fluency. This should be considered when interpreting study findings to date, since the term EF is often used broadly but in fact is formed of many, rather different, components. Thus precise use of terminology is critical.

Furthermore, the focus of previous research has been on ‘cool’ EFs, involving abstract problems without a clear emotional component, whereas some aspects of EF are more motivation-driven (or ‘hot’), as in tasks involving reward or punishment [63]. Studies using hot EF tasks in DCD have required participants to make decisions based on their weighting of positive and negative potential outcomes [39], or to inhibit responses to different emotionally-laden cues, specifically happy or fearful face stimuli [40]. Children with DCD were more likely to choose options that had a high immediate reward but negative long-term outcomes than were controls [39], and were more distracted by stimuli with positive emotional content (i.e., happy faces) than typically-developing children, making more inhibition errors in this condition [40]. This is important because hot EFs are related to self-control and emotional regulation, and therefore have implications for the individual's psychosocial functioning outside of the laboratory [39]. For example, a disruption of hot EFs could affect decision-making with emotional components and, potentially, the ability to behave appropriately in social situations and develop relationships with peers [40]. Hot EFs may develop later than cool EFs [63], and so an understanding of a number of measures of EF at different stages of development will be important for future research, since this may point to the putative underlying causes of any EF difficulties in those with DCD (e.g., immature *vs.* different development).

On a related note, a second methodological point to consider when designing future studies will be the age of participants and the approaches used to identify age-related changes in EFs. Given the extended development of EF over childhood and into adulthood [8,14], it is important to consider age during study design rather than focusing only on DCD *vs.* control comparisons, especially when the groups cover a wide age range (e.g., 7-14 years). Ideally, this would be addressed through the use of longitudinal designs, assessing children over development on key EF measures, as in the study by Michel and colleagues [30] with children screened for motor difficulties in preschool. Longitudinal studies on a shorter time frame, involving an intervention between two visits, have also provided promising results: studies implementing an exercise intervention with children with DCD have reported to both improve behavioural performance and change neural responses in tasks of visuospatial working memory and inhibition [34,35].,The use of cross-sectional developmental trajectories in DCD research will also be of great benefit, ensuring that age is a factor taken into account at the outset of the study.

It will also be important to assess EF in both children and adults using similar (age-appropriate) tasks, as in the studies by Wilmut and colleagues [36, 38]. Currently, the recruitment of adults with DCD is challenging due to two main factors: first, the availability of standardised tests of motor ability for this age range is limited compared to those that are regularly used with children and younger adults [64]; second, practitioners in the past had relatively limited knowledge of DCD or motor disorders, which led to under-diagnosis of DCD in childhood and thus an under-representation of DCD in those who are now adults, at least in the UK. However, the difficulties associated with DCD persist into adulthood [58-61, 65, 66], and therefore following the development of EF into later years will be important in future research. Understanding the development of EFs in DCD will have important implications for treatment, helping us to identify key times for intervention and to understand the impact of earlier difficulties on later EF and psychosocial functioning, and so studies in which age is a central factor in the design are vital for future research in this area.

The final methodological point that stands out from this review of EF in DCD, and in the EF literature as a whole, is the correspondence between laboratory tasks and 'real-life' measures of EF. As a first step, it is important that we use both rating scales and performance measures of EF across samples of children and adults in order to assess the relationship between behaviour in constrained and unconstrained situations in DCD. This will not only be important for identifying the EF areas that may benefit from the most support in the classroom or the workplace, but will also demonstrate how useful a particular task might be in training EF skills for use in these contexts. However, another important step will be to conduct some more ecologically-valid performance-based tasks, providing individuals with DCD with more unconstrained situations in which they have to set their own goals and respond flexibly and creatively, as they would in their everyday lives. Although this type of approach may raise more challenges than the standard behavioural paradigms, it will help us to reconcile findings that may differ across performance-based and rating scale measures of

EF, and will significantly improve our understanding of EF in DCD in more realistic situations than are often seen in laboratory tasks.

Conclusions

The importance of motor skills for the development of social and cognitive abilities is becoming increasingly recognised, and this is occurring alongside improvements in the identification and understanding of DCD. Extensive overlap between neural circuits related to motor and cognitive control [3, 4], along with self-reported weaknesses in planning and organisation in adults with DCD [58], provide a clear rationale for investigating EF in this neurodevelopmental disorder. While research in this area has identified wide-ranging difficulties in EF in DCD, future studies should begin to delve deeper into the underlying causes of these performance differences, carefully considering methodological factors such as the domains and samples tested, the informational constraints of the task, as well as the neural underpinnings of the behaviour observed. Once these issues are addressed, it might be possible to identify particular subgroups of individuals with DCD who have EF difficulties and to understand any risk or protective factors involved in their development. Most importantly, future research should always keep in mind the EF performance of individuals with DCD in ‘real-life’ contexts, allowing appropriate support to be provided for these individuals in the classroom or the workplace.

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Fig. 1. Behavioural measures of executive function (EF) in Developmental Coordination Disorder and individuals with motor difficulties, according to the EF component investigated and the domain assessed by the task. Individual studies are represented by the number of the reference in the reference list, and may appear more than once if they study more than one EF component or compare performance across different domains. *Note* studies that appear between two EF components used a combined measure of working memory and inhibition.