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Toward a Hybrid Model of Developmental Coordination Disorder

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Abstract
Purpose This paper discusses the merits of a hybrid model of developmental coordination disorder (DCD), one that integrates cognitive neuroscience and ecological systems approaches. More specifically, we present an integrative summary of recent empirical work on DCD that enlist behavioural and neuroimaging methods and propose a theoretical interpretation through the lens of a hybrid model.

Findings The review identifies two current hypotheses of DCD that find consistent support: the internal modelling deficit (IMD) and mirror neuron system (MNS) accounts. However, motor performance and brain activation patterns are not expressed in a uniform way under these hypotheses—motor deficits are manifested variously as a function of specific task and environmental constraints and condition severity. Moreover, we see evidence of compensatory processes and strategies.

Summary Taken together, results support the broad hypothesis that children with DCD show distinct motor control deficits and differences in neural structure and function compared with typically developing children. However, researchers still have difficulty ascribing causation. The proposed hybrid (multi-component) model of DCD can help researchers generate novel hypotheses about specific mechanisms, explaining the constellation of deficits that is shown experimentally and observed clinically. This model can be applied to cognate disorders of childhood that affect movement and design of intervention.

Keywords Developmental coordination disorder · Internal modelling deficit · Mirror neuron system · Disorders of motor · Review

Introduction

Overview

Problems of motor skill learning in children (or developmental coordination disorder—DCD) is a commonly occurring issue that can have quite profound effects on academic achievement, participation and well-being [1, 2]. While motor difficulties are observed by parents and teachers from an early age, they are not explained by known medical conditions such as brain injury, muscular dystrophy and so on [3]. However, the latest DSM-V now categorises DCD as a neurodevelopmental disorder which opens up further debate on its causal roots. While DCD continues to attract significant research, our explanatory models of DCD have not developed in synchrony. We see several hypotheses about the underlying basis of the motor issues, but it is fair to say that a unifying account of DCD has been elusive. In Kuhnian terms, it cannot be said that new methods from cognitive neuroscience have seen a full paradigm shift [4]; rather, a tension still exists between neuroscience and ecological approaches (see Wade and Kazeck 2017 [5*]; Wilson et al. 2017). This paper is designed to
address this tension by providing a brief summary of the recent experimental literature on DCD, identifying some unifying themes across various areas of research. Based on this review (and recent work on related developmental motor disorders), a tentative multi-component model of performance in DCD is proposed to help direct hypothesis testing over the coming years. This model endeavours to blend cognitive neuroscience and ecological approaches to understanding motor behaviour. The hope is to provide a principled, conceptual foundation for experimental researchers in the field of DCD, one that supports novel testable hypotheses about mechanisms of DCD and related neurodevelopmental disorders.

Introducing a Hybrid Model of Motor Skill Development

For the first time, the field of DCD research has a body of behavioural and brain-based experimental evidence that can help theorists build linkages across levels of explanation—brain, cognition and motor behaviour [6]. From this body of work, we propose a (hybrid) multi-component model of performance which is based on ecological systems theory and advances in cognitive neuroscience (see Fig. 1). The three core components of the model derive from systems theory—here, motor performance emerges from the interaction of individual, task and environmental constraints [7, 8]. At the individual level, there is an interactive set of constraints that bias our response capabilities at any given point in development [9, 10]. At a basic biological level, genetic factors set in play maturational processes that determine physical structures of the system including brain networks, neuromuscular system and biomechanical linkages [11, 12]. At the same time, however, environmental factors are needed to trigger certain genes—phenotypic expressions are also the product of “nature via nurture”. These basic structures support a range of internal processes: cognition (e.g. executive functions), motor control processes (e.g. internal modelling) and motor learning1 (e.g. procedural learning), all of which can be modified over time by physical activity. In turn, these structures and processes set constraints on the (latent) movement abilities of the individual.2 Task constraints refer to factors external to the body and specific to the task at hand—the goals, rules and equipment associated with a particular activity [13]. Environmental constraints refer to those external factors that shape the (physical) performance environment, e.g. ambient temperature, presence of distractors such as other people, parents/significant others who

provide opportunities for participation and even the broader social context of the activity. Motor behaviour is thus determined by the dynamics of the interaction between the individual, a given task and environmental workspace, i.e. particular movement patterns and degrees of skill performance are constrained by this interaction.3

Recent Experimental Research on DCD and Its Implications for a Hybrid Approach

The increasing volume and high quality of experimental work on DCD over recent years [14] enable us to compare results across studies with more confidence and to consider how findings might be integrated under a more unified framework (Fig. 2). This framework involves thinking about action constraints more broadly (e.g. as task conditions) and about experimental paradigms as a way of testing exactly how different components of the system interact to express poorly coordinated movement. A number of important conclusions can be made about the body of evidence, mainly related to the themes of predictive motor control, action representation, perceptual-motor coupling, task complexity, co-occurring cognitive issues, compensation and persistence into adulthood.

In a recent systematic review, we grouped studies according to the two dominant approaches to DCD research: (1) cognitive neuroscience or (2) ecological/dynamical systems [14]. The review spanned 6 years (2011 to 2016) and included a final sample of 106 studies. The review showed continued progress of work in the field of DCD. Most notably, there was further development in work on motor control (e.g. internal modelling and mirror neuron system—MNS), constraints testing from an ecological perspective, postural control under different constraints, executive function and neuroimaging approaches.

The review showed that research bearing on the internal modelling deficit (IMD) hypothesis and function of the MNS continued to grow [15–17] but certain caveats were necessary. The IMD hypothesis holds that the movement difficulties in DCD are associated with deficits in the ability to enlist predictive control when planning and executing movements. This is inferred by poor motor simulation (e.g. motor imagery), slower adjustments to target perturbations when reaching, less anticipatory postural adjustments when initiating movement and so on. Support for this hypothesis was reasonably strong across effector systems [18], but deficits were more pronounced on tasks of higher complexity or demanding more endpoint precision [19]. For example, on simple endpoint

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1 Motor learning refers to processes associated with practice or experience that lead to a relatively permanent change in movement skill or capacity.

2 Motor abilities are relatively enduring motor traits or capacities that have biological origins and that bias skill learning; biological-environmental interactions determine these abilities. Examples include muscle strength, movement speed and multi-limb coordination.

3 Motor skill refers to a task (with a specific goal) that involves physical movement (e.g. free-throw shooting in basketball, walking on stepping stones, tying shoelaces). Skills are normally refined/learned with practice.
planning tasks, deficits are less readily observed [20]. Related to this, issues with gesture imitation and motor imagery are likely to reflect problems associated with the egocentric representation of space, linked to the function of the MNS. These motor control and body representation issues at the individual level can be embedded within a multi-component
model of DCD (Fig. 2). While we appreciate the importance of structured and unstructured physical activity in modifying motor and cognitive processes over time, the causal relationships that connect different components of the model (and that cut across brain, cognition and behaviour) require further clarification.

In general, performance deficits tend to map to functional and structural issues in a distributed neural network that supports motor control and learning—namely, the MNS, visual-motor mapping and predictive control (e.g. fronto-parietal and parieto-cerebellar structures) and corticospinal tract. Structural diffusion MRI studies show alterations of white matter connectivity, especially in sensorimotor tracts including corticospinal tract, posterior thalamic radiation and parietal sub-region of the corpus callosum [21–23]. Functional MRI studies show reduced activation in DCD across cerebellar, parietal and prefrontal cortices [24–27], areas that support predictive motor control (see also Kashiwagi et al. 2009 [28]). Other data show reduced activity in the MNS during action observation including precentral gyrus, inferior frontal gyrus, precuneus and posterior cingulate [29]. A delay in the maturation of these network structures is possible; however, current data is not conclusive on the neurodevelopmental origins of causation. Moreover, alterations in brain structure and function are also observed in regions that lie outside the IMD/MNS networks, e.g. reduced brain activation in L. superior frontal gyrus. Frontal executive systems support the integration of working memory with attentional resource allocation (called branching) which is important in dual-task performance. Taken together, a (whole-brain) network approach may prove to be more beneficial in modelling developmental differences between DCD and typically developing children [30, 31]. Longitudinal work is needed to clarify the causal connections.

Executive function deficits in DCD are a common finding across measures (experimental, questionnaire and real-world behaviour), persist into early adulthood and are strongly linked to impaired planning and disorganisation in daily life [32, 33]. Executive functions include the short-term maintenance of relevant information, inhibition of irrelevant information/responses and the organisation and scheduling of competing actions. We have shown, in particular, that the combination of cognitive and motor control issues has implications for the control of actions that have a significant cognitive load (e.g. the requirement to inhibit reaching responses to compelling visual cues) [34•]. These individual-level deficits may also extend the rehearsal time required to automate skills, particularly for complex, multi-step tasks. Finally, the relationship between EF and daily organisation is evident not only in DCD but also in other disorders of learning [35].

In general, recent evidence shows that motor control/learning deficits associated with DCD are not expressed uniformly across different task conditions. Performance deficits are more apparent for the following: dual tasks; tasks that demand greater endpoint precision, response complexity, or advanced planning; movements that are subject to an external perturbation and that require in-flight adjustments of the system. For example, manual tracking of a visual target is impaired most when the degree of visual feedback is reduced and when target speed increases [36].

Taken together, these data suggest that atypical individual-level (neurodevelopmental) constraints may underlie DCD including genetic [37•], but their influence varies according to the particular task-environmental conditions that are presented (see Fig. 2). One hypothesis is that delay in the development of sensorimotor and other brain networks may underpin poor internal modeling [28] and observational learning [29] in DCD. Under simple task conditions, slower feedback-based control may be enough for skilled performance. However, when task demands stress the system in a way that requires a fast adaptation, adjustment or advanced planning, then slower feedback control does not suffice—predictive control is needed. Hence, the full complement of task-environment and individual constraints needs to be considered as an interactive system when explaining how coordinated action unfolds (or fails to unfold). The IMD hypothesis, for example, can only be articulated within a hybrid approach. What we still fail to understand, however, are the specific mechanisms by which the behaviour of the individual system is constrained by specific task parameters and how this changes with age and experience. What we do know is that children with DCD can learn fundamental motor skills under the right training conditions, while appreciating that certain skills remain difficult. The important point is that all components of the model must be factored in when building our theoretical models and when designing intervention.

A Unifying, Hybrid Model in Action—Explaining and Predicting Multi-Component Interactions in Related Conditions

Related developmental motor disorders (like cerebral palsy) can also be understood from the perspective of a hybrid model. Collectively, there is sufficient commonality in the science of motor dysfunction in children to support a common theoretical perspective, one that will usefully guide hypothesis testing into the future.

Insights from Other Developmental Motor Disorders (CP)

The issues observed with predictive control share similarities with another developmental motor disorder, cerebral palsy (CP). Children with CP have persistent compromised motor abilities as a consequence of early brain damage (a core individual-level constraint). Recent evidence suggests that
the aetiologies of children with CP and DCD are not as different as were first thought [38], and that they may share a similar underlying neurocognitive deficit that is associated with motor control and skill learning—a compromised ability for motor prediction [39–41]. In recent years, a compelling body of behavioural and neurophysiological evidence has provided important new insights into the motor control system of children with CP [42], but at the same time lays bare some limitations of an IMD hypothesis and the need for a hybrid approach. Behavioural studies where children with CP need to pick up objects for the purpose of manipulating or placing them showed that they do not anticipate the end state of the task. Picking up an object using a comfortable grip did not always enable them to end the task efficiently, indicating a deficit in anticipatory control [43]. Likewise, studies using motor imagery showed that these children were compromised in their abilities to mentally rotate parts of their own body in anticipation of the upcoming task [44].

While these findings hint at a problem with the internal model, this explanation has its limitations. What was also apparent in these studies was the large variation in performance between individual children as a function of task complexity. Similar to the findings in children with DCD, task complexity was shown to affect motor outcome to a large degree, a facet that cannot be explained satisfactorily by the IMD hypothesis only. A major challenge in understanding the nature and basis of CP and DCD is to isolate those control systems that might be compromised in both groups and that may explain common issues in motor skill learning and account for the large variation in performance as a function of task and individual constraints. As well, children with CP have reduced movement experiences due to individual constraints and are often afforded fewer environmental resources/opportunities to engage in play [Imms et al. 2017]. The hybrid model that we propose here does take into account these findings and may help explain the true complexity of motor disabilities in children with DCD and CP. Moreover, the model may provide an overarching framework for future studies that seek to explore commonalities in motor behaviour in CP and DCD; this informs a view that these are not distinct disorders but lie on a continuum of motor impairment [45].

Brain, Cognition and Behaviour: Cross-Level Interactions in DCD and Related Disorders

As indicated above, over the past 5 years, there has been a growing desire in the fields of cognitive neuroscience to understand how DCD influences brain connectivity (for a recent review, see Biotteau et al. 2016) and how brain-based metrics might relate to behavioural manifestations of DCD. Notably, DCD-related reductions in white matter integrity have recently been associated with abnormalities in motor performance. For example, reduction in fractional anisotropy (FA) of the left retrolenticular limb of the internal capsule was associated with poor visuomotor tracing performance (Debrabant et al. 2016). This pattern of association between motor deficits and impairment of sensorimotor pathways corresponds well with other paediatric clinical conditions, such as traumatic brain injury [46, 47] and cerebral palsy [48, 49, 50].

However, the drawback of these regional analyses of structural connectivity is that it does not directly allow the identification of the functional systems or axonal networks that are affected in DCD. Hence, it is impossible to infer how brain dynamics are affected in relation to structural changes. Aware of these challenging limitations, Hagmann and Sporns [51, 52] proposed a conceptual framework where the entire brain connectivity is modelled as a network: the connectome. Connectome analyses rapidly found applications in the clinical neurosciences and have provided biomarkers of specific brain functions or symptoms (for a review, see Griffa et al. 2013) [53]. For example, in our previous study (Debrabant et al. 2016), we found that children with DCD showed reduced network global efficiency (suggesting weaker structural network integration) which correlated significantly with motor deficits. Other paediatric populations, such as brain-injured children [54], ASD children [55] and preterm children [56], exhibit very similar patterns of global network alterations. This integration loss indicates that these paediatric clinical populations share a relative diffuse involvement of the white matter tracts affecting efficacy of long association tracts. These commonalities in altered structural connectivity across different paediatric populations can be embedded within our hybrid model. Future work should examine training-induced changes in brain connectivity in DCD using longitudinal designs, conducted over extended periods of time.

Clinical Implications

The model has some important implications for clinical practice/intervention. The multiple constraint approach encourages scientist-practitioners to think carefully about the co-occurrence of motor and cognitive issues in DCD at the individual level. Indeed, clinicians are encouraged to err on the side of caution and assess broadly across motor and cognitive functions. Associated effects on task organisation and self-regulation also need to be factored in, not just in children but also in adolescents and young adults.

A constraints-led approach to training [57] is consistent with the experimental data on DCD that show that individual-level constraints (like internal modelling and/or EF deficits) can impact performance (or not) in variable ways, especially as a function of task difficulty/type. And work from an ecological perspective that shows difficulties in perception-coupling is consistent with task specificity in intervention. The upshot is that the skill learning context should be scaled...
carefully by manipulating task, informational and environmental constraints, tailored to the needs and capacities of the individual child. Task constraints that make a motor skill difficult are also the constraints that we can modify (or make easier) during intervention: e.g. training single tasks to a certain level of skill before combining them with another task; starting with low accuracy requirements in a task-specific context and then increasing this gradually, or speed requirements, or both together. Some other examples include simplifying the cognitive load by adopting less stringent task rules or using observational approaches to learning, modifying affordances by using equipment that is better scaled to the needs of the child with DCD, or using task instructions that encourage the child to attend to (or predict) the effects of their movement (see also neuromotor task training—NTT) [58]. Environmental constraints that impact performance can also be modified to facilitate learning; for instance, in learning to cycle, you may choose a location out of the wind or without people watching. In terms of the hybrid model, manipulations of these various types may help the child circumvent any apparent individual-level constraints that could impede their ability to plan or implement a movement. Over time, with practice under variable conditions, and perhaps using augmented feedback [59, 60], the child may also improve the fidelity of their motor control system [61].

Conclusion and Future Directions

Recent experimental work on DCD has continued to provide evidence of fundamental issues in motor control and learning in these children (e.g. internal modelling and MNS). At the same time, there has been growth in experimental work on constraints testing from an ecological perspective, postural control under different constraints, executive function and neuroimaging approaches. We have a clearer picture of the constellation of motor and cognitive deficits in DCD, but also critical examples of where specific neurocognitive hypotheses break down because they do not apply across all (even related) paradigms or task conditions. We propose here that a hybrid model of motor development and dysfunction can help reconcile these various findings, accounting for individual differences in performance, the clustering of deficits, variability as a function of task-environmental condition and change with maturation.

A few pointers for future research are in order. No experimental study has yet tracked the dynamic interaction between parametric changes in task condition (viz constraints), the dynamics of motor coordination and real-time functional brain activity, nor tracked over time the impact of training on these factors. Conducted longitudinally, this type of study will advance knowledge of causal processes in DCD; embedded within a hybrid (multi-component) model, findings of this type will have important implications for intervention [62, 63].

Great advances have been made in the study of motor control issues that are associated with DCD and their parallels with CP. Given these converging lines of evidence, there is a compelling case to merge approaches to the study of developmental motor disorders (aka hybrid model), the endgame being to fully understand the interplay between individual, task and environmental constraints in their expression. Put another way, a hybrid model is an important next step in explaining the vast variability in the manifestation of motor control issues in CP and DCD and may further provide insight in the commonalities between DCD and CP, even suggesting that these disorders may not be distinct, but rather expressed on a continuum. There is, indeed, a clinical viewpoint that supports this approach. Longitudinal studies are needed to map developmental trajectories in these groups and to understand whether parallel growth exists on key measures.

Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no competing interests.

Human and Animal Rights and Informed Consent This article does not contain any studies with human or animal subjects performed by any of the authors.

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