School of Psychology and Speech Pathology Curtin Health Innovation Research Institute

# Understanding motor coordination and its cognitive, academic, and psychosocial correlates in an adolescent normative sample

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This thesis is presented for the Degree of Doctor of Philosophy of Curtin University

July 2012

# DECLARATION

To the best of my knowledge and belief this thesis contains no material previously published by any other person except where due acknowledgment has been made. This thesis contains no material which has been accepted for the award of any other degree or diploma in any university.

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# ABSTRACT

Over the past three decades, increasing attention has been paid to the importance of motor competence in relation to other areas of a child's development, including cognitive functioning, academic achievement, and emotional outcomes. For example, a number of studies now show that children with motor difficulties are at increased risk for internalising problems such as anxiety, and may also experience deficits in complex cognitive processes, namely, executive functions. Furthermore, evidence suggests that without intervention, these motor difficulties and associated problems may continue, yet research in older age-groups is limited. The primary aim of this thesis was to explore the relationships between motor coordination and the cognitive, academic, and psychosocial domains in an adolescent sample aged 12 to16 years. This is imperative given the changes that occur, particularly in social and cognitive domains, during this developmental period. Furthermore, given that most of the existing studies have identified these relationships in groups of children with motor problems, this points to the need to investigate whether the close links between these areas exist along a continuum, extending to individuals without problems in the motor domain. The series of papers presented in this thesis therefore examine the association between motor coordination and these areas from a normative perspective.

The first two papers of this thesis relate to the link between motor ability and emotional outcomes. Although the number of studies on this topic has increased, longitudinal evidence is limited. Therefore, the first study is based on a longitudinal project examining the predictive relationship between early motor development, from infancy to early childhood, and later emotional outcomes at school-age (i.e., 6 to 12 years old). The paper aims to present a preliminary analysis of the association between motor development and emotional functioning. Using parent-rated questionnaires, it was found that the stability of early gross motor development predicted later anxiety and depressive symptomatology. Importantly, the study provides an indication of the possible causal relationship between these areas which is important when considering the focus of the second paper in a normative sample of adolescents. Although it is plausible that the relationship between motor problems and internalising difficulties may be explained by organic factors, it has been suggested that the negative environmental experiences associated with motor problems may in turn, lead to negative self-appraisals and subsequently, increased risk for internalising problems. Thus, the second paper examined whether selfperceived competencies in social, academic, and physical areas play an important mediating role in the relationship between motor coordination and emotional functioning in adolescents. A standardised motor performance test (namely, Movement Assessment Battery for Children-2) provided indicators of motor coordination; manual dexterity, aiming and catching, and balance. Questionnaires on self-perceptions, anxiety, and depressive symptomatology were completed by adolescents. Structural equation modeling revealed that motor coordination had an indirect link with emotional functioning, through the mediating influence of selfperceptions. It also appeared that aiming and catching, and balance skills (and not manual dexterity) were important for the emotional outcomes of these adolescents.

The focus of the following two papers aimed to further understand the relationship between these motor components and cognitive areas, including executive functions and academic achievement. The third paper examined possible specific relationships between the different motor components and various executive functions, namely working memory, inhibition, and set-shifting. Attention deficit hyperactivity (ADHD) symptomatology were also taken into account which is important given the close association between ADHD and both executive function and motor problems. Specific relationships between motor coordination components and the various executive functions were found which may suggest possible shared neural processes, including cerebellar mechanisms. Importantly, this study reveals relationships that may have been masked in studies that involved an overall measure of motor performance or groups of children with overall motor impairment.

Given the significant links found between motor coordination and executive functions, it is plausible that certain executive functions may play an important role in understanding the relationship between motor problems and academic underachievement. Recently, working memory difficulties in children with motor problems were found to be important when understanding the academic underachievement often displayed by these children. In the fourth paper, structural equation modeling was used to examine whether the relationship between motor coordination and academic achievement (word reading, spelling, and numerical operations) is mediated by working memory in adolescents, whilst controlling for covariates such as ADHD symptomatology, verbal ability, and socio-economic status. It was found that motor coordination, specifically aiming and catching skills, has an indirect impact on these learning outcomes via working memory.

In the final paper, the importance of identifying those adolescents at risk of motor problems is highlighted, particularly given the associated difficulties in the cognitive and psychosocial domains. Consequently, the fifth paper evaluates the revised Developmental Coordination Disorder Questionnaire (DCDQ), a parent-rated screening tool designed to assess motor difficulties, using the MABC-2 as a criterion standard. The DCDQ was found to have high internal consistency and demonstrated a relationship with the MABC-2. However, although the DCDQ appeared to meet the recommended standard for sensitivity, this was not met for specificity. Preliminary results for the psychometric properties of the revised DCDQ were promising suggesting that it may suitable for initial screening particularly in large samples, however, further assessment using a standardised motor performance test is warranted for those identified at risk of motor problems.

The ultimate aim of this thesis, presented over five papers, was to increase awareness and recognition of the possible associated problems of motor coordination difficulties. The results across the five studies highlight the importance of motor ability in relation to cognitive and psychosocial areas in adolescents, with important implications for assessment and intervention. Furthermore, the papers provide information on the theoretical understanding of the links between these areas with some insight into possible underlying processes explaining the relationships.

#### ACKNOWLEDGEMENTS

Firstly, I wish to express my sincere gratitude to my supervisor, Professor Jan Piek. I could not have asked for a more supportive, encouraging, and inspiring supervisor and mentor. Thank you for your invaluable guidance and for the amazing research opportunities that you have provided over the years. I am also very thankful for introducing me to this exciting area of research which I look forward to pursuing in my future research and clinical practice.

To my associate supervisors, Dr Robert Kane and Professor Jaap Oosterlaan, thank you for your support, advice and assistance, particularly throughout the publication process. To my associate supervisor, Dr Melissa Davis, thank you for your support and encouragement throughout my PhD, and for reading my final draft. Thank you to Dr Andrea Loftus for also providing me with valuable feedback on my final draft.

Especial thanks to the amazing families who participated in the research, making it all possible. Your time, support, and contribution are greatly appreciated.

Thank you to my very special friends, who have been my personal cheer squad throughout my studies. Especial thanks to Maryanne and Danilo for brightening up my days. Your friendships mean so much to me and I am truly grateful.

I am also very grateful for the friends I have made during this process. To Carly, we have shared every moment of this roller coaster ride together. Thank you for always being there for me. Greer, Felicity, Max, and Sue, thank you for your support and for the friendships that I know will continue into the future.

To my amazing parents, who have fed me, sheltered me, kept me company when I studied late into the night and put up with me during my most stressful times. I could not have done this without you. Your endless love, support, advice, and encouragement have brought me to where I am today and I am forever grateful. Carlo, thank you for being the best brother to your sometimes crazy sister. Your love, support, company, and the laughs have helped me so much throughout my studies. Thank you also for your amazing assistance with proof reading.

# DEDICATION

This is dedicated to my parents and my brother, for their love and support. I also dedicate this to my Nonna for the countless times she has expressed her immense love and belief in me.

# LIST OF PUBLICATIONS INCLUDED AS PART OF THESIS

- Piek, J. P., Barrett, N. C., Smith, L. M., Rigoli, D., & Gasson, N. (2010). Do motor skills in infancy and early childhood predict anxious and depressive symptomatology at school age? *Human Movement Science*, 29(5), 777-786.
- Rigoli, D., Piek, J. P., & Kane, R. (2012). Motor coordination and psychosocial correlates in a normative adolescent sample. *Pediatrics*, 129(4), e892-e900.
- Rigoli, D., Piek, J. P., Kane, R., & Oosterlaan, J. (2012). An examination of the relationship between motor coordination and executive functions in adolescents. *Developmental Medicine and Child Neurology*, 54(11), 1025-1031.
- Rigoli, D., Piek, J. P., Kane, R., & Oosterlaan, J. (2012). Motor coordination, working memory, and academic achievement in a normative adolescent sample: Testing a mediation model. *Archives of Clinical Neuropsychology*. Advance online publication.
- Pannekoek, L., Rigoli, D., Piek, J. P., Barrett, N. C., & Schoemaker, M. (2012). The revised DCDQ: Is it a suitable screening measure for motor difficulties in adolescents? *Adapted Physical Activity Quarterly*, 29(1), 81-97.

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# STATEMENT OF AUTHOR CONTRIBUTION

The nature and extent of the intellectual input by the candidate and co-authors has been validated by all authors, and can be found in Appendix E.

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Daniela Rigoli (Candidate) Jan Patricia Piek (Supervisor)

# LIST OF ADDITIONAL PUBLICATIONS

- Pearsall-Jones, J. G., Piek, J. P., Martin, N. C., Rigoli, D., Levy, F., & Hay, D. A. (2008). A monozygotic twin design to investigate etiological factors for DCD and ADHD. *Journal of Pediatric Neurology*, 6(3), 209-219.
- Pearsall-Jones, J. G., Piek, J. P., Rigoli, D., Martin, N. C., & Levy, F. (2009). An investigation into etiological pathways of DCD and ADHD using a monozygotic twin design. *Twin Research and Human Genetics*, 12(4), 381– 391.
- Pearsall-Jones, J. G., Piek, J. P., Rigoli, D., Martin, N. C., & Levy, F. (2011) Motor disorder and anxious and depressive symptomatology: A monozygotic cotwin control approach. *Research in Developmental Disabilities*, 32(4), 1245– 1252.
- Piek, J. P., Rigoli, D., Pearsall-Jones, J. G., Martin, N. C., Hay, D. A., & Bennett, K. S. (2007). Depressive symptomatology in child and adolescent twins with attention-deficit hyperactivity disorder and/or developmental coordination disorder. *Twin Research and Human Genetics*, 10(4), 587–596.
- Piek, J. P., & Rigoli, D. Psychosocial and behavioural problems in children with DCD. In Cairney, J. (Ed.), *Developmental coordination disorder in children: Consequences, concurrent problems and associated co-morbidities.* University of Toronto Press. (in press).

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#### **INTRODUCTION**

Koziol, Budding and Chidekel (2011) highlighted the importance of motor competence when it was argued that the fundamental purpose of all humans is to survive through environmental interactions. From infancy to adulthood, this requires ongoing learning of the effective and efficient movement skills that are integral to face the challenges of our dynamic environment (Gallahue & Ozmun, 2006; Koziol et al., 2011). During childhood in particular, movement experiences (e.g., through play) are crucial as they provide a child with important learning opportunities, including opportunities to learn about themselves and the world around them (Gallahue & Ozmun, 2006). These experiences, in turn, promote the development of skilful movement that is required for a large proportion of a child's daily activities, such as eating, drawing, and playing games, thus, demonstrating the significant role that movement competence may have in other areas of a child's development.

There is accumulating evidence from child and adolescent research of the important links between motor development and the cognitive and emotional domains. For example, Iverson (2010) argued that although the relationship between motor and language development is neither simple nor directional, early motor acquisition is a key contributor to the process of language acquisition. Also, longitudinal research has shown that these early motor milestones are significant predictors of cognitive functioning in primary school-aged children (Piek, Dawson, Smith, & Gasson, 2008). In terms of emotional development in this domain is clear from early childhood such as when a young child experiences the satisfaction of mastering the skill of walking. At school age, the impact of motor competence on academic and social areas becomes more evident as motor demands increase in the classroom (e.g., writing) and on the playground. Furthermore, it is at this age that children may begin to stand out from their peers if they are unable to catch or throw a ball.

In adolescence, the importance of motor competence and its relationship with physical activity and sports is emphasised given that adolescence is a significant period for socialisation and affiliation (Gallahue & Ozmun, 2006). In addition, there is

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longitudinal evidence showing that movement difficulty in earlier childhood is related to increased risk for emotional problems (e.g., anxiety) in the adolescent years (Sigurdsson, van Os, & Fombonne, 2002). Therefore, findings have shown that movement competence is integral for promoting the positive development of cognitive, social, and emotional functioning in childhood and adolescence.

However, traditionally, research in the area of motor development was given much less attention than cognitive and affective domains. Although the study of motor development dates back to the early part of the twentieth century, it was not until the 1970's that inquiry into this area began to escalate (Gallahue & Ozmun, 2006). Since then, studies detailing the underlying processes of movement and its numerous and varied implications have proliferated. It is now recognised that human development involves a complex interaction between motor, cognitive and affective development, and that these areas cannot be separated.

Consequently, over the past 30 years or so, increasing attention has been paid to those children who do not meet the motor demands of their environment but who have otherwise had appropriate opportunities for skill acquisition (i.e., children with Developmental Coordination Disorder, DCD; American Psychiatric Association, 2000). Since the early 1900's, these children have been discussed under a myriad of labels including minimal brain dysfunction, developmental dyspraxia, and clumsy child syndrome (Henderson & Barnett, 1998; Missiuna & Polatajko, 1995). It was not until 1987 that the term Developmental Coordination Disorder was introduced in the American Psychiatric Association's Diagnostic and Statistical Manual, Revised Third Edition (DSM-III-R; American Psychiatric Association, 1987). Conversely, the World Health Organisation's classification system, the International Classification of Diseases and Related Health Problems-10, (ICD-10; World Health Organisation, 1992) uses the term Specific Developmental Disorder of Motor Function to refer to children with motor difficulties. Although the definitions differ only slightly, DCD is the most commonly accepted term and at 1994 and 2005 consensus meetings, DCD was endorsed by a group representing the international research community as the term that should be used in research and practice when referring to these children (Polatajko, Fox, & Missiuna,

1995; Sugden, Chambers, & Utley, 2006).

Although motor problems in children were initially thought to be outgrown, it is now recognised that motor coordination problems continue into adolescence and beyond (Cantell, Ahonen, & Smyth, 1994; Losse et al., 1991). Increased research in the area has also shown that while there is often one feature of these children's difficulties that stands out (i.e., their motor skill difficulties), it is rarely an isolated problem (Kaplan, Wilson, Dewey, & Crawford, 1998). Studies have shown that children with motor problems may experience various negative consequences such as cognitive impairments (Loh, Piek, & Barrett, 2011), academic underachievement (Alloway, 2007), emotional problems such as increased levels of anxiety and depression (Piek et al., 2007; Skinner & Piek, 2001), social difficulties (Smyth & Anderson, 2000), as well as other developmental difficulties such as Attention Deficit Hyperactivity Disorder (ADHD) (Pitcher, Piek, & Hay, 2003). Such findings provide further evidence for the strong links between motor competence and the cognitive and psychosocial domains.

There is increasing evidence showing that DCD is an important, although often misunderstood and unrecognised (Missiuna, Moll, King, King, & Law, 2006), childhood condition with associated negative implications for a child's daily life. Despite this, little is known about the aetiology of motor problems in children. Twin research employing a co-twin control design to investigate possible unique environmental factors has revealed an association between perinatal oxygen perfusion problems and DCD (Pearsall-Jones et al., 2008; Pearsall-Jones, Piek, Rigoli, Martin, & Levy, 2009). Consequently, it has been suggested that DCD may fall along a continuum of movement disorder along with cerebral palsy (Pearsall-Jones, Piek, & Levy, 2010). There is also strong evidence for the involvement of cerebellar processes when understanding movement difficulties (Cantin, Polatajko, Thach, & Jaglal, 2007; Ivry, 2003; O'Hare & Khalid, 2002; Zwicker, Missiuna, & Boyd, 2009). However, given the heterogeneity of DCD, it has been noted that it is unlikely that the cerebellum is the sole underlying mechanism explaining motor problems in children (Zwicker et al., 2009).

Piek et al. (2004) pointed out the way in which developmental disorders mostly appear as recognisable syndromes and tend to be comorbid with particular disorders. It is therefore possible that the high comorbidity rate between developmental difficulties, such as motor skill and attention problems, may reflect a common underlying neurocognitive mechanism (Piek et al., 2004). Following on from this notion, recent research has suggested that deficits in the acquisition and automation of new motor skills may in fact arise from a developmental delay in complex cognitive processes, namely, executive functions (Michel, Roethlisberger, Neuenschwander, & Roebers, 2011). Continued research on the underlying processes involved in movement, as well as its relationship to cognitive and psychosocial development, is crucial in order to inform interventions designed to remediate problems experienced by children with these difficulties.

#### Motor and Cognitive Development

Piaget (1953) argued that cognitive development relies on motor functioning and Bushnell and Boudreau (1993) suggested that motor development may act as a prerequisite or 'rate-limiting factor' for the developmental sequence in which certain perceptual and cognitive abilities emerge. In spite of this, motor and cognitive development have often been examined and discussed separately as they were initially thought to occur across separate domains, along distinct developmental timetables, and underpinned by different neural processes (Davis, Pitchford, & Limback, 2011). Descartes (1596–1650) stated that cognitive processes are entirely different from motor processes. However, in more recent times, there has been accumulating support for the important relationship between motor and cognitive development (Diamond, 2000).

Through neuroimaging and neuroanatomical analysis, Diamond (2000) provided strong evidence for the close interrelation between motor and cognitive development and suggested that the cognitive functions of the prefrontal cortex (i.e., executive functions such as holding information in mind in order to remember what we are supposed to be doing, and inhibiting a frequently used movement in favour of a more appropriate behaviour) are important for skilled motor performance (Diamond, 2000). It was also stated that the cerebellum is important for cognitive functions in addition to motor functions and hence, the association between the motor and cognitive domains may be understood in terms of the close co-activation of the cerebellum and the prefrontal cortex areas. Since Diamond's work, there has been a growing consensus that executive functions are not only involved in the mastery of complex cognitive tasks but may also be implicated with motor skill performance (e.g., Michel et al., 2011; Piek, Dyck, Francis, & Conwell, 2007).

The literature presents a myriad of definitions for executive functions, although it is commonly employed as the umbrella term for the various cognitive processes underlying purposeful, goal-directed behaviour and adaptive responses to complex or novel situations (Hughes, 2011). These include the ability for goal formation and planning, and the effective execution of goal-directed plans (Jurado & Rosselli, 2007). There is also ongoing debate about whether the executive function construct is best considered to be a unitary process or a set of related but separable components. Many have adopted the latter view based on empirical and neuroimaging evidence. For example, executive dysfunction rarely occurs as a global impairment. Instead, specific executive function processes have been linked to distinct prefrontal areas, and these executive function processes have also shown variable developmental profiles (Anderson, 2002; Best, Miller, & Jones, 2009). Factor analytical research in adult populations has revealed three distinguishable but related domains, namely, inhibition (i.e., the ability to inhibit one's automatic, dominant, or prepotent responses), working memory (i.e., the ability to maintain and manipulate information over a brief period of time to support completion of tasks), and switching/shifting between different mental states, rule sets, or tasks (Miyake et al., 2000). Support for these separable domains has also been found at various developmental stages including older children (Lehto, Juujärvi, Kooistra, & Pulkkinen, 2003). Conversely, there is some evidence to suggest a single-factor model of executive functioning in very young children (Wiebe, Espy, & Charak, 2008). Best and Miller (2010) therefore argued that the degree of unity and independence of executive function components may vary developmentally. However, it is important to note that although there are inconsistencies regarding the nature and definition of executive functions, Cartwright (2012) suggests that at the basis of each definition is the notion of control, conscious or unconscious, of one's mental and

physical actions. Thus, it is clear that there are overlapping aspects between conceptualisations of motor control and cognitive control (Roebers & Kauer, 2009).

Executive function tasks are often described as those involving a high level of difficulty (due to complexity or novelty), changing conditions, time constraints and/or an emphasis on accuracy (Hughes & Graham, 2002). Research has shown that children with motor difficulties demonstrate greater coordination problems when tasks are more complex (Piek & Coleman-Carman, 1995) and involve greater demands for speed or accuracy (Vaessen & Kalverboer, 1990). This provides some support for the notion that executive function deficits may be implicated in motor coordination problems. Recent studies have examined this idea further by investigating executive functions in children with motor impairment.

A study investigating executive functions in children (aged 6 to 14 years) with DCD or ADHD revealed that on a task measuring both working memory and behavioural inhibition, the DCD group performed more slowly and had greater variability (but did not produce more errors) than the ADHD or control groups (Piek et al., 2007). It was also found that the DCD group had slower visual inspection times and slower reaction times on a line length discrimination task assessing both processing speed and set-shifting (Piek et al., 2007). Significant differences on this task remained even after visual inspection time was controlled for, suggesting that the children with DCD made a slower motor response and needed more time to discriminate the stimulus when the task became more complex. The authors also suggested that the poorer performance speed and variability found in these studies may be related to cerebellar processes (Piek et al., 2007). Furthermore, given that children with ADHD did not perform more poorly than the control group, it is possible that inconsistencies across studies implicating executive function deficits in ADHD may be the result of comorbid conditions, such as DCD, which has not been addressed in these previous studies (Piek et al., 2007).

In their study of five to seven year old children with and without motor deficits, Michel and colleagues (2011) investigated the executive functions of inhibition and setshifting. They found that children with coordination difficulties were slower in

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performing inhibition and attention shifting tasks, but did not produce more errors than the control group (Michel et al., 2011). However, Michel et al. argued that their results were unlikely to be entirely due to information-processing speed or due to the motor demand of the task, as the children did not perform slower on a simple reaction task that required the same motor response. Furthermore, the motor demands were minimal. It was suggested that children with motor deficits have slower performance due to the complex demands of such tasks, such as the speed-accuracy trade-off component requiring the need to react as fast and accurately as possible (Michel et al., 2011).

These studies have revealed important links between executive functions and motor problems. There has been suggestion that executive function deficits may predict motor problems (Michel et al., 2011), yet longitudinal evidence for the direction of the relationship is limited. There are few studies that have shown that motor ability predicts executive functions. For example, infant gross motor development has shown to predict executive functions in primary school-aged children (Piek et al., 2008) and adults (Murray et al., 2006). A recent longitudinal study has investigated the relationship between motor skills and spatial working memory in preschool children and found that motor skills predicted spatial working memory over a nine-month period, whereas, spatial memory was not found to predict motor performance at follow-up (Niederer et al., 2011). Niederer et al. (2011) suggested that these results may provide an indication of the dominant direction of the relationship between motor skills and executive functions in the relationship between motor skills and executive functions in predict motor performance at cognitive areas. Although, it is important to note that the relationship between motor skills and executive functions may change developmentally.

It is also important to note that examination of the relationship between motor coordination and executive functions is needed using a normative population, as this may provide valuable information, such as whether these relationships exist along the continuum of ability (i.e., from those who perform poorly on both motor and cognitive tasks to those who perform well in both these areas) (Wassenberg et al., 2005). It appears that the few normative studies investigating the association between the motor and cognitive domains have involved younger samples (Roebers & Kauer, 2009;

Wassenberg, 2005). Therefore, further studies are needed to examine whether the relationship extends to older samples, including adolescents.

Continued research in the area is also needed as it may have practical implications for children with DCD, particularly given that executive functions are a significant predictor of various functional outcomes. Research has demonstrated an overlap between DCD and language, reading, spelling, and math problems (Alloway, 2007; Dewey, Kaplan, Crawford, & Wilson, 2002). There is also accumulating crosssectional (e.g., Bull & Scerif, 2001; St Clair-Thompson & Gathercole, 2006) and longitudinal (e.g., Bull, Espy, & Wiebe, 2008; Mazzocco & Kover, 2007) evidence for an important association between executive functions and academic achievement. This suggests that executive functions may have a role in explaining the relationship between motor skills and these learning outcomes. However, although research has shown an association between executive functions and academic achievement across age groups (Best, Miller, & Naglieri, 2011) and in both clinical (e.g., Miller & Hinshaw, 2010) and normative samples (Best et al., 2011), the causal relationship between the two areas is still unclear. One suggestion is that executive functions directly impact on academic attainment.

Investigation into the possible causes of low academic achievement has consistently revealed a close relationship between working memory performance and indicators of academic achievement, with the understanding that poor working memory may directly impede the acquisition of knowledge and the ability to learn complex skills (Gathercole, Lamont, & Alloway, 2006). It seems clear that working memory skills would be important in explaining individual differences in learning, as poor working memory is known to hamper the ability to remember instructions, perform mental calculations as well as various other academic tasks such as sentence writing (Best et al., 2009; Gathercole et al., 2006). Given findings of an important link between motor skills and working memory (e.g., Alloway, 2007; Piek et al., 2008; Roebers & Kauer, 2009), it is therefore plausible that working memory mediates the relationship found between motor and learning problems.

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# Motor Skills, Working Memory, and Academic Achievement

There is growing evidence for a strong link between working memory measures and cognitive ability. Consequently, researchers have argued that working memory may be the crucial underpinning of the psychometric concept of general intelligence (Miyake, Friedman, Rettinger, Shah, & Hegarty, 2001). A considerable body of research suggests that working memory is a reliable predictor of general fluid intelligence (Engle, Tuholski, Laughlin, & Conway, 1999), reading and mathematics (de Jong, 1998; Gathercole, Alloway, Willis, & Adams, 2006; McLean & Hitch, 1999), language comprehension (Nation, Adams, Bowyer-Crane, & Snowling, 1999), and attentional control (Kane, Bleckley, Conway, & Engle, 2001). Associations between working memory and learning have also been shown in children with special educational needs (Gathercole & Pickering, 2001) and using national assessments in literacy and numeracy (Gathercole & Pickering, 2000). Importantly, the significant associations between working memory and achievement have been found to persist even after differences in IQ were statistically controlled (Cain, Oakhil, Bryant, 2004; Gathercole et al., 2006).

Given extensive research linking learning outcomes to both motors skills (e.g., Dewey et al., 2002) and working memory (e.g., Gathercole et al., 2006), and given increasing evidence for an important association between working memory and motor skills (e.g., Piek et al., 2008), it is therefore plausible that working memory may have a crucial role in the relationship between motor skills and academic outcomes. Recent research has highlighted the causal link between working memory impairments and learning difficulties and investigated whether this relationship extends to children who exhibit primary deficits in motor coordination (Alloway, 2007).

Alloway (2007) investigated a sample of 55 children with DCD, aged between 5 to 11 years, to further understand the working memory profile of these children and whether there would be links between verbal and visuospatial memory (short-term and working memory) impairments and learning. Approximately half of the children with DCD demonstrated difficulty with verbal short-term and working memory, whereas slightly more children demonstrated poor visuospatial short-term and working memory, with up to 60% scoring low on the visuospatial working memory task. Results also

revealed that visuospatial short-term and working memory deficits were significantly worse than verbal short-term memory performance, although there was no significant difference between visuospatial memory and verbal working memory (Alloway, 2007). Therefore, it appears children with DCD struggle on tasks that require movement planning (involved in visuospatial memory tasks) as well as simultaneous processing and storage of information (required in both verbal and visuospatial working memory tasks) (Alloway, 2007). In terms of learning outcomes, over half of the sample demonstrated literacy and numeracy difficulties, with 56% and 51% performing one standard deviation below the mean respectively. It was also found that those children with low visuospatial memory skills (based on a composite visuospatial short-term and working memory score) performed significantly worse with respect to learning (literacy and numeracy) when compared to a high visuospatial memory group, even after verbal and performance IQ were taken into account (Alloway, 2007). This result is consistent with the findings that working memory skills are dissociable from IQ in predicting learning ability (e.g., Gathercole et al., 2006). Alloway also found a significant difference in learning outcomes between low verbal working memory and high verbal working memory groups, but not when performance IQ was controlled for. This study provides preliminary evidence for understanding the relationship between memory (in particular, working memory) and motor skills and how this may affect learning. It was suggested that difficulties with processing and storing information may underlie the learning problems found in children with DCD. However, more research is needed.

Although the volume of research in this area has increased, the mechanisms underlying the relationship between motor skills and learning outcomes remain unclear. It has been noted that this may be due, in part, to the substantial heterogeneity in the cognitive profiles of children with DCD (Alloway & Temple, 2007). Further research is therefore needed in normative samples, particularly in adolescents, extending Alloway's (2007) research on these relationships in children with DCD. Alloway also noted that the increased overlap between DCD and attention difficulties, which was not addressed in her particular study, merits further attention in relation to its possible implications for the relationship between motor skills, memory, and learning. Continued research on the relationship between motor skills and learning outcomes is also important, as academic underachievement may have a detrimental effect on the psychosocial outcome of these children. Both children with DCD and those with learning difficulties have demonstrated poorer psychosocial outcomes such as depression, anxiety and lower self esteem (Li & Morris, 2007; Piek et al., 2007; Skinner & Piek, 2001). Cairney and colleagues (2010) proposed an Environmental Stress Hypothesis, stating that the associated negative experiences of DCD (including difficulties in the classroom) may play an important role in the aetiology of mental health difficulties in this population. This highlights the importance of further understanding the relationships between motor, cognitive, and emotional domains in order to improve prognosis in these children.

# **Motor and Emotional Functioning**

Recognition of the importance of motor competence for positive emotional development is increasing given the number of studies that are now showing a significant link between these two domains. Piek and colleagues (2007) found that child and adolescent twins with DCD demonstrated higher levels of depressive symptomatology than their co-twin without DCD. In this study, DCD and depressive symptomatology were ascertained by parent-rated questionnaires. A measure for ADHD was also employed to control for its confounding effects. In another study of 7 to 11 year old children, results showed that those with mild to severe DCD (based on a standardised performance measure) reported increased depressive symptoms compared with age and gender matched control children (Francis & Piek, 2003). More recently, a population-based study revealed that 11 year old children (77 boys and 82 girls) with parent-rated motor problems, reported more depressive symptoms than their peers without motor coordination difficulties (Campbell, Missiuna, & Vaillancourt, 2012). These accumulating findings are of concern, given that depression is associated with negative functional outcomes such as interpersonal difficulties (Garber, 2006; Puig-Antich et al., 1985).

Research has also demonstrated an important relationship between motor problems and anxiety symptoms. Skinner and Piek (2001) found increased levels of selfrated anxiety in children and adolescents with DCD (scoring below the 15th percentile on a motor performance test) compared with controls. These findings were supported by a twin study, which also reported increased levels of anxiety in individuals with DCD compared with their non-DCD co-twins (Pearsall-Jones, Piek, Rigoli, Martin, & Levy, 2011). A more recent study investigated the level and profile of anxiety in children with a clinical diagnosis of DCD (based on DSM-IV criteria), whilst also controlling for overlapping conditions such as ADHD (Pratt & Hill, 2011). The results suggested that children with DCD not only show increased levels of anxiety, according to parentreport, but also experience greater difficulty in the domains of panic/agoraphobic anxiety, social phobia, and obsessive compulsive anxiety than children in a typically developing group. Up to 50% of the DCD group experienced elevated levels of panicrelated symptoms (Pratt & Hill, 2011).

Children with anxiety disorders have shown increased motor skill difficulties (Ekornas, Lundervold, Tjus, & Heimann, 2010; Skirbekk, Hansen, Oerbeck, Wentzel-Larsen, & Kristensen, 2012). Ekornas et al. (2010) screened for children with 'pure' anxiety disorders (i.e., without comorbidities such as ADHD) from a population based sample and compared them with a matched control group with no psychiatric diagnosis. In this study, children aged 8 to 11 years with anxiety disorder displayed increased risk for motor skill impairment compared to the control group (44% versus 11% below the 5th percentile respectively). Anxious boys in particular demonstrated motor problems, with up to two-thirds of these boys demonstrating motor difficulty at or below the 5th percentile (Ekornas et al., 2010). These findings were also supported by a clinical study of 8 to 13 year old children (Skirbekk et al., 2012). Children diagnosed with an anxiety disorder, without comorbid ADHD, exhibited increased motor impairment compared with controls, with a total of 46% of the anxious children scoring below the 5th percentile on the motor performance measure.

It is now increasingly accepted that there is an important relationship between motor and emotional functioning, and that motor problems may have significant implications for the emotional development of a child. However, the causal relationship remains unclear. A shared organic basis for the association is plausible. This notion partly stems from suggestions of a specific link between elevated anxiety and balance problems in children (Erez, Gordon, Sever, Sadeh, & Mintz, 2004) and adults (Balaban & Jacob, 2001). Brain structures such as the cerebellum and basal ganglia have been implicated in balance control (Lalonde & Strazielle, 2007). Recently, a repetitive transcranial magnetic stimulation study implicated the cerebellum in the regulation of emotion and mood (Schutter & van Honk, 2009). Interestingly, Stins, Ledebt, Emck, van Dokkum, and Beek (2009) noted that balance problems may also result from increased activity in the limbic structures that underlie emotionality. It has therefore been theorised that the link between motor and emotional domains is mediated by common underlying neuronal networks (e.g., Stins & Beek, 2007; Stins et al., 2009). Research has proposed various cortical and subcortical loops that provide an interface between the emotion and motor control systems (Stins & Beek, 2007). For example, limbic structures (e.g., the anterior cingulate cortex and the orbitofrontal cortex) receive input from the amygdala, and send projections to the basal ganglia via the limbic loop (Stins & Beek, 2007). Thus, the basal ganglia may be involved in the bodily expression of emotions, as well as involuntary bodily movements. Skirbekk and colleagues (2012) caution, however, that the assumption of a 'specific' relationship between anxiety and balance problems is based primarily on the presence of balance problems in individuals with elevated anxiety, rather than on findings of increased balance problems relative to other motor areas in this group.

Based on their twin study, Moruzzi and colleagues (2010) argued that the relationship between motor problems and anxiety may be explained by shared genetic factors rather than a direct causal effect in either direction. However, it is important to consider the limitations of this study when interpreting the results. For example, motor problems were not measured by a standardised questionnaire or performance-based measure, but rather using a three item scale based on selected items from the Child Behavior Checklist (i.e., "Gets hurt a lot, accident-prone", "Poorly coordinated or clumsy", "Underactive, slow moving, or lacks energy") (Achenbach & Rescorla, 2001). Moreover, a study has shown that the Child Behavior Checklist items of "Gets hurt a lot, accident-prone" and "Poorly coordinated or clumsy", lack predictive validity in

identifying mild to moderate motor impairment in non-clinical populations (Piek et al., 2010).

It has been noted that psychosocial problems are generally thought to be secondary consequences to motor problems and tend to appear once a child is challenged by social and peer demands in the school years (Piek, Bradbury, Elsley, & Tate, 2008). The idea that emotional problems may follow motor coordination difficulties is also partly supported by longitudinal studies, which have shown that motor skill difficulties earlier in childhood are related to later emotional problems (Lingam et al., 2012; Losse et al., 1991; Shaffer et al., 1985; Sigurdsson et al., 2002). Conversely, there does not appear to be any evidence that anxiety precedes later motor impairment. It is also important to note that a significant association has been found between motor coordination and anxious/depressed behaviour in preschool age children as young as three and four years of age (Piek et al., 2008). This highlights how from a very young age, the ability to move through space and manipulate objects effectively and efficiently is of great importance and may potentially contribute to social and emotional difficulties if a child is unable to play appropriately with their peers (Piek et al., 2008).

Pratt and Hill (2011) argued that given their motor skills difficulties, children with DCD have to navigate an intensely complex world. It therefore makes sense that motor problems may lead to increased risk for emotional problems such as anxiety. It was hypothesised that if a child persistently struggles on the everyday tasks that are, in comparison, carried out relatively easily by their peers, this may lead to anxiety for current and future situations, feelings of panic when attempting such tasks, as well as possible avoidant behaviours. Avoidance may, in turn, impede the development of appropriate skills needed for challenging tasks (Pratt & Hill, 2011).

In their Environmental Stress Hypothesis, Cairney and colleagues (2010) propose that the negative psychosocial consequences (secondary stressors) of motor problems (primary stressor) may then lead to negative self-appraisals which in turn, contribute to increased risk for emotional problems. Indeed, there is much research showing that children and adolescents with DCD report lower self-perceived competence in various domains, including negative perceptions of athletic, scholastic,

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and physical competence as well as perceived lower levels of self-worth and social acceptance and support (Piek, Baynam, Barrett, 2006; Piek, Dworcan, Barrett, & Coleman, 2000; Schoemaker & Kalverboer, 1994; Skinner & Piek, 2001). Skinner and Piek (2001) stated that the low self-perceived competence experienced by children with DCD may be understood in terms of the repeated failure encountered in their daily lives. The term competence refers to one's level of mastery, which is known to be an important contributor to psychological health and well-being (Pearlin, Menaghan, Lieberman, & Mullan, 1981). Hence, for individuals with movement problems, their own sense of mastery may play a significant role in the emotional outcomes of these children. Skinner and Piek also highlighted how these children often try to avoid participation in such activities for fear of failure and/or peer criticism. Subsequently, by avoiding participation, children limit their opportunity to practise skills and to participate in a social environment, creating a vicious circle. This demonstrates the importance of motor skill competency and its relationship to a child's self-perceptions, social functioning and, ultimately, emotional adjustment.

Using a co-twin control design, Piek et al. (2007) and Pearsall-Jones et al. (2011) provided support for the Environmental Stress Hypothesis. It was suggested that their findings of increased anxiety and depressive symptomatology found in monozygotic twins with DCD, compared with co-twins without DCD, may be understood in terms of nonshared environmental experiences, such as the negative peer relationships and academic difficulties experienced by those with DCD. Skinner and Piek (2001) also provided partial support for the Environmental Stress Hypothesis, as they found that children and adolescents with DCD perceived themselves as less competent (e.g., physically, socially, and scholastically) and as having lower social support than their peers without DCD. Those with DCD also reported lower global self-worth and higher state and trait anxiety compared with the control group. Therefore, it is plausible that lower self-perceptions of competency, self-worth, and social support may mediate the relationship between motor and anxiety problems. Skinner and Piek (2001) noted that further research is needed to investigate the interplay between these variables.

Recently, Lingam and colleagues (2012) employed a longitudinal design with a

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large birth cohort sample in order to explore the association between 'probable' DCD and mental health difficulties, whilst also taking into account potential mediating factors. It was found that probable DCD at 7 years of age was associated with increased risk of self-reported depressive symptoms and parent-rated mental health and behavioural difficulties (i.e., emotional symptoms, conduct, hyperactivity and inattention, and peer relationship difficulties) at 9 or 10 years. The significant association remained after controlling for child-related (e.g., gender, age, and stressful life events) as well as parent and environmental-related confounding factors (e.g., socio-economic status, parent mental health). Furthermore, it was found that verbal IQ, social communication, bullying, self-esteem, and scholastic competence mediated the relationship, with problems in these areas increasing the risk of mental health difficulties in children with DCD.

There is some support for the Environmental Stress Hypothesis in the literature, however, these studies are scarce. Therefore more research is needed in order to further examine the potential mediating and moderating factors influencing the relationship between motor and emotional problems. It is also important to note that the influence of certain mediating variables may also depend on a child's age (e.g., Lingam et al., 2012). Factors that contribute to the mental health outcomes of younger children may be different to those which are important in explaining the emotional outcomes of adolescents.

Ultimately, although there has been a proliferation of research on the outcomes of motor difficulties in children, misconceptions prevail (Missiuna et al., 2006). For example, that these children will outgrow their motor difficulties and thus do not warrant special attention. Furthermore, research has shown that motor problems continue into adolescence (e.g., Cantell et al., 1994; Losse et al., 1991), yet research in this age-group is limited. Further investigation of these relationships, particularly in adolescents, is integral for increasing awareness and recognition of the possible associated problems of motor coordination difficulties.

# **Aims and Rationale**

The primary aim of this thesis was to further understand the relationship between

motor coordination and the cognitive, academic, and emotional domains in adolescents.

Accumulating research suggests that motor and emotional problems are linked and that difficulties in the motor domain may be predictive of internalising problems, including anxiety and depression (Pearsall-Jones et al., 2011). However, longitudinal studies are limited. Further longitudinal research is therefore needed to provide information on the possible causal relationship between these areas. Previous work has identified a relationship between school-age motor ability and anxiety problems in adolescence (e.g., Shaffer et al., 1985; Sigurdsson et al., 2002). Hence, a specific aim of this thesis was to provide further longitudinal evidence on the relationship between motor skills and emotional outcomes by examining whether early motor development (i.e., in infancy to early childhood) predicted later emotional functioning at school-age. This is important as it provides an empirical basis for exploring the Environmental Stress Hypothesis (i.e., that it is the negative environmental experiences associated with motor problems that may result in emotional difficulties) (Cairney et al., 2010) in an older sample.

Studies providing a direct exploration of the way in which motor ability may impact on emotional outcomes are still very limited. Cairney and colleagues (2010) highlighted the need to investigate multiple pathways, incorporating both risk and protective factors, when linking coordination difficulties to internalising problems. Previous research examining the potential mediating factors in the relationship between these areas has involved children with motor problems (e.g., Lingam et al., 2012). Therefore, information regarding the relationship is needed from a normative perspective and in an older sample. Research in adolescent populations is important given that this age-group has shown to experience greater anxiety and lower selfperceived social support than younger groups (e.g., Skinner & Piek, 2001). Also, motor problems may have a significant impact during adolescence as it is a time when increased importance is placed on affiliation and social support. This is concerning for those with motor difficulties given evidence for social and physical withdrawal in these individuals (e.g., Cairney et al., 2005; Schoemaker & Kalverboer, 1994). It has been suggested that access to important socialisation processes and coping experiences available in physical activities (e.g., sporting games) may be limited for those with poor motor skills, ultimately hampering their self-perceived competencies and emotional well-being (Ekornas et al., 2010).

A specific aim of this thesis was to investigate the association between motor coordination and psychosocial functioning in a normative sample of adolescents, specifically, to determine whether the link between motor coordination and emotional symptoms (depression and anxiety) is mediated by the self-perceived competencies in these adolescents. Clarification of the potential mediating factors in the relationship between motor coordination and emotional functioning is crucial particularly given the functional impairments associated with internalising problems. For example, research has suggested that children with elevated anxiety and depressive symptoms also demonstrate difficulties in the cognitive domain, including executive functions (e.g., Emerson, Mollet, & Harrison, 2005). Recently, research showing a link between executive functions and motor problems has also increased.

Studies have shown that executive function difficulties exist in children with DCD. However, research is also needed in normative samples. Previous research employing normative samples have involved younger children aged 5 or 7 years (e.g., Roebers & Kauer, 2009; Wassenberg et al., 2005), or a mixed sample of children and adolescents (Piek et al., 2004). Best et al. (2009) notes that most research on executive function and its correlates has focused on young children, and therefore highlights the importance of further studies in older age-groups given that significant improvements in executive functions are seen into adolescence and even adulthood. Therefore, it is plausible that the relationship between motor ability and executive functions may also change developmentally. Normative research has also revealed specific relationships between these areas, that is, that specific components of motor coordination may have a relationship with certain aspects of executive function, whereas other components may not (e.g., Roebers & Kauer, 2009). For example, Roebers and Kauer (2009) found a significant association between working memory and postural flexibility but not between working memory and a fine motor pegboard task in their normative study of 7 year old children. It is plausible that these specific relationships may be understood in

terms of common underlying neural processes. Continued research is needed. This thesis therefore aimed to examine whether specific relationships between different components of motor coordination (manual dexterity, aiming and catching skills, balance) and executive function (working memory, inhibition, and set-shifting) are also found in adolescents. Examination of the association between motor ability and executive functions is important as executive functions have significant implications for various outcomes particularly in the academic domain (Best et al., 2011). This also suggests that executive functions may have an important role in explaining the academic underachievement seen in individuals with motor problems.

Although there is much research showing that motor problems are related to difficulties in various academic domains, the processes underlying the relationship are still unclear. Recently, it has been found that working memory is important when understanding the increased risk for academic underachievement in children with DCD (Alloway, 2007). Therefore, an aim of this thesis was to extend Alloway's (2007) findings by establishing whether working memory plays a mediating role in the relationship between motor coordination and academic achievement in a normative sample of adolescents. Furthermore, previous research did not account for the confounding influence of ADHD symptomatology (Alloway, 2007). Therefore, a specific aim of this thesis was to control for such potentially confounding variables in the relationship between motor skills, working memory, and learning. The findings of such research may have practical implications in the assessment and treatment of individuals who present with motor and learning problems, as targeting working memory difficulties in intervention may have important benefits in improving learning outcomes.

The findings from this thesis aim to shed further light on the relationships between the motor, cognitive, and psychosocial domains with important implications for those who may experience difficulties in these areas. Based on figures in the literature, it appears that motor coordination difficulties are common in the general population, with the American Psychiatric Association (2000) suggesting a DCD prevalence rate of approximately 6% for 5 to 11 year old children and figures in the literature ranging from

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approximately 1.4% to 19% (Tsiotra et al., 2006; Wright & Sugden, 1996). There is a lack of epidemiological research on the prevalence of this condition in adolescent years. This may be partly due to the difficulties in diagnosing DCD including, the shortage of appropriate norm-referenced motor skill tests for this age-group. Although, importantly, longitudinal evidence has shown that DCD persists into adolescence (e.g., Cantell et al., 1994; Hellegren et al., 1993; Losse et al., 1991). This is concerning given the significant cognitive, academic, and psychosocial difficulties seen in those with movement difficulties. It is clear that identification of these individuals in the community is imperative in order to prevent and target the associated negative outcomes. Recently, attention has been paid to evaluating screening questionnaires that may provide a cost and time effective way of identifying those at risk for motor problems. For example, the original parent-rated Developmental Coordination Disorder Questionnaire (DCDQ) was designed to assess motor difficulties in children, and recently extended its age range to include adolescents. Given that, to the authors knowledge, an evaluation of the revised DCDQ in this older age group has not be carried out, a final aim of this thesis was to investigate the suitability of this measure in screening for motor difficulties in a community-based sample of adolescents.

# Outline of papers included in this thesis

The first paper (Paper 1) in this thesis presents a longitudinal study of the relationship between motor development and emotional outcomes. The participants in this study were recruited in the first few months of life as part of a larger study investigating the relationship between preterm birth and early spontaneous activity. A subsample of the participants from the larger study agreed to the follow-up testing stage once reaching school-age. As part of the larger study, parent-rated developmental screening questionnaires were used to assess fine and gross motor performance from 4 months to 4 years of age. At follow-up, these children aged 6 to 12 years were then assessed on their levels of anxious and depressive symptomatology. This paper extends previous research as it examines whether early fine and gross motor development, from infancy, predicts anxious and depressive symptomatology at school-age. The aim of this

paper is to provide support for the Environmental Stress Hypothesis, which is then the focus of the second paper (Paper 2).

The second and subsequent papers present a series of cross-sectional studies, involving a normative sample of adolescents aged 12 to 16 years. These adolescents were tested on a large assessment battery comprising motor coordination, cognitive functioning, academic achievement, and psychosocial measures. These data were collected for the purposes of the main aim of the doctoral thesis, which was to investigate motor coordination and its psychosocial and cognitive correlates in an adolescent population and from a normative perspective.

Paper 2 provides an examination of possible factors that may mediate the relationship between motor and emotional functioning. Specifically, it explores the association between motor coordination and emotional outcomes (i.e., depressive and anxious symptomatology) in adolescents, and examines whether the relationship is mediated by self-perceived competencies in various domains such as academic, social, and physical. Importantly, the study takes into account potential confounding factors in the relationship, including ADHD symptomatology, verbal ability, and socio-economic status.

In addition to the psychosocial implications of motor problems, research has also suggested an important link between motor skills and executive functions (Diamond, 2000). Therefore, the third paper (Paper 3) provides an investigation into motor and executive functioning in a sample of adolescents, exploring how different components of motor coordination may be related to various aspects of executive functioning, including working memory, inhibition, and set-shifting. This study also controls for factors such as ADHD symptomatology, socio-economic status, and verbal ability. Findings of specific relationships between these areas may provide information on the possible underlying neural mechanisms of this relationship. It may also point to possible neurocognitive mechanisms that could underlie the associations between motor ability and academic outcomes, the focus of the fourth paper.

The fourth paper (Paper 4) aims to further understand the strong links found between motor performance and learning outcomes. It extends previous findings by employing an adolescent normative sample to examine whether working memory performance mediates the relationship between motor coordination and academic outcomes (namely, word reading, numerical operations, and spelling) in this age-group. Importantly, the paper also extends previous work by controlling for the confounding effects of ADHD symptomatology.

Finally, the fifth paper (Paper 5) examines the internal consistency and validity of the revised DCDQ in a sample of adolescents. Evaluation of the DCDQ, in adolescents, is important as such a screening measure may prove valuable in identifying those at risk of motor difficulties who are in need of further assessment. This could have significant implications for preventing and addressing the possible negative cognitive and psychosocial outcomes described above.

Ultimately, theoretical and practical implications for the relationships found in these studies are discussed throughout the thesis. For example, possible underlying mechanisms that may explain the relationships are highlighted as well as the importance of these findings in relation to the assessment and intervention of children who experience difficulties in these domains. PAPER 1

Piek, J. P., Barrett, N. C., Smith, L. M., Rigoli, D., & Gasson, N. (2010). Do motor skills in infancy and early childhood predict anxious and depressive symptomatology at school age? *Human Movement Science*, *29*(5), 777–786.

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PAPER 2

Rigoli, D., Piek, J. P., & Kane, R. (2012). Motor coordination and psychosocial correlates in a normative adolescent sample. *Pediatrics*, *129*(4), e892-e900.

# Motor Coordination and Psychosocial Correlates in a Normative Adolescent Sample

**WHAT'S KNOWN ON THIS SUBJECT:** Research has highlighted an important relationship between motor coordination and emotional functioning in children and adolescents. Few studies have provided support for this idea; research is therefore needed to further understand the relationship between the motor and emotional domains.

**WHAT THIS STUDY ADDS:** The results suggest that the relationship between motor coordination and emotional functioning (anxious and depressive symptoms) in an adolescent sample may be understood in terms of a mechanism whereby motor coordination has an indirect impact on emotional functioning via self-perceptions.

## abstract

**OBJECTIVES:** Previous research has revealed an important relationship between motor coordination difficulties and internalizing problems such as anxiety and depressive symptoms. However, further research is needed to understand the potential mediating factors in this relationship. The aim of the current study was to examine whether the association between motor coordination and emotional functioning is mediated by self-perceptions in a normative sample of adolescents.

**METHODS:** Participants included 93 adolescents aged 12 to 16 years. The Movement Assessment Battery for Children–2 provided 2 indicators of motor coordination; the Mood and Feelings Questionnaire and Spence Children's Anxiety Scale provided 2 indicators of emotional functioning; and the Self-Description Questionnaire–II provided 6 indicators for self-perceived competence.

**RESULTS:** Structural equation modeling revealed that motor coordination affects emotional functioning via self-perceptions.

**CONCLUSIONS:** These results suggest that the relationship between motor coordination and emotional functioning in adolescents from a normative sample may be understood in terms of a mechanism by which motor coordination has an indirect impact on emotional outcomes through various self-perception domains. These findings have important implications for increasing awareness and developing appropriate treatment programs for motor coordination and emotional difficulties. *Pediatrics* 2012;129:e892–e900

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#### **KEY WORDS**

adolescents, anxious symptoms, depressive symptoms, motor coordination, normative sample, self-perceptions

#### ABBREVIATIONS

ADHD—attention-deficit/hyperactivity disorder DCD—developmental coordination disorder MABC-2—Movement Assessment Battery for Children–2 MFQ—Mood and Feelings Questionnaire SCAS—Spence Children's Anxiety Scale SDQ-II—The Self-Description Questionnaire–II SES—socioeconomic status SWAN—Strengths and Weaknesses of ADHD Symptoms and Normal Behavior Scale VCI—Verbal Comprehension Index WISC-IV—Wechsler Intelligence Scale for Children–IV

www.pediatrics.org/cgi/doi/10.1542/peds.2011-1237

doi:10.1542/peds.2011-1237

Accepted for publication Nov 29, 2011

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PEDIATRICS (ISSN Numbers: Print, 0031-4005; Online, 1098-4275).

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**FINANCIAL DISCLOSURE:** The authors have indicated they have no financial relationships relevant to this article to disclose.

FUNDING: No external funding.

**COMPANION PAPER:** A companion to this article can be found on page e882, online at www.pediatrics.org/cgi/doi/10.1542/peds. 2011-1556.

Research has demonstrated an important link between movement difficulties such as developmental coordination disorder (DCD)<sup>1</sup> and increased depressive<sup>2</sup> and anxious symptoms.<sup>3</sup> Similarly, motor coordination difficulties have been found in children with anxiety-related disorders.<sup>4</sup> Although a common neurodevelopmental cause is plausible in some cases, motor coordination deficits themselves may cause internalizing problems.<sup>5</sup> Given the psychosocial implications (eg, academic underachievement,6 decreased participation in play,7 obesity problems8) and negative selfperceptions associated with motor skill difficulties,3 it is not surprising that anxiety and depression are possible emotional outcomes. Few studies, however, have provided support for this hypothesis.9

The current study examined a mediating model of the relationship between motor coordination, self- perceptions, and emotional functioning in an adolescent normative sample. It has been noted that correlational studies using normative samples are important to provide a better understanding of relationships found in children with DCD.<sup>10</sup> For example, methodologic problems associated with the use of clinical samples include overestimating associations between domains. Furthermore, research has also highlighted the value of a dimensional approach in research and practice given that it is often difficult to make clear boundaries between concepts such as developmental disorders.11

The study also aims to control for potentially confounding factors such as attention-deficit/hyperactivity disorder (ADHD) symptoms, verbal ability, socioeconomic status (SES), age, and gender.<sup>12</sup> This is important given the poorer emotional functioning found in individuals with combined ADHD and DCD.<sup>2</sup> Sigurdsson et al<sup>13</sup> found that childhood motor impairment was strongly

associated with persistent anxiety in male adolescents but not females. Furthermore, adolescents (both those with DCD and control groups) have reported significantly lower levels of social support and self-worth, and higher levels of anxiety, than younger children,<sup>3</sup> highlighting the importance of further research in adolescent populations. A partial mediation model is proposed in which motor coordination has both a direct and indirect effect via selfperceptions on emotional functioning. Specifically, better motor coordination will have a positive, direct effect on emotional functioning; in addition, better motor coordination will have a positive effect on self-perceptions, which in turn leads to better emotional functioning. The present correlational data cannot, of course, be used to establish cause-and-effect relationships. Our aim was to determine the degree to which the proposed causal model had the capacity to generate our correlational data.

#### **METHODS**

#### **Participants**

Recruitment occurred across 5 randomly selected secondary schools and through public advertisements. Ninetythree adolescents, 38 girls and 55 boys, aged 12 to 16 years (mean  $\pm$  SD: 14.2  $\pm$ 1.1 years) participated in the study. Participants had a minimum Verbal Comprehension Index (VCI) of 70, as measured by using the Wechsler Intelligence Scale for Children-IV (WISC-IV). This criterion excluded any adolescent whose difficulties might be attributed to general delayed development.<sup>14</sup> In addition, none had a physical disability, chronic illness, or a medical condition that affects development. SES scores were derived from the Australian Prestige Scale,15 which uses a 7-point scale with 1 representing high prestige of occupation and 7 representing low prestige. The occupation rated as most prestigious out of mothers' and fathers' occupations was used as the SES score (mean  $\pm$  SD: 3.77  $\pm$  1.00; range: 1.80-6.60).

#### **Measures**

## Movement Assessment Battery for Children–2

The Movement Assessment Battery for Children-2 (MABC-2)<sup>16</sup> is a standardized test used for the identification and description of children with movement difficulties and is suitable for age bands 3 to 6, 7 to 10, and 11 to 16 years. Age-based standard scores are derived for manual dexterity, aiming and catching, and balance domains and for the total test score. A score between the fifth and 15th percentile indicates a child is "at risk" of having a movement difficulty, and a total test score at or below the fifth percentile indicates significant movement difficulty. The age-standardized manual dexterity, aiming and catching, and balance component scores were used for this study.

The original MABC<sup>17</sup> is well established, and preliminary evidence suggests favorable psychometric properties for the recently revised MABC-2. The MABC-2 manual reports a reliability coefficient of 0.80 for the MABC-2 total test score and coefficients ranging from 0.73 to 0.84 for the individual component scores, as well as preliminary results demonstrating criterion-related and discriminative validity.<sup>16</sup>

#### Wechsler Intelligence Scale for Children–IV

The Wechsler Intelligence Scale for Children–IV (WISC-IV)<sup>18</sup> assesses cognitive ability in children aged 6 to 16 years 11 months. The WISC-IV subtests yield a full-scale IQ and 4 subtest indexes; namely, VCI, perceptual reasoning, working memory, and processing speed. For the current study, the VCI was used as a potential control variable and to exclude any adolescent whose difficulties might be attributed to general delayed development. The widely used WISC-IV has excellent internal consistency, test-retest reliability, criterion validity, and construct validity.<sup>18</sup>

#### Strengths and Weaknesses of ADHD Symptoms and Normal Behavior

The Strengths and Weaknesses of ADHD Symptoms and Normal Behavior (SWAN)<sup>19</sup> is a parent-rated questionnaire based on the 18 ADHD symptoms listed in the *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition.* It involves observations based on the last month with reference to other children of the same age. Scores for each item range from "far below average" (scored as 3) to "far above average" (scored as -3). An overall SWAN score, calculated by averaging the total of the 18 items, was used in this study.

Polderman et al<sup>20</sup> found the SWAN rating scale to yield a normal distribution of scores, therefore making it a useful instrument for examining variation of (hyper) activity and attention in the general population. Furthermore, Martin et al<sup>21</sup> found that the prevalence rate of ADHD, as assessed by using the SWAN, was comparable to those reported in previous studies. Cronbach's  $\alpha$  for the current study was high (.97), which is often indicative of item redundancy, but in this case it may reflect the relatively large number of items contained in the scale.<sup>22</sup>

#### Mood and Feelings Questionnaire

The Mood and Feelings Questionnaire (MFQ)<sup>23</sup> was developed as a screening tool to assess depression in children and adolescents aged 8 to 16 years. Items are derived from diagnostic criteria for depression and dysthymia specified within the *Diagnostic and Statistical Manual of Mental Disorders, Revised Third Edition.* The questionnaire

asks the child to rate depressive symptoms in the past 2 weeks as "not true" (0), "sometimes true" (1), or "true" (2). The MFQ has demonstrated high internal consistency and test—retest reliability and has also shown to reliably identify major depressive episode and other mood disorders in youth with diverse demographic and clinical characteristics.<sup>24</sup>

#### Spence Children's Anxiety Scale

The Spence Children's Anxiety Scale (SCAS)<sup>25</sup> assesses anxiety symptoms in children and consists of 6 subscales: panic attack and agoraphobia, separation anxiety disorder, social phobia, physical injury fears, obsessivecompulsive disorder, and generalized anxiety disorder. Respondents are asked to indicate frequency with which each symptom occurs on a 4-point scale ranging from never (scored 0) to always (scored 3). A total SCAS score, obtained by summing scores of the 38 anxiety symptom items, was used for the purposes of this study. The scale has shown high internal consistency for the total score and for each subscale, and strong psychometric properties have been reported with adolescent samples aged up to 19 years.<sup>26</sup>

#### The Self-Description Questionnaire-II

The Self-Description Questionnaire–II (SDQ-II)<sup>27</sup> is a 102-item self-report scale designed to measure multiple dimensions of self-concept for adolescents aged 12 to 18 years, including physical ability, physical appearance, samegender and opposite-gender peer relations, parent relations, mathematics, reading, school in general, and a global perception of self, as well as emotional stability and honesty/trustworthiness. The items are structured on a 6-point Likert scale ranging from "not like me at all" (1) to "it is very much like me" (6). The physical ability, physical appearance,

same-gender and opposite-gender peer relations, parent relations, school, and global self-concept subscale scores were used for the purposes of this study because these self-concept domains are commonly associated with motor difficulties.<sup>3,28,29</sup> The subscale scores represent the mean of each subscale's total score.

The SDQ-II demonstrates good internal consistency ranging from 0.83 to 0.91.<sup>27</sup> Byrne<sup>30</sup> stated that the SDQ-II is the most validated self-concept measure for use with adolescents.

#### **Procedure**

This study followed the National Health and Medical Research Council of Australia ethical guidelines and was granted approval from the Curtin University Human Research Ethics Committee and the participating schools' representative bodies. Written consent was provided by interested adolescents and their parents; participants were then individually tested by a trained examiner. Duration of testing was 4.5 hours, which was broken into 2 sessions. Measures were administered in a standard order. Adolescents completed the selfreport MFQ and SCAS scales and parents completed the SWAN and a developmental history questionnaire. Sessions were conducted at the family home or Curtin University, according to family preference.

#### **Data Analysis**

Structural equation modeling, with maximum likelihood estimation, was used to determine the extent to which self-perceptions mediate the relationship between motor coordination and emotional functioning. The analysis was implemented by using LISREL version 8.54 (Scientific Software International Inc, Lincoln, IL).<sup>31</sup>

For relatively simple models such as our 1-mediator model, sample sizes between 100 and 150 have been recommended.<sup>32</sup> Our current sample size of 93 falls just short of this recommendation but should be sufficient to provide stable estimates of the path coefficients.

#### RESULTS

#### **Descriptive Factors**

Means, SDs, and ranges for the variables measuring motor coordination, self-perceptions, and emotional functioning are presented in Table 1.

Five adolescents performed at or below the fifth percentile on the MABC-2 and 2 scored between the sixth and 15th percentile (regarded as at risk). The prevalence of significant movement difficulty (ie, at or below the fifth percentile) was 5.4%, which is comparable to previous estimates of 6%.1 Ten adolescents scored at or above the MFQ cutoff point of 29 (recommended by Daviss et al<sup>24</sup>). In terms of anxiety symptoms as measured by the SCAS, 7 adolescents were subclinical (1 SD above the mean) and 5 demonstrated clinical (1.5 SDs above the mean) levels.<sup>33</sup> One adolescent considered at-risk on the MABC-2 scored in the subclinical range on the SCAS, and 2 adolescents scoring at the fifth percentile on the MABC-2 scored in the clinical range for both the SCAS and MFQ.

#### **Correlations**

Potential control variables included, age, gender, SES, ADHD symptoms, and VCI. These variables did not significantly correlate with the outcome variables of anxiety and depressive symptoms and therefore were not included in the analysis.

Indicators driven by the same latent construct will necessarily correlate. For the self-perception construct, however, the opposite-gender peer relations subscale did not correlate with the parent relations and school subscales and was therefore not included in the model as an indicator of this construct. Similarly, the MABC-2 manual dexterity and aiming and catching subscales were not significantly correlated, although both correlated with balance. Because indicators of the same construct must be correlated, this pattern of correlations suggests 2 potential measurement models: 1 in which motor coordination is measured by manual dexterity and balance, and another in which motor coordination is measured by aiming and catching and balance.

TABLE 1	Means,	SDs,	and	Range	of	Scores	(N =	93)
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Scale	Mean	SD	Range
MABC-2 manual dexterity <sup>a</sup>	9.57	2.47	3.0 to 15.0
MABC-2 aiming and catching <sup>a</sup>	11.03	2.73	4.0 to 16.0
MABC-2 balance <sup>a</sup>	11.42	2.98	4.0 to 14.0
MFQ depressive symptoms <sup>b.c</sup>	13.49	10.43	1.0 to 48.0
SCAS anxiety symptoms <sup>b/c</sup>	21.91	12.36	1.0 to 67.0
SDQ-II physical ability <sup>b,d</sup>	4.54	1.23	1.25 to 6.00
SDQ-II physical appearance <sup>b.d</sup>	4.10	0.89	1.88 to 5.88
SDQ-II same-gender peer relations <sup>b.d</sup>	4.99	0.77	3.00 to 6.00
SDQ-II opposite-gender peer relations <sup>b,d</sup>	4.33	0.93	1.63 to 6.00
SDQ-II parent relations <sup>b,d</sup>	4.76	1.02	1.00 to 6.00
SDQ-II school <sup>b·d</sup>	4.70	0.93	2.00 to 6.00
SDQ-II general self <sup>b/d</sup>	4.46	0.71	2.00 to 5.50
SWAN ADHD symptoms <sup>b/e</sup>	-0.99	1.02	-3.0 to 1.22
WISC-IV VCIa	106.63	11.25	81.0 to 132.0
SES <sup>b,f</sup>	3.77	1.00	1.80 to 6.60

<sup>a</sup> Standard score

<sup>b</sup> Raw score.

<sup>c</sup> Total scores are calculated by summing the item scores.

<sup>d</sup> Scores are calculated by averaging the total of the SDQ-II subscale items.

e Scores are calculated by averaging the total of the 18 ADHD items.

f The occupation rated as most prestigious out of mothers' and fathers' occupation

An important underlying premise for mediation states that the independent variable (motor coordination) must be correlated with both the mediator (self-perceptions) and the outcome variable (emotional functioning).<sup>34</sup> Because manual dexterity was not correlated with the outcome measures, including this as an indicator of motor coordination would immediately compromise the mediator model. We therefore opted for a measurement in which motor coordination is measured by using the aiming and catching and balance subscales.

#### **LISREL Analysis**

In the current study, the 10 structural equation modeling indicators were not multivariate normal. It was therefore decided to use the Spearman correlation as an index of association among the indicators.<sup>35</sup> The Spearman correlations are reported in Table 2.

The parameter estimates for the measurement model (ie, the confirmatory factor model without the structural pathways among the latent variables) are given in Fig 1. Fit indices providing an indication of the overall fit of the measurement model are reported in Table 3. The fit statistics suggest an acceptable fit to the data. The  $\chi^2/df$ ratio was <3, the comparative fit index was >0.90, and the standardized root mean square residual was  $< 0.10^{.36}$ Although the root square mean square error of approximation is above the desired cutoff, Tabachnick and Fidell<sup>37</sup> note that this index may be less preferable with smaller samples due to the tendency to overreject the true model. Overall, the results indicate an acceptable fit for the measurement model.

The saturated structural model is depicted with its path coefficients in Fig 2A. The path from motor coordination to emotional functioning was not significant (when controlling for self-perceptions). The hypothesis that

TABLE 2 Spearman's Correlation Matrix for the Key Variables (N	atrix for the Ke	sy Variables (N =	93)								
Scale	Mapual	MABC-2 Aimine and	MABC-2 Balance	MFQ Depressive	SCAS Anvietv	SDQ-II Physical	SDQ-II Physical	SDQ-II Same-Gender	SDQ-II Darent	SDQ-II School	SDQ-II General
	Dexterity	Catching	Dalalloc	Symptoms	Symptoms	Ability	Appearance	Peer Relations	Relations	1001100	Self
MABC-2 manual dexterity <sup>a</sup>	1.00										
MABC-2 aiming and catchinga	0.048	1.00									
MABC-2 balancea	0.214e	0.367 <sup>f</sup>	1.00								
MFQ depressive symptoms <sup>b.c</sup>	-0.085	-0.239e	-0.297 <sup>f</sup>	1.00							
SCAS anxiety symptoms <sup>b.c</sup>	-0.047	-0.318f	-0.303f	0.748 <sup>f</sup>	1.00						
SDQ-II physical abilityb	0.169	0.460 <sup>f</sup>	0.424 <sup>f</sup>	-0.341 <sup>f</sup>	-0.371f	1.00					
SDQ-II physical appearance <sup>b.d</sup>	0.012	0.347 <sup>f</sup>	$0.265^{e}$	-0.391 <sup>f</sup>	—0.427 <sup>f</sup>	0.608 <sup>f</sup>	1.00				
SDQ-II same-gender peer relations <sup>b.d</sup>	0.095	0.123	0.272 <sup>f</sup>	-0.400f	-0.327 <sup>f</sup>	0.569 <sup>f</sup>	0.515 <sup>f</sup>	1.00			
SDQ-II parent relations <sup>b.d</sup>	0.003	0.052	0.142	-0.493 <sup>f</sup>	−0.242 <sup>e</sup>	0.296 <sup>f</sup>	0.329 <sup>f</sup>	0.459 <sup>f</sup>	1.00		
SDQ-II schoolbd	0.245 <sup>e</sup>	0.148	$0.207^{e}$	-0.484 <sup>f</sup>	-0.369f	0.239 <sup>e</sup>	0.385 <sup>f</sup>	0.377 <sup>f</sup>	0.487 <sup>f</sup>	1.00	
SDQ-II general self <sup>b.d</sup>	0.116	0.286 <sup>f</sup>	0.351 <sup>f</sup>	-0.621 <sup>f</sup>	-0.490 <sup>f</sup>	0.534 <sup>f</sup>	0.669 <sup>f</sup>	0.570 <sup>f</sup>	0.643 <sup>f</sup>	0.710 <sup>f</sup>	1.00
<sup>a</sup> Standard score.											
<sup>b</sup> Raw score.											
<sup>c</sup> Total scores are calculated by summing the item scores.	e item scores.										
<sup>d</sup> Subscale scores represent the mean of each subscale's total score.	ch subscale's total	score.									
$^{\rm e}$ P < .05 (2-tailed).											
$^{\rm f}$ P < .01 (2-tailed).											

motor coordination would have a direct impact on emotional functioning in this model was therefore not supported. All other hypotheses were supported. Specifically, the path from motor coordination to self-perceptions was significant, as was the path from self-perceptions to emotional functioning, indicating that motor coordination has an indirect effect on emotional functioning through self-perceptions.

Fit indices providing an indication of the overall fit of the saturated model can be found in Table 3. Using the cutoffs identified earlier when testing the measurement model, the fit statistics for the saturated model suggest an acceptable fit to the data. Once again, the root square mean square error of approximation is above the desired cutoff, but as we noted earlier, this index has a tendency to overreject the true model when the sample size is small.

The test of the saturated model suggested that, when self-perception is controlled, the magnitude of the path coefficient for the direct pathway from motor skills to emotional functioning is trivial. This pathway was therefore dropped from the model. The fit of the resulting mediator model was acceptable (Table 3). Furthermore, the fit of the mediator model was not significantly different from the fit of the saturated model ( $\chi^2_{\text{difference}}$ : -1.21; P = .271). The more parsimonious mediator model was therefore selected. The mediator model is depicted with its path coefficients in Fig 2B. LISREL estimated the indirect effect of motor coordination on emotional functioning to be -0.37. The mediator model explained 45.14% of the variance in emotional functioning.

#### DISCUSSION

The current results revealed that in a normative sample of adolescents, motor coordination did not have a

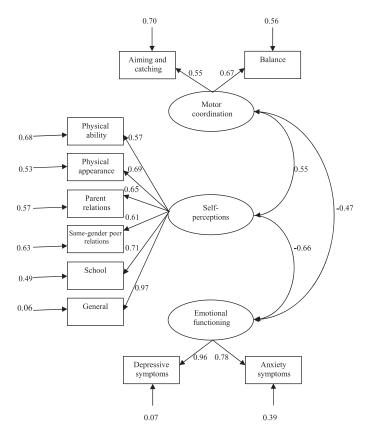


FIGURE 1 The measurement model with correlations, factor loadings, and measurement errors.

 
 TABLE 3
 Summary of Relevant Model Fit Indices for the Measurement Model and the Structural Models of the Relationship Between Motor Coordination, Self-Perceptions, and Emotional Functioning

Model	$\chi^2/df$	Comparative Fit Index	Standardized Root Mean Square Residual	Root Mean Square Error of Approximation
Measurement model Structural model	95.58/32 = 2.99	0.92	0.087	0.15 (90% CI: 0.110- 0.180)
Saturated Mediator	86.84/32 = 2.71 88.05/33 = 2.58	0.92 0.92	0.087 0.089	0.15 (90% CI: 0.120- 0.180) 0.15 (90% CI: 0.110- 0.180)

 $\chi^2$  difference = -1.21, P = .271. Cl, confidence interval.

direct impact on emotional functioning; rather, it affected emotional functioning through self-perceived competence in various domains. Few studies have attempted to understand the mechanisms through which motor coordination may be related to internalizing domains<sup>3,38</sup>; therefore, the current study is important in adding to existing research.

One study found that child and adolescent twins with DCD demonstrated significantly higher levels of depressive symptoms compared with their monozygotic co-twins without DCD, suggesting that the findings may be attributed to the effects of unique environmental factors.<sup>2</sup> Cairney et al<sup>5</sup> name the environmental stress hypothesis as a plausible explanation for the relationship between motor coordination and internalizing problems. Specifically, as children with coordination difficulties are exposed to the cascade of negative psychosocial consequences,<sup>39</sup> this in turn leads to negative self-appraisals, which in turn, may lead to anxiety and/or depression.<sup>5</sup> Consequently, negative self-perceptions may play a crucial role in understanding the nature of the relationship between coordination deficits and emotional difficulties. The current study provides support for this idea. First, the current study supports previous research linking selfperceived competence with emotional outcomes such as depression and anxiety.40,41 Furthermore, the results also support previous findings on DCD,<sup>3,28</sup> as significant positive correlations were found for the MABC-2 motor components and SDQ-II self-perceived physical ability, physical appearance, general school, and same-gender peer relations subscales.

Few longitudinal studies have identified the relationship between early motor difficulties and later anxiety and depressive symptoms at school-age and adolescence.13,42,43 Shaffer et al43 found that the relationship between motor development at 7 years of age and later anxiety difficulties at age 17 years remained even when there was no evidence of anxiety at the early age. Consequently, it is plausible that the relationship may be better explained through environmental rather than biological factors. The current study provides evidence that the relationship between motor skills and internalizing symptoms in adolescents may be understood in terms of a mechanism whereby motor coordination has an indirect impact on emotional functioning via self-perceptions. However, it is important to note that given the crosssectional nature of the current study, possible biological factors underlying the relationship cannot be ruled out. For example, cerebellar dysfunction has been associated with both motor coordination<sup>44</sup> and emotional regulation.<sup>45</sup> In fact, suggestion of a possible

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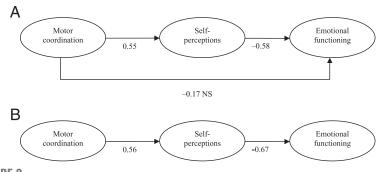


FIGURE 2 Path coefficients. A, the saturated structural model; B, the nested mediator model.

common neurologic cause stems from research demonstrating a specific link between balance problems and anxiety disorders in children.<sup>46</sup> Interestingly, aiming and catching and balance were the only MABC-2 components related to the construct of emotional functioning, which may partly support previous literature linking anxiety and balance problems.<sup>46</sup>

The manual dexterity subscale, a measure of fine motor ability, failed to demonstrate significant correlations with the outcome measures of depressive and anxious symptoms. It is possible that some individuals may have improved aspects of their motor skills (eg, fine motor) as a result of intervention in childhood or acquired some skills from prolonged practice.<sup>47</sup> Consequently, these areas may not have a significant impact on emotional functioning in adolescence.

Kirby et al<sup>47</sup> noted how at older ages, individuals may have learned coping mechanisms such as adapting or avoiding situations or specific tasks. The tendency of individuals with motor coordination difficulties to avoid participating in sporting domains that often require gross motor skills, such as ball throwing,<sup>48</sup> may have an important role in understanding the relationships found in this study, particularly given the associated risk for social isolation and the increased need for belongingness during adolescence. Research has also highlighted the positive impact of sports participation on the emotional well-being of adolescents.49 Furthermore, it has been noted that in Western society, sporting competence and the ability to play games with friends are highly regarded,<sup>39</sup> which may also be important when considering the relationship between gross motor ability and emotional functioning in the current study.

#### **CONCLUSIONS**

The results of this study suggest that the relationship between motor coordination and emotional functioning in an adolescent sample may be best understood in terms of a mediational association, although this crosssectional study cannot imply causality. Furthermore, although the current study aimed to control for possible confounding factors such as ADHD symptoms and verbal ability, other associated problems (such as language difficulties<sup>50,51</sup>) may also have a role in the relationship between motor coordination and emotional outcomes. The current findings, however, highlight the importance of assessing emotional outcomes in individuals who present with motor difficulties. Similarly, assessment of motor coordination in those referred for emotional problems is important given research showing a high rate of motor problems in children (particularly, boys) diagnosed with an anxiety disorder.<sup>4</sup> In addition, it may be important to assess the individual's self-perceived competencies. For example, if an adolescent also presents with self-perceived difficulties in the peer domain, a treatment plan aimed to promote social competency may also work to reduce or avoid possible emotional difficulties. Future research is needed to further elucidate the nature of the relationship between motor coordination deficits and emotional outcomes.

#### **ACKNOWLEDGMENTS**

We are very grateful to the parents and adolescents who were willing to participate in this study. We also thank Sean Piek, Linda Pannekoek, and Eva Kuhry for their assistance with data entry.

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PAPER 3

Rigoli, D., Piek, J. P., Kane, R., & Oosterlaan, J. (2012). An examination of the relationship between motor coordination and executive functions in adolescents. *Developmental Medicine and Child Neurology*, *54*(11), 1025-1031.

## An examination of the relationship between motor coordination and executive functions in adolescents

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#### This article is commented on by Michel on page 971 of this issue.

#### PUBLICATION DATA

Accepted for publication 29th May 2012. Published online 31st July 2012.

#### ABBREVIATIONS

DCD	Developmental coordination disorder
MABC-2	Movement Assessment Battery
	for Children-2
SES	Socio-economic status
SWAN	Strengths and Weaknesses of
	ADHD Symptoms and Normal
	Behaviour
VCI	Verbal Comprehension Index
WISC-IV	Wechsler Intelligence Scale for
	Children-IV
WMI	Working Memory Index

**AIM** Research suggests important links between motor coordination and executive functions. The current study examined whether motor coordination predicts working memory, inhibition, and switching performance, extending previous research by accounting for attention-deficit– hyperactivity disorder (ADHD) symptomatology and other confounding factors, in an adolescent normative sample.

**METHOD** Ninety-three adolescents (38 females, 55 males) aged 12 to 16 years (mean age 4y 2mo, SD 1y 1mo) were assessed on the Movement Assessment Battery for Children-2 (MABC-2), Wechsler Intelligence Scale for Children-IV, N-back task, the inhibition subtest from the NEPSY-II: A Developmental Neuropsychological Assessment, second edition, and the parent-rated Strengths and Weaknesses of ADHD Symptoms and Normal Behaviour Questionnaire.

**RESULTS** The MABC-2 total score accounted for a significant proportion of the variance in visuospatial working memory (p=0.041) but not for verbal working memory. The MABC-2 aiming and catching component, however, was found to account for unique variance in both verbal (p=0.019) and visuospatial working memory (p=0.016). The MABC-2 total score was found to account for a significant proportion of the variance in inhibition total completion time (p=0.017). Finally, balance skills accounted for unique variance in a NEPSY-II inhibition total errors variable (p=0.020). **INTERPRETATION** The results provide support for an overlap between motor coordination and executive functions, which has important practical implications. The study also suggests shared mechanisms underpinning the relationship between these areas, including possible cerebellar involvement.

It has been noted that motor control involves cognitive processes such as inhibiting frequently used movements, anticipating and updating aspects of the task to allow forward planning, resisting interference due to automatic postural control and fatigue, and the monitoring and correction of incorrect movements.<sup>1</sup> However, although there is some suggestion that complex cognitive processes (i.e. executive functions) affect motor performance, causal evidence regarding the direction of the relationship is limited.

The notion that motor development may predict cognitive functioning is partly supported by research highlighting that it is the sensory and motor functioning regions of the brain that are typically the first to mature.<sup>2</sup> Furthermore, longitudinal studies have found that early motor development predicts later performance on complex cognitive tasks, including working memory.<sup>3</sup> Conversely, in a study of preschool children, Niederer et al.<sup>4</sup> found that baseline memory was not associated with an improvement in motor skills 9 months later.

Diamond<sup>5</sup> argued that the close association between motor and cognitive development is mediated by the coactivation of the cerebellum and the prefrontal cortex. It is also important to note the role of individual differences when understanding this relationship.<sup>6</sup> For example, there are a number of studies suggesting that physical activity and high levels of aerobic fitness during childhood may enhance neurocognition.<sup>7</sup> This provides further evidence that motor coordination may predict executive functions.

Evidence for the relationship between motor performance and executive functions also exists from behavioural studies. Normative studies<sup>1</sup> as well as those examining developmental coordination disorder (DCD)<sup>8</sup> have demonstrated a link between motor coordination and working memory. Baddeley's<sup>9</sup> model of working memory comprises separable components for the temporary storage of verbal (i.e. the phonological loop) and visuospatial (i.e. visuospatial sketchpad) information, and research in the area of DCD has found that motor coordination may be more closely linked to visuospatial working memory than to verbal working memory.<sup>8</sup> This may be partly understood in terms of the visuospatial processing deficit found in individuals with DCD.<sup>10</sup> Regarding other executive function domains, studies have found that children with coordination difficulties are slower in performing inhibition and attention shifting tasks but are not less accurate than typically developing children.<sup>6</sup> It is possible that this reflects an automatization deficit in children with motor impairments, suggesting that cerebellar mechanisms may be implicated in the slower performances on these tasks.

The available literature on the relationship between motor functioning and executive functions leaves a number of issues needing to be addressed. First, it is important to control for attention and/or hyperactivity-impulsivity (attention-deficithyperactivity disorder [ADHD]) symptomatology as ADHD has been linked with motor problems<sup>11</sup> as well as executive function areas such as working memory and inhibition.<sup>12</sup> Very few studies have employed normative samples of children. Normative studies are important given methodological problems associated with clinical samples such as overestimating associations between domains.<sup>1</sup> In addition, as there is evidence from normative samples<sup>1</sup> and studies examining motor impairment<sup>6</sup> that specific components of motor coordination have a relationship with certain aspects of executive function, whereas others do not, it is important to examine these components separately. Furthermore, research is needed in adolescent samples given that previous studies have involved vounger children or a mixed sample of children and adolescents.13,14

The current study examined the relationship between motor coordination (namely overall motor performance, manual dexterity, aiming and catching, and balance) and executive functions (namely working memory, response inhibition, and switching) in an adolescent normative sample, whilst controlling for ADHD symptomatology, age, gender, socioeconomic status (SES), and verbal ability. It is hypothesized that motor coordination will show a significant relationship with working memory, and this may be stronger for visuospatial working memory than for verbal working memory.<sup>8</sup> It is also hypothesized that a significant relationship will be found between motor coordination and the timing measures from the response inhibition and switching tasks, but not with motor coordination and the accuracy variable.

#### METHOD

#### Participants

Recruitment occurred across five randomly selected secondary schools and through public advertisements (e.g. community newspapers). Adolescents aged 12 to 16 years were eligible for inclusion and had a minimum Verbal Comprehension Index (VCI) of 80 as measured by the Wechsler Intelligence Scale for Children-IV (WISC-IV), in order to exclude any adolescent whose difficulties might be attributed to general delayed development.<sup>15</sup> Furthermore, a parent-rated developmental history questionnaire was used to ascertain the absence of physical disability, chronic illness, pervasive developmental disorder, and neurological disorder. Ninety-four participants responded and consented to the project; however, one participant with undiagnosed hand tremor was excluded. The final sample included 93 adolescents (38 females and 55 males) with

#### What this paper adds

- The results show that motor coordination may be more closely linked to visuospatial working memory than to verbal working memory.
- `Aiming and catching' skills may be linked to both verbal and visuospatial working memory.
- Motor coordination is related to performance speed on inhibition tasks.
- Balancing skills are related to interference control.

a mean age of 14 years 2 months (SD 1y 1mo). The Australian Prestige Scale<sup>16</sup> was used to provide SES scores. The scale assesses the prestige of occupations, with scores ranging from 1 (reflecting high prestige) to 6.9 (reflecting low prestige). For the current study, the occupation rated as most prestigious out of mother's and father's occupation was used as the SES score (mean 3.8, SD 1.0, range 1.8–6.6).

#### Measures

#### Movement Assessment Battery for Children-2 (MABC-2)

The MABC-2 is a standardized test used for the identification and description of children with movement difficulties.<sup>17</sup> Age-based standard scores for manual dexterity, aiming and catching, and balance components and a total test score are provided (mean 10, SD 3), with higher scores demonstrating better performance. A total test score at or below the 5th centile indicates significant movement difficulty, whereas a score between the 5th and 15th centile indicates that a child is 'at risk'.

Henderson et al.<sup>17</sup> provide evidence suggesting favourable psychometric properties for the MABC-2. A reliability coefficient of 0.80 for the total test score and coefficients ranging from 0.73 to 0.84 for the individual component scores are reported.<sup>17</sup>

#### Wechsler Intelligence Scale for Children-IV – Australian

The WISC-IV is a measure of cognitive ability for children aged 6 years to 16 years 11 months.<sup>18</sup> The 10 core subtests yield a full-scale IQ and four indices of verbal comprehension (i.e. VCI), perceptual reasoning, working memory (i.e. WMI), and processing speed. For the current study, the VCI was used as a potential control variable and to exclude any adolescent whose difficulties might be attributed to general delayed development. The WMI was employed as measure of verbal working memory. The WISC-IV is widely used and has excellent internal consistency, test–retest reliability, criterion validity, and construct validity.<sup>18</sup>

#### N-back task

The N-back task was used to assess visuospatial working memory, designed after Gevins and Cutillo<sup>19</sup> and Jansma et al.<sup>20</sup> The task has also been adapted to make it more attractive and appropriate for children.<sup>21</sup> An apple with four holes from which a caterpillar appears is presented on the computer screen. Respondents are required to press one of the four buttons that corresponds spatially with the hole from which the caterpillar emerged. There are four conditions of graded difficulty requiring the respondent to indicate where the caterpillar was one move back, two moves back, three moves back, or four moves back. Each condition comprises a practice block

(10 trials) and an experimental block in which performance is measured (32 trials). Respondents move to the next level of difficulty only if they score a minimum of eight correct responses (indicating performance above chance levels) in the experimental blocks. For this study, task performance is measured by the total number of correct responses across the conditions (maximum raw score of 128). The N-back task is a widely used measure of working memory, and in a study involving a sample of adolescents test–retest reliabilities of 0.70 and 0.66 were reported for 3- and 4-back, respectively.<sup>21</sup>

#### NEPSY-II: a developmental neuropsychological assessment

The NEPSY-II provides a comprehensive neuropsychological assessment for children and adolescents aged 3 to 16 years.<sup>22</sup> The 'naming', 'inhibition', and 'switching' sections of the inhibition subtest were administered for the purposes of this study. These sections assess, respectively, simple naming skills, the ability to inhibit automatic responses in favour of novel responses, and the ability to switch between response types. The age-standardized total completion time scaled score for the inhibition and switching sections were utilized for this study, with higher scores representing faster completion times. A total errors scaled score was also used, which combines errors across all sections in the inhibition subtest (namely naming, inhibition and switching sections). A higher total errors scaled score corresponds to better performance (i.e. fewer errors made).

The inhibition subtest has shown adequate to high internal consistency, for example average reliability coefficients for the inhibition and switching combined scaled scores range from 0.73 to 0.87 for ages 12 to 16 years.<sup>22</sup> The NEPSY-II also demonstrates adequate stability across time, as well as good content, construct, and criterion-related validity.<sup>22</sup>

## Strengths and Weaknesses of ADHD Symptoms and Normal Behaviour (SWAN)

The parent-rated SWAN scale includes 18 items based on ADHD symptoms listed in DSM-IV.<sup>23</sup> Parents are asked to rate the items based on observations from the last month and with reference to age-matched peers. Scores for each item range from +3 (i.e. 'far below average') to -3 (i.e. 'far above average'). For the current study, attention and hyperactivity/impulsivity scores were calculated by averaging the total of the nine corresponding items, with positive scores indicating presence of symptoms and negative scores indicating absence of symptoms.

The SWAN scale has been found to yield a normal distribution of scores, making it useful for examining variability in (hyper)activity and (in)attention in the general population.<sup>24</sup> Martin et al.<sup>25</sup> found the prevalence rate of ADHD, as assessed using the SWAN scale, to be similar to what has been reported in previous studies.

#### Procedure

The Curtin University Human Research Ethics Committee granted approval for the project and National Health and Medical Research Council of Australia ethical guidelines were followed. Approval was also granted from the participating schools' representative bodies and, subsequently, from interested school principals in Perth, Western Australia. Adolescents and their parents provided written consent and were then individually tested by a trained examiner at home or at the university, depending on family preference. Testing duration was 4.5 hours over two sessions. Measures were administered in a standard manner. Parents completed questionnaires including a developmental history questionnaire and the SWAN scale.

#### Statistical analysis

A series of hierarchical regressions were conducted to determine whether the MABC-2 total score or its component scores (manual dexterity, aiming and catching, and balance) accounted for incremental variance in working memory (N-back accuracy, WISC-IV WMI), inhibition (NEPSY-II inhibition total completion time scaled score), switching (NE-PSY-II switching total completion time scaled score), and the NEPSY-II total errors scaled score, after controlling for covariates (WISC-IV VCI, SWAN attention and hyperactivity/impulsivity symptoms). In a hierarchical regression analysis,  $\Delta R^2$  represents the increase in the proportion of variance in the criterion variable explained from step N-1 to step N. The term  $sr^2$  represents the unique amount of variance that a predictor brings to the model. In a hierarchical regression analysis where just one predictor is added at step N, then the  $\Delta R^2$  from step N-1 to step N will be equivalent to the sr<sup>2</sup> for the added predictor.

The most complex regression model included three control variables and three primary predictors. Our sample size of 93 was sufficient to detect moderate relationships (i.e.  $f^2=0.12$ ) between the criterion variables and the primary predictors.<sup>26</sup>

#### RESULTS

#### Descriptives

Table I shows the means, standard deviations, and ranges for the study variables. Five adolescents scored at or below the 5th centile on the MABC-2 total score, indicating significant movement difficulty. The prevalence of significant movement difficulty was 5.4%, which is similar to previous estimates of 6%.<sup>27</sup> Two adolescents scored between the 6th and 15th centiles, suggesting that they were 'at risk' of movement difficulty.

#### **Bivariate correlations**

The correlations between the criterion variables, predictors, and control variables are shown in Table II.

#### Multiple linear regression analyses

As expected, there were strong correlations between the MABC-2 total score and each of its component scores (see Table II). Because the MABC-2 total score was a reliable predictor of the component scores, it was included as a proxy for the component scores in the primary analysis, thereby reducing the complexity of the regression model and optimizing statistical power. Because there was no correlation between the

Table I: Means, SDs,	and range of scores	for the study variables
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	Mean	SD	Range
MABC-2 total score <sup>a</sup>	10.63	2.56	3.0–16.0
MABC-2 manual dexterity <sup>a</sup>	9.57	2.47	3.0-15.0
MABC-2 aiming and catching <sup>a</sup>	11.03	2.73	4.0-16.0
MABC-2 balance <sup>a</sup>	11.42	2.98	4.0-14.0
WISC-IV Working Memory Index <sup>a</sup>	103.75	12.47	59.0-141.0
N-back accuracy <sup>b,c</sup>	88.17	19.69	6.0-124.0
NEPSY-II inhibition completion time <sup>a</sup>	10.68	2.9	4.0-19.0
NEPSY-II switching completion time <sup>a</sup>	10.64	2.42	3.0-16.0
NEPSY-II total errors <sup>a</sup>	8.88	3.19	1.0–16.0
SWAN attention <sup>b,d</sup>	-0.83	1.17	-3.0 to 2.33
SWAN hyperactivity/impulsivity <sup>b,d</sup>	-1.17	1.03	-3.0 to 1.11
WISC-IV Verbal Comprehension Index <sup>a</sup>	106.63	11.25	81.0–132.0
SES <sup>b,e</sup>	3.77	1.00	1.80–6.60

<sup>a</sup>Age-standardized score. <sup>b</sup>Raw score. <sup>c</sup>Total number of correct responses. <sup>d</sup>Scores are calculated by averaging the total of the nine attention or hyperactivity/impulsivity items. <sup>e</sup>The occupation rated as most prestigious out of mother's and father's occupations. MABC-2, Movement Assessment Battery for Children-2; WISC-IV, Wechsler Intelligence Scale for Children-IV; SWAN, Strengths and Weaknesses of ADHD Symptoms and Normal Behaviour; SES, socioeconomic status.

MABC-2 aiming and catching and manual dexterity components (*r*=0.071), individuals with the same total score can be fundamentally different at the component level. It was important to conduct secondary regression analyses that replaced MABC-2 total score with its component scores. Only those outcomes that were significantly associated with the MABC-2 score total, or at least one component score, were analysed. The VCI was entered first, followed by SWAN attention and hyperactivity/impulsivity (Table II indicates that these were the only covariates), and then the MABC-2 total score or its component scores.

#### Working memory

After controlling for the three covariates, the MABC-2 total score explained a significant 4.2% of the variance in N-back accuracy ( $\Delta R^2$ =0.042; *p*=0.041). When the MABC-2 total score was replaced by its component scores, however, the combined scores explained no additional variance over and above that already explained by the covariates ( $\Delta R^2$ =0.069; *p*=0.077), although aiming and catching uniquely explained a significant 5.8% of the variance in N-back accuracy (*sr*<sup>2</sup>=0.058; *p*=0.016).

After controlling for the three covariates, the MABC-2 total score explained no additional variance ( $\Delta R^2$ =0.011; *p*=0.272) in WISC-IV WMI performance. Similarly, when the MABC-2 total score was replaced by its component scores, the combined scores explained no additional variance over and above that already explained by the covariates ( $\Delta R^2$ =0.050; *p*=0.123), although aiming and catching uniquely explained a significant 4.8% of the variance in WMI performance (*sr*<sup>2</sup>=0.048; *p*=0.019), and VCI uniquely explained 12.2% of the variance (*sr*<sup>2</sup>=0.122; *p*<0.001). Table III summarizes the regression results for the N-back and WISC-IV WMI tasks.

#### Inhibition and switching

After controlling for two of the three covariates (VCI was not correlated with inhibition completion time), the MABC-2 total score explained a significant 6.1% of the variance in inhibition completion time ( $\Delta R^2$ =0.061; *p*=0.017). When the MABC-2 total score was replaced by its component scores, however, the combined scores explained no additional variance over and above that already explained by covariates ( $\Delta R^2$ =0.063; *p*=0.120).

After controlling for covariates, the MABC-2 total score explained no additional variance in switching completion time ( $\Delta R^2$ =0.023; p=0.128). Similarly, when the MABC-2 total score

	MABC-2 total score	MABC-2 manual dexterity	MABC-2 aiming and catching	MABC-2 balance	SWAN attention	SWAN hyperactivity/ impulsivity	VCI	Age	Sex	SES
MABC-2 total score										
MABC-2 manual dexterity	0.657 <sup>a</sup>									
MABC-2 aiming and catching	0.656 <sup>a</sup>	0.071								
MABC-2 balance	0.780 <sup>a</sup>	0.264 <sup>b</sup>	0.423 <sup>a</sup>							
SWAN attention	-0.178	–0.252 <sup>b</sup>	-0.052	-0.106						
SWAN hyperactivity/impulsivity	0.020	-0.070	0.093	0.007	0.724 <sup>a</sup>					
VCI	0.152	0.075	0.048	0.155	-0.382 <sup>a</sup>	–0.209 <sup>b</sup>				
Age	-0.114	-0.069	-0.066	-0.095	0.077	-0.036	-0.167			
Sex	-0.069	0.235 <sup>b</sup>	–0.397 <sup>a</sup>	-0.007	–0.251 <sup>a</sup>	–0.184 <sup>b</sup>	-0.018	0.021		
SES	-0.074	-0.122	0.032	-0.099	0.192	0.000	-0.384 <sup>a</sup>	0.244 <sup>b</sup>	-0.083	
WISC-IV WMI	0.201	0.113	0.251 <sup>b</sup>	0.112	-0.298 <sup>a</sup>	-0.135	0.442 <sup>a</sup>	-0.089	-0.020	-0.174
N-back accuracy	0.271 <sup>a</sup>	0.129	0.281ª	0.146	–0.242 <sup>b</sup>	-0.113	0.253 <sup>b</sup>	0.150	-0.051	-0.039
NEPSY-II inhibition completion time	0.276 <sup>a</sup>	0.229 <sup>b</sup>	0.176	0.163	-0.077	0.027	0.142	0.008	-0.092	0.061
NEPSY-II switching completion time	0.237 <sup>b</sup>	0.189	0.215 <sup>b</sup>	0.101	-0.182	0.025	0.219 <sup>b</sup>	-0.158	-0.102	-0.017
NEPSY-II total errors	0.190	0.096	0.036	0.259 <sup>b</sup>	-0.325 <sup>a</sup>	–0.229 <sup>b</sup>	0.222 <sup>b</sup>	-0.076	0.163	0.007

<sup>a</sup>p<0.01 (two-tailed). <sup>b</sup>p<0.05 (two-tailed).

MABC-2, Movement Assessment Battery for Children-2; SWAN, Strengths and Weaknesses of ADHD Symptoms and Normal Behaviour; VCI, Verbal Comprehension Index; SES, socio-economic status; WISC-IV, Wechsler Intelligence Scale for Children-IV; WMI, Working Memory Index.

Table III: Step 3 statistics for hierarchical multiple regression analyses predicting working memory performance from MABC-2 total score or component scores (*n*=93)

				Working mem	nory outcome	s		
		W           B         95% Cl           0.41         0.18, 0.63           -2.13         -5.27, 1.01           1.03         -2.31, 4.36           0.52         -0.42, 1.46           0.43         0.20, 0.65           -1.82         -4.99, 1.35           0.61         -2.71, 3.93           0.23         -0.77, 1.23           1.12         0.19, 2.06           -0.30         -1.18, 0.58				N-ba	ck	
	В	95% CI	sr <sup>2</sup>	<i>p</i> -value	В	95% CI	sr <sup>2</sup>	<i>p</i> -value
Model 1 predictors								
VCI	0.41	0.18, 0.63	0.114	0.001 <sup>a</sup>	0.29	-0.09, 0.66	0.023	0.134
Attention	-2.13	-5.27, 1.01	0.016	0.181	-2.86	-8.13, 2.40	0.011	0.282
Hyperactivity/impulsivity	1.03	-2.31, 4.36	0.003	0.543	0.76	-4.83, 6.36	0.001	0.787
MABC-2 total score	0.52	-0.42, 1.46	0.011	0.272	1.65	0.07, 3.22	0.042	0.041 <sup>b</sup>
Total R <sup>2</sup>			0.197	< 0.001 <sup>c</sup>			0.097	0.011 <sup>b</sup>
Model 2 predictors								
VCI	0.43	0.20, 0.65	0.122	< 0.001 <sup>c</sup>	0.32	-0.06, 0.70	0.028	0.094
Attention	-1.82	-4.99, 1.35	0.011	0.257	-2.67	-8.04, 2.71	0.009	0.327
Hyperactivity/impulsivity	0.61	-2.71, 3.93	0.001	0.717	0.37	-5.25, 5.99	0.000	0.895
Manual dexterity	0.23	-0.77, 1.23	0.002	0.649	0.52	-1.17, 2.22	0.004	0.540
Aiming and catching	1.12	0.19, 2.06	0.049	0.019 <sup>b</sup>	1.95	0.37, 3.54	0.058	0.016 <sup>b</sup>
Balance	-0.30	-1.18, 0.58	0.004	0.502	-0.20	-1.70, 1.29	0.001	0.788
Total R <sup>2</sup>			0.221	< 0.001 <sup>c</sup>			0.104	0.016 <sup>b</sup>

<sup>a</sup><0.01. <sup>b</sup><0.05. <sup>c</sup><0.001.

WMI, Working Memory Index; B, unstandardized regression coefficient; CI, confidence interval; *sr*<sup>2</sup>, the part correlation squared, VCI, Verbal Comprehension Index; MABC-2, Movement Assessment Battery for Children-2.

**Table IV:** Step 3 statistics for hierarchical multiple regression analyses predicting inhibition completion time, switching completion time, and total errors from MABC-2 total score or component scores (*n*=93)

						Executive fu	nctionin	g				
		Inhibit	time			Switch	time			Total e	rrors	
	В	95% CI	sr <sup>2</sup>	<i>p</i> -value	В	95% CI	sr <sup>2</sup>	<i>p</i> -value	В	95% CI	sr <sup>2</sup>	<i>p</i> -value
Model 3 predictors												
VCI	-	-	-	-	0.03	-0.02, 0.08	0.016	0.205	0.03	-0.03, 0.09	0.009	0.340
Attention	-0.24	-0.99, 0.51	0.004	0.521	-0.60	-1.25, 0.05	0.033	0.070	-0.62	-1.48, 0.23	0.021	0.148
Hyperactivity/impulsivity	0.26	-0.58, 1.10	0.004	0.537	0.61	-0.08, 1.30	0.030	0.082	-0.13	-1.04, 0.77	0.001	0.769
MABC-2 total score	0.29	0.05, 0.53	0.061	0.017 <sup>a</sup>	0.15	-0.04, 0.35	0.023	0.128	-0.17	-0.09, 0.42	0.016	0.199
Total R <sup>2</sup>			0.050	0.056			0.088	0.016 <sup>a</sup>			0.094	0.013 <sup>a</sup>
Model 4 predictors												
VCI	-	-	-	_	0.04	-0.01, 0.08	0.022	0.141	0.02	-0.04, 0.09	0.005	0.457
Attention	-0.19	-0.96, 0.59	0.002	0.630	-0.53	-1.19, 0.14	0.024	0.121	-0.75	-1.62, 0.12	0.028	0.089
Hyperactivity/impulsivity	0.23	-0.62, 1.09	0.003	0.588	0.55	-0.15, 1.25	0.024	0.120	-0.03	-0.94, 0.87	0.000	0.947
Manual dexterity	0.23	-0.03, 0.49	0.032	0.084	0.13	-0.08, 0.34	0.015	0.218	-0.06	-0.33, 0.22	0.002	0.676
Aiming and catching	0.14	-0.10, 0.38	0.014	0.259	0.17	-0.03, 0.37	0.029	0.090	-0.11	-0.36, 0.15	0.006	0.409
Balance	0.05	-0.18, 0.27	0.002	0.683	-0.06	-0.24, 0.13	0.004	0.549	0.29	0.05, 0.53	0.054	0.020 <sup>a</sup>
Total R <sup>2</sup>			0.031	0.173			0.088	0.029 <sup>a</sup>			0.114	0.011 <sup>a</sup>

B, unstandardized regression coefficient; Cl, confidence interval; *sr*<sup>2</sup>, the part correlation squared, VCl, Verbal Comprehension Index; MABC-2, Movement Assessment Battery for Children-2.

<sup>a</sup><0.05.

was replaced by its component scores, the combined scores explained no additional variance over and above that already explained by the covariates ( $\Delta R^2$ =0.043; *p*=0.236).

p=0.020). Table IV summarizes the regression results for the inhibition and switching tasks.

After controlling for covariates, the MABC-2 total score explained no additional variance in the total errors score ( $\Delta R^2$ =0.016; *p*=0.199). Similarly, when the MABC-2 total score was replaced by its component scores, the combined scores explained no additional variance over and above that already explained by the covariates ( $\Delta R^2$ =0.055; *p*=0.137), although the MABC-2 balance component uniquely explained a significant 5.4% of the variance in total errors (*sr*<sup>2</sup>=0.054;

#### DISCUSSION

In the current study, the MABC-2 total score accounted for a significant proportion of the variance in a visuospatial working memory task but not in verbal working memory. These results suggest that motor coordination may be more closely related to visuospatial working memory than to verbal working memory, supporting previous findings of a specific deficit in visuospatial memory in children with DCD.<sup>8</sup>

However, aiming and catching (but not manual dexterity or balance) accounted for statistically significant unique variance in both visuospatial and verbal working memory. This supports previous research revealing specific relationships between working memory and certain aspects of motor coordination. For example, Piek and colleagues<sup>3</sup> found a relationship between early gross motor (but not fine motor) development and later working memory ability in a normative sample of school-aged children. Although speculative in nature, it is possible that the specific association found in the current study may be partly explained by shared underlying cerebellar processes. The lateral zone of the cerebellum is important for the rapid, aimed movements required in aiming and catching tasks.<sup>28</sup> Research has also implicated the cerebellum in working memory.<sup>29</sup> Furthermore, Diamond<sup>5</sup> highlighted the close coactivation of the cerebellum and prefrontal cortex when understanding the relationship between complex motor and cognitive domains. Consequently, it is also possible that the complex nature of ball skills assessed in the current study coactivates greater prefrontal cortex activity than the tasks solely assessing manual dexterity or balance skills.

Research has also demonstrated how individuals with motor difficulties tend to avoid participation in sporting domains.<sup>30</sup> Given the reported cognitive benefits of physical activity through physiological (e.g. increased cerebral blood flow) and learning/developmental mechanisms,<sup>7</sup> it is possible that the lack of opportunity to learn and practise the skills associated with aiming and catching games may play an important role in understanding the link found in the current study.

The MABC-2 total score was also found to account for a significant proportion of variance in inhibition completion time, supporting previous research in normative and motor impairment samples.<sup>6,14</sup> The slower performance speed on inhibition tasks for children with DCD may be understood in terms of an automatization deficit, most likely linked to cerebellar dysfunction.<sup>6</sup> Ouerne and colleagues<sup>31</sup> also demonstrated slower responses for children with DCD and showed that children with DCD demonstrated abnormal hemispheric lateralization for attentional and inhibitory functions. It is also important to note that the current findings of a significant association between motor coordination and inhibition completion time may simply reflect the involvement of speed in both tasks, as, upon inspection of the MABC-2 components, manual dexterity (comprising two timed tasks) appeared to be the strongest predictor in explaining this relationship. However, Michel and colleagues,<sup>6</sup> who reported slower performance in children with motor impairment, argued that their results were unlikely to be due to differences in information processing speed, as the children with motor impairment did not perform more slowly in a simple reaction time task. Rather, it was suggested that the slower performance of children with motor impairments was due to the complex demands of the task.<sup>6</sup>

In the current study, a non-significant relationship between motor coordination and switching completion time was found. This may suggest possible differences in the neural processes underlying inhibition and switching. Switching is a complex task requiring various cognitive processes in addition to the inhibitory demands inherent in the task.<sup>22</sup> Thus, the switching task may require the recruitment of additional prefrontal regions and/or may be mediated by different cortical areas. In fact, functional magnetic resonance imaging studies have shown that localization within the frontal cortex is task dependent.<sup>32</sup> This may explain the divergent findings between inhibition and switching in the current study.

An unexpected result in the current study was the specific link found between balancing ability and total errors (a composite score including inhibition and switching errors). This does not support previous research linking motor coordination to only the timing components of such tasks. However, it is important to note that these previous studies involved a composite score of movement ability<sup>14</sup> or a group of children defined by fine motor difficulties.<sup>6</sup>

The link between balance and total errors supports accumulating evidence for the attentional requirements of young and older children during postural tasks.<sup>33</sup> Woollacott and Shumway-Cook<sup>34</sup> argued that postural control requires significant attentional resources depending on the complexity of the postural task and the individual's age and balance abilities. The NEPSY-II inhibition subtest used in the current study is based on the Stroop paradigm and, thus, examines interference control (i.e. the ability to ignore irrelevant information). Therefore, this suggests that interference control may be important when understanding balancing ability.

The current study has some limitations. The study cannot provide information on the directional relationship between the motor and cognitive domains. There is some evidence to suggest that motor development may predict cognitive performance; however, further longitudinal research is needed.<sup>3,4</sup> In addition, performance accuracy was measured by a variable combining simple naming, inhibition, and switching errors, which introduces the problem of process specificity. It is also important, in attempting to interpret the results of the present study, to note that other variables may have played a role (e.g. processing speed, motivation). In addition, the digit-span forward component of the WISC-IV WMI and the 1-back level of the N-back task may be considered measures of storage rather than of the processing component of working memory. However, Unsworth and Engle<sup>35</sup> have suggested that shortterm memory and working memory tasks largely measure the same basic processes and therefore argue against the notion that short-term memory and working memory are different constructs.

Finally, although the current sample size was sufficient to detect important relationships, upon closer inspection of the predictors for inhibition completion time, manual dexterity appeared to be the strongest predictor, although this did not reach statistical significance (p=0.084). This suggests that future research could benefit from examining these relationships with a larger sample.

#### CONCLUSION

The results of this adolescent normative study suggest specific relationships between aspects of motor coordination and exec-

utive functions. It is possible that the specific relationships found in the current study (e.g. between aiming and catching skills and working memory) may be understood through shared neural mechanisms, namely, cerebellar processes.

The current results have practical implications when considering interventions for motor and/or executive functioning difficulties. For example, the current study highlights the importance of assessing executive functions in individuals who present with motor difficulties and, subsequently, tailoring the

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intervention accordingly. Similarly, it may also be important to screen for motor difficulties in those who present with executive function problems.

#### **ACKNOWLEDGEMENTS**

We are very grateful to the parents and adolescents who were willing to participate in this study. We also thank Sean Piek, Linda Pannekoek, and Eva Kuhry for their assistance with data entry.

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PAPER 4

Rigoli, D., Piek, J. P., Kane, R., & Oosterlaan, J. (2012). Motor coordination, working memory, and academic achievement in a normative adolescent sample: Testing a mediation model. *Archives of Clinical Neuropsychology*. Advance online publication.



## Motor coordination, working memory, and academic achievement in a normative adolescent sample: Testing a mediation model

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Accepted 11 June 2012

#### Abstract

The aim of the present study was to examine whether the relationship between motor coordination and academic achievement is mediated by working memory (WM) in a normative adolescent sample. Participants included 93 adolescents aged 12–16. The Movement Assessment Battery for Children-2 provided three indicators of motor coordination (Manual Dexterity, Aiming and Catching, and Balance), the WM Index of the Wechsler Intelligence Scale for Children-IV and the N-back paradigm provided two indicators of WM, and the Wechsler Individual Achievement Test-II provided three indicators of academic achievement (Word Reading, Spelling, and Numerical Operations). Structural equation modeling, controlling for verbal comprehension, attention deficit hyperactivity disorder symptoms, and socioeconomic status, suggested that the association between motor coordination and academic achievement may be best understood in terms of a mechanism whereby motor coordination (specifically, Aiming and Catching skills) has an indirect impact on academic outcomes via WM. These findings have important implications for the assessment and treatment of motor coordination and learning difficulties as well as in increasing the understanding of the possible neural mechanisms underpinning the relationship between these areas.

Keywords: Motor coordination; Working memory; Academic achievement; Adolescents; Normative sample

#### Introduction

There is extensive evidence linking motor coordination and learning outcomes. Research has shown that children with motor difficulties display significant problems in language, reading, spelling, and arithmetic (Alloway, 2007; Archibald & Alloway, 2008; Dewey, Kaplan, Crawford, & Wilson, 2002) and children with learning disabilities, such as dyslexia, have shown a high rate of motor difficulties (Fawcett & Nicholson, 1995). Furthermore, studies have found motor coordination in young children to be a unique, significant predictor of later achievement in reading and mathematics (Kurdek & Sinclair, 2001). Consequently, it has been argued that motor coordination may be crucial in identifying children at risk for academic underachievement (Son & Meisels, 2006), although the nature of this relationship remains unclear. Recent research, however, has suggested an important link between motor coordination, working memory (WM), and learning outcomes (Alloway, 2007).

WM refers to the ability to store and manipulate information over a brief period of time (Baddeley & Hitch, 1974). According to the widely used and accepted Baddeley (2000) model, WM comprises four components. The central executive controls resources and monitors information processing, as well as being responsible for various regulatory functions (Baddeley & Hitch, 1974). The central executive system is supported by separable components for the temporary storage of verbal (i.e., the phonological loop) and visuospatial (i.e., the visuospatial sketchpad) information. Finally, the episodic buffer is responsible for integrating information from the different components of WM and long-term memory (Baddeley, 2000). A substantial body of research now suggests that WM capacity is a reliable predictor of various cognitive skills

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such as general fluid intelligence (Engle, Tuholski, Laughlin, & Conway, 1999) as well as academic skills such as reading and mathematics (Alloway, 2009) and language comprehension (Nation, Adams, Bowyer-Crane, & Snowling, 1999).

Recently, WM has been linked to motor coordination. For example, Piek and colleagues (2004) found that after controlling for age, gender, and verbal IQ, motor coordination was significantly associated with WM in children aged 6–15. In this study, motor coordination was operationalized by a composite score comprising both fine (e.g., beads in a box and nut and bolt activities) and gross (e.g., balancing on one foot, jumping) motor tasks. Therefore, differential relationships between WM and certain aspects of motor coordination were not examined. In a later study (Piek, Dyck, Francis, & Conwell, 2007), children with developmental coordination disorder (DCD) were slower than attention deficit hyperactivity disorder (ADHD) and control groups on the same WM task used in Piek and colleagues' (2004) study, but also performed less accurately on another measure of WM. DCD group composition was not known in this study, that is, the proportion of children experiencing mainly fine motor or gross motor difficulties, or a combination of both.

Conversely, in a study that identified "motor impaired" children by using a cutoff below the 10th percentile on the Movement Assessment Battery for Children (MABC) Manual Dexterity subscale (consisting of tasks such as threading, drawing, posting coins in a box, and pegboard), it was found that these children did not perform worse on a WM task of Backwards Color Recall when compared with those without motor impairments (Michel, Roethlisberger, Neuenschwander, & Roebers, 2011). Furthermore, correlations revealed that although the WM task was correlated with Manual Dexterity performance in the motor-impaired group (even after controlling for intelligence), interestingly, this association was not apparent in the control group (Michel et al., 2011). These results suggest the possibility of specific relationships between WM and certain aspects of motor coordination. For example, Manual Dexterity may not have an important association with WM.

In a normative study investigating the relationship between different aspects of motor coordination and cognitive control in 7-year-old children, a significant association was found between Backwards Color Recall and postural flexibility, whereas no significant association was found between Backwards Color Recall and a fine motor pegboard task (Roebers & Kauer, 2009). Longitudinal research examining the predictive ability of motor skills on later WM has also revealed an important relationship between gross motor skills and WM. Piek and colleagues (2008) found a relationship between early gross motor (but not fine motor) development (assessed by the parent-rated ages and stages questionnaire from 4 months to 4 years of age and includes items such as "does your child usually pick up a small toy with only one hand?" and "does your child climb onto furniture?") and later school-aged WM ability. In another study, Murray and colleagues (2006) found early gross motor development (i.e., age of learning to stand without support) to be related to adult executive functioning, including WM. Similarly, in relation to the link between academic outcomes and certain aspects of motor coordination, Gaysina, Maughan, and Richards (2010) did not find any significant association between fine motor skills and academic difficulties in the reading domain at age 15.

Evidence suggesting important links between certain aspects of motor coordination and outcomes of cognitive functioning (namely, WM and academic achievement) provides support for specific neural mechanisms underlying these relationships. The pyramidal motor system provides a direct pathway for projections from the motor areas of the cortex to go to the muscles via the spinal cord (Piek, 2006). The corticospinal tract forms part of the pyramidal system and consists of axons of cortical neurons which are concentrated in the primary motor cortex of the frontal lobe (Carlson, 2010). Axons of the lateral corticospinal tract form synapses with motor neurons which control muscles of the distal limbs that move arms, hands, and fingers. Thus, the lateral corticospinal tract is said to be important for Manual Dexterity (Carlson, 2010). Conversely, the indirect pathway for projections from the motor areas of the cortex involves the structures of the extrapyramidal system such as the cerebellum (Piek, 2006). The cerebellum is crucial for motor control as it is associated with functions such as timing, motor learning, and regulation of muscle tone which are important for smooth and coordinated movement (Piek, 2006). Furthermore, certain parts of the cerebellum are said to be associated with specific aspects of motor control, for example, the vermis has been linked to postural reflexes (important for balance), whereas the lateral zone of the cerebellum has been linked to the control of independent limb movements particularly rapid, skilled movements (Carlson, 2010) such as Aiming and Catching skills.

Diamond (2000) highlighted the important role of the cerebellum (specifically, the lateral portion of the cerebellum, namely, the neocerebellum) not only in subserving motor function but also in cognitive functioning. Nicolson and colleagues (2001) propose a cerebellar deficit hypothesis when attempting to explain the reading and motor problems often seen in children with dyslexia. In addition to their observed motor deficits (e.g., balance and muscle tone problems), these children have also demonstrated difficulties with time estimation and skill automatization, pointing to a deficit of the cerebellum. Nicolson and colleagues gues also provide direct evidence for this theory through imaging studies. In fact, Rae and colleagues' (1998) study of metabolic abnormalities in developmental dyslexia provided evidence for lateral cerebellum involvement in dyslexic dysfunction.

In Nicolson and colleagues' model (2001), it is argued that the cerebellum contributes to cognitive processes that rely on internal speech, namely, verbal short-term memory or WM. According to Baddeley (2003), articulatory rehearsal mechanisms are important to retain verbal items in store. Nicolson and colleagues' cerebellar deficit hypothesis proposes that articulation

difficulties, resulting from the mild motor difficulties of cerebellar dysfunction, then lead to verbal short-term or WM difficulties, through its impact on subvocal rehearsal. It is further suggested that the resulting problems of cerebellar dysfunction, namely, difficulties in automation of skills and production of inner speech, then lead to deficits in automating word recognition processes and in phonological awareness (Nicolson et al., 2001), thus providing a framework for the involvement of the cerebellum in reading difficulties, as well as in WM.

Other studies have also implicated the cerebellum in WM (Ravizza, McCormick, Schlerf, Justus, & Ivry, 2006) and other academic areas such as mathematics (Feng, Fan, Yu, Lu, &Tang, 2008). Consequently, it appears that the cerebellum may play an important role when understanding the relationships found between specific aspects of motor coordination and cognitive areas such as WM and academic achievement. Evidence for the close co-activation of the cerebellum and prefrontal cortex (which has a well-established role in complex cognitive functions such as WM) in functional neuroimaging (Diamond, 2000) provides further evidence for a relationship between motor coordination and cognitive outcomes including WM.

Ultimately, in light of the increasing evidence of a link between motor coordination and cognitive outcomes such as WM and academic achievement, what role does WM play in the relationship between motor coordination and academic achievement? Alloway (2007) separated a DCD sample based on high and low visuospatial memory ability scores (averaged across short-term and WM tasks) and found that the low visuospatial memory ability group performed significantly worse on literacy and numeracy compared with the high visuospatial memory group. This finding remained after controlling for Vocabulary and Block Design (a nonverbal IQ task involving a motor component) scores, suggesting that the link between visuospatial memory and learning outcomes in children with DCD can be explained by more than just general ability and the motor components of such visuospatial memory tasks (Alloway, 2007). Thus, it is possible that the combined storage and processing component of the memory tasks is important when understanding how memory and learning outcomes are linked in children with DCD.

This is further supported by a recent intervention study involving children with DCD and comorbid learning difficulties (Alloway & Warner, 2008). Following the 13-week program of task-specific motor exercises, motor coordination and visuospatial WM showed improvement, but there was no improvement in verbal WM or reading and math scores. First, the results suggest that motor coordination may be more important in predicting visuospatial WM than verbal WM which is not surprising given that visuospatial processing (with or without a motor component) was found to be the greatest deficit in a meta-analysis examining the information processing deficits characterizing DCD (Wilson & McKenzie, 1998). The improvement in visuo-spatial WM in Alloway and Warner's intervention study may be understood in terms of the movement planning and control components of such visuospatial WM tasks, which can be improved by movement training. However, given that neither verbal WM nor reading and mathematics scores improved, this may suggest that it is the processing and storage component of the memory tasks (which is dissociable from the motor component) that influences learning outcomes in children with DCD (Alloway & Warner, 2008). Therefore, such findings suggest that motor coordination is not directly related to learning outcomes rather, the relationship may be mediated by the ability to simultaneously process and store information (i.e., WM ability).

Although preliminary evidence provides important insights into the relationship between motor coordination, WM, and learning outcomes, a number of issues need to be addressed. First, the present study controls for the confounding influence of ADHD symptomatology. This is important given that ADHD has been linked to motor problems (Pitcher, Piek, & Hay, 2003), WM (Martinussen, Hayden, Hogg-Johnson, & Tannock, 2005), and learning outcomes (Semrud-Clikeman et al., 1992). Also, previous research investigating motor coordination, WM, and academic outcomes has involved atypical population groups. Therefore, further investigation using a normative population is needed. It has been noted that correlational studies using normative samples are important in order to provide a better understanding of relationships found in children with DCD (Roebers & Kauer, 2009). This is important given the methodological problems associated with the use of clinical samples, for example, overestimating associations between domains (Roebers & Kauer, 2009). In addition, research in the area has involved younger samples aged 5–11 (e.g., Alloway & Warner, 2008). Thus, it is important to examine whether these findings extend to an adolescent population, particularly since recent findings have demonstrated how relationships between ability domains differ across age cohorts of 3–14 years of age (Dyck, Piek, Kane, & Patrick, 2009) and that the dimensional structure of executive functions also appears to undergo developmental changes, with the underlying processes being less distinguishable in the earlier years (Miyake et al., 2000).

The present study examined a mediating model of the relationship between motor coordination, WM, and academic achievement in adolescents from a normative sample whilst controlling for potentially confounding factors such ADHD symptoms, verbal ability, socioeconomic status (SES), age, and gender. It was hypothesized that motor coordination (as measured by Manual Dexterity, Aiming and Catching, and Balance) would have a positive direct effect on academic achievement (as measured by Numerical Operations, Word Reading, and Spelling); motor coordination would have a positive effect on WM (as measured by verbal and visuospatial WM) through a direct path; WM would have a positive direct effect on academic

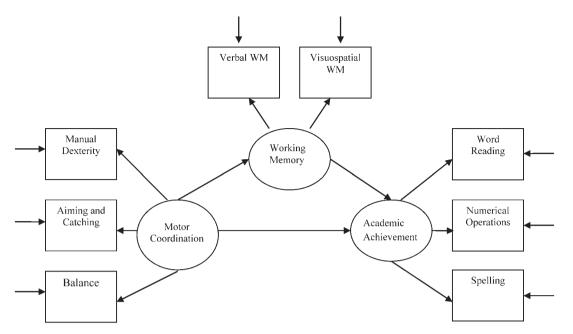


Fig. 1. Diagrammatic representation of the proposed mediating model.

achievement; and motor coordination would have a positive effect on academic achievement through an indirect path with WM mediating this relationship. Fig. 1 provides a diagrammatic representation of the proposed mediating model.

Finally, the directional nature of the relationship between motor and cognitive domains remains unclear given the very few longitudinal studies in the area (Murray et al., 2006; Piek, Dawson, Smith, & Gasson, 2008). Studies have provided initial evidence that motor coordination predicts performance on complex cognitive tasks including WM (Murray et al., 2006; Piek et al., 2008). However, given that complex cognitive and motor development display equally protracted developmental courses continuing into early adulthood and both the prefrontal cortex and cerebellum reach maturity late (Diamond, 2000), this may suggest that motor performance affects cognitive functioning and vice versa. Therefore, the current study also investigated an alternative model whereby the meditational role of motor coordination in the relationship between WM and academic achievement was examined. It is also important to note that the present correlational data cannot, of course, be used to establish cause-and-effect relationships. Therefore, the aim of the study was to determine the degree to which the proposed causal model had the capacity to generate our correlational data.

#### Method

#### **Participants**

Sixty government, private, and independent secondary schools were randomly selected from available lists. These schools were from varying areas of SES, in order to ensure a representative sample of the population. From these schools, five schools (representing these various school groups) consented to promote the project. Participants were also recruited through public advertisements in community newspapers, radio, and snowballing (i.e., existing participants recruit future participants through their associations). Inclusion criteria for the study were adolescents aged 12–16. Exclusion criteria included a minimum Verbal Comprehension Index (VCI) of 80 as measured by the Wechsler Intelligence Scale for Children-IV (WISC-IV) in order to exclude any adolescent whose difficulties might be attributed to general delayed development (Henderson & Barnett, 1998; Piek et al., 2004), as well as no presence of a physical disability, chronic illness, or a medical condition that affects development (such as neurological disorder and Down syndrome, ascertained by a parent report). The final sample included 93 adolescents, 38 girls and 55 boys, with a mean age of 14.2 (SD = 1.1). The SES scores were derived from the Australian Prestige Scale (Daniel, 1983) which rates the prestige of occupations in Australia, with scores ranging from 1 (reflecting high prestige) to 6.9 (reflecting low prestige). The occupation rated as most prestigious out of mother's and father's occupation was used as the SES score (M = 3.77, SD = 1.00, range = 1.80–6.60).

*Movement Assessment Battery for Children-2.* The three subscales from the MABC-2 (Henderson, Sugden, & Barnett, 2007) were utilized to provide the observed variables for the construct motor coordination. The MABC-2 is a standardized test used for the identification and description of children with movement difficulties. It consists of tasks suitable for three age bands (i.e., age band 3-6, 7-10, and 11-16 years) and tasks are grouped into the subscales: Manual Dexterity, Aiming and Catching, and Balance. For the 11-16 years age band, Manual Dexterity comprises three tasks including turning pegs with preferred and non-preferred hand, a bimanual task to make a triangle with nuts and bolts, and a drawing trail. The Ball Skill tasks include aiming and throwing at a wall target, and catching a ball with one hand. The Balance subscale involves a two-board balance task, walking toe-to-heel backwards, and a zigzag hopping task. Age-based standard scores are derived for the three subscales (M = 10, SD = 3) and for the Total Test Score (TTS; M = 10, SD = 3). A TTS of 67 (equivalent to a standard score of 7 on the MABC-2) and above (i.e., >15th percentile) suggests no evidence of movement difficulty, a score between 57 and 67 (6–15th percentile) suggests that the child is "at risk" of having a movement difficulty, and a TTS up to and including 56 (i.e., equivalent to a TTS standard score from 1 to 5) indicates significant movement difficulty ( $\leq$ 5th percentile). The age-standardized Manual Dexterity, Aiming and Catching, and Balance subscale scores were used for the purposes of this study.

The original MABC (Henderson & Sugden, 1992) is well established as a research tool and has favorable psychometric properties (Henderson et al., 2007). Reliability coefficients range from 0.73 to 0.84 for the subscale scores and 0.80 for the MABC-2 TTS. There is also evidence demonstrating criterion-related and discriminative validity (Henderson et al., 2007). Schulz and colleagues (2011) provided recent evidence for the structural validity (i.e., factor structure) of the MABC-2 across the three age-bands. Based on their findings in a large normative sample, the authors also noted that confidence in the structural validity of the three MABC-2 components becomes stronger for older children (i.e., age band 11–16 years).

Wechsler Individual Achievement Test-II. The Wechsler Individual Achievement Test-II (WIAT-II) Australian (Wechsler, 2007) is an individually administered test of achievement in individuals aged 4–85, assessing academic skills in the domains of reading, writing, mathematics, and oral language. In the present study, the age-standardized Word Reading, Spelling, and Numerical Operations subtest scores (M = 100, SD = 15) were used to provide observed variables for the construct academic achievement. These academic areas were chosen because they comprise essential aspects of academic achievement and have been examined previously in studies investigating the relationship between motor, WM, and academic outcomes (e.g., Alloway, 2007). The Word Reading subtest involves reading aloud from a graded word list. Numerical Operations assesses the ability to solve written calculation problems and simple equations involving the basic operations of addition, subtraction, multiplication, and division. The Spelling subtest assesses the ability to spell dictated words.

The WIAT-Australian has demonstrated an overall total composite reliability of 0.98, and test-retest reliabilities varying from 0.80 to 0.96 for subtests (Wechsler, 2007). The WIAT-II Australian also has good content, construct, and criterion-related validity (Wechsler, 2007).

*WISC-IV: Australian.* The WISC-IV (Wechsler, 2003) measures cognitive ability in children aged 6 to 16 years 11 months. The 10 core subtests yield a Full-Scale IQ and are organized to yield four composite scores (M = 100, SD = 15), namely: VCI, Perceptual Reasoning Index, WM Index (WMI), and Processing Speed Index. For the purposes of this study, the VCI was used as a control variable and to exclude any adolescent whose difficulties might be attributed to general delayed development. The age-standardized WMI score (comprising digit span and letter-number-sequencing (LNS) subtests to assess verbal WM) was used to provide an observed measurement for the construct, WM. The WISC-IV is a widely used measure of intelligence in children and has excellent internal consistency, test-retest reliability, criterion validity, and construct validity (Wechsler, 2003).

*N-back task.* The N-back task assesses visuospatial WM and was used to provide the second observed variable for the construct WM. This task involves a visuospatial variant of the N-back task, designed after Gevins and Cutillo (1993) and Jansma and colleagues (2000), and has been adapted to make it more attractive and appropriate for children (van Leeuwen, van den Berg, Hoesktra, & Boomsma, 2007). An apple is presented on the computer screen which has four holes from which a caterpillar appears. Respondents are instructed to stop the caterpillar from eating the apple by pressing one of the four buttons that corresponds spatially with the hole the caterpillar appeared from. There are four conditions of graded difficulty in which respondents are required to indicate where the caterpillar was one move back, two moves back, three moves back, or four moves back, respectively. The caterpillar appears on the screen for 1 s and is then followed by a warning tone which prompts children to respond. Each condition consists of a practice block (10 trials) and a block in which performance

is measured (32 trials). The task was discontinued if participants performed below chance levels, that is, 8 or less correct trials on a condition. Task performance was measured by the total number of correct responses on all trials administered (maximum score of 128 correct responses over the four conditions), with higher scores indicating better visuospatial WM thereby capturing the full dimension of visuospatial WM performance. For the purposes of the present study, the raw score of total number of correct responses was converted to a *z*-score. The N-back task is a widely used measure of WM and in a study examining a sample of adolescents with the current version of the N-back task, test–retest (carried out 2–3 weeks after initial assessment) reliabilities of 0.70 and 0.66 were reported for the 3- and 4- back conditions, respectively (van Leeuwen et al., 2007). For such tasks measuring specific abilities, it has been noted that reliabilities of 0.7 or higher are considered satisfactory, whereas reliabilities of 0.5 and 0.6 may be considered as modest (Kuntsi, Stevenson, Oosterlaan, & Sonuga-Barke, 2001; van Leeuwen et al., 2007).

Strengths and weaknesses of ADHD symptoms and normal behavior. The parent-rated strengths and weaknesses of ADHD symptoms and normal behavior (SWAN) scale (Swanson et al., 2001) is based on the ADHD symptoms listed in the Diagnostic and Statistical Manual of Mental Disorders-IV and involves observations based on the last month with reference to other children of the same age. The first nine items of the scale describe symptoms relating to inattention, while the second nine items relate to hyperactive/impulsive behaviors. Items are phrased in order to sample the full dimension of a particular behavior. An example of an item is: "How does this child pay attention to detail?" Scoring for each item ranges from "far below average" (scored as +3) to "average" (scored as 0), and "far above average" (scored as -3) in order to reflect both strengths and weaknesses. An overall SWAN score was calculated by averaging the scores on the 18 items. For the present study, the raw overall SWAN score was converted to a *z*-score. Hay and colleagues (2007) found the SWAN to be an accurate reflection of the ADHD phenotype, and Polderman and colleagues (2007) found that the SWAN rating scale yields a normal distribution of scores, making it a useful instrument for examining variation of (hyper) activity and attention in the general population. The Cronbach  $\alpha$  for current study was 0.97, demonstrating excellent internal reliability.

*Australian Prestige Scale*. Daniel's Prestige Scale (Daniel, 1983) rates occupational status on a scale of 1 (representing higher prestige) to 6.9 (representing lower prestige). High prestige occupations reflect power and privilege and require educational qualifications as well as high earning capacity. The occupation of "housewife," "student," or "unemployed" has no code on the scale. Occupational prestige based on parental occupation was coded as a continuous score and was used as an indicator of SES in the current study. When both parents were working, the most prestigious occupation was used. Daniel's scale has been widely used in health and social research (Smith, Owen, & Baghurst, 1997).

#### Procedure

This study followed the ethical guidelines of the National Health and Medical Research Council of Australia and was granted approval from the Curtin University Human Research Ethics Committee and from the representative bodies for the participating schools. Principals were contacted by mail seeking permission to recruit via their school and the project was then promoted in school newsletters. Interested adolescents and their parents provided written consent for participation. Participants were individually tested by a single trained examiner using standardized instructions. Testing time was 4.5 h which was broken into two sessions, with the MABC-2 and WISC-IV (respectively) administered in the first session and the WIAT-II and N-back (respectively) administered in the second session. Parents completed the SWAN questionnaire. Testing sessions were carried out at the family home or Curtin University, depending upon family preference. Most sessions occurred at the family home; however, it was ensured that distractions in both settings were kept to a minimum.

#### Data Analysis

Structural equation modeling (SEM), with maximum likelihood estimation, was used to determine the degree to which WM mediates the relationship between motor coordination and academic achievement. The analysis was implemented through LISREL (Version 8.54; Jöreskog, & Sörbom, 2004). For relatively simple models such as our one-mediator model, sample sizes between 100 and 150 have been recommended (Hair, Black, Babin, Anderson, & Tatham, 2006). Our current sample size of 93 falls just short of this recommendation, but should still be sufficient to provide stable estimates of the path coefficients. Furthermore, a sample size of 93 provides approximately seven participants for each parameter in the saturated model, which exceeds the minimum requirement of five participants per parameter recommended by Kline (2005). The assumption of multivariate normality was met.

#### **Results**

#### **Descriptives**

Table 1 provides the means, standard deviations, and ranges for the variables measuring motor coordination, WM, and academic achievement.

Five adolescents scored at or below the 5th percentile on the MABC-2 total score (indicating significant movement difficulty) and two scored between the 6th and 15th percentile (regarded as "at risk"). The prevalence of significant movement difficulty (<5th percentile) was 5.4%, which is comparable with previous estimates of 6% (APA, 2000). The numbers of adolescents scoring below the 25th percentile (Shafrir & Siegal, 1994) on the Word Reading, Numerical Operations, and Spelling subtests of the WIAT-II were 7, 12, and 5, respectively. Two participants with significant movement difficulty (<5th percentile) also demonstrated learning difficulties ( $\leq$ 25th percentile on the WIAT-II). One participant demonstrated Spelling and Numerical Operations difficulties, and the other, Word Reading and Numerical Operations difficulties.

#### **Correlations**

Potential control variables included, age, gender, SES, ADHD symptoms, and VCI. All indicators, except for the N-back task (z-score), are represented by age-standardized scores. Given that no significant correlation was found between the N-back task and age (r = .15, p = .151), age was not retained as a control variable. The VCI, SWAN, and SES variables significantly correlated with indicators of WM and/or academic achievement and were thus retained as control variables. A covariance structure analysis was conducted to determine whether the partial correlations among the eight indicators (after controlling for SES, ADHD symptoms, and VCI) varied as a function of gender. As they did not, gender was ignored in all further analyses of these partial correlations,  $\chi^2$  (36) = 35.03, p = .51.

Indicators that are "driven" by the same latent construct will necessarily correlate. In the present study, however, two of the MABC-2 subscales-Manual Dexterity and Aiming and Catching-were not significantly correlated and therefore could not appear in the same model as indicators of the same latent construct. It was therefore decided to test three separate mediator models; one for each of the three MABC-2 subscales (namely, Manual Dexterity, Aiming and Catching, and Balance). An important correlational assumption underlying mediation states that the independent variable (motor coordination as measured by each of the three MABC-2 subscales) must be significantly correlated with both the mediator (WM) and the outcome variable (academic achievement). The model using Aiming and Catching satisfied all correlational assumptions described above and, thus, met this underlying premise to mediation testing (Baron & Kenny, 1986). However, the models with Manual

	Mean	SD	Range
MABC-2 Manual Dexterity <sup>a</sup>	9.57	2.47	3.0-15.0
MABC-2 Aiming and Catching <sup>a</sup>	11.03	2.73	4.0-16.0
MABC-2 Balance <sup>a</sup>	11.42	2.98	4.0-14.0
WISC-IV Working Memory Index <sup>a</sup>	103.75	12.47	59.0-141.0
N-back <sup>b,c</sup>	88.17	19.69	6.0-124.0
Z N-back <sup>d</sup>	0.00	1.00	-4.17 to $1.82$
WIAT-II Word Reading <sup>a</sup>	107.44	10.62	77.0-128.0
WIAT-II Numerical Operations <sup>a</sup>	106.85	14.73	63.0-139.0
WIAT-II Spelling <sup>a</sup>	107.19	11.68	67.0-129.0
SWAN <sup>b,e</sup>	-0.9989	1.02	-3.0 to 1.22
ZSWAN <sup>d</sup>	0.00	1.00	-1.95 to $2.16$
WISC-IV Verbal Comprehension Index <sup>a</sup>	106.63	11.25	81.0-132.0
SES <sup>f</sup>	3.77	1.00	1.80-6.60

Table 1. Means, sta

Notes: MABC-2 = Movement Assessment Battery for Children-2; WISC-IV = Wechsler Intelligence Scale for Children-IV; WIAT-II = Wechsler Individual Achievement Test-II; SWAN = Strengths and weaknesses of ADHD symptoms and normal behavior.

<sup>a</sup>Age-standardized score.

<sup>b</sup>Raw score.

<sup>c</sup>Total number of correct responses.

<sup>e</sup>Scores are calculated by averaging the total of the 18 ADHD items.

<sup>f</sup>The occupation rated as most prestigious out of mothers' and father's occupation.

<sup>&</sup>lt;sup>d</sup>z-score.

Dexterity and Balance did not satisfy these assumptions, leading to the immediate rejection of these models. The measurement error associated with the Aiming and Catching subscale was fixed at one minus its reliability coefficient, and its factor loading was fixed at the square root of its reliability coefficient (*see* Goodwin & Plaze, 2000, p. 286).

Finally, Spelling was removed because, unlike Word Reading and Numerical Operations, it did not correlate with motor coordination, and its inclusion rendered the pathway between motor coordination and academic achievement non-significant (Table 2).

#### LISREL Analysis

Pearson's correlations (controlling for ADHD symptoms, VCI, and SES) were input to LISREL for structural equation modeling. The parameter estimates and standard errors for the saturated model are given in Fig. 2. The path from motor coordination to academic achievement was not significant. The hypothesis that motor coordination would have a direct impact on academic achievement in this model was therefore not supported. All other hypotheses were supported. Specifically, the path from motor coordination to WM was significant, as was the path from WM to academic achievement. This indirect pathway was significant (p = .003), indicating that motor coordination has an indirect effect on academic achievement through WM.

Fit indices providing an indication of the overall fit of the model can be found in Table 3. The fit statistics for this model suggest a good fit to the data— $\chi^2(3) = 5.12$ , p = .16; a non-significant  $\chi^2$  value ( $p \ge .05$ ; Kline, 2005); the  $\chi^2/df$  ratio is below 2 (Kline); the Comparative Fit Index is >0.90 (Kline, 2005); and the Standardized Root Mean Square Residual is <0.10 (Kline, 2005). Although the Root Square Mean Square Error of Approximation for the saturated model is above the desired 0.05 level and above the more liberal cutoff of 0.08 (i.e., 0.092), Tabachnick and Fidell (2001) note that this index may be less preferable with smaller samples due to the tendency to over-reject the true model. Overall, the results indicate good data-model fit.

The test of the saturated model indicated that, when WM is controlled, the magnitude of the path coefficient for the direct pathway from motor coordination to academic achievement is trivial. The direct pathway can therefore be dropped from the model without significantly reducing model fit— $\chi^2_{diff}(1) = 0.00$ , p = .99) or changing parameter estimates (Fig. 3). The more parsimonious mediator model was therefore selected. The fit indices for the mediator model are reported in Table 3; the parameter estimates for the mediator model are given in Fig. 3.

There is a plausible alternative model in which motor coordination mediates the impact of WM on academic achievement. The previous analysis, however, indicated that the pathway from motor coordination to academic achievement is non-significant. According to our data, therefore, the alternative model is not viable.

Finally, in the proposed measurement model for the current study, all four N-back conditions (i.e., 1-back to 4-back) are presumed to load on a visuospatial WM factor, while the digit-span forward (DSF), digit-span backward (DSB), and LNS tasks from WISC-IV WMI are presumed to load on a verbal WM factor. Previous research has argued for separation of short-term memory and WM (e.g., Baddeley, 2000; Kail & Hall, 2001), which suggests a plausible alternative measurement model for the data in which three of the N-back conditions (2-back, 3-back, and 4-back), the DSB, and LNS tasks load on a WM factor, while the 1-back and DSF measures load on a short-term memory factor. Confirmatory factor analyses was conducted to compare the alternative measurement model (in which 2-back to 4-back, DSB, and LNS load on WM; while 1-back and DSF load on short-term memory) with the proposed measurement model (in which 1–4-backs load on visuospatial WM, while DSF, DSB, and LNS load on verbal WM). A comparison of the fit statistics (Table 4) indicated that the proposed model provides the better fit. These results are in line with previous research, suggesting that simple (i.e., Short-Term Memory [STM]) and complex (i.e., WM) span tasks largely measure the same basic processes and also have correlations with higher order cognitive abilities that are similar in magnitude (Unsworth & Engle, 2007). Unsworth and Engle argue against the notion that STM and WM are different constructs.

#### Discussion

Research supporting the relationship between motor coordination and academic achievement has accumulated without any clear understanding of the nature of this relationship. The aim of the current study was to advance this understanding. The results indicate that, after controlling for VCI, ADHD symptoms, and SES, WM (verbal and visuospatial WM) mediated the relationship between motor coordination (specifically, MABC-2 Aiming and Catching) and academic achievement (specifically, Word Reading and Numerical Operations). In SEM terms, motor coordination did not have a direct impact on academic achievement; instead, it impacted on academic achievement via WM.

There is extensive evidence demonstrating WM as a reliable predictor of a range of cognitive skills and academic areas, including reading and mathematics (Alloway, 2009). The current study adds to these findings by revealing a very strong

Table 2. Zero-order correlation matrix for the key and control variables

	MABC-2 Manual Dexterity	MABC-2 Aiming and Catching	MABC-2 Balance	WISC-IV WMI	ZN-back	WIAT-II Word Reading	WIAT-II Numerical Operations	WIAT-II Spelling	Gender	VCI	SES	ZSWAN
MABC-2 Manual Dexterity <sup>a</sup>	1.00											
MABC-2 Aiming and Catching <sup>a</sup>	.071	1.00										
MABC-2 Balance <sup>a</sup>	.264*	.423**	1.00									
WISC-IV WMI <sup>a</sup>	.113	.251*	.122	1.00								
ZN-back <sup>b</sup>	.129	.281**	.146	.431**	1.00							
WIAT-II Word Reading <sup>a</sup>	.037	.280**	.128	.453**	.410**	1.00						
WIAT-II Numerical Operations <sup>a</sup>	.173	.229*	.146	.632**	.400**	.545**	1.00					
WIAT-II Spelling <sup>a</sup>	.113	.121	.164	.566**	.269**	.714**	.688**	1.00				
Gender	.235*	397**	007	020	051	128	041	.116	1.00			
WISC-IV VCI <sup>a</sup>	.075	.048	.155	.442**	.253*	.512**	.513**	.535**	018	1.00		
SES <sup>c</sup>	122	.032	099	174	039	246*	196	252*	083	384**	1.00	
ZSWAN <sup>b</sup>	179	.017	057	238*	196	208*	406**	360**	236*	324**	.110	1.00

*Notes:* MABC-2 = Movement Assessment Battery for Children-2; WISC-IV = Wechsler Intelligence Scale for Children-IV; WMI = Working Memory Index; WIAT-II = Wechsler Individual Achievement Test-II; SWAN = Strengths and weaknesses of ADHD symptoms and normal behavior; VCI = Verbal Comprehension Index.

<sup>a</sup>Age-standardized score.

<sup>b</sup>z-score.

<sup>c</sup>The occupation rated as most prestigious out of mothers' and father's occupation.

\*p < .05 (two-tailed).

\*\*p < .01 (two-tailed).

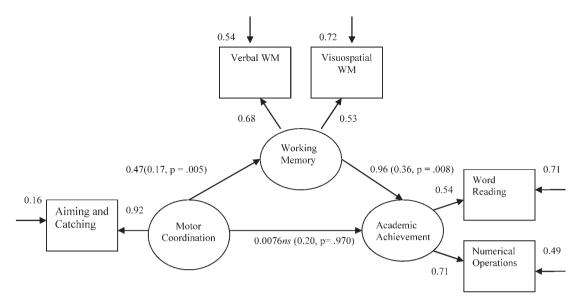


Fig. 2. Parameter estimates for the saturated model.

Table 3. Summary of model fit indices for the saturated and mediator models of the relationship between motor coordination, WM, and academic achievement

Model	$\chi^2$	df	<i>p</i> -value	RMSEA	CFI	SRMSR
Saturated model	5.28	3	.15	0.095	0.98	0.041
Mediator model	5.28	4	.26	0.063	0.99	0.041

Notes: CFI = Comparative Fit Index; SRMSR = Standardized Root Mean Square Residual; RMSEA = Root Square Mean Square Error of Approximation.

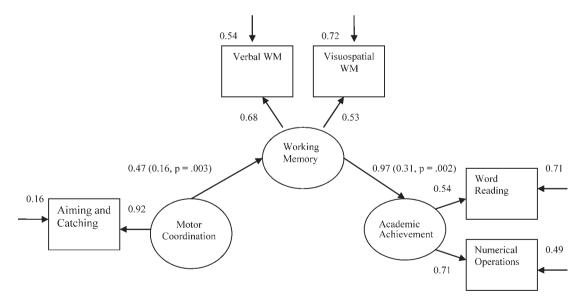


Fig. 3. Parameter estimates for the mediator model.

link between WM and academic outcomes in an adolescent normative sample. The current results also support recent research suggesting a link between WM and motor coordination (Piek et al., 2004; Wassenberg et al., 2005).

Importantly, the results from this study suggest that the relationship between motor coordination and academic achievement can be understood in terms of a mechanism whereby motor coordination has an indirect impact on learning outcomes via WM.

Table 4. Summary of model fit indices for alternative measurement models of the WISC-IV WMI and the ZN-back

Model	$\chi^2$	df	<i>p</i> -value	RMSEA	CFI	SRMSR	Model AIC
Model 1 1BACK-4BACK = VSWM DSF DSB LNS = VWM Model 2	16.90	13	.20	0.058	0.95	0.070	46.90
2BACK-4BACK DSB LNS = WM 1BACK DSF = STM	26.97	13	.013	0.120	0.85	0.084	59.97

*Note:* CFI = Comparative Fit Index; SRMSR = Standardized Root Mean Square Residual; RMSEA = Root Square Mean Square Error of Approximation; Model AIC = Akaike's Information Criterion (smaller is better); VSWM = Visuospatial Working Memory; VWM = Verbal Working Memory; WM = Working Memory; STM = Short-Term Memory.

Alloway and Warner (2008) provided evidence that learning outcomes may not be directly impacted by motor skills in children with DCD, but rather, it is difficulties with combined processing and storage of information that may underlie learning outcomes in these children. This argument is consistent with a mediation model in which motor coordination impacts on learning via WM. The present study extends from these findings by establishing the viability of this model in an adolescent normative sample.

It is important to note that in the present study, "motor coordination" was operationalized with just one of the three MABC-2 motor skill components, namely, Aiming and Catching. The three models (i.e., Aiming and Catching, Manual Dexterity, and Balance) were initially examined separately given that the association between Aiming and Catching and Manual Dexterity subscales was found to be non-significant for this sample of adolescents. This result is in line with Haga, Pedersen, and Sigmundsson's (2007) study which found weak correlations among the MABC motor tasks in a sample of 4-year-old children. The authors of the study explained their findings in terms of task-specific skills and argued for the importance of identifying the skills that are necessary and important for children to learn (Haga et al., 2007).

In the present study, the models with Manual Dexterity and Balance were subsequently dropped because they failed to demonstrate significant correlations with the mediator and the outcome measures. This is consistent with Gaysina and colleagues (2010) study, which did not find any significant association between fine motor skills and academic difficulties in the reading domain at age 15. Similarly, Michel and colleagues (2011) found that "motor impaired" children, identified by having Manual Dexterity difficulties, did not perform worse on a WM task of Backwards Color Recall when compared with those without motor impairment. In another study, Backwards Color Recall did not significantly correlate with fine motor skills as measured by a pegboard task in a normative sample of 7-year olds (Roebers & Kauer, 2009). However, significant correlations were found with a postural flexibility task (Roebers & Kauer, 2009).

The current study demonstrates an important relationship between Aiming and Catching games, WM, and academic achievement (specifically, Word Reading and Numerical Operations), supporting previous research of a specific relationship between aspects of motor coordination and these cognitive areas. The specific relationship found between the Aiming and Catching games, WM, and academic outcomes may be explained by shared underlying neural processes. Ball games such as those used in the current study (e.g., throwing a ball against a wall and then catching it with one hand upon return) require the control of independent limb movements, including rapid skilled movements. Carlson (2010) notes that the lateral zone of the cerebellum is important in calculating the complex, closely timed sequences of muscular contractions required for such rapid skilled movements. Consequently, it is possible that the specific associations found in the current study may be explained by cerebellar mechanisms, specifically, involvement from the lateral cerebellum. Therefore, the current results provide some support for the cerebellar deficit hypothesis proposed by Nicolson and colleagues (2001). Their framework suggests a causal relationship between cerebellar dysfunction and reading problems, which may be understood in terms of the cerebellar contributions to automation of skills and production of inner speech. An important link between the cerebellum and verbal WM is also suggested which is important when understanding the resulting reading problems (Nicolson et al., 2001). The results of the current study also provide support for previous evidence which demonstrates the role of the lateral cerebellum in developmental dyslexia (e.g., Rae et al., 1998). The present results also support other studies implicating the cerebellum in WM (Ravizza et al., 2006) and in other academic areas such as mathematics (Feng et al., 2008).

In addition, the basal ganglia may also play a role in the present findings as it has it been associated with the ability to modulate force of movement (Lundy-Ekman, Ivry, Keele, & Woollacott, 1991) which is a skill needed for the fast, goal-directed movements involved in ball throwing activities. The basal-ganglia forms part of the extrapyramidal system (along with the cerebellum) and has also been implicated in cognitive functions such as WM (Voytek & Knight, 2010). However, it is also important to note the complex interactions between the motor areas of the brain and other parts of the central nervous system such as the cerebellum, resulting in continuous interplay among these structures (Piek, 2006). Diamond (2000) highlighted the close co-activation of the cerebellum (specifically, the neocerebellum which forms part of the lateral cerebellum) and prefrontal cortex when understanding the relationship between complex motor and cognitive domains. In addition to the important role of the cerebellum, it is possible that the complex nature of ball skills assessed in the current study co-activates greater prefrontal cortex activity than the tasks assessing solely fine motor (Manual Dexterity tasks) or Balance skills. The prefrontal cortex plays an important role in WM (Crone, Wendelken, Donohue, van Leijenhorst, & Bunge, 2006) and has been implicated in both mathematics (Ansari & Dhital, 2006; Rivera, Reiss, Eckert, & Menon, 2005) and reading performance (Backes et al., 2002; Maguire, Frith, & Morris, 1999). This may, in part, explain the specific links found in the current study.

In addition, it is likely that children who experience difficulty in executing the complex combination of motor skills involved in ball games will subsequently avoid participating in such tasks (Cairney et al., 2005). Children also typically require partners to practice with in order to develop ball skills which may be a problem for individuals with movement difficulties given the associated difficulties in the social domain (Smyth & Anderson, 2000). It is possible that the resulting lack of opportunity to learn and practice the skills needed for ball games may play a significant role in understanding the current findings.

Best (2010) highlighted the protracted period of cognitive and brain development into adolescence and argued that since executive functions and the underlying neural circuitry are still immature during this time, complex cognitive functions (such as WM) may be sensitive to the effects of a child's experiences and plausibly enhanced by certain experiences (Best, 2010). In fact, there is increasing research demonstrating the positive impact of physical activity on cognitive and academic functioning (Tomporowski, Davis, Miller, & Naglieri, 2008). Sibley and Etnier (2003), in their meta-analysis, suggest that the mechanisms underlying the relationship between physical activity and cognition may be explained by two broad categories including physiological and learning/developmental mechanisms. Physiological mechanisms, induced by exercise, include physical changes such as increased cerebral blood flow, structural changes in the central nervous system, alterations in brain neurotransmitters, and arousal levels (Sibley & Etnier, 2003). Conversely, learning/developmental mechanisms suggest that movement and physical activity provide learning experiences which enhance, and may be essential for, cognitive development (Sibley & Etnier, 2003). For example, active games may require similar cognitive processes to those involved in EF tasks such as strategic and goal-directed behavior when faced with a novel game experience. Thus, the skills gained during participation in such games may also transfer to EF tasks (Best, 2010).

Research has also suggested that the more complex forms of physical exercise, requiring greater cognitive engagement as well as coordination of complex bodily movements, are more likely to enhance EF than simpler exercises (Budde, Voelcker-Rehage, Pietrabyk-Kendziorra, Ribeiro, & Tidow, 2008; Pesce, Crova, Cereatti, Casella, & Bellucci, 2009). Therefore, it is likely that games involving Aiming and Catching motor skills (e.g., basketball) require this complex cognitive engagement which may prove important in transferring to and enhancing EF skills. Ultimately, individuals with motor coordination difficulties may not be provided with the same opportunity to enhance these areas given their tendency to withdraw from physical participation.

This study has some limitations. It is important to note that the current study investigated the academic domains of Word Reading, Numerical Operations, and Spelling only. Consequently, it is possible that motor areas, such as Manual Dexterity, may be important in predicting other academic outcomes in adolescence such as writing. The present study did not include other potential mediating variables, such as processing speed or motivation, which may also be important in understanding the nature of the relationship between motor coordination and academic achievement. Furthermore, an important area of future research appears to be addressing the potential mediating influence of physical participation/fitness levels in the relationship between motor coordination and academic outcomes. Examining the role of individual factors may be important in attempting to further understand the relationship between motor functioning, WM, and academic achievement. For example, it would be interesting to study children with motor coordination difficulties who show significant strengths in WM and academic achievement. It should also be noted that although researchers made effort to minimize all distractions in the testing setting, those sessions conducted at the family home (according to family preference) may have been more susceptible to such distractions, potentially confounding the results (particularly, on cognitive measures). However, despite these limitations and to the best of our knowledge, this is the first study to reveal the important relationship between motor coordination, WM, and academic achievement in an adolescent normative sample, highlighting the significance of these findings. Additionally, the present study is cross-sectional in nature and cannot conclude the directional relationships between the motor and cognitive domains. Further research is needed to elucidate the directional nature of the relationships. Finally, given that our findings provide some support for Unsworth and Engle (2007) who argue against the notion that STM and WM are different constructs, it is recommended that future studies attempt to further examine this notion and compare it with Baddeley's model which argues for a domain independent central executive.

#### Conclusion

Overall, the results of this study suggest that the association between motor coordination and academic achievement in an adolescent normative sample can be best understood in terms of a mechanism whereby motor coordination, specifically Aiming and Catching skills, has an indirect impact on learning outcomes via WM. These findings have important implications for the early assessment and treatment of motor coordination and learning difficulties. For children with movement difficulties, for example, strategies aimed at reducing excessive WM loads in the classroom may prove useful in enhancing their capacity to achieve in these academic areas. Finally, the current results revealing an important association between Aiming and Catching skills, WM, and academic outcomes (specifically Word Reading and Numerical Operations) suggest that the association between motor coordination and such cognitive outcomes may be understood in terms of common underlying mechanisms in the lateral cerebellum.

#### **Conflict of Interest**

None declared.

#### Acknowledgements

We are very grateful to the parents and adolescents who were willing to participate in this study. We also wish to thank Sean Piek, Linda Pannekoek, and Eva Kuhry for their assistance with data entry.

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PAPER 5

Pannekoek, L., Rigoli, D., Piek, J. P., Barrett, N. C., & Schoemaker, M. (2012). The revised DCDQ: Is it a suitable screening measure for motor difficulties in adolescents? *Adapted Physical Activity Quarterly*, 2(1), 81-97. Adapted Physical Activity Quarterly, 2012, 29, 81-97 © 2012 Human Kinetics, Inc.

### The Revised DCDQ: Is It a Suitable Screening Measure for Motor Difficulties in Adolescents?

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The parent-rated Developmental Coordination Disorder Questionnaire (DCDQ) has been revised to incorporate a wider age range, including adolescence. In this exploratory study, internal consistency and validity of the DCDQ-2007 was assessed using a community-based sample of 87 adolescents. Psychometric properties of the DCDQ-2007 were investigated and concurrent validity, sensitivity, and specificity were assessed with the MABC-2 as a criterion standard. The results demonstrated high internal consistency for the DCDQ-2007 and a relationship with the MABC-2 was found. The DCDQ-2007 met the recommended standard for sensitivity, although the confidence interval was large; however, it failed to meet the recommended standard for specificity. This has important implications concerning the suitability of the DCDQ-2007. Although promising psychometric properties were found within the current study, the applicability of the DCDQ-2007 as a screening measure for motor difficulties requires careful consideration.

Keywords: DCDQ-2007, motor difficulties, screening, adolescents, MABC-2

The definition of Developmental Coordination Disorder (DCD) in short, is "poor motor performance in daily activities that is not consistent with the child's age and intelligence, and is not due to medical condition" (APA, 2000). According to the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV), the prevalence of DCD in the age band of 5–11 years is about 6% (APA, 2000). However, prevalence rates as low as 2% have been reported in research studies that have used more stringent application of DCD criteria and cut-off scores (e.g., Lingam, Hunt, Golding, Jongmans, & Emond, 2009; van Dellen & Geuze, 1988; Wright &

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Sugden, 1996). Prevalence estimates for adolescents are still unknown (Cantell, Smyth, & Ahonen, 1994; Cantell, Smyth, & Ahonen, 2003).

The lack of movement experience that is often seen in individuals with motor difficulties can have a negative impact on behavioral, cognitive, social, emotional, and motor domains (Cantell et al., 1994; Cantell et al., 2003; Losse, et al., 1991; Skinner & Piek, 2001). Many of the negative effects are interrelated and are often more profound in adolescents compared with younger children (Skinner & Piek, 2001). As a vicious circle, the motor difficulties seen may lead to more avoidance of motor activities at older ages (Cantell, Crawford, & Doyle-Baker, 2008). Age-appropriate physical fitness levels are often not reached, resulting in greater risk of overweight and obesity, negative long-term effects on fitness, and other health risks (Cantell et al., 2008).

The few studies that have investigated the outcome of DCD have found that in about 50% of individuals identified with DCD in childhood, poor motor skills persist throughout adolescence and into adulthood (Cantell et al., 1994; Cantell et al., 2003; Losse et al., 1991); however, diagnosing DCD is problematic and there is little consistency in the procedures used (Geuze, Jongmans, Schoemaker, & Smits-Engelsman, 2001). The DSM-IV lists four criteria for the diagnosis of DCD (see Table 1); however, these are not well defined, with little information on how to assess motor performance (Geuze et al., 2001; Smits- Engelsman, Fiers, Henderson, & Henderson, 2008). This is particularly the case for children older than age 11 years where there is a lack of appropriate norm-referenced motor skill tests (Cantell et al., 1994). Measures designed specifically to assess younger children have been used to screen for motor difficulties in adolescents (Cousins & Smyth, 2005; Losse et al., 1991). Caution is warranted when using these measures with older age groups, however, as adolescents might score in the upper limit, producing a ceiling effect (Geuze & Borger, 1993; Losse et al., 1991). This makes the validity of the assessment questionable (Cousins & Smyth, 2005).

A valid, multidimensional measure that reflects an individual's developmental level appropriately is required to gain more insight into the nature and course of the disorder. The Movement Assessment Battery for Children (MABC; Henderson &

Criterion	Description
A	Performance in daily activities that require motor coordination is substan- tially below that expected, given the person's chronological age and mea- sured intelligence. This may be manifested by marked delays in achiev- ing motor milestones, dropping things, 'clumsiness', poor performance in sports, or poor handwriting.
В	The disturbance in criterion A significantly interferes with academic achievement or activities of daily living (self-care activities).
C	The disturbance is not due to a general medical condition (e.g., cerebral palsy, hemiplegia, or muscular dystrophy) and the child does not meet criteria for pervasive developmental disorder.
D	If intellectual delay is present, the motor difficulties are greater than would be expected, given the level of delay.

#### Table 1 DSM-IV Criteria for DCD

Sugden, 1992), a standardized measure for the identification of motor difficulties, is the most commonly used measure and is currently recognized as most suitable for identifying children with DCD (Brown & Lalor, 2009; Geuze et al., 2001). The original MABC was revised and restandardized, resulting in the publication of the MABC-2 in 2007 (Henderson, Sugden, & Barnett, 2007). The basic structure of the measure was maintained, but the scoring has changed from the use of impairment scores (i.e., lower scores indicating a better performance) to higher scores, indicating better performance. Therefore, the MABC-2 now covers the entire range of motor ability. The age range was extended, covering three age bands: 3–6 years, 7–10 years, and 11–16 years. As a result, the MABC-2 is one of the few motor assessment measures that includes adolescence (Henderson et al., 2007).

Motor performance measures such as the MABC require one-on-one testing of the individual. This is time consuming and expensive. Parent questionnaires may form an efficient alternative to screen large numbers of individuals for DCD (Schoemaker et al., 2006). Those found at risk by the initial screening can consequently be assessed with a standardized motor test to decide whether they meet the DSM-IV criteria (two-step procedure; Schoemaker et al., 2006).

The Developmental Coordination Disorder Questionnaire (DCDQ) is a parent report measure, designed to assess motor difficulties in children (Wilson, Kaplan, Crawford, Campbell, & Dewey, 2000). The original version of the questionnaire has been reported as reliable and valid for identifying DCD in children (Civetta & Hillier, 2008; Schoemaker et al., 2006; Wilson, Kaplan, Crawford, & Roberts, 2007). The DCDQ was revised in 2007 and now has an extended age coverage, which gives it good prospects as a measure for initial community-based screening of adolescents (Wilson et al., 2007). Furthermore, in the revised measure, all items are positively worded, with a higher score reflecting a better performance, in contrast to a lower score reflecting a better performance as in the original DCDQ.

To our knowledge the DCDQ-2007 has not been evaluated in an adolescent sample, and information on the psychometric properties of this revised measure is sparse. The current exploratory study used an existing dataset to analyze the performance of the DCDQ-2007 as a screening measure for motor difficulties in a community-based sample of adolescents aged 12–15 years.

The DCDQ-2007 cannot be used to diagnose DCD as the measure can only give an indication of DCD (Schoemaker et al., 2006). Therefore, the term motor difficulties will be used in the following sections of this manuscript instead of DCD. Although no gold standard for the diagnosis of motor difficulties currently exists (Crawford, Wilson, & Dewey, 2001), the MABC-2 was chosen as a criterion measure for this study, given its promising properties as described above. The original MABC has been used previously as a gold standard in studies examining the validity of a new measure (Rosenblum 2006; Schoemaker, Flapper, Reinders-Messelink, & de Kloet, 2008).

The DCDQ-2007 and the MABC-2 assess a similar construct, motor difficulties, and both are suitable for use in older age groups. It was therefore hypothesized that when used to screen for motor difficulties in adolescents, the DCDQ-2007 will to a large extent identify the same adolescents as the MABC-2. Significant positive correlations between the two measures were expected, supporting concurrent validity. In addition, if the sensitivity, specificity, and predictive values are high, this would provide psychometric evidence for the use of the DCDQ-2007 as an initial screening instrument for motor difficulties in a community-based sample.

# Method

#### **Participants**

This study is part of a larger study examining the relationship between motor skills, academic achievement, cognitive skills, and psychosocial outcomes in adolescence. Participants were recruited from randomly selected schools in areas of varying levels of socioeconomic status, through snowballing and public advertisement in community newspapers across a Western Australian city. Inclusion criteria for participation were between 12 and 15 years of age and no diagnosed physical disability, chronic illness, or medical condition that affects development (e.g., Down Syndrome). A minimum Verbal Comprehension Index of 70 on the Wechsler Intelligence Scale for Children–Fourth Edition (WISC-IV) was applied to exclude participants whose difficulties might be attributed to a general delayed development. All participants obtained a WISC-IV VCI score above 70 (*M* 106.6, min 81.0, max 132.0), indicating that none had intellectual disability.

From 87 adolescents (35 girls, 52 boys; age M 14.1 y, SD 0.99 y) a complete dataset was obtained, which was used for the statistical analyses performed in the current study. A parent-rated developmental history questionnaire was employed to screen for previous diagnoses of Attention Deficit Hyperactivity Disorder (ADHD), Learning Disability (LD), motor difficulties, or any other disability. A diagnosis of ADHD, LD, motor difficulties, or a combination of these disorders was present in 14 adolescents. Only two adolescents were diagnosed with ADHD, of which one had comorbid motor difficulties. Three adolescents were diagnosed with both LD and motor difficulties. Six adolescents were identified with motor difficulties only, and three adolescents were identified with LD only.

#### Measures

Movement Assessment Battery-2 (MABC-2; Henderson, Sugden, & Barnett, 2007).

The MABC-2 evaluates eight motor skill tasks, which are grouped into three components: manual dexterity, aiming and catching, and balance. For the three different age bands, the test items are similar but age-adjusted. The age band of 11–16 years was used for the purpose of the current study (Henderson et al., 2007).

Standard scores are calculated for the test items with the use of age-adjusted normative data. Subsequently, standard scores and percentiles for the three test components and the Total Test Score (TTS; sum of the eight item standard scores) are determined. Scores between 57 and 67 (6–15th percentile) indicate "at risk," and a TTS of 56 or lower ( $\leq$  5th percentile) is considered indicative of significant motor difficulties (Henderson et al., 2007). In the current study, the 15th percentile was applied as the criterion for motor difficulties (TTS < 67); as for research purposes, a 15th percentile cut-off is recommended on motor tests to prevent the exclusion of children with mild DCD (Geuze et al., 2001). Unless otherwise specified, the MABC-2 test results are applied for the identification of motor difficulties throughout this manuscript.

Psychometric evidence regarding the MABC-2 is limited; however, the MABC-2 manual reports good to excellent reliability and validity (Henderson et al., 2007). Test-retest reliability of r = 0.80 for the TTS and correlations ranging between r = 0.73 and r = 0.84 for individual component scores have been documented for all three age bands (n = 60; Henderson et al., 2007). The authors argue that as the general structure of the MABC did not change, and the content is regarded as sufficiently similar, previous findings regarding the validity of the original MABC remain relevant (Henderson et al., 2007). Using videotaped performances of children from 4 to 12 years old, good interrater reliability has been demonstrated for the original MABC, with kappa values of 0.95–1.00 (Smits-Engelsman et al., 2008). Good concurrent validity has been reported for the original measure, with a correlation between the TTS of the original MABC and the Bruininks-Oseretsky Test of Motor Proficiency (BOTMP) composite score of r = -.53 (n = 63, age range 4–12 y; Henderson & Sugden, 1992).

**Developmental Coordination Disorder Questionnaire-2007 (DCDQ-2007; Wilson, Kaplan, Crawford, & Roberts, 2007).** The DCDQ-2007 is a 15-item questionnaire comprising three subscales: Control During Movement, Fine Motor/Handwriting, and General Coordination. The DCDQ-2007 is self-administered by parents using a 5-point Likert scale, ranging from 1 (*not at all like your child*) to 5 (*extremely like your child*). Parents are requested to compare their child's motor performance with that of peers. By adding the 15 item scores, a Total Score (TS) is calculated (range 15–75; Wilson et al., 2007). Whilst the items are the same for all ages, different cut-off scores for motor impairment are given for the three age bands. For the purpose of this study, the third age band (10–15 years) was used, with a TS of 57 or lower indicative of motor difficulties (Wilson et al., 2007).

Information concerning the psychometric properties of the DCDQ-2007 is sparse; however, Wilson et al. (2009) reported a Cronbach's alpha of 0.89 and item-total correlations of r = 0.42 to r = 0.67, indicating good internal consistency. Good internal consistency was also found in another study (Cairney, Missiuna, Veldhuizen, & Wilson, 2008). Although evidence concerning test-retest reliability of the DCDQ-2007 is limited, a Chinese translated version of the DCDQ-2007 revealed a test-retest correlation of r = 0.94 (Tseng, Fu, Wilson, & Hu, 2010).

Strong construct validity has been reported for the DCDQ-2007, with DCD and suspect DCD groups scoring significantly lower compared with a non-DCD group (Wilson et al., 2009). Furthermore, no gender differences have been found for DCDQ-2007 scores (Wilson et al., 2009). Significant correlations between the total scores on the DCDQ-2007, the original MABC, and the Beery-Buktenica Developmental Test of Visual-Motor Integration (VMI) have been reported (respectively r = -.55 and r = 0.42), suggesting concurrent validity (Wilson et al., 2009).

Considering factor analytic validity, results are less consistent. A four-factor structure has been reported for the original DCDQ (Schoemaker et al., 2006; Wilson et al., 2000), while three factors are suggested to underlie the DCDQ-2007 (Cairney et al., 2008; Wilson et al., 2009). A poor level of fit for the proposed three factor structure has been reported. Studies either found unacceptably high interfactor correlations (Cairney et al., 2008), could not demonstrate adequate simple structure (Wilson et al., 2009), or found very low standardized parameter estimates (Tseng et al., 2010). This indicates that the instrument is best used as a measure of general motor difficulties and not to discriminate between specific kinds of motor difficulties.

**The Wechsler Intelligence Scale for Children–Fourth Edition (WISC-IV, Wechsler, 2003).** The WISC-IV is suitable for the assessment of intelligence of children between the ages 6 years and 16 years, 11 months. The scale contains 15 subtests, 10 of which form the core battery and yield a Full-Scale IQ. The 10 core subtests are organized to form four indexes, namely, Verbal Comprehension (VCI), Perceptual Reasoning (PRI), Working Memory (WMI), and Processing Speed (PSI). For the purpose of the current study only the VCI was used (Wechsler, 2003).

The WISC is one of the most widely used measures of intelligence in children, in both clinical and research populations. It has excellent internal consistency, test-retest reliability, criterion validity, and construct validity (Wechsler, 2003). The reliability values of the WISC-IV Australian Composite Scores averaged across age range from 0.85 (Processing Speed) to 0.95 (Full-Scale; Wechsler).

## Procedure

Ethical approval was acquired from the University Ethics Committee and representative education bodies, and written consent for participation was obtained from the parents and adolescents. The MABC-2 and the WISC-IV were individually administered, either at the university or at the family home. The DCDQ-2007 was completed by one of the caregivers. In 89.5% of the cases, this was the mother; in 8.1% of the cases, the father; and in 2.3% of the cases, the grandmother, who in these instances was the caregiver of the child.

## Statistical Analyses

SPSS version 16.0 was used for the statistical analyses. Statistical significance was set a priori at p < .05. To obtain an estimate of the internal consistency of the DCDQ-2007, Cronbach's alpha was calculated. This was calculated on the full scale and for the subscales of the DCDQ-2007 separately. An overall alpha coefficient of 0.70 was applied as the criterion for sufficient homogeneity among test items (Bland & Altman, 1997). Corrected item-total correlations of the DCDQ-2007 were taken into consideration to evaluate the homogeneity of the DCDQ-2007. In addition, correlations between the individual items and intersubscale correlations of the DCDQ-2007 were calculated. Correlations between r = 0.25 and r = 0.5 were considered fair, while correlations ranging from r = 0.5 to r = 0.75 were regarded as moderate to good (Portney & Watkins, 2009).

Concurrent validity of the measures was investigated by (a) conducting Spearman rank order correlations between the DCDQ-2007 and the MABC-2 total and subscale scores and (b) Relative Improvement Over Chance (RIOC) to examine case agreement at the 15th percentile level (Copas & Loeber, 1990). RIOC is indicative of the relative improvement in allocation of adolescents as with or without motor difficulties over chance that can be achieved by using the DCDQ-2007. Outcomes are expressed as a percentage. Otherwise, the RIOC may be interpreted in the same manner as kappa statistics. That is, 100% represents perfect agreement, whereas 0% represents no agreement. The RIOC was favored above kappa, as kappa has the tendency to underestimate agreement, especially when the 2 by 2 tables are unbalanced, which was the case in the current study. RIOC is able to correct for this tendency. Therefore, failure to consider the RIOC may lead to the conclusion that an instrument has poor predictive capability, even though agreement is relatively high (Cairney & Streiner, 2011).

To investigate the discrimination accuracy of the DCDQ-2007, sensitivity and specificity along with the positive and negative predictive values were determined, using the MABC-2 as the criterion standard. A value of 80% is warranted for sensitivity, while for specificity, 90% is preferable (APA, 1985).

# Results

## **Descriptive Data**

Seven adolescents (8.0%) were identified with motor difficulties according to the MABC-2. Of these seven adolescents, five scored at or below the fifth percentile, indicating significant motor difficulties. The DCDQ-2007 in contrast identified 24 adolescents (27.6%) with motor difficulties. The means and standard deviations of the scores on the MABC-2 and DCDQ-2007 are presented in Tables 2 and 3, respectively. The large standard deviations of the scores on both measures indicate large variability in motor performance.

	MABC-2 (Criterion Measure)			
	TTS ª (SD)	Manual Dexterity <sup>a</sup> (SD)	Aiming and Catching <sup>a</sup> (SD)	Balance <sup>a</sup> (SD)
Overall $(n = 87)$	10.6 (2.6)	9.5 (2.5)	11.0 (2.8)	11.4 (3.0)
No motor difficulties $(n = 80)$	11.1 (2.0)	9.8 (2.3)	11.4 (2.5)	11.9 (2.6)
Motor difficulties $(n = 7)$	4.9 (1.1)	6.4 (3.0)	7.1 (2.9)	5.4 (0.8)

# Table 2Mean MABC-2 Scores for Total Sample and IndividualsWith and Without Motor Difficulties

Notes. Cut-off score for motor difficulties used: 15th percentile of the MABC-2. a Standard Score Mean.

# Table 3Mean DCDQ-2007 Scores for Total Sample and IndividualsWith and Without Motor Difficulties

	DCDQ-2007			
	TS ª (SD)	Control During Movement <sup>a</sup> (SD)	Fine Motor/ Handwriting <sup>a</sup> (SD)	General/ Coordination <sup>a</sup> (SD)
Overall $(n = 87)$	61.4 (12.9)	24.7 (5.6)	16.6 (3.7)	20.1 (4.8)
No motor difficulties $(n = 80)$	63.4 (10.3)	25.5 (4.4)	17.1 (3.2)	20.8 (4.0)
Motor difficulties $(n = 7)$	39.0 (18.8)	15.4 (9.1)	11.9 (5.6)	11.7 (5.7)

Notes. Cut-off score for motor difficulties used: 15th percentile of the MABC-2. a Mean score

# Internal Consistency

A high Cronbach's alpha was found for the DCDQ-2007 15 item full-scale ( $\alpha = 0.95$ ). The alpha coefficient did not increase significantly if any of the items were deleted, indicating that none of the items seemed to be problematic for the measure and that the removal of no one item would consolidate the DCDQ-2007. The internal consistency of the subscales was also found to be high:  $\alpha = 0.94$  for the six items tapping Control During Movement,  $\alpha = 0.88$  for the four items that tap Fine Motor/Handwriting, and  $\alpha = 0.85$  for the five General Coordination items. Corrected item total correlations were all significant and positive (p < .001), ranging from r = 0.62 to r = 0.82. All reached a value of > 0.30, which is the minimum value as suggested by Streiner & Norman (1995). Three items (items 1, 2, and 4, all belonging to the Control During Movement subscale) reached a value of r > 0.80.

Fair to moderate positive correlations were found between items, ranging from  $r_s = 0.31$  to  $r_s = 0.86$ , all reaching significance (p < .001). Correlations between items belonging to the same subscale were on average higher than correlations between items from different subscales. Intersubscale correlations of the DCDQ-2007 were all positive and significant. See Table 4.

# **Concurrent Validity**

A fair but significant correlation was found between the total scores on the MABC-2 and the DCDQ-2007 ( $r_s = 0.34$ , p = .001). The correlations between subscales of both measures were all significant, except for the Fine Motor/Handwriting subscale of the DCDQ-2007, which did not show a significant correlation with any of the MABC-2 subscales. See Table 5.

Table 4 Inte	ersubscale Correlation	ns of the DCDQ-2007
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	Control During Movement	Fine Motor/ Handwriting	General Coordination
Control During Movement	$r_{\rm s} = 1.00^{**}$	$r_{\rm s} = 0.52^{**}$	$r_{\rm s} = 0.77 * *$
Fine Motor/ Handwriting	$r_{\rm s} = 0.52^{**}$	$r_{\rm s} = 1.00^{**}$	$r_{\rm s} = 0.73^{**}$
General Coordination	$r_{\rm s} = 0.77^{**}$	$r_{\rm s} = 0.73^{**}$	$r_{\rm s} = 1.00^{**}$

\*\**p* < .01, two- tailed.

# Table 5Spearman's Correlations Between DCDQ-2007 andMABC-2 (TTS and Subscale Scores; n = 87)

DCDQ-2007	MABC-2 (Criterion Measure)			
	TTS	Manual Dexterity	Aiming and Catching	Balance
TS	$r_{\rm s} = 0.34^{**}$	$r_{\rm s} = 0.29^{**}$	$r_{\rm s} = 0.22^*$	$r_{\rm s} = 0.21$
Control During Movement	$r_{\rm s} = 0.42^{**}$	$r_{\rm s} = 0.24*$	$r_{\rm s} = 0.37^{**}$	$r_{\rm s} = 0.27*$
Fine Motor/Handwriting	$r_{\rm s} = 0.02$	$r_{\rm s} = 0.16$	$r_{\rm s} =08$	$r_{\rm s} = 0.02$
General Coordination	$r_{\rm s} = 0.35^{**}$	$r_{\rm s} = 0.28^{**}$	$r_{\rm s} = 0.24*$	$r_{\rm s} = 0.23^*$

\* p < .05, two- tailed. \*\*p < .01, two- tailed.

#### **Discrimination Accuracy**

The numbers of adolescents identified with or without motor difficulties by the DCDQ-2007 and MABC-2 are displayed in Table 6. A RIOC of 81% was obtained (95% CI [45%, 117%]). High sensitivity was found for the DCDQ-2007 (85.7%) due to a low number of false negatives; however, the confidence interval was wide (95% CI [42.0%, 99.2%]). Of the participants without motor difficulties, 77.5% were accurately identified by the DCDQ-2007 (specificity, 95% CI [66.5%, 85.8%]). The positive predictive value was 25.0% (95% CI [10.6%, 47.1%]), as a consequence of the large number of false positives (75.0%) in the identification of motor difficulties according to the DCDQ-2007. A negative predictive value of 98.4% was reached (95% CI [90.3%, 99.9%]).

No complete agreement in the identification of motor difficulties in this adolescent sample was found for the DCDQ-2007 and the MABC-2. The DCDQ-2007 failed to identify one of the adolescents, who had been identified with motor difficulties by the MABC-2. Of the 24 participants that were identified with motor difficulties by the DCDQ-2007, only six were identified by the MABC-2. A plausible explanation for the inconsistencies in test outcome could be that the measures differ in their sensitivity around the cut-off point. However, in the 18 cases where the MABC-2 did not indicate the presence of a motor difficulty, but the DCDQ-2007 did, the total score on both the DCDQ-2007 and the MABC-2 did not fall close to the cut-off point (see Appendix). Evidently, no motor difficulties were indicated by the MABC-2 in these 18 inconsistent cases. This plausible explanation is thereby ruled out.

	MABC-2 (Criterion Measure)*		
	Motor Difficulties	No Motor Difficulties	
DCDQ-2007**			
Motor difficulties	6 (a)	18 (b)	
No motor difficulties	1 (c)	62 (d)	

Table 6Adolescents Identified With or Without Motor DifficultiesAccording to the MABC-2 and DCDQ-2007 (n = 87)

*Notes. a* true positive; *b* false positive; *c* false negative; *d* true negative.

sensitivity [a/(a+c)]; specificity [d/(b+d)];

positive predictive value [a/(a+b); negative predictive value [d/(c+d)].

\* cut-off score: 15th percentile. \*\* cut-off score: ≤ 57.

#### Discussion

A suitable measure to screen for DCD in different age groups is warranted. The original DCDQ is reported to be a promising candidate for this (Schoemaker et al., 2006). In the current exploratory study, the DCDQ-2007 appeared to be a measure with sufficient reliability (internal consistency) and scores on the DCDQ-2007 were related to those on the MABC-2.

The good internal consistency that was found in the current study replicates findings of other studies investigating the DCDQ-2007 (Cairney et al., 2008; Prado, Magalhães, & Wilson, 2009; Wilson et al., 2009) and is consistent with previous studies involving the original version of the DCDQ in younger age groups (Civetta & Hillier, 2008; Loh, Piek, & Barrett, 2009; Schoemaker et al., 2006; Wilson et al., 2000).

Good intersubscale correlations were found for the DCDQ-2007. Items belonging to the same subscale of the DCDQ-2007 were more closely related than items from different subscales. Nevertheless, correlations between all individual items were significant. This suggests that the items measure a similar construct (i.e., motor difficulties) but that items of the different subscales measure a different aspect of this construct. Three items of the Control During Movement subscale of the DCDQ-2007 (items 1, 2, & 4) showed a very close relationship with the total score (r > 0.80). Control during movement might be a general construct underlying all aspects of motor ability. It can be questioned whether a separate subscale are possibly not specific enough for the investigation of a distinct aspect of motor ability. Using factor analysis, however, Wilson et al. (2009) found that Control During Movement emerged as a separate factor, suggesting that the items of this subscale do form a distinctive component.

The RIOC indicated that using the DCDQ-2007 improves the allocation of adolescents with or without motor difficulties over chance alone by 81%. The 95% CI that was found (45%, 117%) exceeds 100% and is thereby unrealistic. Such impossible values are reported to be likely in the case of small sample sizes (Copas & Loeber, 1990). This forms a major concern when conditions with a low prevalence such as DCD are studied, as small samples are often encountered in this instance. The formula to calculate the standard error for RIOC is, therefore, applied to small samples. Alternative tests of agreement exist; however, these are also influenced by small sample sizes (Cairney & Streiner, 2011). When the goal is to evaluate the efficacy of a measure, such as the DCDQ-2007, to identify a smaller subset of possible cases from a larger population of subjects, insight in the improvement of allocation over chance as indicated by the RIOC is of critical importance (Cairney & Streiner, 2011).

The relationship between the total scores on the DCDQ-2007 and the MABC-2 revealed a moderate but statistically significant correlation ( $r_s = 0.35$ ). This supports findings from other studies that correlated the earlier versions of the DCDQ and MABC in younger children (Civetta & Hillier, 2008; Schoemaker et al., 2006; Wilson et al., 2000). Although the correlation is smaller than that found previously between the DCDQ-2007 and the original MABC (i.e., r = -.55) by Wilson et al. (2009), this may be due to the smaller number of participants identified with motor difficulties by the MABC-2 in the current study. The sample in the study of Wilson et al. (2009) consisted of participants of younger age, with a high prevalence of developmental and learning problems (including DCD). Children with learning and attention problems frequently demonstrate motor difficulties (Kaplan, Wilson, Dewey, & Crawford, 1998). Therefore, more variation in motor skill performance might have been present, resulting in a higher correlation between the DCDQ-2007 and the original MABC. The sample used in the current study was considered a typical sample, however, with the number of adolescents identified with motor difficulties in the expected prevalence range (APA, 2000; Tan, Parker, & Larkin, 2001; Wilson et al., 2007). The DCDQ-2007 may prove to be a useful instrument in the screening of motor difficulties in the general population and, therefore, its performance should be examined using such a sample.

A nonsignificant relationship was found between the DCDQ-2007 TS and the Balance subscale of the MABC-2. This result is inconsistent with previous studies using the original versions of both measures, which did reveal significant correlations (Civetta & Hillier, 2008; Schoemaker et al., 2006). Low variability in MABC-2 Balance scores in the current study may have played a role in the nonsignificant relationship that was found. In addition, the DCDQ-2007 does not contain items that specifically represent balance.

Furthermore, nonsignificant correlations were found between the Fine Motor/ Handwriting subscale of the DCDQ-2007 and all of the MABC-2 subscales. Although one would expect the MABC-2 Manual Dexterity- and the DCDQ-2007 Fine Motor/Handwriting subscale to measure similar constructs, the results suggest that the concepts tapped by the DCDQ-2007 Fine Motor/Handwriting subscale are not reflected in the MABC-2. In the DCDQ-2007, three of the four items in the Fine Motor/Handwriting subscale specifically concern handwriting. The MABC-2, in contrast, only contains one item that asks the adolescent to use a pencil (i.e., the Drawing Trail). The issue is whether the skill tested here is representative of handwriting. It has been reported previously that the original MABC does not identify children with handwriting difficulties (Geuze et al., 2001). In contrast, Loh et al. (2009) did find a relationship between the Fine Motor/Handwriting subscale of the original DCDQ and the bimanual dexterity subscale of the McCarron Assessment of Neuromuscular Development (MAND), supporting concurrent validity.

To be useful as a screening tool, the DCDQ-2007 should, for a large part, replicate the identification of motor difficulties in adolescents according to a standardized motor test. Considering that the DCDQ-2007 should only be used for the initial screening, before assessment with a more detailed motor test like the MABC (Schoemaker et al., 2006), sensitivity seems to be more important than specificity (Wilson et al., 2009). Sensitivity was found to be satisfactory and reached a level (85.7%) similar to what is reported in the manual for this age band (88.5%).

The prevalence of motor difficulties in the sample needs to be considered when investigating sensitivity (Goodman & Scott, 1999), as a high prevalence increases the sensitivity and positive predictive value (Loh et al., 2009). In the current study, the prevalence of motor difficulties at or below the fifth percentile as indicated by the MABC-2 (i.e., 5.7%) was in the expected prevalence range. A somewhat lower than expected prevalence was found (8.1%) in reference to the 15th percentile criterion; however, this number is in the same range as reported previously in literature. Overall, the prevalence in the current study was considered typical for the occurrence of the condition, that is, low. The small sample size and especially the low occurrence of motor difficulties do not appear to have influenced sensitivity largely; however, they are expected to have played an important role in the wide CI that was found for sensitivity. The lower bound of 42.0% indicates that a large portion of the CI fell below the criterion for acceptability of 80%, which undermines the credence of the sensitivity inferences. Good sensitivity is also reported in previous studies investigating the DCDQ-2007 using larger samples, however, with improved sensitivity compared with the original DCDQ (Wilson et al., 2009).

This suggests that there is no reason to believe that a similar sensitivity value with a narrower 95% CI would not be found with larger samples. Further studies with larger samples are warranted to confirm this result.

Agreement in identification of motor difficulties between the DCDQ-2007 and the MABC-2 was not satisfactory, as the DCDQ-2007 seemed to carry compromised specificity. In the current study, a specificity of 77.2% was found for the DCDQ-2007, which is consistent to what is reported in the manual for this age band (75.6%; Wilson et al., 2007). This is lower than the preferred specificity level of 90% (APA, 1985). Using the DCDQ-2007, more adolescents were identified with motor difficulties (27.5%) as expected according to the cut-off score (15%) in the current study, which is likely to be due to the compromised specificity. Other studies demonstrated that parents generally express more concerns than needed about their children's motor behavior, exaggerating the prevalence of DCD (Green et al., 2005; Kroenke 2001; Loh et al., 2009; Schoemaker et al., 2006). The rating of motor performance by parents is influenced by other difficulties. In contrast to false negatives, which were nearly absent in the current study (n = 1), this would cause false positives, influencing specificity in a negative way.

One might question whether the criterion for specificity (i.e., 90% agreement) is too stringent in case of the DCDQ-2007. Wilson et al. (2009) intentionally favored a higher sensitivity over specificity, which is supported by the current results; specificity is sacrificed to obtain a high sensitivity. This high sensitivity makes the DCDQ-2007 a good candidate for the initial screening for motor difficulties in adolescents. The false positives would likely be corrected in later confirmatory diagnostic testing with a norm referenced standardized motor test, bringing down the prevalence, which is reflected in the lower prevalence of adolescents identified by the MABC-2 in the current study.

The negative predictive value of 98.4% shows that almost all adolescents identified as without motor difficulties by the DCDQ-2007 were identically allocated by the MABC-2. Only in one instance, the DCDQ-2007 failed to identify an adolescent with a score between the fifth and the 15th percentile on the MABC-2. This adolescent scored below the 15th percentile on the MABC-2 Balance subscale only (and above the 15th percentile on the other two MABC-2 subscales), the subscale for which no significant correlation with the DCDQ-2007 TS was found.

The positive predictive value indicated that 25.0% of the adolescents identified with motor difficulties by the DCDQ-2007 had motor difficulties according to the MABC-2. A low positive predictive value was also found by Schoemaker et al. (2006; 44%). This again may imply that parents express more concerns than needed about their children's motor performance, an issue inherent to questionnaire-based screening, which generally exaggerates a condition owing to over-endorsement bias (Kroenke, 2001).

The current study did not find complete agreement in the classification of motor difficulties by the MABC-2 and the DCDQ-2007. Different levels of discriminative ability around the cut-off points were ruled out as a cause of the disagreement. The existence of discrepancy in test outcomes might indicate that the measures assess overlapping but distinct constructs. This is also reflected in the low but significant correlations between both measures. The MABC-2 is designed to assess Criterion A of the DSM-IV criteria for DCD and the DCDQ-2007 is intended to assess

Criterion B (Wilson et al., 2009). For this reason a very high correlation between the two measures should not be expected. Parent responses on the DCDQ-2007 are suggested to represent performance (i.e., how a participant acts in his or her natural environment). This is partly but probably not entirely reflected in the isolated observation from a single moment of the MABC-2, which represents motor capability (Civetta & Hillier, 2008). The subjectivity of parent report might play a role. Different measures assess different aspects of motor performance; a multilevel approach to motor assessment has been recommended (Schoemaker et al., 2006). The DCDQ-2007 is not designed to replace the clinical assessments of individuals referred for motor difficulties (Schoemaker et al., 2006; Wilson et al., 2000).

#### Limitations of Study

Results of this study must be interpreted in light of the small sample size, as it was an exploratory study utilizing an existing data set. Future studies are warranted, with a larger sample and a greater spread of scores on the measures to enable the potential for more compelling conclusions about the sensitivity of the DCDQ-2007 and to reduce the 95% CIs.

The original DCDQ is reported to have potential value for initial communitybased screening for motor difficulties (Wilson et al., 2000). To analyze the performance of the DCDQ-2007 with this purpose in mind, a sample that closely resembles the general population is preferable, as was the case with the current study. The prevalence of motor difficulties in the current sample according to the MABC-2 was within the prevalence range reported in literature (APA, 2000; Tan et al., 2001; Wilson et al., 2007). If the sample size was increased largely, the prevalence of motor difficulties). Due to the low prevalence of the condition in general, positive predictive value will always be negatively affected. Results of the current study were in line with expectations and comparable to earlier research about the original and revised measures. Most adolescents were correctly identified, and misidentifications mainly concerned false positives.

One drawback of the validity of the DCDQ-2007 is the factor structure of the measure. Issues related to the factor structure of the original DCDQ have not been resolved. Therefore, until further evidence concerning the factor structure of the revised questionnaire arises, the DCDQ-2007 should not be used to discriminate between specific kinds of motor difficulties (fine or gross). Instead, it is best used to identify general motor difficulties (Cairney et al., 2008). Although the problems with the factor structure certainly are a limitation of the questionnaire, this does not mean that the questionnaire is to be discarded. The original purpose of the questionnaire should, however, be clearly kept in mind: initial screening for motor difficulties. As argued by Borsboom, Mellenbergh, & van Heerden (2004), the focus of validity research should return back to the original, mere question of whether one measures what one intends to measure. Beyond a confirmed factor structure, reliability, and predictive adequacy are also important properties. Which psychometric testing procedure is preferred depends on the specific situation, goals, and resources in the form of time and money that are available. Good internal consistency, sensitivity, and predictive value were found, providing evidence that the DCDQ-2007 measures what it intends to measure.

#### **Recommendations for Future Research**

Only two studies have been performed regarding the factor structure of the DCDQ-2007, and as mentioned before, there appear to be some issues with the factor structure that warrant further investigation. Test-retest and interrater reliability should be investigated to rule out that errors in the identification of motor difficulties may have been a function of unreliability in assessment. As no clear relationship between the Fine Motor/Handwriting subscale of the DCDQ-2007 and the MABC-2 was found, this subscale in particular should be further investigated.

# Conclusion

To our knowledge this study presents the first evaluation of the DCDQ-2007 for the identification of motor difficulties in a community-based sample of adolescents. The low specificity of the DCDQ-2007 indicates that it cannot be used as the only measure to identify motor difficulties. The sensitivity of the DCDQ-2007 was found to be high, but the CI was wide, so caution should be taken with the interpretation of this seemingly positive result.

The DCDQ-2007 did seem to pick up most adolescents with probable motor difficulties, and a positive correlation with scores on the MABC-2 was found. Although preliminary results concerning psychometric properties are rather promising, the underlying factor structure of the DCDQ-2007 has not yet been identified with consistency. As mentioned by Wilson et al. (2007), it should be kept in mind that the DCDQ-2007 alone cannot be used to diagnose DCD. It can only give an indication of DCD. Further assessment with a more detailed motor test is warranted for those identified with motor difficulties by the DCDQ-2007.

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# Appendix

DCDQ: TS	Difference Between Cut-Off and TTS DCDQ (i.e., TTS—57)	MABC-2: TTS	Difference Between Cut-Off and TTS MABC-2 (i.e., TTS—67)
32	-25	84	17
46	-11	75	8
41	-16	73	6
46	-11	83	16
52	-5	80	13
57	0	89	22
41	-16	85	18
51	-6	83	16
41	-16	74	7
54	-3	86	19
57	0	83	16
56	-1	72	5
54	-3	68	1
46	-11	94	27
40	-17	90	23
54	-3	77	10
47	10	77	10
50	_7	81	14

#### TS for Cases Identified With Motor Difficulties by the DCDQ-2007 Only

*Note.* Cut-off scores for classifying a child as having motor difficulties are TTS DCDQ-2007  $\leq$  57; TTS MABC-2  $\leq$  67 as per test manuals.

#### DISCUSSION

For many years, motor coordination problems in children were overlooked, as there was a widespread belief that these children, who did not perform well at sports and/or had difficulty with writing, would eventually outgrow these difficulties. Numerous studies now show increased risk for various negative outcomes in children with motor impairment, including executive function, academic, and internalising problems. There is also evidence that the motor skill difficulties and associated problems do not disappear in older years. Thus, it is clear that motor competence is crucial for successful functioning in a number of domains and that these difficulties should not be ignored.

Although knowledge regarding the implications of motor coordination problems has increased, misconceptions prevail. Therefore, continued research regarding the relationships between motor coordination and its cognitive, academic, and psychosocial correlates is needed to promote further awareness and recognition in the educational and health settings. For example, there are important social and cognitive developmental changes from childhood to adolescence, yet studies of motor coordination and the associated cognitive and psychosocial domains are limited in adolescent samples. Also, much of the research investigating the association between these areas has been carried out in groups of children with motor impairment. It has been noted that although these studies provide a basis for understanding these relationships, findings from these samples may not extend to a typically developing population (Pangelinan et al., 2011). Also, the heterogeneity of motor skill impairment is well known, however, it is not often considered when examining the relationship between motor difficulties and various outcomes. Therefore, investigation of the possibility of specific relationships between these areas has been frequently disregarded.

To address these knowledge gaps, this thesis presented a series of published papers investigating the relationship between motor coordination and the psychosocial, executive function, and academic domains in a normative adolescent sample. The results of these studies highlight that the importance of motor coordination in relation to these functional areas also extends to the adolescent years. This has significant implications, particularly when considered in terms of the psychosocial functioning of adolescents, as there is evidence that they are at greater risk, socially and emotionally, when compared with younger children (Skinner & Piek, 2001). Using a normative sample, the findings from these studies also demonstrated that the relationships between these areas are not just confined to individuals with movement difficulty. Rather, they appear to be located on a continuum, ranging from individuals who demonstrate difficulty in the motor and associated cognitive and emotional areas to those individuals who perform well across these domains.

In their study on the relationship between motor and cognitive control, Roebers and Kauer (2009) suggested that normative correlational research may shed light on the specific nature of the relationships between these domains, whilst also suggesting shared underlying processes. Although the association between motor ability and academic and emotional areas has been established, little has been discussed in terms of the underlying mechanisms explaining the relationships. The findings from this thesis revealed specific links between certain motor components and aspects of cognitive and emotional functioning. For example, aiming and catching skills (and not balance or manual dexterity) demonstrated a significant association with working memory and the academic areas of word reading and numerical operations. This leads to hypotheses regarding shared cerebellar mechanisms underlying the relationship found between these areas. Additionally, the paper examining the relationship between motor coordination and psychosocial correlates provided partial support for previous suggestions of a specific link between balance control and emotional functioning, which has also been explained in terms of neuronal networks common to both these areas (Stins & Beek, 2007).

It is plausible that the specific associations found between motor coordination, and the cognitive and emotional domains may be a result of shared aetiological processes. The cerebellar contributions to motor coordination and postural control are well established. There is also growing behavioural evidence suggesting cerebellar dysfunction in DCD. For example, these children demonstrate difficulties in motor adaptation, postural control, and timing of movements (see Zwicker, Missiuna, & Boyd, 2009 for a review). Recently, neurobiological research has provided some support for this suggestion. Using functional magnetic resonance imaging (fMRI), Zwicker and colleagues (2011) mapped brain activity during skilled motor practice in a small sample of children with DCD. Compared with typically developing children, results suggested that children with DCD may be underactivating the cerebellar-parietal and cerebellar-prefrontal networks (Zwicker et al., 2011).

Regarding the relationship between the motor and cognitive domains, Diamond (2000) argued that development in these areas may be fundamentally interrelated, which is strongly supported by functional neuroimaging findings of the close co-activation of the cerebellum and the prefrontal cortex during performance on cognitive tasks. It is further suggested that the prefrontal cortex is important for movement given its close communication with subcortical and cortical regions directly involved in motor functions (Diamond, 2000). In terms of the underlying processes of academic domains, cerebellar contributions to academic areas such as reading have also been found using fMRI analysis (Baillieux et al., 2009). Indeed, Nicolson and colleagues (2001) proposed the cerebellar deficit hypothesis when attempting to explain the motor and reading problems displayed by individuals with dyslexia. There is also recent evidence, using repetitive transcranial magnetic stimulation, that the cerebellum is implicated in the regulation of emotion and mood (Schutter & van Honk, 2009).

Interestingly, recent neurobiological research on DCD highlighted the constellation of cognitive and affective symptoms often associated with the disorder, and suggested that DCD may be closely related to 'cerebellar cognitive affective syndrome' (CCAS; characterised by executive dysfunction, visual-spatial and linguistic impairments, and affective dysregulation which results from acquired or developmental cerebellar disorder) (Mariën, Wackenier, De Surgeloose, De Deyn & Verhoeven, 2010). Mariën and colleagues (2010) presented structural and functional neuroimaging as well as neuropsychological findings of a young patient with diagnosed DCD. Findings suggested disruption of the cerebello-cerebral network important for the execution of planned actions, visuo-spatial cognition, and affective regulation. Consequently, it was hypothesised that insufficient maturation or underdevelopment of the cerebro-cerebellar network may account for the constellation of motor, cognitive and affective symptoms seen in DCD. Future neurobiological research in larger samples is imperative.

Although there is some evidence to suggest that the relationship between the motor, cognitive and emotional domains may be explained by biological processes, it is important not to rule out the environmental contributions to these relationships. For example, it is also plausible that the negative social and interpersonal

experiences associated with motor problems places one at increased risk for anxiety and depressive symptoms. This possible causal relationship between motor coordination and emotional outcomes was the focus of the first two papers presented in this thesis.

The first paper of this thesis provided a preliminary investigation of the causal relationship between motor skills and psychosocial functioning by presenting a longitudinal study of the predictive ability of early motor development on later school-age anxiety and depressive symptomatology. It was found that the stability of early gross motor development, from infancy to early childhood, predicted later anxiety and depressive symptomatology. Therefore, the first paper presented in this thesis lends some support to the hypothesis that motor problems are causally linked to internalising symptoms.

The Environmental Stress Hypothesis, proposed by Cairney and colleagues (2010), provides a causal framework for understanding the internalising problems often displayed by individuals with movement skill difficulties. Children and adolescents with movement problems are faced with everyday stressors in the home, classroom, and playground environments. Not only do they experience ongoing difficulty in the physical interactions with the world around them (interfering with activities of daily living such as self-care) (e.g., Summers, Larkin, & Dewey, 2008) but are also at increased risk for peer relationship difficulties (e.g., social withdrawal and exclusion) (Livesey, Lum Mow, Toshack, & Zheng, 2011; Smyth & Anderson, 2000) and academic failure (Alloway, 2007). Cairney et al. emphasise the core role that these negative environmental experiences play in the aetiology of internalising problems in DCD. It was suggested that these 'secondary stressors' then lead to negative self-appraisals, which in turn, lead to increased risk for anxiety and depression (Cairney et al., 2010).

Thus, the second paper provided an examination of this hypothesis by testing the mediating role of self-perceptions on the relationship between motor coordination and emotional functioning, specifically, anxiety and depressive symptomatology. In an adolescent normative sample, it was found that motor coordination did not directly impact emotional functioning, rather, motor coordination had an indirect relationship with emotional functioning through the mediating influence of selfperceived competence in peer-relation, school, and physical domains. Therefore, this paper provided some support for the Environmental Stress Hypothesis, which may

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have important implications when considered in the context of the prevention and intervention of negative emotional outcomes in individuals with movement difficulties. The finding that self-perceptions play a significant role in explaining the relationship between motor coordination and internalising problems supports previous research on competency-based models of child and adolescent anxiety and depression (Cole, Martin, & Powers, 1997; Smári, Pétursdóttir, & Porsteinsdóttir, 2001). It is also important to note the influence of self-perceptions on social and physical participation, as this may provide further information on the possible pathways linking motor coordination to emotional outcomes.

Using Harter's competence motivation theory, Skinner and Piek (2001) suggested that if people perceive themselves to be physically incompetent, they will have less motivation to practice motor skills, resulting in reduced participation in the physical and social domains. Indeed, there is accumulating evidence for a link between motor problems and sedentary behaviour (Rivilis et al., 2011) and studies have found that negative self-perceptions contribute to the decreased levels of participation shown by individuals with movement problems (Cairney et al., 2005; Poulsen, Ziviani, & Cuskelly, 2008). Participation in structured leisure activities, such as team sports, may provide opportunities for supportive interpersonal relationships and may consequently, promote a sense of affiliation and positive psychological functioning. Thus, compared with their well coordinated counterparts, individuals with movement problems may not be provided with the same opportunity to enhance their emotional well-being given evidence of social and physical withdrawal in this population. In fact, team sports participation has been found to significantly mediate the inverse relationship between loneliness and physical coordination (Poulsen, Ziviani, Cuskelly, & Smith, 2007). Furthermore, it is possible that adolescents with motor difficulties may be at a particular risk for poor emotional outcomes considering the importance placed on belongingness during this developmental period.

In addition to the social benefits of participation, research has noted the possible role of physiological mechanisms underlying the association physical activity and mental health outcomes. For example, Nabkasorn et al. (2006) found that physical exercise improved depressive symptoms and reduced neuroendocrine stress hormone levels in an adolescent female sample. It was suggested that although the mechanisms mediating the relationship between exercise and improved mental

health are still unclear (e.g., other factors such as an increase in self-efficacy and mastery may also contribute to improvement in depressive symptoms), improvements in the hormonal response to stress may play a role in the psychological benefits of exercise.

Recently, a number of studies have also pointed to the physiological benefits of physical activity on cognition, including executive functions (Sibley & Etnier, 2003). Physiological mechanisms underlying the relationship between physical activity and cognition include exercise induced physical changes such as increased cerebral blood flow, structural changes in the central nervous system, as well as alterations in brain neurotransmitters and arousal levels (Sibley & Etnier, 2003). Sibley and Etnier (2003) also noted the learning experiences associated with physical activity (e.g., many sporting games require increased cognitive engagement, promoting skills which may then transfer to cognitive tasks) and that these may also be important in enhancing cognitive development. Given the role of negative selfperceptions in explaining the decreased participation levels of individuals with movement difficulties, it is plausible that these negative self-perceptions may ultimately have an indirect negative impact on cognitive functioning through the mediating role of physical inactivity. Thus, this provides a framework which links the psychosocial and cognitive difficulties often displayed by those with motor coordination difficulties.

Also, there is growing evidence for cognitive dysfunction (including executive functioning deficits) in depressive and anxiety disorders (Castaneda, Tuulio-Henriksson, Marttunen, Suvisaari, & Lönnqvist, 2008; Cataldo, Nobile, Lorusso, Battaglia, & Molteni, 2005; Günther, Holtkamp, Jolles, Herpertz-Dahlmann, & Konrad, 2004). Although the direction of this relationship remains unclear, there is some evidence that executive functioning deficits are symptomatic of current anxiety or depression (Micco et al., 2009). Micco and colleagues (2009) found that in a sample of children at increased risk for depression or anxiety (because of parental depressive or anxiety disorder), current anxiety or depression in these children was associated with poor performance on executive function measures. Conversely, children at increased risk for depression or anxiety, but without current symptoms, did not show poor executive functioning. Therefore, it was argued that executive function difficulties appear to be symptomatic of current anxiety or depressive problems, rather than serving as trait markers for developing these disorders. This is supported by Eysenck and colleagues' (2007) attentional control theory which attempts to explain the effects of anxiety on cognition. It is proposed that anxiety impairs the processing efficacy needed for cognitive performance by decreasing attentional control and increasing attention to threat-related stimuli. Taken together, these findings suggest that emotional functioning may have a mediating role in the relationship between motor coordination and executive functions, which is important when considered in the context of executive function difficulties reported in the DCD literature.

The third paper of this thesis extended previous research by demonstrating that the important relationship between motor coordination and executive functions also exists in a normative adolescent sample. The results revealed significant positive relationships between motor coordination and the executive functions of working memory and inhibition, supporting findings from samples of children with motor impairment (e.g., Alloway, 2007; Michel et al., 2011). These significant results remained after controlling for the potentially confounding effects of ADHD symptomatology. This is important as Wassenberg et al. (2005) questioned whether the relationship between cognitive and motor performance is direct, or mediated by other factors such as attention. In fact, Wassenberg et al. argued against a global relation between the motor and cognitive domains, supported by their findings of a nonsignificant association between motor performance and an estimate of general cognitive performance in a sample of 5 to 6 year old children. Rather, their results demonstrated more specific links between the motor and cognitive areas. Findings from a normative study of 7 year old children also revealed significant relations between some motor areas and aspects of executive function, but not others (Roebers & Kauer, 2009). The results from this thesis also demonstrated specific associations between aspects of cognitive and motor performance and extended these previous studies by revealing these associations in an older age-group.

It has been noted that the overlap between the motor and cognitive domains may be explained by shared underlying processes, specifically, possible domaingeneral control processes (Roebers & Kauer, 2009). Conversely, it has been argued that skilled motor performance involves complex cognitive processes (Diamond, 2000). Causal information regarding this relationship is limited, however, longitudinal studies examining the predictive relationship between motor and cognitive development have suggested that movement experiences in early childhood that facilitate environmental interactions are important for cognitive development (Piek et al., 2008). It is clear that further research is needed to clarify the nature of this relationship as well as elucidating possible mediating factors, including the impact of emotional functioning. Understanding the relationship between motor and executive functioning is crucial, particularly given the functional implications of executive function deficits, which was the focus of the fourth paper presented in this thesis.

The relationship between motor ability and academic outcomes has been established in the literature, yet little has been examined regarding the factors that may mediate the association between these areas. Previous findings from DCD research have suggested a possible indirect relationship between motor coordination problems and academic underachievement (Alloway, 2007). Specifically, these studies have found that in children with DCD, poorer performance in reading and mathematics may be explained in terms of their difficulties in storing and processing information (i.e., working memory) (Alloway, 2007; Alloway & Warner, 2008). Thus, the fourth paper of this thesis presented a mediating model of the relationship between motor coordination, the executive function of working memory, and academic achievement in a normative sample of adolescents, whilst controlling for confounding factors such as verbal ability and SES. Importantly, the study also controlled for ADHD symptomatology and allowed for an investigation of specific relationships between working memory, academic outcomes, and various motor components which is important given increasing evidence for specific links between these areas. Structural equation modeling revealed that the association between motor coordination and academic achievement may be best understood in terms of a mechanism whereby motor coordination (specifically, aiming and catching skills) has an indirect impact on academic outcomes (specifically, word reading and numerical operations) via working memory. In addition to pointing to possible shared cerebellar processes, the results also suggested that improving aiming and catching skills may have a beneficial effect on working memory and in turn, academic achievement. This is concerning for individuals with movement difficulties who tend to avoid participating in such tasks and therefore, may not have the same opportunity to enhance these cognitive areas. Consequently, these results also provide invaluable information when considering intervention for these children.

#### Implications for prevention, clinical intervention, and classification systems

The results of the research presented in this thesis demonstrate a number of significant findings highlighting the importance of motor coordination in relation to cognitive and psychosocial areas. Specifically, the results suggest that poor motor coordination may place an individual at greater risk for executive function and academic difficulties, poor self-perceptions, and anxiety and depressive symptoms. It seems crucial to increase awareness of these associations in the community as this may ultimately have important practical implications. Firstly, identification of individuals with movement difficulties is imperative. However, identifying motor difficulties from a community-based perspective can be time consuming and expensive. The fifth paper of this thesis provides information on the suitability of the revised Developmental Coordination Disorder Questionnaire (DCDQ) as a community-based screening tool for motor difficulties in adolescents. The DCDQ demonstrated high internal consistency and concurrent validity. The DCDQ also met recommended standards for sensitivity suggesting that it may be a suitable and efficient method for screening large number of individuals for motor problems. However, it is important to note that the DCDQ did not meet recommended standards for specificity. Therefore, although the measure may prove valuable for initial community-based screening, further detailed assessment is warranted for those identified as at risk of having motor problems according to the DCDQ.

Secondly, in the assessment of individuals identified with movement problems, the possibility of associated emotional and cognitive difficulties should not be overlooked. In addition, when an individual presents with problems in the cognitive or emotional domain, the results reported this thesis suggest that they should also be screened for possible motor difficulties. Similarly, the results also have important implications for research, for example, experimental studies employing motor impairment samples should consider the impact of the cognitive domain in terms of task demands that may affect performance in this sample, and vice-versa for studies on cognitive impairment. Regarding intervention, the interrelation between cognitive and motor development suggests that intervention in one domain may support development of the other domain. It is plausible that enriched motor or cognitive experiences may promote improvements across the two domains as well as influencing the development of neural mechanisms underlying these areas (Pangelinan et al., 2011). This is in line with research showing that increased physical activity may enhance cognitive development (Sibley & Etnier, 2003).

When addressing the possible psychosocial implications of motor difficulties, it appears that self-perceived competencies may play an important role in buffering anxiety and depressive symptoms. For example, increasing self-perceived competency in the social domain may have a positive impact on the emotional functioning of individuals with movement difficulties. Targeting self-perceived competencies in treatment is also important given the contribution of negative selfperceptions to the decreased participation levels often shown by this population.

Finally, most research investigating these relationships has been carried out in atypical samples. Davis, Pitchford, and Limback (2010) pointed out that investigating the interrelations between motor and cognitive domains in typically developing children is integral in order to be able to then identify deviations from the normal pathway in atypical populations, as well as to be able to differentiate between cases of developmental delay (i.e., similar strength of association between the areas but at a depressed performance) versus developmental deviance (i.e., normal/advanced performance in one domain with depressed performance in the other domain). Therefore, the normative findings presented in this thesis may also have important implications in the definition of developmental disorders. Dyck, Piek, Kane, and Patrick (2009) argue that the use of discrepancy criteria when defining developmental disorders neglects to consider how relationships between ability domains change across age cohorts. Given that previous studies examining the relationships between motor and cognitive development have involved younger samples or a mixed sample of children and adolescents, the results from this thesis, which present an examination of relationships between these domains in adolescence, adds to our current understanding of normal development in these areas. Dyck et al. (2009) state that if relationships between abilities are not constant across age-groups in normal development, definitions of developmental disorders need to be amended to reflect this. Consequently, the investigation of these relationships in a normative sample of adolescents represents a significant strength of this thesis. However, it is important to note that the cross-sectional nature of these studies impedes the ability to directly examine developmental effects.

#### Strengths

Whilst most of the studies presented in this thesis were cross-sectional in nature, the first paper employed a longitudinal design to investigate the relationship between early motor development and later emotional outcomes. This represents a significant strength, given the paucity of longitudinal research in the area. Importantly, the first paper provided an indication of the possible causal relationship between motor ability and emotional functioning and thus, provided empirical support for investigating the impact of motor coordination on emotional outcomes in the second paper, which employed a cross-sectional design.

Although there is much research suggesting an important relationship between motor coordination and the areas of academic achievement and emotional functioning, very few studies have provided a direct investigation of the possible pathways through which motor coordination impacts these areas. Therefore, the mediational models presented in this thesis add valuable information to the literature as the findings revealed that self-perceptions and working memory, respectively, are important factors when understanding the internalising problems and academic underachievement often displayed by individuals with DCD.

In addition, much of the existing literature has failed to take into account confounding factors such as ADHD symptomatology as well as the impact of heterogeneity of motor skill impairment on these associated areas. The studies from this thesis control for such confounding factors and also allow for the investigation of specific relationships between different motor skills components and the emotional and cognitive domains. Furthermore, these investigations were carried out using widely used and accepted measures of motor coordination, academic, psychosocial and executive functioning, with established psychometric properties. **Limitations** 

# Cross-sectional research cannot provide information on causality and the directional nature of the relationships. In addition, although the first paper presented a longitudinal study of the association between motor and emotional development, causality was not established, as it is difficult to measure internalising symptoms such as anxiety during infancy. Consequently, this thesis was unable to determine whether the motor, cognitive, and emotional domains coexist through biological factors or are better explained through environmental contributions. Longitudinal research over multiple time points can also provide a direct investigation of the

developmental changes between these relationships. Therefore, the studies were also unable to investigate possible differences in the nature and strength of these associations across development.

The studies presented in this thesis accounted for potentially confounding variables whilst also investigating possible mediating factors in the relationship between motor coordination and cognitive and emotional domains. However, there are other variables, not measured in these studies, which may have influenced the relationships. A factor that was not measured in this thesis is the role of physical activity. There is increasing evidence that physical activity is important for both cognitive and emotional development. Given the strong links between motor coordination and physical participation, this suggests that physical activity may provide a significant pathway linking motor coordination to emotional and cognitive areas.

Finally, the sample size of the studies was sufficient to detect important relationships. However, future research could benefit from examining these relationships with a larger sample. In particular, given the small sample size (87 participants) of the study investigating the psychometric properties of the revised DCDQ, it is noted that the results should be interpreted with this in mind. The exploratory study provides some psychometric evidence for the suitability of the revised DCDQ in screening for motor difficulties, although larger studies with a greater spread of scores are warranted.

#### **Future directions**

Given that causal evidence for the relationship between motor coordination and the associated emotional and cognitive domains is scarce, future longitudinal research is needed. These studies should provide measurements across the domains from baseline and at multiple points over time in order to provide information on directionality as well as possible developmental changes in the nature of the relationships. Longitudinal research is also necessary to investigate the role of environmental factors in influencing development in these domains, in both typically developing and atypical populations. In particular, it would be interesting to determine the mediating role of physical activity in enhancing cognitive and emotional development in individuals with movement difficulties.

Intervention studies investigating the effects of physical activity training programs may also prove useful. For example, it would be interesting to examine the

benefits of physical activity on the self-perceptions and emotional functioning of individuals with movement difficulties. Such an intervention may work to increase a sense of mastery which may in turn, promote positive self-perceived competence and consequently, emotional functioning. Furthermore, in addition to the possible physiological mechanisms through which physical activity may enhance psychological functioning, there may be social mechanisms that play a role in mediating the relationship between physical activity and emotional functioning in individuals with movement problems. Therefore, it may also be important to investigate the differential impact of various types of physical interventions on emotional functioning, for example, individual versus group format. It is plausible that participation in team activities may prove more beneficial than individual activities given the increased social isolation reported in the DCD population. Furthermore, given research findings of a significant association between motor coordination and anxious/depressed behaviour in preschool age children (Piek et al., 2008), this highlights the importance of future research on early intervention for these children which may work to avoid possible future internalising problems.

Similarly, the effects of different types of physical activity on cognitive outcomes, such as working memory, appears to be an important area for future DCD research, given evidence that complex forms of physical exercise are more likely to enhance executive functions than simpler exercises (Budde, Voelcker-Rehage, Pietrabyk-Kendziorra, Ribeiro, & Tidow, 2008). Also, given that working memory has shown to have an important mediating role in the relationship between motor coordination and academic outcomes, it is plausible that intervention programs aimed at training working memory in individuals with motor difficulties may also work to improve academic areas.

Finally, although there is suggestion that motor, cognitive, and emotional domains rely on the development of the same neural mechanisms, there are very few neurobiological studies investigating brain development with respect to the relationships between motor, cognitive, and emotional development. This represents an important knowledge gap in need of future research.

#### Conclusions

The series of studies presented in this thesis highlight the significance of motor coordination in relation to psychosocial, cognitive, and academic areas. From the findings, it is clear that motor competence is important for effective functioning in cognitive and emotional domains and that the relationships between these areas continue into adolescence. The studies investigated these relationships from a normative perspective which provides an important means for comparison with individuals with developmental disorders. Mediational models suggested that, for individuals with movement difficulties, negative self-perceptions and working memory deficits may be important in explaining the increased risk for internalising problems and academic underachievement, respectively. This has important practical implications. Finally, the specific relationships found between certain motor components and aspects of cognitive and emotional functioning may point to possible shared neural processes underlying these relationships.

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## **APPENDIX** A

Pearsall-Jones, J. G., Piek, J. P., Martin, N. C., Rigoli, D., Levy, F., & Hay, D. A. (2008). A monozygotic twin design to investigate etiological factors for DCD and ADHD. *Journal of Pediatric Neurology*, *6*(3), 209-219.

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## **Original Article**

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# A monozygotic twin design to investigate etiological factors for DCD and ADHD

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Received 27 February 2008 Revised 9 April 2008 Accepted 27 April 2008

Abstract. The high level of comorbidity between Developmental Coordination Disorder (DCD) and Attention Deficit Hyperactivity Disorder (ADHD) suggests that these disorders may have a shared etiology. We used a co-twin control design to study monozygotic (MZ) twins concordant and discordant for DCD and ADHD. In a total of 922 sets of MZ twins, 866 sets were eligible. We found equal numbers of DCD concordant and discordant sets; more ADHD concordant than discordant sets; nine sets in which both twins met criteria for DCD+ ADHD; 773 sets of twins did not meet criteria for either DCD or ADHD. The only significant sex difference between groups was for ADHD discordant sets, with more males than females. For DCD groups there were no significant sex differences, with slightly more girls than boys affected in both groups. There was a greater number of oxygen perfusion complications in DCD affected than unaffected twins, suggesting that, given equal numbers of DCD concordant and discordant sets and a similar number of DCD girls and boys, the role of pre- and perinatal environmental factors is stronger in the etiology of DCD than in ADHD. Factors such as placental difficulties and hypoxia have also been related to cerebral palsy, which suggests that DCD may fall on the upper end of a continuum of movement disorder that includes cerebral palsy. The results suggest different etiological pathways for DCD and ADHD. Second born twins were at greater risk for oxygen perfusion difficulties in sets concordant for DCD, ADHD, and unaffected for either.

Keywords: Monozygotic twin, genetic, discordant, concordant, ADHD, DCD, etiology

#### 1. Introduction

Comparisons between monozygotic (MZ) and dizygotic (DZ) twins for concordance for disorder provide a strong indication of the role played by genes in phenotypic variation, as the greater the concordance in MZ relative to DZ twins, the greater the role attributable to genes [1,2]. A logical extension of the classical twin study approach is to explore MZ twins discordant for disorder. This co-twin control, or twin differences design [3], in conjunction with comparisons between MZ twins concordant for disorders, enables us to better examine the interaction between genetic, epigenetic and environmental factors in bio behavioral and genetic disorders.

This stated, interpretations drawn from twin research are traditionally based on the assumption that MZ twins are genetically identical, whereas DZ twins share on

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average only half their genes. Although there is generally a higher concordance rate for wellness and illness in MZ than DZ twins, there is seldom 100% concordance. More sophisticated technology in molecular genetics and increased knowledge of epigenetic processes have challenged the assumption that MZ twins are always genetically identical, and it has been recognized that developmental discordance in MZ twins is not always the result of environmental effects [4,5]. In some instances there is discordance for phenotype in twins with the same genotype.

Reasons for prenatal discordance are numerous [6]. These may arise from post zygotic genetic effects, for instance, unequal splitting of the zygote [7]; different patterns of methylation maintenance [8]; and tissue specific patterns of methylation [6]. They may also arise from differences in placental placement and nutrition [9]; whether the twins are MZ monochorionic or dichorionic or MZ monochorionic monoamniotic or diamniotic [9]; MZ monochorionic twins suffering ischemic damage due to acute or chronic episodes of hemodynamic instability as a result of twin-to-twin transfusion syndrome [10]; different exposure or response to toxins in utero [11]; whether they present vertex, breech or transverse [12]; birth order [13,14]; and exposure to infections, such as the human immunodeficiency virus, at birth [15]. Discordant outcome has been reported when both twins were infected in utero, for instance with rubella, resulting in defects in only one twin at birth [16].

Developmental Coordination Disorder (DCD), which affects approximately 6% of children aged five to 11 years [17], is a movement disorder characterized by: a marked impairment in the development of motor coordination; significant interference with academic achievement or activities of daily living; the movement disorder must not be due to a medical condition such as cerebral palsy (CP) (which by definition indicates neurological origin of movement disorder), hemiplegia or muscular dystrophy; if mental retardation (MR) is present, motor deficits must be more than anticipated with MR. As would be expected, due to lack of physical activity, children with DCD are at increased risk for coronary vascular disease, including a reduction in cardio-respiratory fitness [18], and increased body fat [19].

There is very little literature on the etiology of DCD. Foulder-Hughes and Cooke [20] assessed children (average age 7.5 years) using the Movement Assessment Battery for Children. They found that 30.7% of those who were born preterm possibly met criteria for DCD, compared to 6.7% born at term. They found no significant differences for DCD between males and females, unlike other studies that have found a higher incidence of DCD in boys [21–23]. It remains unclear whether there is a difference in prevalence for boys and girls.

DCD has been linked to damage to the developing or immature brain less severe than that generally associated with CP. Jongmans et al. [24] examined the relationship between the duration of 'flares' (echodensities in the periventricular white matter) in preterm infants and motor competence at six years of age. They found that the longer the duration of the flares, the greater the decrease in motor performance, particularly in the lower limbs [24]. Jongmans et al. [25] found that premature infants with extensive perceptual-motor difficulties were more likely to have a shorter gestational age (GA) and to have shown a brain lesion soon after birth.

Motor difficulties experienced during childhood have been found to remain into adolescence [26] and adulthood [27]. In their study of 21 adults (range 19-63 years, average 39.6 years) with developmental coordination impairments, Cousins and Smyth [27] noted that affected adults performed more poorly and slowly than did controls on virtually all motor measures, including simple tasks. Their poor motor development and problems in learning skills excluded a large number of subjects from activities of daily living such as driving, which in turn limited their social and employment opportunities [27].

Attention Deficit Hyperactivity Disorder (ADHD) has both attentional and movement characteristics. The DSM-IV-TR noted that for a diagnosis of ADHD: inattention and/or hyperactivity must be more frequently displayed and be more severe than is typical for the person's developmental stage; symptoms must be present before seven years of age; impairment must be present in two or more settings – school, and/or work, and/or home; it must cause clinically significant problems in at least one of these settings; the disturbance must not occur exclusively with various other disorders, such as Pervasive Developmental Disorder [17].

DSM-IV-TR has separate diagnostic criteria for symptoms of inattention and hyperactivity/impulsivity, with ADHD diagnosable as three subtypes: predominantly Inattentive (ADHD I); predominantly Hyperactive/Impulsive (ADHD H/I); and Combined (ADHD C) [17]. Both ADHD H/I and ADHD I subtypes require that at least six of nine symptoms specific to those domains be present for diagnosis. ADHD C requires a minimum of six symptoms of both ADHD H/I and ADHD I for a diagnosis. Although some estimate the prevalence rate of AD-HD as low as 1% [28], it is generally thought to affect some 3–10% of school age children [29]. Mannuzza and Klein [30] looked at the long-term prognosis of ADHD, and found that in early and middle adolescence about two thirds to three quarters of the individuals studied still experienced problematic symptoms of ADHD. The behavioral characteristics in ADHD might remain into adulthood, although symptoms at later ages may not be sufficient to meet DSM-IV-TR criteria [31].

ADHD has been found to have a high genetic etiology. Based on findings from their Australian Twin ADHD Project, Levy et al. [32] concluded that AD-HD is one of the most heritable disorders of childhood. They found that approximately twice as many MZ as DZ twin pairs were concordant for ADHD [32]. This would be anticipated for a heritable disorder, as MZ twins are genetically identical, whereas DZ twins share only half their genes.

In their study of MZ twins discordant and concordant for ADHD, Lehn et al. [33] noted that the estimated 60% heritability of ADHD left 40% to environmental factors. They found that low birth weight and delayed motor development were markers for ADHD in infancy. Affected twins experienced more adverse birth outcomes, such as low birth weight and more time in an incubator, than did unaffected twins [33]. Herskovits et al. [34] have associated closed head injury with secondary ADHD, which they argued had a different etiology to that of developmental ADHD. Environmental factors were also noted by Sharp et al. [35], Castellanos et al. [36], and van 't Ent et al. [37] in their studies of MZ twins discordant for ADHD. In their structural magnetic resonance imagining (MRI) study of MZ twins concordant and discordant for ADHD, van 't Ent et al. [37] found that different areas of the action-attentional network were affected in concordant high risk sets compared to high risk twins in discordant sets. In concordant high risk sets, there was volume loss in the orbitofrontal subdivisions of the brain, and a compromised posterior corpus callosum. In the highrisk twin in discordant sets, there was loss of volume in the right inferior dorsolateral prefrontal cortex. The authors concluded that these patterns indicated differences in areas of loss of volume in twins with a genetic etiology of ADHD (concordant high risk sets) compared to twins with environmental etiology of AD-HD (high risk twins in discordant sets). Seidman et al. [38] conducted a structural MRI study of 24 adults with ADHD. They too found diminished brain volume overall compared to controls, and smaller dorsolateral

prefrontal and anterior cingulate cortex volumes. Krain and Castellanos [39] have reviewed studies of brain development in ADHD.

The rate of co-occurrence of DCD and ADHD has been found to be close to 50% [40]. Gillberg [41] has published widely on Deficits in Attention, Motor Control and Perception (DAMP). Piek et al. [42] found an association between inattentive symptomatology and motor ability, fine motor coordination in particular, and Martin et al. [43] found a strong-shared additive genetic component between most subtypes of DCD and AD-HD, to the subtypes of the other disorder. Under differential diagnosis for DCD, the DSM-IV-TR specified that, in ADHD, symptomatology involving movement was limited to increased motor activity, and noted that children with ADHD may fall or knock things over, but the authors attributed this to impulsivity and distractibility rather than to motor impairment per se [17]. However, Pitcher et al. [44] provided evidence to suggest that motor deficits in ADHD were a result of poor motor ability rather than of ADHD symptomatology.

A number of studies have concluded that at delivery the second born twin is at higher risk of poor outcome – such as requiring resuscitation or intubation, suffering respiratory distress syndrome, and having a lower 5 min Apgar score [13]. Second born twins are also at higher risk for mortality [14]. Some studies have indicated increased neonatal death and morbidity in second born twins if there is a vertex-non-vertex cesarean delivery of the second twin after vaginal delivery of the first, particularly if there was a birth weight 1500–4000 g [45]. Cesarean delivery also reduced the chances of a 5 min Apgar score less than four for neonates < 2000 g [46]. This is important, as low 5 min Apgar scores have been associated with neonatal encephalopathy and minor academic impairments at school age [47].

The aims of this study were to further explore the etiology of DCD and ADHD; and establish whether second born twins were at higher risk for DCD and/or ADHD.

#### 2. Materials and methods

#### 2.1. Participants

This study is part of the Australian Twin ADHD Project [48,49]. Using parental questionnaire, the Twin and Sibling Questionnaire, four categories of identical twins were investigated: (1) only one twin scored positive for DCD or ADHD (discordant co-twin control design); (2) both twins scored positive for DCD or ADHD only; (3) both twins scored positive for both DCD+ADHD; (4) neither twin scored positive for DCD or ADHD. Given that parent report was the only means for identification in this study, due to the large size of the study sample, it should be noted that although the terms DCD and ADHD are used throughout, these 'diagnoses' do not fulfill all criteria specified in the DSM-IV-TR [17].

Participants were recruited from 3180 families of twins aged 6–18 years registered on the voluntary Australian Twin Registry (ATR). A total of 2122 Twin and Sibling Questionnaires were returned (66.73%) by major caregivers, primarily mothers. Of these, 47 families were excluded as their twins were outside the study's age range when parents completed the questionnaire. The total eligible sample of 2075 twin pairs was separated into two groups, MZ and DZ, based on parent report of prior DNA testing or on a zygosity questionnaire. As a result, 922 sets of MZ twins were identified.

Using the Developmental Coordination Disorder Ouestionnaire (DCD-Q) [50], and the Strengths and Weaknesses of ADHD Symptoms and Normal Behavior (SWAN) [51], which formed part of the Twin and Sibling Questionnaire, twins who met the questionnaire criteria for DCD and ADHD were identified. We excluded all twins on whom we had insufficient data to establish whether they met criteria for DCD or AD-HD, those who had DCD+ADHD (except for 9 sets in which both twins were rated as DCD+ADHD), and those who had mild CP, hemiplegia or were on a disability pension. There remained 866 sets of twins, who formed the sample for the current study: 23 sets discordant for DCD; 23 sets concordant for DCD; 16 sets discordant for ADHD; 22 sets concordant for ADHD; nine sets concordant for both DCD and ADHD; and 773 sets unaffected for DCD or ADHD.

#### 2.2. Measures

#### 2.2.1. Developmental Coordination Disorder Questionnaire (DCD-Q)

The DCD-Q parent rated questionnaire features 17 items on a five point scale ranging from 1 "Not at all like this child" to 5 "Extremely like this child" [50]. The final seven questions are reverse-scored. The DCD-Q includes four subtypes: general coordination; control during movement; gross motor/planning; and fine motor/handwriting. Parents are asked to complete the questionnaire by comparing their child to children of the same age. The total score is 85. Because the Twin and Sibling Questionnaire has a four point scale, to make it easier for parents completing the combined questionnaire, the 3 was omitted to make a 4 point scale of 1, 2, 4, 5. For inter-item reliability, Cronbach's alpha was 0.88 for the full scale and from 0.86-0.88 for each item if deleted [43]. Rather than using a fixed score to assign individuals as affected or unaffected, for this measure the cut-off score was calculated using the formula:

Cut-off score = Mean –  $[1.65 \times \text{standard deviation}$ (SD)]. On this scale a low score assigns the participant to the 'affected' group, a high score to the 'unaffected' co-twin control group [43,52].

Concurrent validity has been established by Wilson and colleagues [50], by correlating the DCD-Q with the Movement Assessment Battery for Children (r = -0.59, P < 0.0001) and the Bruininks-Oseretsky Test of Motor Performance (r = -0.46-0.54, P < 0.0001). Wilson et al. [50] demonstrated the construct validity by finding significant differences in scores between a group without DCD, a group of suspected DCD, and a group with DCD.

#### 2.2.2. Strengths and Weaknesses of ADHD Symptoms and Normal Behavior (SWAN)

Symptoms for ADHD were assessed using the parent-rated SWAN scale [51], which is based on the 18 ADHD symptoms listed in the DSM-IV-TR [17]. Observations are based on the child's behavior over the previous month compared to other children of the same age. Scores range from "Far below average" (scored as +3) to "Far above average" (scored as -3), reflecting both strengths and weaknesses. The scores are summed then divided by nine for the inattention and hyperactivity/impulsivity subscales, and by 18 for the combined subscale to obtain a mean score for each subtype. The SWAN cut-offs between affected and unaffected for inattention and hyperactivity/impulsivity are calculated from the distribution of scores using the formula:

#### Cut-off score = Mean + $(1.65 \times SD)$

A high score indicates status as 'affected', a low score indicates 'unaffected' [43,52]. Importantly, this scale is one of few that measures strengths as well as weaknesses. Martin et al. [43] reported that the SWAN reflected the ADHD phenotype more accurately than did scales based on DSM-IV-TR [17]. In their study, Cronbach's alpha was 0.95 for both the inattention and hyperactive/impulsive scales, which indicated good internal reliability.

Groups	n (pairs)	Mean age (years)	SD	Range	Male (pairs)	Female (pairs)
DCD discordant	23	11.91	3.7	6.45-16.69	10 (43.5%)	13 (56.5%)
DCD concordant	23	12.89	3.29	6.51-18.02	11 (47.8%)	12 (52.2%)
ADHD discordant	16	13.12	3.43	6.44-16.67	12 (75%)	4 (25%)
ADHD concordant	22	14.51	1.75	11.95-17.49	13 (59.1%)	9 (40.9%)
DCD+ADHD	9	11.12	3.96	6.52-15.62	6 (66.7%)	3 (33.3%)
Unaffected	773	13.34	3.26	6.41-18.76	378 (48.9%)	395 (51.1%)
All monozygotic twins	866	13.29	3.27	6.41-18.76	430 (49.7%)	436 (50.3%)

 Table 1

 Age and gender of twins concordant and discordant for DCD and ADHD, unaffected and all monozygotic twins

DCD = Developmental Coordination Disorder; ADHD = Attention Deficit Hyperactivity Disorder.

#### 2.3. Procedure

The project was approved by the Curtin University of Technology Human Research Ethics Committee and by the ATR. The Twin and Sibling Questionnaire was completed by the mother of the twins, or if that was not possible, by a carer who knew all of the children in the family. Questionnaires were mailed to families. Respondents were asked that, if any of their children were taking medication for behavioral or attention difficulties, they complete the form as reflecting the child's behavior when not on medication.

#### 3. Results

#### 3.1. Prevalence

Prevalence of DCD and ADHD in the full sample of 922 sets of MZ twins was almost 6% for both DCD and ADHD. We found more ADHD I than ADHD H/I or ADHD C, in both ADHD concordant (54.5%; 20.5%; 25%), and discordant (68.8%; 12.5%; 18.7%) groups. However, because of the small number of twins, all subtypes were pooled as 'ADHD affected' for purposes of data analysis.

The age and sex of the different groups of twins can be seen in Table 1. ANOVA revealed a significant group difference in ages (F(5,860) = 24.905, P = 0.039). Pair-wise comparisons revealed that AD-HD concordant children were significantly older than the DCD+ADHD concordant group (P = 0.009), and DCD discordant group (P = 0.008). The unaffected group was significantly older than the DCD discordant group (P = 0.039), and the DCD+ADHD concordant group (P = 0.042). There were no other significant group differences.

Differences in the prevalence of male and female twins in each group were examined using Chi-square. The only significant difference was for the ADHD discordant group  $(\chi^2(1, n = 16) = 4, P = 0.05)$ , where more males were identified than females. For both of the DCD groups, there were no significant sex differences with slightly more girls than boys affected in both concordant (52.2%) and discordant (56.5%) twins.

#### 3.2. Birth history

Table 2 shows the GA, birth weight and Apgar scores for each group. There were no significant differences in 1 and 5 min Apgar scores or birth weight in unaffected or affected twins. DCD discordant twins were on average heavier than DCD concordant twins, and ADHD concordant twins were heavier than ADHD discordant twins, but these differences were non-significant.

ANCOVA was used to examine the GA for each group. There was a significant group effect (F(5,841) = 2.302, P = 0.043). Pairwise comparisons showed that the GA for the ADHD discordant group was significantly shorter than for the ADHD concordant group (P = 0.003), the DCD discordant group (P = 0.034) and the unaffected group (P = 0.013).

In no concordant nor discordant group was one twin delivered vaginally and the other by cesarean; in the 773 unaffected sets there were nine sets in which the second twin only was delivered by cesarian section. More second born than first born twins were affected in discordant twin pairs for both DCD (43.5% vs. 56.5%) and ADHD (43.8% vs. 56.2%), although these were not significantly different. There were no significant differences in mode of delivery (forceps, breech, cesarian) in any but the group of 773 unaffected sets of twins, in which significantly more second than first born twins presented in the breech position (41: 5.3% vs. 162: 21%;  $\chi^2(1, n = 773) = 74.641$ , P < 0.001).

The prevalence of oxygen perfusion complications can be found in Table 3. Although there appeared to be significantly more oxygen perfusion complications in affected than unaffected twins for DCD discordant (9 vs. 3), this just failed to reach significance

				Gestatic	Gestational age, birth weight and Apgar scores	h weight and	l Apgar scor	es				
Parameters	DCD di	discordant	ADHD discordant	iscordant	DCD concordant	ncordant	ADHD o	ADHD concordant	DCD+ADHD	ADHD	No DCD or ADHD	or ADHD
	Affected	Unaffected	Affected	Affected Unaffected	Twin 1	Twin 2	Twin 1	Twin 2	Twin 1	Twin 2	Twin 1	Twin 2
Gestational age	36	36.20	34	34.29	35.43	43	æ	36.99	34.89	89	36.03	33
SD	2	2.24	3.11	=	3.01	11	2.63	53	2.6	2	2.69	6
Range	31-39 (	(n = 22)	28-39 (	28–39 (n = 15)	28-41 (n = 23)	n = 23)	32-44 (	32-44 (n = 22)	31–38 (n = 9)	n = 9)	23-42 (n = 756)	= 756)
Birth weight (g)	2466	2514	2159	2218	2234	2256	2470	2650	2133	2234	2376	2353
SD	404	481	643	514	513	675	660	629	656	697	551	540
Range	1580-3523	1448-3523	1065-2948	1325-3005	1220-3100	1030-3420	1695-3820	1695-3820 1720-3810	1442-2900	3100-2234	530-3850	540-3800
	(n = 22)	(n = 22)	(n = 13)	(n = 13)	(n = 15)	(n = 15)	(n = 12)	(n = 12)	(9 = U)	(9 = u)	(n = 472) $(n = 471)$	(n = 471)
Apgar I	6.77	7.15	6.8	6.7	7.08	6.15	8.29	7.33	8.25	7	7.66	6.98
minute	SD = 2	SD = 2.1	SD = 1.95	SD = 2.5	SD = 2.4	SD = 3	SD = 1.98	SD = 1.63	SD = 1.71	SD = 1.83	SD = 1.83 SD = 1.73 SD = 2.18	SD = 2.18
Range	1-8	2–9	4-9	6-1	6-1	2–9	4-10	5-9	6-10	5-9	1-10	1-10
ı	(n = 13)	(n = 13)	(n = 12)	(n = 12)	(n = 13)	(n = 13)	(1 = 1)	(9 = U)	(n = 4)	(n = 4)	(n = 414) $(n = 412)$	(n = 412)
Apgar 5	8.15	8.62	8.50	8.58	8.64	6	9.22	88.8	9.20	8.80	6	8.9
minutes	SD = 2.1	SD = 0.9	SD = 0.91	SD = 1	SD = 1.1	SD = 1	SD = 0.67	SD = 0.64	SD = 0.84	SD = 1.1	SD = 0.99 SD = 1.1	SD = 1.11
Range	2-10	7–10	6-10	6-9	6-10	6-10	8-10	8-10	8-10	7-10	4-10	1-10
	(n = 13)	(n = 13)	(n = 12)	(n = 12)	(n = 14)	(n = 13)	(n = 9)	(n = 8)	(n = 5)	(n = 5)	(n = 416)	(n = 412)
DCD = Developmental Coordination Disorder; ADHD = Attention Deficit Hyperactivity Disorder.	mental Coo	rdination Dis	order; ADHI	D = Attentic	n Deficit Hy	peractivity I	Disorder.					

					Table 3							
				Birth c	Birth complications	suo	1					
Parameters	DCD4	DCD discordant	ADHD (	ADHD discordant DCD concordant ADHD concordant DCD+ADHD No DCD or ADHD	DCD co	ncordant	ADHD c	oncordant	DCD+	ADHD	No DCD	or ADHD
	(23	(23 pairs)	(16]	(16 pairs)	(23 pairs)	airs)	(22]	(22 pairs)	(9 pairs)	irs)	(773	(773 pairs)
Oxygen perfusion problems Affected Unaffected Unaffected Twin 1 Twin 2 Twin 1 Twin 2 Twin 1 Twin 2 Twin 1	Affected	Unaffected	Affected	Unaffected	Twin 1	Twin 2	Twin 1	Twin 2	Twin 1	Twin 2	Twin 1	Twin 2
Asphyxia	4	-	S	5	3	4	1	2	2	ŝ	52 (6.7%)	52 (6.7%) 115 (14.8%)
Placenta	I	-	7	2	0		0	0	-	-	37 (4.8%)	37 (4.8%) 43 (5.6%)
Umbilical cord	4	-	7	-	ę	4	0	3	2	1	22 (2.9%)	22 (2.9%) 43 (5.6%)
Total*	6	3	6	8	9	6	1	5	5	5	111	201

\*Some twins experienced more than one oxygen perfusion problem. DCD = Developmental Coordination Disorder; ADHD = Attention Deficit Hyperactivity Disorder.

Table 2

 $(\chi^2(1, n = 23) = 3.00, P = 0.08)$ . There was no difference for ADHD discordant affected and unaffected twins (9 vs. 8)  $(\chi^2(1, n = 16) = 0.06, P = 0.81)$ . Although there were no significant differences in oxygen perfusion complications between twin 1 and twin 2 for the DCD concordant (6 vs. 9) and DCD+ADHD concordant (5 vs. 5) (ADHD concordant (1 vs. 5) could not be tested due to too few cases), significantly more oxygen perfusion complications were found in second born than first born twins who had neither DCD nor ADHD (111 vs. 201:  $\chi^2(1, n = 773) = 26.00, P < 0.001)$ .

#### 4. Discussion

The primary aim of this study was to utilize the twin differences design to examine the etiology of DCD and ADHD, and to explore whether the second born twin was at higher risk than the first born twin for adverse events at birth.

#### 4.1. Etiology of DCD and ADHD

In this study of twins discordant and concordant for DCD and ADHD, the prevalence of DCD and ADHD in the full sample of 922 sets of MZ twins was almost 6% for both DCD and ADHD. This is similar to prevalence rates reported in DSM-IV-TR [17]. For DCD, there were equal numbers of affected concordant (23 sets) and discordant (23 sets). In terms of the hypothesis that in disorders with a genetic etiology, there would be more affected concordant than discordant sets of twins, the present data would suggest that DCD has an environmental rather than genetic etiology. For ADHD there were more concordant (22 sets) than discordant (16 sets), which we would expect if the etiology of ADHD was largely genetic.

There were significantly more affected males than females for ADHD discordant twins, which is consistent with reports in the literature on ADHD. The similar number of males and females for DCD discordant and concordant is consistent with the findings of Foulder-Hughes and Cooke [20], but does not support previous suggestions that more boys are affected than girls [21– 23]. Females and males were equally represented also suggesting an environmental etiology for DCD rather than a genetic basis.

Age differences were identified for the different groups, with the DCD discordant group and the DCD+ADHD concordant group being significantly younger than the ADHD concordant group and the unaffected group. This suggested that a greater number of younger twins were identified in these groups. The more severe symptoms for the combined DCD+ADHD group, and the clearer difference between the discordant twins with and without motor problems may explain these findings.

When GA was examined, the ADHD discordant group had a significantly shorter mean GA than AD-HD concordant twins. This suggested a different etiology for the two types of ADHD groups, with ADHD concordant being developmental and more genetically influenced, compared with ADHD discordant affected, with more environmental effects. Equal numbers of oxygen perfusion difficulties were associated with both affected and unaffected twins in the ADHD discordant group. In contrast, there were more oxygen perfusion difficulties in affected than in unaffected twins in DCD discordant pairs, although this just failed to reach significance. In their study of preterm babies with spastic diplegia (SD), Koeda and Takeshita [53] attempted to clarify the relationship between visual perceptual impairment - which is often attributed to motor deficits or strabismus - and cerebral lesions. They used MRI to show that all subjects in their study had a reduced amount of peritrigonal white matter without a delay in myelination. They proposed that the relationship between these lesions and CP and visual perceptual impairment may not be limited to SD, but may also include spastic CP in preterm birth. They suggested that the hemodynamics specific to preterm infants might cause these lesions, and noted that many of the children participating in their study showed perinatal oxygen perfusion episodes, such as respiratory distress syndrome, asphyxia, dyspnea and pneumonia [53]. Miyahara et al. [54] studied preterm twins to test whether twins were more at risk for adverse developmental outcome at school age than singletons. They concluded that brain lesions and GA, rather than twinning, predicted outcome.

Kaplan et al. [55] argued that developmental disorders such as DCD and ADHD are not discrete diagnostic categories, but have a single etiology, Atypical Brain Development (ABD), which is why they so commonly co-occur. Kaplan et al. [56] further argued that ABD reflected developmental variation of brain structure and function, with a spectrum of effects, positive and negative, on brain-based skills. Using MRI, Shaw et al. [57] found that on average children with ADHD had a thinner right orbitofrontal/inferior and posterior parietal cortex than did controls, especially in the superior prefrontal and precentral as well as medial regions of the brain. They found that there was a thinner left medial prefrontal cortex at baseline in ADHD children with poorer outcome, compared to ADHD children with better outcome and controls [57]. Shaw et al. [58] found that in children with ADHD, the developmental trajectory of cortical maturation was typical, but reached its peak thickness at the mean age of 10.5 years rather than the mean age of 7.5 years in normally developing children. This was particularly the case in prefrontal regions of the brain, which are important in the control of attention and motor planning. Those ADHD children who had the DRD4 7-repeat allele, which is frequently associated with ADHD, had a thinner cortex during childhood in brain regions important to attentional control, but to a great extent this resolved during adolescence, and these children generally had a better clinical outcome [59]. This suggests that genes play an important role in brain development in children with developmental ADHD. In affected twins in MZ discordant twins, environmental factors may play a role. For instance, as in the present study Lehn et al. [33] found that affected twins in discordant sets were of lower birth weight. They also showed delayed motor development and physical growth. In view of these and similar findings, the relationship between ADHD and motor deficits might need revision by the authors of DSM-IV-TR.

It is argued that in DCD, environmental factors such as flares [24] and brain lesions [25] are likely to be the major cause of changes in the immature brain; whereas in developmental ADHD genetic factors are the major contributors to patterns of maturation of the brain and outcome [36,57-59], rather than ABD, thus the etiology of the two disorders is different. This is important for clinical practice, as treatments may differ depending on etiology. For instance, if DCD is related to oxygen perfusion difficulties, there might be a role for neonatal induced hypothermia, which has been used since the 1950s [60]. In the case of neonatal hypoxicischemic encephalopathy, an estimated 20-30% of survivors had long term neurodevelopmental sequelae, including CP [61]. It has been shown that head cooling and minimal hypothermia - reduction of the temperature of the brain by 2-5°C for 72 hours, commencing within 6 hours after birth - improves outcome for neonates experiencing perinatal asphyxia [60,62].

If flares and brain lesions – which are general medical conditions – are a primary etiology of DCD, it is questionable whether mild CP or hemiplegia should be exclusionary criteria for DCD, as DCD and CP may fall on a continuum of movement disorder. This has important implications for at least two reasons. First, DSM-IV-TR diagnostic criteria specify that CP is an exclusionary criterion for a diagnosis of DCD because it is a medical condition [17], and if we are correct, this might need to be revised. Second, and consequent to this, in some instances a diagnosis of mild CP, rather than DCD, may make children with DCD and their families eligible for services currently reserved for children with CP and their families. Such a diagnosis could facilitate treatment commencement at the earliest possible time, as recommended in Piek [63].

#### 4.2. First vs. second born twins

There were more oxygen perfusion complications in the second born twin for ADHD and DCD concordant twins, and for twins with neither ADHD nor DCD. This is consistent with studies such as those of Hartley and Hitti [13] and Smith et al. [14], who found that the second born twin was at greater risk than the first born twin. There were equal numbers of oxygen perfusion complications in first and second born twins who both had DCD+ADHD. This has implications for obstetricians and midwives delivering twins, as noted previously regarding neonatal induced hypothermia utilizing a cooling cap or cooling blanket [61,62]. Significantly more second born twins in the unaffected group were breech presentations. Breech birth of the second born twin has been described as a safe alternative to a cesarean section in twins born less than 24 weeks and  $\leq$ 1500 g [64], although there is some controversy about this [65].

#### 5. Conclusions

Given the strong heritability of ADHD, it was extremely difficult to find a large sample of MZ twins discordant for ADHD. This was also the experience of Lehn et al. [33], Sharp et al. [35] and van 't Ent et al. [37] in their studies of ADHD discordant MZ twins. Were numbers greater in our study it might have been possible to make a more robust demarcation between affected and unaffected twins in discordant pairs. Questionnaire data does not constitute a clinical diagnosis of DCD nor ADHD, so results may differ from those found in a clinical sample.

Concordant and discordant MZ twins have an important place in further advancing our knowledge and understanding of the mechanisms of DCD and ADHD, with implications for clinical practice and future research. For instance, studies of genes such as the DRD4 7-repeat allele, involving MZ twins concordant and discordant for ADHD, to explore genetic and epigenetic factors that may play a role, as has been done in, for instance, MZ twins discordant for a caudal duplication anomaly [66] and schizophrenia [67].

The results of our study strengthen the argument that environmental factors, such as obstetric complications relating to hypoxia, may be responsible for many cases of DCD. These factors are also known to be the etiology of at least some instances of CP. An area for research would be to examine whether DCD and CP are movement disorders on a continuum rather than different conditions with different etiologies. It also strengthens the argument in a number of our previous publications that movement disorders, unless taken into consideration, can confound the results of studies of ADHD.

Greater insight into etiology of DCD and ADHD will assist in clarifying causal pathways, pathophysiology, progression, and outcome of the present disorders, contributing to management, and perhaps in some instances, prevention. Studies of twins concordant and discordant for a number of disorders should provide important indicators of factors leading to different phenotypes in individuals with the same genotype.

#### Acknowledgements

This research was funded by the National Health and Medical Research Council of Australia. The authors would like to thank Grant Baynam for his assistance with data collection and entry, the Australian Twin Registry, and the many families who kindly gave their time to be involved in this study. Thanks too to Professor F. Xavier Castellanos for his comments on drafts of this paper.

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## **APPENDIX B**

Pearsall-Jones, J. G., Piek, J. P., Rigoli, D., Martin, N. C., & Levy, F. (2009). An investigation into etiological pathways of DCD and ADHD using a monozygotic twin design. *Twin Research and Human Genetics*, *12*(4), 381–391.

# An Investigation Into Etiological Pathways of DCD and ADHD Using a Monozygotic Twin Design

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le previously described a co-twin control design using questionnaire data on monozygotic twins discordant and concordant for developmental coordination disorder (DCD) and attention deficit hyperactivity disorder (ADHD). Our results suggested that DCD and developmental ADHD had different causal pathways, and that second-born twins were at higher risk for oxygen perfusion problems than first-born twins. In the current study we further explored our findings using DNA confirmed zygosity and assessments of 4 female and 10 male sets of monozygotic twins, aged 8 to 17 years, from the first study. Using the McCarron Assessment of Neuromuscular Development (MAND), twice as many second- as first-born twins met criteria for DCD. Second-born twins attained significantly lower scores on 1-minute Apgar, MAND Gross Motor, Bimanual Dexterity and Neuromuscular Development Index. Seven of the nine twins who met criteria for DCD experienced perinatal oxygen perfusion problems. This supported findings in the first study of an association between perinatal oxygen perfusion problems and DCD, and our hypothesis that DCD and cerebral palsy have similar causal pathways. We found similar numbers of males and females discordant for DCD. On telephone interview using the Diagnostic Interview Schedule for Children Parent Interview, the only first-, and all five second-born twins who met criteria for ADHD had an inattentive component - three Inattentive; three Combined. All twins positive for ADHD were male. This adds support to our hypothesis that ADHD symptoms found in some participants may reflect secondary ADHD associated with environmental factors, rather than developmental ADHD.

**Keywords:** monozygotic twin, genetic, discordant, concordant, ADHD, DCD, etiology, oxygen perfusion, environmental

Into the 21st century, behavior genetic twin research has become pivotal in attaining estimates for the relative contribution of genetic and environmental factors to disorders such as Developmental Coordination Disorder (DCD) and Attention Deficit Hyperactivity Disorder (ADHD). However, there is increasing interest in epigenetic processes and other external and internal factors that might lead to different phenotypes in individuals with the same genotype. The co-twin control, or twin-differences design (Martin et al., 1997), with its focus on monozygotic (MZ) twins discordant for wellness and disorder, provides a unique means by which to control for potentially confounding factors such as genotype, gender, age, socioeconomic status, and shared family environment.

Factors leading to discordance in MZ twins include the stage (Race et al., 2006) and equality of splitting of the zygote (Sperber et al., 1994). Monochorionic monoamniotic twins are at high risk for twin-to-twin transfusion syndrome (TTTS), which may result in acute or chronic episodes of hemodynamic instability at various stages during pregnancy, and can cause ischemic damage (Pharoah, 2006). Lifelong discordance for phenotype can also result from different vascular patterns in the placenta of monochorionic MZ twins (Machin et al., 1996).

During the prenatal period, discordance may also result from post zygotic epigenetic effects such as tissue specific patterns of methylation (Bennett et al., 2008); varying patterns of methylation maintenance (Brown & Robinson, 2000); and chromatin remodeling (Wilson, 2008). Fraga and colleagues (2005) found that although in their younger years MZ twins were epigenetically indistinguishable, as they aged epigenetic differences, such as disease susceptibility, became more divergent.

Different exposure or response to toxins and infections may occur in utero (Iwayama et al., 2007) and at birth, sometimes affecting only one twin (Duliège et al., 1995; Forrester et al., 1966). Timing of exposure to infections is important, and may have long-term

Received 25 December, 2008; accepted 1 June, 2009.

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cumulative effects; for example, increased sensitization to hypoxia at birth and in later life (Gunn & Bennet, 2008). Discordant outcome may also be a result of birth complications and presentation (Bjelic-Radisic et al., 2007) and birth order (Hartley & Hitti, 2005; Smith et al., 2007).

Developmental coordination disorder (DCD), or specific developmental disorder of motor function, are described respectively in both the Diagnostic and Statistical Manual of Mental Disorders - Fourth Edition Text Revision (DSM-IV-TR - American Psychiatric Association (APA), 2000) and the International Statistical Classification of Diseases and Health Related Problems, 10th Revision, 2nd Edition (ICD-10 World Health Organization (WHO), 2004). DCD, which is defined as motor coordination significantly lower than expected for the child's age and intellect, that interferes significantly with activities of daily living, affects approximately 6% of children aged five to 11 years (Maeland, 1992). Both DSM-IV-TR and ICD-10 state that the movement disorder must not be due to a medical (e.g., neurological) condition such as cerebral palsy (CP). If the movement disorder has a neurological component, it is coded on Axis III as a General Medical Condition, rather than on Axis I as a Clinical Disorder. We previously found similar numbers of females and males with DCD (Pearsall-Jones et al., 2008; Skinner & Piek, 2001), as did Foulder-Hughes and Cooke (2003), although other earlier studies have reported a higher incidence of DCD in males (Kadesjö & Gillberg, 1999; Maeland, 1992).

Some 'clumsy' children — especially those with fewer deficits — appear to 'grow out of it' (Cantell et al., 2003); however, others continue to experience difficulties into adulthood (Missiuna et al., 2008). Children with DCD also have been shown to experience psychosocial and academic difficulties (Skinner & Piek, 2001).

DCD has been linked to insult to the developing or immature brain and to premature birth. Foulder-Hughes and Cooke (2003) found that 30.7% of children born preterm possibly met criteria for DCD, compared to 6.7% of children born at term. Jongmans et al. (1998) found that premature infants with extensive perceptual-motor difficulties at 6 years of age were more likely than term infants to have shown a brain lesion shortly after birth. They related this to 'flares', echodensities, or cysts, in the periventricular white matter, which are diagnostic hallmarks of periventricular leukomalacia (PVL), and found that the longer the duration of flares, the worse the motor performance (Jongmans et al., 1993). Episodes of preand perinatal hypoxia have also been associated with PVL as a major cause of CP (Wang et al., 2008).

The DSM-IV-TR (APA, 2000) and ICD-10 (WHO, 2004) also have similar classification categories for ADHD and Hyperkinetic Disorder respectively. DSM-IV-TR notes that for a diagnosis of ADHD, inattention and/or hyperactivity must be more frequently displayed and be more severe than is typical for the

person's developmental stage, and symptoms must be present before 7 years of age; there must be impairment in two or more settings, and clinically significant impairment must be present in at least one of these academic, occupational or social functioning.

ADHD has both attentional and movement components. DSM-IV-TR (APA, 2000) has separate diagnostic criteria for symptoms of inattention and hyperactivity/ impulsivity, with ADHD diagnosable as three subtypes: Inattentive (ADHD I); Hyperactive/Impulsive (ADHD H/I); and Combined (ADHD C). Both ADHD H/I and ADHD I subtypes require that at least 6 of 9 symptoms specific to those domains be present for diagnosis. ADHD C requires a minimum of 6 symptoms of both ADHD H/I and ADHD I for a diagnosis. It is generally thought to affect some 3% to 10% of school-age children (Wender, 2000), although some estimate the prevalence rate as low as 1% (Swanson et al., 1998). It has been widely reported that more males than females are affected, with estimates in clinical samples varying from 2:1 to 9:1 (APA, 2000).

Investigating the long-term prognosis of ADHD, Mannuzza and colleagues (2000) found that approximately two-thirds to three-quarters of their study participants experienced problematic symptoms of ADHD into early and middle adolescence. Behavioral characteristics of ADHD may remain into adulthood, although symptoms of ADHD may be insufficient to meet DSM-IV-TR (APA, 2000) criteria (Faraone et al., 2006).

Developmental ADHD has been estimated as one of the most heritable disorders of childhood, with approximately twice as many MZ as DZ twin pairs concordant for ADHD (Levy et al., 1997). However, environmental factors, such as brain damage caused by intrauterine infection, forceps delivery, hypoxia or anoxia during pregnancy or at birth, often go unrecognized (Henderson-Smart, 1995; Lou, 1994), and have been linked to secondary ADHD. Lehn et al. (2007) studied MZ twins discordant and concordant for ADHD, and observed that despite the high heritability of ADHD, estimated at 60%, this left 40% to environmental factors. They identified delayed motor development, low birthweight and increased time in an incubator as markers for ADHD in infancy.

Attention deficits and movement disorders have been found to co-occur in close to 50% of cases (Barkley, 1990). For example, Deficits in Attention, Motor Control and Perception (DAMP), has been widely described (Gillberg, 2003). Piek and colleagues (1999) reported an association between inattentive symptomatology and motor ability, particularly fine motor coordination.

The DSM-IV-TR (APA, 2000) specified that, in ADHD, symptomatology involving movement was limited to increased motor activity, and noted that children with ADHD may fall or knock things over, but that this related to impulsivity and distractibility rather than to motor impairment. However, Pitcher and colleagues (2003) provided evidence to suggest that the motor deficits in ADHD were a result of poor motor ability rather than ADHD symptomatology.

The second-born twin has been found to be at higher risk than the first for poor outcome, for instance, requiring resuscitation or intubation, suffering respiratory distress syndrome, and having a lower 5-minute Apgar (Hartley & Hitti, 2005). Adverse effects of preeclampsia, which has been found to be two or three times more common in twin than in singleton pregnancies (Sibai et al., 2000), have been associated with the second-born twin. Blickstein, Ben-Hur and Borenstein (1992) examined 25 twin pregnancies in which the mother had mild pre-eclampsia, 19 in which pre-eclampsia was severe, and 44 controls matched for gestational age (GA). They found that babies whose mothers had severe pre-eclampsia had a significantly shorter GA and lower birthweight compared to those with mild preeclampsia. There were three deaths, all second-born twins of mothers with severe pre-eclampsia.

In an earlier study (Pearsall-Jones et al., 2008), we examined questionnaire data on 866 sets of MZ twins, consisting of 23 sets discordant for DCD, 23 sets concordant for DCD, 16 sets discordant for ADHD, 22 sets concordant for ADHD, nine sets in which both twins had both DCD and ADHD, and 773 sets in which neither twin met criteria for either disorder. Given the size of this data set, only questionnaire information could be used as individual assessment of 866 sets of twins was not possible. The aim of the current study was to confirm zygosity using DNA, and to further explore the etiology of movement and attention problems in a smaller sample of MZ twins, using individually administered assessments to confirm diagnosis rather than rely on information from parent questionnaires. Based on the previous study, it was hypothesized that different etiological pathways would be identified for DCD and ADHD, with a relationship found between DCD and perinatal oxygen perfusion problems but not between developmental ADHD and oxygen perfusion problems. Furthermore, it was expected that the second-born twin would be at higher risk for morbidity than the first.

#### **Materials and Method**

#### **Participants**

Participants were recruited from a large cohort of 2075 sets of twins, of which 866 sets were rated by either parent completed questionnaire, or by DNA, as MZ, or identical twins. From this sample, families with twins discordant or concordant for motor problems or attention problems on the Developmental Coordination Disorder Questionnaire (DCD-Q; Wilson et al., 2000) and the Strengths and Weaknesses of ADHD Symptoms and Normal Behavior (SWAN; Swanson et al., 2001) questionnaire respectively (Martin et al., 2006; Pearsall-Jones et al., 2008; Piek et al., 2007), who lived within approximately a 3-hour drive of any capital city in Australia, were sent expressions of interest for participation in a

further stage of the study. Those who consented to participate were interviewed by telephone, during which the ADHD component of the Diagnostic Interview Schedule for Children IV - Parent Interview (DISC-IV-P; Shaffer et al., 2000) was administered to the major caregiver to establish the ADHD status of each young person. After screening for ADHD using the DISC-IV-P and for DCD using questionnaire data from the DCD-Q, 16 sets of twins were eligible to participate. That is, they were considered identical and were aged between 6 and 17 years; they were either discordant or concordant for DCD or ADHD on at least one of the measures, and lived within a 3-hour drive of a capital city. To confirm zygosity, buccal cell DNA was collected and amplified using the ABI profiler plus HID kit (9 DNA markers + sex marker, co-amplified) and separated on a capillary electrophoresis platform using the ABI 3100 instrument. Two female sets of the 16 sets were found to be dizygotic, or fraternal twins, leaving 14 eligible sets.

The average GA at birth was 34.64 weeks, (SD = 2.44; range 31-38 weeks). Nine sets were born  $\leq 36$  weeks GA, and four sets were born  $\leq 32$  weeks GA. Average age at time of interview was 13.1 years (SD = 3.65; range 8.17-17.85). Four of the 14 sets were female and 10 sets male. In our study, both twins in a set were delivered either vaginally or by cesarean section. Four sets of twins were born by cesarean delivery. Three mothers reported pre-eclampsia, two of whom had both their twins delivered by cesarean section. Two mothers, including one with severe pre-eclampsia, reported TTTS.

It was decided that regardless of each twin's status on the DCD and ADHD screening instruments, for this study status for disorder would depend on fixed scores as set out in the McCarron Assessment of Neuromuscular Development (MAND; McCarron, 1997) and DISC-IV (Shaffer et al., 2000) manuals. We did this for two reasons. First, on the initial study, rather than use fixed scores on the DCD-Q and SWAN to calculate status as affected or unaffected, individual scores relative to the entire sample of twins determined status (Martin et al., 2006; Pearsall-Jones et al., 2008; Piek et al., 2007). This was not an appropriate method for this small sample. Second, because there was a considerable time lapse between assessments on the two sets of measures, participants who met criteria initially might not at the later date, so discrepancies for status may have reflected actual differences in number of symptoms rather than the validity of the measures.

On the Wechsler Intelligence Scale for Children — 4th Edition (WISC-IV — Weschler, 2003), the scores of all twins fell within the Low Average (n = 3), Average (n = 18), High Average (n = 3), Superior (n = 3) or Very Superior (n = 1) range of intellectual functioning.

On face-to-face assessment for DCD using the MAND, five sets of twins, three male, two female, were discordant for DCD, with 6 or more points dif-

ference (mean = 9.8; SD = 3.27; range 6–15) between co-twin scores. In two other female sets, the first-born twin did not meet criteria for DCD, and the secondborn had a score of 86, 1 point above the MAND cut-off. Two sets, both male, were concordant for DCD. The concordant twins had the lowest scores on the MAND (mean = 63.5; SD = 13.53; range 48–78).

On a parental telephone interview for ADHD using the DISC-IV-P (Shaffer et al., 2000), two male sets of twins were discordant for ADHD, in that one met criteria for a DISC-IV-P diagnosis and the other had neither a *Positive* nor an *Intermediate* diagnosis. Three sets, all male, were neither concordant nor discordant on the DISC-IV-P as either one met criteria for a *Positive* diagnosis and the co-twin met *Intermediate* criteria, or one twin met *Intermediate* criteria and the other was unaffected.

#### Measures

#### **Developmental Coordination Disorder Questionnaire (DCD-Q)**

The DCD-Q (Wilson et al., 2000), a parental report of their child's movement abilities, was included in the Twin and Sibling Questionnaire, the measure initially mailed to families. It includes four subtypes: general coordination; control during movement; gross motor/ planning; and fine motor/handwriting. Parents were asked to complete the questionnaire by comparing their child to children of the same age. The total score is 85. Because the Twin and Sibling Questionnaire has a four point scale, to make it easier for parents completing the questionnaire that included the DCD-Q, the 3 was omitted to make a 4-point scale of 1, 2, 4, 5. For inter-item reliability Cronbach's alpha was .88 for the full scale and from .86-.88 for each item if deleted (Martin et al., 2006) Rather than use a fixed score to assign individuals as affected or unaffected, for this measure the cut-off score was calculated using the formula:

Cut-off score = Mean –  $(1.65 \times SD)$ .

On this scale a low score assigns the participant to the 'affected' group, a high score to the 'unaffected' cotwin control group (Martin et al., 2006; Pearsall-Jones et al., 2008; Piek et al., 2007).

#### **McCarron Assessment of Neuromuscular Development (MAND)**

To further explore movement ability, the MAND (McCarron, 1997) was administered during face-to-face assessments. The MAND is a standardized measure developed to assess fine and gross motor development in children aged 3.5 years to young adulthood and above. The measure incorporates five measures of fine motor coordination (e.g., Beads in a Box; Nut and Bolt; Finger Tapping) and five measures of gross motor coordination (e.g., Heel to Toe Walk; Stand on One Foot; Jumping). The scaled scores on each of these are added and the age norms, provided for children aged 3.5 years to young adulthood and above, are used to determine a Neuromuscular Development Index (NDI) with a mean of 100 and standard deviation of 15. A score below 55

is classified as a *severe disability*, 55 to 70 a *moderate disability* and 71 to 85 a *mild disability*. Scores are categorized into four factors: Persistent Control (PC), Muscle Power (MP), Kinesthetic Integration (KI) and Bimanual Dexterity (BD). Test–retest reliabilities after a month interval over the 10 tasks ranged from .67 to .98 (McCarron). Tan et al. (2001), using an Australian sample, found the MAND to have good specificity, good sensitivity and to be a valid measure for the identification of motor impairment.

#### Strengths and Weaknesses of ADHD Symptoms and Normal Behavior (SWAN)

The SWAN (Swanson et al., 2001), is a parental report of their child's attention, impulse control and activity. This instrument is based on the 18 ADHD symptoms listed in the DSM-IV (1994) and involves observations based on the last month in comparison with other children of the same age. Scores for each item range from *Far below average* (scored as +3) to *Far above average* (scored as -3) in order to reflect both strengths and weaknesses. The scores are summed and then divided by nine for the Inattention and Hyperactivity/ Impulsivity scale, and by 18 for the Combined subscale, resulting in an average score for each subtype. The cutoffs between individuals affected and unaffected for inattention and hyperactivity/impulsivity are calculated from the distribution of scores using the formula:

Cut-off score = Mean +  $(1.65 \times SD)$ .

A high score indicates status as 'affected', a low score indicates 'unaffected' (Hay et al., 2007; Martin et al., 2006; Pearsall-Jones et al., 2008; Piek et al., 2007).

#### The Diagnostic Interview Schedule for Children IV — Parent Interview (DISC-IV-P)

The DISC-IV-P (Schaffer et al., 2000), is a standardized measure designed by the National Institute of Mental Health to assess child and adolescent psychiatric diagnoses by ascertaining presence or absence of symptoms in children and young people aged six to 17 years. The computerized version we used was administered telephonically to the major caregiver, asking questions about the young person. It incorporated a number of impairment questions, to measure the degree to which symptoms significantly impaired or distressed the individual, a criterion required for diagnosis. The DISC-IV-P has eight modules. Only the E (Disruptive Behavior Disorders) ADHD module was administered for purposes of this study. If there were sufficient symptoms causing significant severity/impairment, a DSM-IV (1994) Positive Diagnosis was generated. If symptoms were present, but were insufficient or not sufficiently severe to meet DSM-IV diagnosis, an Intermediate Diagnosis was made. It has good reliability and validity (Hersen, 2004).

#### Wechsler Intelligence Scale for Children-IV (WISC-IV) — Australian

The WISC-IV (Weschler, 2003) measures cognitive ability in children aged 6 to 16 years 11 months. This

#### Table 1

Mean GA, Status on the DISC-IV-P and MAND, and Birth Complications

		Twin 1			Twin 2	
M GA <i>SD</i> range	DISC-IV-P	MAND (NDI)	Birth history	DISC-IV-P	MAND (NDI)	Birth history
DCD discordant						
33.4 (2.51) 31–37	Ν	N (93)	Mother pre-eclampsia	Ν	Y (78)	Mother pre-eclampsia
	Ν	N (89)		Ν	Y (80)	Oxygen perfusion problems
	Ν	Y (84)	Oxygen perfusion problems	I	N (93)	
	I	N (93)		Ν	Y (83)	
	Ν	N (88)	Oxygen perfusion problems	Ι	Y (82)	Oxygen perfusion problems
DCD concordant						
33.5 (3.54) 31–36	Ι	Y (78)	Oxygen perfusion problems; TTTS	Y (CT)	Y (71)	Oxygen perfusion problems; TTTS
	I	Y (57)	Oxygen perfusion problems	Y (IT)	Y (48)	Oxygen perfusion problems
MAND second twin 86						
35.5 (2.12) 34–37	Ν	N (104)		I	N (86)	Oxygen perfusion problems
	I	N (94)	Mother pre-eclampsia	I	N (86)	Mother pre-eclampsia
ADHD Discordant						
36 (2.83) 34–38	Ν	N (101)		Y (IT)	N (87)	
	Ν	N (111)		Y (CT)	N (94)	
ADHD Intermediate and/or Positive diagnoses						
36 (1.73) 34–37	Ι	N (95)	Oxygen perfusion problems; TTTS; mother severe pre-eclampsia	Y (CT)	N (90)	Oxygen perfusion problems; TTTS; mother severe pre-eclampsia
	Y (IT)	N (99)		I	N (95)	
	I	N (111)		Ν	N (111)	

Note: Y = diagnosis present; N = diagnosis absent; I = intermediate diagnosis; CT = combined type; IT = inattentive type

was administered to ensure that participants met DSM-IV-TR (2000) criteria for DCD, which requires motor coordination to be significantly lower than expected for the child's intellect, and to ascertain whether there were significant differences in intellectual ability between first- and second-born twins. The 10 core subtests yield four subtest indices: Verbal Comprehension (VCI), Perceptual Reasoning (PRI), Working Memory (WMI), and Processing Speed (PSI). These subtests were administered for the purposes of this study.

The WISC-IV has excellent internal consistency, test-retest reliability, and criterion and construct validity. Reliability coefficients for the WISC-IV Australian subtests averaged from .75 to .89.

#### Behavioral Assessment System for Children Structured Developmental History (BASC-SDH)

Mothers were asked to complete the BASC SDH (Reynolds & Kamphaus, 2004). This was to gather more extensive information on birth complications and developmental history. This survey provides a detailed birth, medical and developmental history, contextualising the young person's behavior.

#### Procedure

The project was approved by the Curtin University of Technology Human Research Ethics Committee and by the Australian Twin Registry (ATR). Following written consent by parents and assent by young people the parents were contacted and the DISC-IV-P (Schaffer et al., 2000) was administered telephonically. Appointments were made to visit the homes of 15 sets of the 16 sets identified prior to DNA confirmation of zygosity. One set chose to be interviewed at Curtin University of Technology.

Twins in a set were interviewed by different researchers — one assessing one twin whilst the other assessed the other. Assessors were blind to the participant's DCD and ADHD status. Administration time varied between five and seven hours, with several breaks in between. Three sets of twins had their status as monozygotic confirmed by DNA analysis prior to our study. The remaining 13 sets were mailed kits to collect buccal cells for DNA analysis. The DNA of two sets of female twins indicated that they were dizygotic or fraternal twins. Data on these twins were excluded from data analysis.

#### **Results**

#### DCD and ADHD

Table 1 shows case by case twin status for DCD and ADHD on the DISC-IV-P and MAND, and provides the relevant birth history. It is broken down into

#### Table 2

Comparisons Between First- and Second-Born Twins for Birthweight and 1- and 5-Minute Apgar Dcores Using 1-Tailed Paired *t* Tests; and on the MAND, C-DISC IV, and WISC-IV Using 2-Tailed Paired *t* Tests

		First-born	Second-born	Both twins		
Measure	Variable	M (SD)	M (SD)	Range	t	р
Birth demographics	Weight (gms)	2208 (475)	2228 (401)	1448–3005	26	.4
	1 min Apgar	8.22 (.97)	7.11 (1.27))	6–9	1.89	.048*
	5 min Apgar	8.8 (.79)	8.6 (.52)	7–10	1	.17
MAND	Fine motor	41.64 (13.19)	36.93 (12.31)	5–65	1.77	.1
	Gross motor	44.08 (8.6)	39.62 (11.51)	9–68	2.9	.012*
	NDI	92.64 (13.91)	84.57 (14.15)	48–111	4.26	.001*
	PC	18.43 (5.16)	17.21 (6.96)	0–27	.69	.5
	MP	17.29 (6.56)	15.5 (8.22)	2–35	1.23	.24
	KI	19 (5.81)	16.86 (7.15)	0–26	2.15	.05
	BD	17.21 (6.24)	14.36 (5.75)	0–25	2.4	.032*
DISC-IV-P	Number ADHD symptoms	4 (3.64)	6.29 (5.98)	0–18	1.35	.2
WISC-IV	VCI	110.21 (12.81)	108.14 (15.84)	81–138	.77	.45
	PRI	104.29 (11.73)	102.36 (14.93)	67–133	.51	.62
	WMI	104.36 (19.94)	104.14 (11.26)	86-146	.05	.96
	PSI	95.07 (10.37)	91.93 (9.79)	75–112	.88	.39
	FSIQ	105.43 (12.17)	102.86 (12.74)	84–138	.93	.37

groups with age means: DCD Discordant (13.3 years; SD = 2.93; range 8.42–15.58); DCD Concordant (12.8 years; SD = 5.54; range 8.33–16.67); MAND second twin 86 (9.13 years; SD = .64; range 8.67–9.58); ADHD discordant (13.63 years; SD = 1.29; range 13.17–14.09); and ADHD sets with *Positive* and *Intermediate* diagnoses (13.95 years; SD = 5.11; range 8.17–17.85). Because of the small numbers in the groups, mean age comparisons were not made.

Of the four sets of twins born  $\leq 32$  weeks GA, two first-born twins (one of whom was born with a cord around the neck), and three second-born twins, had MAND scores < 85. At birth, three first-born twins and five second-born twins required oxygen supplements for a period longer than 2 hours to 11 weeks (some twins required oxygen for a 'few minutes' to 1 or 2 hours — this was not regarded as indicating oxygen perfusion problems).

All three first-born twins with DCD on the MAND experienced perinatal oxygen perfusion problems (two required oxygen at birth, one for 11 weeks; the mother of one reported pre-eclampsia; a third was born with a cord around the neck). Four of the six second-born twins with DCD on the MAND experienced perinatal oxygen perfusion problems (three required oxygen at birth, one for 8 weeks; the fourth had breathing difficulties); one mother reported pre-eclampsia. Two second-born twins had scores of 86 on the MAND — the mother of one reported pre-eclampsia; the other twin was reported as having severe breathing difficulties and required supplementary oxygen. Both twins in both sets concordant for DCD experienced perinatal oxygen perfusion problems, and one set experienced TTTS. Both second-born twins in these sets also met criteria for a *Positive* diagnosis of ADHD (one Combined, one Inattentive), and both firstborn twins' scores placed them in the *Intermediate* range for ADHD.

A total of six twins were diagnosed with ADHD based on the DISC-IV-P, one first-born (Combined) and five second-born twins (two Inattentive, three Combined). Three of these (all second-born twins) experienced oxygen perfusion problems. An additional 11 twins — the co-twins of four of whom were *Positive* for ADHD — had DISC-IV-P scores placing them in the *Intermediate* range for ADHD, indicating that they had attention problems which were insufficient to warrant a *Positive* diagnosis. Six were first-born and five second-born twins, and only two of these, both second-born, experienced perinatal oxygen perfusion problems. The only twins who met criteria for both DCD and ADHD were the two second-born twins in the DCD concordant group.

#### First vs. Second Twin

Table 2 shows first and second-born twin average birthweights and scores on Apgar (one-tailed paired ttests) and scores on the MAND, DISC-IV-P, and WISC-IV (two-tailed paired t tests). There were statistically significant differences between first- and second-born twins on 1-minute Apgar, and on the MAND Gross Motor, Bimanual Dexterity and Neuromuscular Developmental Index, in favor of first-born twins.

#### Discussion

This study was designed primarily to further examine the etiology of DCD and ADHD, and to explore whether the second-born twin was at higher risk than the first-born for adverse events pre- and perinatally.

There were similar numbers of males and females in the DCD discordant group. We previously found similar numbers of males and females with DCD (Pearsall-Jones et al., 2008; Skinner & Piek, 2001), as did Foulder-Hughes and Cooke (2003), although other studies have found more males than females (Kadesjö & Gillberg, 1999; Maeland, 1992). There were only two sets of twins, both male, in the DCD concordant group.

All six twins who met criteria for a *Positive* diagnosis of ADHD were male. We previously found more males than females with ADHD (Pearsall-Jones et al., 2008), as has been reported elsewhere (Rhee et al., 2001). Rhee and colleagues concluded that males were more likely than females to be affected by ADHD as, genetically, males have a lower threshold for the required liability to express ADHD.

All three first-born twins and four of the six second-born twins with DCD on the MAND experienced birth complications including oxygen perfusion problems perinatally, in another the mother reported pre-eclampsia. Of the two second-born twins who had scores one point above the DCD cut-off on the MAND, one mother reported pre-eclampsia; the other affected twin had severe perinatal breathing difficulties and required supplementary oxygen. The current study thus found an association between perinatal oxygen perfusion problems and DCD, as we found previously on a larger study using questionnaire data only (Pearsall-Jones et al., 2008). In our studies it was not clear whether movement difficulties in these twins resulted from perinatal oxygen perfusion problems, or whether, as proposed by Gunn and Bennet (2008), exposure to infections in utero cumulatively sensitized them to hypoxia at birth, or because of prenatal cardiac or lung problems leading to perinatal oxygen perfusion problems (Morley, 2005). In neonatal hypoxic-ischemic encephalopathy, 20-30% of survivors were estimated to have long term neurodevelopmental sequelae, including CP (Vannucci & Perlman, 1997).

Although there is little literature on the etiology of DCD, most of what is available suggests that environmental factors, for instance brain lesions (Jongmans et al., 1998) and flares (Jongmans, 1993), are likely to be the major cause of damage to the developing brain. There were no reports from parents participating in our study of brain scans on their twins shortly after birth, so the possibility of brain lesions and 'flares' could not be explored. In our study, of the three mothers who reported pre-eclampsia, two delivered at 34 weeks, the other at 35 weeks. Previous research has shown that mothers with twin pregnancies with gestational hypertension (hypertension without proteinuria) had more twins delivered < 37 weeks and < 35 weeks, and more babies who were small for GA. Bdolah and colleagues (2008) found that the larger placenta of a

twin compared to a singleton pregnancy resulted in higher rates of angiogenic proteins, and that this contributed to the risk for pre-eclampsia. One of the two sets of DCD concordant twins experienced TTTS in utero, which can cause ischemic damage at various stages of pregnancy (Pharoah, 2006).

If neurological conditions are a primary etiology of DCD, then DCD and CP may fall on a continuum of movement disorder. This is important, as DSM-IV-TR (APA, 2000) and ICD-10 (WHO, 2004) diagnostic criteria specify that if a general medical (e.g. neurological) condition is present, the movement disorder is coded on Axis III as a General Medical Condition, rather than on Axis I as a Clinical Disorder. With advances in neuroimaging and other scientific developments, the etiology of wellness and illness is less mysterious than was the case even relatively recently. If DCD is a result of a medical condition, DSM-IV-TR and ICD-10 diagnostic criteria might need to be revised. Second, and consequent to this, in some instances a diagnosis of mild CP, rather than DCD, may make young people with DCD eligible for services currently reserved for young people with CP and their families. This could facilitate early treatment, as recommended in Piek (2006).

None of the five sets of twins discordant for DCD also met criteria for a *Positive* diagnosis of ADHD, although three twins met criteria for an *Intermediate* diagnosis. Of the two sets of twins who were concordant for DCD, both second-born twins also met criteria for a *Positive* diagnosis for ADHD (one Combined, one Inattentive), and both first-born twins' scores placed them in the *Intermediate* range for ADHD. By contrast, previous studies have shown a close association between movement disorders and ADHD (Gillberg, 2003; Martin et al., 2006; Piek et al., 1999).

One first and five second-born twins met the DISC-IV-P (Shaffer et al., 2000) criteria for ADHD, and four of their co-twins were Intermediate for ADHD. None of the twins discordant for ADHD experienced perinatal oxygen perfusion problems. Of the six twins who met criteria for ADHD, only the two second-born DCD concordant sets had co-occurring ADHD, and both experienced perinatal oxygen perfusion problems. This raises the issue of secondary ADHD, with environmental etiology, as opposed to developmental ADHD with a genetic etiology, and whether the etiology of ADHD in the DCD concordant twins is secondary to intrauterine infection and anoxia or hypoxia at birth (Henderson-Smart, 1995; Lou, 1994), as has been previously suggested (Pearsall-Jones et al., 2008). The DCD concordant twins were the most severely affected in terms of their motor deficits, further supporting the view that there was more extensive damage that may also be associated with ADHD. This suggests that it is important to further explore the nature of movement deficits associated with developmental and secondary ADHD, and the nature of attention deficits associated with DCD. All of the young people in our study with ADHD had an Inattentive component. In cases in which discordance for ADHD cannot be associated with birth complications or other medical factors in affected twins, epigenetic processes affecting only one twin might be a possibility, as found by Bennett and colleagues (2008) in hemophilia A.

On paired t tests, the 1-minute Apgar score of the second-born twin was significantly lower than that of the first-born twin. Lower 5-minute Apgar scores in second- than first-born twins have previously been found (Hartley & Hitti, 2005), with the second-born twin at higher risk for requiring resuscitation or intubation, and for suffering respiratory distress syndrome. Montassir and colleagues (in press) have associated perinatal oxygen perfusion difficulties and low 1minute Apgar with hypoglycemic brain lesions and CP. Three first- and six second-born twins met MAND criteria for DCD, suggesting that second-born twins were at higher risk for movement disorder than first-born twins. Second-born twins performed at a significantly lower level than first-born twins on MAND Gross Motor, Bimanual Dexterity and the Neuromuscular Developmental Index. Previous studies have concluded that second-born twins were at higher risk for morbidity than first-born twins (Hartley & Hitti, 2005). In our study this could not be linked with type of delivery, as in no case was the first twin delivered vaginally and the second by cesarean delivery.

There were no significant differences between firstand second-born twins on the WISC-IV, so secondborn twins were not significantly more intellectually compromized than first-born twins. This is consistent with previous studies (Ramsey et al., 2000; Tinca et al., 2006).

#### **Future Research**

Establishing the etiology of DCD and ADHD, and the role of environmental and epigenetic processes in discordant MZ twins, has significant implications for clinical practice and perhaps for prevention. Assessment, using the MAND, of young people with mild CP will help clarify whether the patterns of movement disorder in mild CP and DCD are similar. Current research underway in Australia is investigating the association between movement disorders, specifically CP, genes and susceptibility to infections in utero. If such genes are identified, a project could be undertaken to establish whether children with DCD have similar genetic vulnerabilities, which may be related to oxygen perfusion difficulties at birth. Whether the relationship is causal or by association, for instance reflecting a prenatal insult or aberration involving the lungs or brain which depresses ability to oxygenate at birth, is beyond the scope of this paper.

Some neonates who have experienced perinatal asphyxia have developed movement disorders, including CP (Vannucci & Perlman, 1997; Morley, 2005; Westin, 2006). Neonatal induced hypothermia might also be further explored, as this has been shown to improve outcome for neonates experiencing perinatal asphyxia (Jacobs et al., 2007; Lin et al., 2006). Permissive hypercapnia has also been found to be of benefit in neonates with respiratory disease and brain injury who experience severe hyper and hypocapnia (Zhou & Liu, 2008).

Creation of, and access to, twin registers and databases, for instance the WA Twin Child Health (WATCH) register, with links to databases such as the Western Australia Maternal and Child Health Database (Croft et al., 2002), would provide a more detailed birth history to supplement caregiver recall.

#### Conclusions

In the current study we used a co-twin control design to investigate etiological pathways for DCD and ADHD. This was found to be a very effective design as it identified unique environmental factors that could be linked to specific disorders. In particular, DCD was found to be associated with perinatal oxygen perfusion problems that were not present for developmental ADHD. Furthermore, being a secondborn twin was also linked with poorer motor outcome but not on average with increased inattention nor hyperactivity/impulsivity. However, given the small number of twins that can be identified using this approach, these findings need to be examined further with studies using both the discordant co-twin control design as well as other suitable approaches.

#### Acknowledgments

This research was partially funded by the National Health and Medical Research Council of Australia. The authors would like to thank Grant Baynam and Alison Scott for their assistance with data collection and entry, the Australian Twin Registry, and particularly the many twins and their families who kindly gave their time to be involved in this study.

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## **APPENDIX C**

Pearsall-Jones, J. G., Piek, J. P., Rigoli, D., Martin, N. C., & Levy, F. (2011) Motor disorder and anxious and depressive symptomatology: A monozygotic co-twin control approach *Research in Developmental Disabilities*, 32(4), 1245– 1252.

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### **APPENDIX D**

Piek, J. P., Rigoli, D., Pearsall-Jones, J. G., Martin, N. C., Hay, D. A., & Bennett, K. S. (2007). Depressive symptomatology in child and adolescent twins with attention-deficit hyperactivity disorder and/or developmental coordination disorder. *Twin Research and Human Genetics*, *10*(4), 587–596.

# Depressive Symptomatology in Child and Adolescent Twins With Attention-Deficit Hyperactivity Disorder and/or Developmental Coordination Disorder

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Drevious research has demonstrated a link between attention-deficit/hyperactivity disorder (ADHD), developmental coordination disorder (DCD), and depression. The present study utilized a monozygotic (MZ) differences design to investigate differences in depressive symptomatology between MZ twins discordant for ADHD or DCD. This extends previous research as it controls for genetic effects and shared environmental influences and enables the investigation of nonshared environmental influences. In addition, children and adolescents with comorbid ADHD and DCD were compared on their level of depressive symptomatology to those with ADHD only, DCD only, and no ADHD or DCD. The parent-rated Strengths and Weaknesses of Symptoms and Normal Behavior, ADHD Developmental Coordination Disorder Questionnaire, and Sad Affect Scale were used to assess ADHD, DCD, and depressive symptomatology respectively. The results revealed higher levels of depressive symptomatology in MZ twins with ADHD or DCD compared to their nonaffected co-twins. In addition, children and adolescents with comorbid ADHD and DCD demonstrated higher levels of depressive symptomatology compared to those with ADHD only, DCD only, and no ADHD or DCD. The implications of these findings are discussed with emphasis on understanding and recognizing the relationship between ADHD, DCD, and depression in the assessment and intervention for children and adolescents with these disorders.

It is well recognized that depressive symptoms and depressive disorders represent significant mental health problems during childhood and adolescence (e.g., Compas et al., 2004). In a recent Australia wide survey, 3% of children and adolescents aged 4 to 17 years met the criteria for depressive disorder (Sawyer et al., 2001).

Depression in children and adolescents has been associated with a number of risk factors, including genetic influences (Rice et al., 2002; Thapar & McGuffin, 1994), low self-esteem (Reinherz et al., 1989), cognitive factors (Cole & Jordan, 1995), and deficits in social skills (Altman & Gotlib, 1988). Genetic studies have demonstrated the importance of both genetic and environmental influences, particularly nonshared intra- and extra-familial environmental experiences (Birmaher et al., 1996; Cytryn & McKnew, 1996). For example, the Nonshared Environment and Adolescent Development (NEAD) Project found that adolescents who experienced more maternal negativity than their sibling were more likely to be depressed, independent from genetic factors and shared family environment (Reiss et al., 1994, as cited in Pike & Plomin, 1996). Clinical and epidemiological studies have shown that depression in childhood and adolescence is highly comorbid with other disorders such as anxiety disorder, substance abuse, attention-deficit/hyperactivity disorder (ADHD), and conduct disorder (Angold & Costello, 1993; Costello et al., 2002; Kovacs & Devlin, 1998; Lewinsohn et al., 1993). It has been suggested that comorbid ADHD and depression complicates symptoms and increases the risk of psychiatric and educational problems (Kewley, 2001), and may result in higher risk for suicide compared with children without such comorbid disorders (Biederman et al., 1991).

ADHD is one of the most commonly diagnosed childhood psychiatric disorders with approximately 3% to 5% of school aged children affected (American

Received 1 December, 2006; accepted 11 May, 2007.

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Psychiatric Association, 1994). Research using an Australian sample of 3597 children and adolescents (aged 6-17 years) reported a prevalence rate of approximately 7.5% (Graetz et al., 2001). ADHD is characterized by symptoms of inattention, impulsivity, and hyperactivity which must be persistent, developmentally inappropriate, and maladaptive (American Psychiatric Association, 1994). Studies employing clinical based samples have shown that children and adolescents with ADHD are more likely to be diagnosed with mood disorders such as depression and anxiety than comparison children (Angold & Costello, 1993; Biederman et al., 1991). Studies involving community samples have also linked ADHD with increased levels of depressive symptoms if not a diagnosis of depression per se (Jensen et al., 1993; Kitchens et al., 1999; LeBlanc & Morin, 2004). A recent twin study examining separation anxiety and generalized anxiety in DZ twins discordant for ADHD also identified the twin with ADHD at greater risk of these disorders than the nonaffected twin (McDougall et al., 2006).

A strong link between ADHD and motor problems has been identified. We found that approximately 50% of children with ADHD also have motor deficits severe enough to be diagnosed as developmental coordination disorder (DCD; Pitcher et al., 2003). We have also identified a genetic link between the two disorders (Martin et al., 2006). The Diagnostic and Statistical Manual of Mental Disorders (4th ed.; DSM-IV; American Psychiatric Association, 1994) defines DCD as a significant impairment in the development of motor coordination which is diagnosed when a child's motor coordination is markedly inappropriate given the child's age and intellectual ability. These movement difficulties must significantly interfere with the individual's daily life or academic achievement, and are not due to physical or neurological defects (American Psychiatric Association, 1994). According to the DSM-IV, approximately 6% of children aged 5 to 11 years experience motor problems that meet the criteria for DCD.

In addition to movement difficulties, children with DCD exhibit behavioral, conduct, and attentional problems (Gillberg & Gillberg, 1989; Piek et al., 1999). Children and adolescents with DCD have been found to experience feelings of lower self-worth, perceived lower levels of social support, and higher levels of anxiety (Sigurdsson et al., 2002; Skinner & Piek, 2001). However, little research has examined the relationship between DCD and depression. In a recent study, young school age children with DCD perceived significantly higher levels of depressive symptomatology compared to children without poor coordination (Francis & Piek, 2003). Perceived athletic competence was shown to have a direct impact on depression, which emphasises the importance of motor ability on emotional functioning.

The first aim of the present study was to understand the nature of the relationship between ADHD,

DCD, and depression by examining the levels of depressive symptomatology in monozygotic (MZ) twins discordant for ADHD or DCD. This is useful given that research on childhood and adolescent depression has identified the influences of both genetic and environmental factors (Rice et al., 2002). The MZ differences design (co-twin control method) is based on the idea that although within-pair similarities between MZ twins can be the result of genetics or postzygotic events (pre-, peri- and postnatal environment), differences between them are generally due to postzygotic events, although in some instances epigenetic factors such as patterns of methylation (Machin, 1996), demethylation and hypermethylation (Reik et al., 2001) may play a role. Consequently, one twin provides a control for examining prenatal and postnatal development, physiology, and life experiences of the co-twin (Phelps et al., 1997). In addition, MZ twins who are reared together also experience the same common or familial environment (Bulik et al., 2001). Therefore, differences between MZ twins, who are reared together, may be attributed to the influence of unique environmental factors (Bulik et al., 2001). Although there have been several family studies examining the influence of common familial vulnerabilities in the relationship between ADHD and mood disorders such as depression (e.g., Faraone & Bierderman, 1997), twin research in this area has been very limited. Based on previous evidence it is hypothesized that the ADHDonly twins will demonstrate significantly higher levels of depressive symptomatology compared to their non-ADHD twins, and the DCD-only twins will demonstrate significantly higher levels of depressive symptomatology compared to their non-DCD twins.

A further aim of the current study was to investigate the relationship between comorbid 'ADHD+DCD' and depression. Studies involving community samples have found that diagnoses of ADHD and DCD often cooccur (Kadesjo & Gillberg, 1999; Piek et al., 1999), with approximately 50% of individuals with ADHD meeting the criteria for DCD and vice versa (Kadesjo & Gillberg, 1999; Pitcher et al., 2003). Gillberg (1995) categorized this overlap between coordination and attention problems as Deficits in Attention, Motor Control and Perception (DAMP). Children with comorbid ADHD and DCD are often at higher risk for psychiatric and personality disorders than controls without ADHD or DCD. For example, Hellgren et al. (1994) reported that more than half of the adolescents with DAMP also had personality or psychiatric disorders (particularly depression), whereas only one tenth of the control group met these diagnoses. A subsequent study was conducted investigating the outcome of these individuals at the age of 22, and results revealed that 58% of the comorbid group had a poorer outcome, with higher rates of psychiatric disorders, drug or alcohol abuse, and low rates of independence compared to those without ADHD or DCD (Rasmussen & Gillberg, 2000).

These studies have demonstrated a poorer psychosocial outcome (including higher rates of depression) in the DAMP group compared to a non-DAMP control group (i.e., without ADHD or DCD; Hellgren et al., 1994). Some studies have also revealed a worse outcome in the comorbid group compared to an ADHD-only or DCD-only group (e.g., greater school dysfunction; Kadesjo & Gillberg, 1999, 2001). However, these studies have not investigated differences in rates of depression or levels of depressive symptoms. Therefore, we compared the levels of depressive symptomatology in children and adolescents with both ADHD and DCD to those with DCD only, ADHD only, and no ADHD or DCD in a large community sample. As the number of twins who were discordant for comorbid ADHD and DCD was very small, this comparison was carried out on the entire sample. To ensure the samples were independent, firstborn and second-born twins were examined in separate analyses. This study extends previous research as it specifically focuses on depressive symptomatology and examines differences between a comorbid group and ADHD-only and DCD-only groups, as well as a control group without ADHD or DCD. It was predicted that the ADHD + DCD group will demonstrate higher levels of depressive symptomatology compared to the DCD-only, ADHD-only group, and no ADHD or DCD group.

#### Method

#### Participants

#### **Co-Twin Comparisons**

Sixteen pairs of MZ twins discordant for ADHD only were identified using the SWAN (Swanson et al., 2001). Their mean age was 13.12 years with a SD of 3.43 (range = 6.44-16.67). There were 12 male pairs and 4 female pairs. Twins who met the cut-offs on the inattentive and/or hyperactivity/impulsivity subscales were classified as having ADHD (11 twins were classified as inattentive type, two were classified as hyperactive/impulsive, and three met criteria for combined type). All those who scored below the cut-offs were assigned to the control twin group. The DCD-Q (Wilson et al., 2000) was used to ensure that the twins did not meet the criteria for DCD.

Twenty-four pairs of MZ twins discordant for DCD-only were identified using the DCD-Q (Wilson et al., 2000). The mean age was 11.91 years with a *SD* of 3.57 (range = 6.45–16.99 years). They consisted of 11 male and 13 female pairs. MZ twins who scored 63 and below (using calculated cut-offs from the distribution of scores) on the DCD-Q were classified as having DCD. The unaffected co-twins who obtained a DCD-Q score of 64 and above were assigned to the control group. The twins were assessed for ADHD symptoms by the SWAN scale in order to ensure they did not meet criteria for ADHD.

There were no significant differences between the MZ twin pairs for either co-twin comparison on birth weight or apgar scores at 1 or 5 minutes.

#### **Full Sample Comparisons**

The full sample of 2040 twin pairs was separated into Twin A (first-born) and Twin B (second-born) groups in order to ensure independence of groups. The twins were then classified into ADHD-only, DCD-only, ADHD + DCD, and control groups. The DCD-only and ADHD-only groups were identified using the same calculated cut-offs described above. Twins had to meet the cut-offs for both DCD and ADHD in order to be classified as having comorbid ADHD and DCD (i.e., ADHD + DCD). Twin A sample comprised of 42 inattentive, 10 hyperactive/impulsive, and 19 combined type in the ADHD-only group. The ADHD + DCD group consisted of 20 inattentive and 13 combined. Twin B sample comprised of 47 inattentive, 15 hyperactive/impulsive, and 19 combined type in the ADHD-only group. The ADHD + DCD group consisted of 18 inattentive and 18 combined.

The cut-off scores for both scales was calculated using the mean and standard deviations (Hay et al.,

#### Table 1

Sample Size, Age, and Sex of the ADHD-only, DCD-only, ADHD+DCD, and Control Groups

	п		Age			Sex	
Group		М	SD	Range	Males	Females	
Twin A							
ADHD only	71	14.08	2.78	6.44-17.49	52	19	
DCD only	92	12.26	3.65	6.45-18.02	46	46	
ADHD + DCD	33	12.85	3.45	6.52-18.09	23	10	
Control	145	14.68	2.71	6.64–18.51	47	98	
Twin B							
ADHD only	81	13.95	2.73	6.41-17.49	64	17	
DCD only	100	12.12	3.55	6.44-18.29	52	48	
ADHD + DCD	36	13.28	3.76	6.52-18.09	27	9	
Control	134	14.67	2.45	7.10–18.70	53	81	

2007; Swanson et al., 2001). For the SWAN scale the cut-off was defined as, mean + 1.65 SD (as a low score indicates unaffected status) while for the DCD-Q it was defined as mean – 1.65 SD (as a high score indicates unaffected status). The control group was defined by those who scored equal to or below –2 ('above average' to 'far above average') on all items of the hyperactive/ impulsive and inattentive scales of the SWAN, and scored equal to or above 80 on the DCD-Q.

These cut-offs were chosen in order to ensure that the control group demonstrated minimal DCD and ADHD symptoms. The characteristics of the groups are presented in Table 1.

A one-way independent group analysis of variance (ANOVA) was conducted to determine whether the groups differed in terms of age and sex. There was a statistically significant difference in age between groups in the Twin A, F(3, 335) = 12.72, p < .001, and Twin B, F(3, 343) = 14.02, p < .001, samples. Furthermore, there was a statistically significant difference in sex between groups for the Twin A, F(3, 336) = 14.57, p < .001, and Twin B, F(3, 346) = 14.33, p < .001, samples.

#### Measures

#### Zygosity

Parents were asked whether zygosity had been previously determined by a DNA or blood test. If the twins had not been tested, parents were asked to complete a twin similarity questionnaire (Cohen et al., 1975). This scale had six questions on similarity of features and six on frequency of confusion by the mother. A description of this scale can be found in Hay et al. (2001). Such questionnaires have demonstrated validity and have shown to have good agreement with results from blood or DNA tests (McGuffin et al., 1994).

#### Strengths and Weaknesses of ADHD Symptoms and Normal Behavior (SWAN)

ADHD symptoms were assessed using the parentrated SWAN scale (Swanson et al., 2001) which is based on the 18 ADHD symptoms listed in the DSM-IV manual and involves observations based on the last month with reference to other children of the same age. Scoring for each item ranges from 'Far below average' (scored as +3) to 'Far above average' (scored as -3) in order to reflect both strengths and weaknesses. The scores are totalled and then divided by 9 for the inattention and hyperactivity/impulsivity scale, or by 18 for the combined subscale (resulting in an average score for each subtype). The cut-offs between affected and unaffected for inattention and hyperactivity/impulsivity are calculated from the distribution of scores using:

Cut-off score = Mean +  $(1.65 \times \text{standard deviation}; \text{Martin et al., 2006})$ 

The calculated cut-offs for the inattention and hyperactivity/impulsivity subtypes were 1.11 and 1.03 respectively. In order the meet a diagnosis for the combined subtype, the cut-offs for both the inattention and hyperactivity/impulsivity had to be met.

Martin et al. (2006) found the prevalence rate of ADHD, as assessed by the SWAN, to be comparable to those reported in previous studies. Hay et al. (2007) found the SWAN to be a more accurate reflection of the ADHD phenotype than DSM-IV based scales. In the present study, Cronbach's alpha was .95 for both the inattention and hyperactive/impulsive scales, demonstrating good internal reliability.

#### Developmental Coordination Disorder Questionnaire (DCD-Q)

The DCD-Q (Wilson et al., 2000) is a 17-item parentrated questionnaire, designed to differentiate between children with and without motor problems. The questionnaire includes four subtypes, namely, general coordination, control during movement, gross motor/planning, and fine motor/handwriting as revealed by previous factor analysis using clinical and community samples. This study utilized a similar cutoff calculation to the SWAN (Martin et al., 2006). The cut-off between affected and unaffected is calculated from the distribution of scores using:

Cut-off score = Mean –  $(1.65 \times \text{standard deviation})$ As a result, a cut-off score of 63 and below indicates affectedness, and a cut-off score of 64 and above indicates no DCD. Martin et al. (2006) found that this method produced a more reliable prevalence estimate of 8% (as opposed to 2% when using the Canadian cut-offs) which is comparable to the prevalence rate reported in the DSM-IV of 6%.

The DCD-Q has sound reliability and validity with studies reporting good sensitivity and specificity (Crawford et al., 2001; Green et al., 2005). The DCD-Q has demonstrated high internal consistency of the items with reliabilities of .87 to .88 as measured by Cronbach's alpha (Wilson et al., 2000). These reliabilities are comparable to those reported by Martin et al. (2006). The DCD-Q has acceptable concurrent validity as it has been shown to significantly correlate with scores on the Movement Assessment Battery for Children (r = -.59), a standardized test designed to identify motor difficulties in children (Wilson et al., 2000). The present study also identified good internal reliability, with a Cronbach's alpha of .84.

#### Depressive Symptomatology ('Sad Affect')

The Twin and Sibling Questionnaire also includes 12 items relating to 'sad affect' which assess depressive symptomatology (Hartman et al., 2001). These items were taken from a larger questionnaire which includes the 'sad affect' construct among other childhood internalizing and externalizing problems (Hartman et al., 2001). The responses for the 12 items are rated on a 4-point scale and are totalled to produce a 'sad affect' score with a total possible score of 36, with higher scores indicating greater depressive symptoms. Research involving a twin sample from an earlier wave of the Australian Twin ADHD Project reported acceptable internal reliability for the 12-item 'sad

#### Table 2

Mean Scores and Standard Deviations of Depressive Symptomatology for the ADHD-Only, DCD-Only, ADHD + DCD, and Control groups

Twin Samples	Group	п	М	SD
A	ADHD only	71	5.87	6.08
	DCD only	92	4.47	4.01
	ADHD + DCD	33	7.00	5.09
	Control	145	2.07	3.01
В	ADHD only	81	5.22	4.25
	DCD only	100	4.92	4.17
	ADHD + DCD	36	8.97	4.81
	Control	134	1.86	2.09

affect' scale with a Cronbach's alpha of .72 and an alpha for each item if deleted, ranging between .68 and .72 (Levy, Bennett, et al., 2005). In the current study, Cronbach's alpha was .77 which demonstrates acceptable internal reliability for the 'sad affect' scale.

#### Procedure

The current research was carried out as a part of a larger study (The Fourth Wave of the Australian Twin ADHD Project) investigating child and adolescent twins aged between 6 and 17 years and was approved by both the Curtin Human Research Ethics Committee and the Australian Twin Registry. The recruitment procedure used by the Australian Twin ADHD Project (ATAP) is described in Levy and Hay (2001) and Bennett et al. (2006). Following written consent, families were sent the Twin and Sibling Questionnaire which consisted of questions on ADHD, DCD and sad affect. Parents were also requested to provide information on current medication use which revealed that some of the participants comprising the groups were on medication at the time the questionnaires were completed. In this case, parents were asked to rate behavior when the child is off medication. Additionally, the questionnaire was used to ensure that the participants did not meet the exclusion criteria, namely a physical disability, chronic illness, or a medical condition affecting development (e.g., Down Syndrome). Those who met the exclusion criteria were not included in the final sample.

#### Data Analysis

To determine whether there was a statistically significant difference between the MZ co-twins, a one-tailed Wilcoxon signed rank test was conducted. This nonparametric test was a more suitable analysis as the skewed nature of depressive symptoms (Hankin et al., 2005) violated the stringent assumptions of the related samples t test.

To determine whether there was a statistically significant difference in depressive symptomatology between the ADHD + DCD, DCD-only, ADHD-only, and control groups, a Kruskal-Wallis test was used, as opposed to ANOVA. This nonparametric test was used due to the skewed nature of depressive symptoms (Hankin et al., 2005). Planned comparisons (three Mann-Whitney U tests) were carried out in order to determine which specific groups were statistically significantly different.

#### **Results**

#### **Co-Twin Comparisons**

For the 16 ADHD-only twins, the mean score for depressive symptomatology was 6.75 (SD = 6.18), and for their co-twins, the mean was 4.31 (SD = 5.12). For the 24 DCD-only twins, the mean score for depressive symptomatology was 5.21 (SD = 4.44), and for the co-twins the mean was 3.75 (SD = 3.73). A statistically significant difference was found between the ADHD-only and co-twins, Z = -2.16, p = .016, and between the DCD-only and control twins, Z = -2.83, p = .003, indicating that the twin with ADHD or DCD demonstrated higher levels of depressive symptomatology compared to their non-ADHD or non-DCD co-twin.

#### **Full Sample comparisons**

The mean scores and standard deviations of depressive symptomatology for the ADHD-only, DCD-only, ADHD + DCD, and control groups are presented in Table 2.

The relationship between group membership and depressive symptomatology was found to be statistically significant for both the Twin A sample,  $\chi^2$  (3, N = 341) = 65.28, p < .001, and Twin B sample,  $\chi^2$  (3, N = 351) = 94.26, p < .001. Planned comparisons (Mann-Whitney) between the DCD-only and ADHD + DCD groups were found to be significant for both the Twin A sample, z = -2.70, p = .004, and Twin B sample, z = -4.53, p < .001, indicating that the ADHD + DCD groups had higher levels of depressive symptomatology than the DCD-only groups. The comparisons between the ADHD-only and ADHD + DCD groups were found to be statistically significant for Twin A sample, z = -1.65, p = .05, and Twin B sample, z = -3.98, p < .001. This indicates that the ADHD + DCD group demonstrated higher levels of depressive symptomatology compared to the ADHD-only group. Finally, the comparisons between the ADHD + DCD and control groups were found to be statistically significant for both the Twin A sample, z = -5.85, p < .001, and Twin B sample, z = -7.76, p < .001, indicating that the ADHD +DCD group demonstrated higher levels of depressive symptomatology than the control group.

Pearson's correlations revealed that the variables of sex and age did not significantly correlate with the 'sad affect' scores for any of the groups in the Twin A sample. In the Twin B sample there was a weak relationship between age and sad affect for the Twin B DCD-only group (r = .22, p < .05), and a moderate relationship between age and sad affect for the Twin B ADHD + DCD group (r = .59, p < .01). However, given that the results were identical for both Twin A and Twin

B samples, these relationships for the Twin B sample did not appear to have influenced the findings.

#### Discussion

The MZ-differences design provides a powerful tool to identify nongenetic risk factors for depression. In the current study, twins with ADHD demonstrated higher levels of depressive symptomatology compared to their non-ADHD co-twins. These findings concur with previous studies which have also indicated that children and adolescents with ADHD are more likely to experience increased levels of depressive symptoms compared to controls without ADHD (Kitchens et al., 1999; LeBlanc & Morin, 2004). However, this study extends from these findings as the significant differences between the twins can be attributed to the effects of unique environmental factors (Phelps et al., 1997).

Previous research suggests that children and adolescents with ADHD experience a number of social, emotional, and behavioral difficulties such as relationship difficulties, school failure, and low self-esteem (Slomkowski et al., 1995). Authors have argued that depression could represent a secondary disorder to ADHD due to the difficulties that the children face (Jensen et al., 1993: Schmidt et al., 1998). Consequently, it is conceivable that the twins with ADHD experience higher levels of depressive symptomatology compared to their co-twins without ADHD because of unique environmental experiences such as academic, behavioral, and social difficulties. It has been noted, however, that estimates of nonshared environmental influences also involve measurement error (Plomin et al., 2001). Consequently, this can also make co-twins differ. However, this would affect the error variance as much as the means and thus cannot be the explanation of consistent co-twin differences. Furthermore, the cross-sectional nature of this study cannot ascertain whether the twins manifested higher levels of depressive symptomatology before or after the ADHD.

A difference in depressive symptomatology was also found between the twins discordant for DCD, indicating that the twins with DCD demonstrated significantly higher levels of depressive symptomatology compared to their co-twins without DCD. These results support the findings of Francis and Piek (2003) who also found increased levels of depressive symptomatology in young children with DCD compared to children without poor motor coordination. Furthermore, research has demonstrated a link between DCD and higher levels of anxiety (Sigurdsson et al., 2002; Skinner & Piek, 2001). This study extends from previous research as it involved a MZ differences design which controls for the effects of genes and shared environmental factors. Thus, the finding of a significant difference in the level of depressive symptomatology between the twins can be attributed to the effects of unique environmental factors. Children and adolescents with DCD experience a number of psychosocial problems such as poor self-perceptions, academic underachievement, perceived lower levels of social support, and negative peer relations (Cratty, 1994; Gillberg et al., 1983; Losse et al., 1991; Skinner & Piek, 2001). It has been argued that the motor problems and the associated psychosocial implications experienced by children with DCD, predispose them to many of the risk factors for depression (Francis & Piek, 2003). Consequently, it is plausible that the twins with DCD experience higher levels of depressive symptomatology compared to their non-DCD co-twins due to unique environmental experiences such as negative social feedback and academic difficulties.

The final analysis in this study indicated higher levels of depressive symptomatology in children and adolescents with comorbid ADHD and DCD compared to those with ADHD only, DCD only, and no ADHD or DCD. Research assessing the prognosis of individuals with comorbid ADHD and DCD has suggested a poor outcome. For example, children and adolescents with the comorbid condition are at greater risk for negative long-term outcomes such as psychiatric disorders, school dysfunction, personality disorders, and neurodevelopmental problems compared to individuals without ADHD or DCD (Hellgren et al., 1994). Depression is one of the main psychiatric problems that appears to be more common in children with DAMP compared to children without DAMP (Hellgren et al., 1994). Consequently, the results of this study support research suggesting a poorer emotional functioning in children and adolescents with comorbid ADHD and DCD. Furthermore, the findings of this study extend from those of previous studies as the comorbid group was also compared to an ADHD-only and DCD-only group. Previous studies have compared a comorbid group to an ADHD-only or DCD-only group on outcomes such as school dysfunction (Kadesjo & Gillberg, 1999, 2001). However, these studies have not addressed the emotional functioning of these individuals. Consequently, this study provides further support for the DAMP model and the associated poorer emotional functioning demonstrated in individuals with the comorbid condition. It should be noted that the control groups were older and included more girls than boys, in contrast to the affected groups. Given that research demonstrates that older girls are more likely to have higher levels of depression (e.g., Angold et al., 1998), these demographic differences may result in an underestimation of the depressive symptoms of the affected groups.

Hellgren et al. (1994) suggest that the high rate of associated depression in individuals with DAMP may be the result of 'biological/ genetic factors that predispose to/show as attention problems and motor clumsiness ... and major depression' (p. 1268). Alternatively, individuals with comorbid ADHD and DCD may experience rejection by peers, teachers, and relatives, which may ultimately result in feelings of unhappiness, isolation, and consequently, depression (Hellgren et al., 1994). Furthermore, it is possible that the combination of ADHD and DCD complicates the symptoms experienced by the individual, increasing the risk of psychiatric, educational, and other problems. As a result, they may experience increased levels of depression. The findings of increased levels of depression. The findings of increased levels of depressive symptomatology in MZ twins with ADHD or DCD compared to their co-twins without ADHD or DCD provides indirect evidence that the association between higher levels of depressive symptomatology and comorbid ADHD and DCD is not entirely due to common genetic factors.

There are various limitations that should be taken into account when considering the results of this study. Firstly, ADHD and DCD symptoms were assessed using parent-rated questionnaire measures, which do not produce a 'true' DSM-IV clinical diagnosis. However, these screening measures for ADHD and DCD have produced comparable prevalence rates to those reported in the DSM-IV manual (Martin et al., 2006). The use of parent-rated measures also introduces the issue of parental rating biases and contrast effects. The issue of reliability of parental reports when examining trait symptoms in MZ and DZ twins introduces a bias referred to as 'rater contrast'. This occurs when parents try to make their twins appear more similar (as is the case for MZ twins) or more different (for DZ twins) than they really are, based on their knowledge of the twin's zygosity rather than on their actual behavior (Levy, Hay et al., 2005; Rice et al., 2002). Consequently, this bias may influence the parental responses which may be less likely to occur with twins using self-report measures. Additionally, research has suggested that parents are less likely to report the presence of internalizing symptoms such as depression compared to externalizing problems (Howells Wrobel & Lachar, 1998). Despite these issues, differences in depressive symptomatology were identified between twin pairs in the current study, suggesting that these factors did not influence the findings. Furthermore, the present study involved a cross-sectional design and is therefore unable to specify whether the twins manifested higher levels of depressive symptomatology before or after the ADHD or DCD. It is also possible that certain environmental experiences or life events, unrelated to DCD, may have contributed to the differences in depressive symptomatology between the twins (e.g., stressful life events). Future research should implement a longitudinal design in order to investigate the direction of the relationship between ADHD, DCD, and depressive symptoms.

#### Conclusions

The results from the study indicate increased levels of depressive symptomatology in MZ twins with ADHD or DCD compared to their co-twin without ADHD or DCD. Furthermore, the results revealed a higher level of depressive symptomatology in children and adolescents with comorbid ADHD and DCD compared to those with ADHD only, DCD only, or no ADHD or DCD. The MZ differences design enables the differences in depressive symptomatology between the discordant twins to be attributed to unique environmental influences. Consequently, it is plausible that the twins with ADHD or DCD face unique environmental experiences such as negative self-perceptions, poor relationships with peers, behavioral problems, negative social feedback, and academic underachievement, which may consequently predispose them to many of the risk factors for increased levels of depressive symptomatology.

The present findings suggest that children and adolescents with ADHD or DCD are more likely than controls to be experiencing higher levels of depressive symptomatology. Therefore, such research could have important implications for the evaluation and treatment of children and adolescents with ADHD or DCD. While the study did not specifically identify the source of the increased levels of depressive symptoms, it highlights the importance of not overlooking aspects of emotional functioning in the evaluation process for children and adolescents with ADHD or DCD. This is crucial as it may point to the need for addressing issues such as depressive symptoms in the treatment of these children and adolescents, in addition to managing the primary ADHD (i.e., inattention and hyperactivity/impulsivity) or DCD (i.e., motor coordination problems) symptoms.

The findings of increased levels of depressive symptoms in children and adolescents with comorbid ADHD and DCD compared to those with ADHD only, DCD only, or no ADHD or DCD also have important clinical implications. Given that ADHD and DCD co-occur at a rate of approximately 50% (Piek et al., 1999), these findings emphasise the importance of addressing psychosocial issues such as emotional problems in the evaluation and treatment of these children and adolescents. In addition to the poorer prognosis associated with comorbid ADHD and DCD, children and adolescents with the comorbid condition may respond differently to treatment compared to individuals with ADHD only or DCD only. This highlights the need for the assessment of motor coordination as a standard practice for children and adolescents with ADHD and vice versa. It is important to acknowledge and explore the various symptoms of each overlapping disorder, as well as associated emotional problems, in the assessment and intervention of these individuals.

#### Acknowledgments

This research was funded by the National Health and Medical Research Council of Australia. The authors would like to thank Grant Baynam for his assistance with data collection and entry, the Australian Twin Registry, and the many families who kindly gave their time to be involved in this study. Jan P. Piek, Daniela Rigoli, Jillian G. Pearsall-Jones, Neilson C. Martin, David A. Hay, Kellie S. Bennett, and Florence Levy

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## **APPENDIX E**

**Confirmation of author contributions** 



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19 July 2012

School of Psychology and Speech Pathology, Curtin Health Innovation Research Institute, Curtin University, GPO Box U1987 Perth, Western Australia, 6845

#### To Whom It May Concern,

I, Daniela Rigoli, made a significant contribution to the following paper:

Do motor skills in infancy and early childhood predict anxious and depressive symptomatology at school age? Piek, J. P., Barrett, N. C., Smith, L. M., Rigoli, D., & Gasson, N. (2010). *Human Movement Science*, *29*, 777-786.

I coordinated and implemented the data collection for the school-age component of the project, entered and processed data, contributed to the analysis and provided input into the interpretation of the findings. I also assisted with the drafting of the manuscript. This paper, with lead author Professor Jan Piek, examined the longitudinal relationship between early motor development and later emotional outcomes at school-age.

#### Daniela Rigoli

I, as Co-Author, endorse that this level of contribution by the candidate indicated above is appropriate.

Jan Patricia Piek

Nicholas C. Barrett

Leigh M. Smith

Natalie Gasson

## Curtin University

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#### To Whom It May Concern,

I, Daniela Rigoli, was the major contributor to the conceptualisation, coordination, and implementation of my PhD project, *Understanding motor coordination and its cognitive, academic, and psychosocial correlates in an adolescent normative sample.* I am lead author, and it was primarily my responsibility to conceptualise, draft, and proofread the paper entitled:

Motor coordination and psychosocial correlates in a normative adolescent sample. Rigoli, D., Piek, J. P., & Kane, R. (2012). *Pediatrics, 129*, e892- e900.

This paper is based on data collected for my PhD thesis which involved individual assessments with each participant, for which I was the examiner. This paper investigated the relationship between motor coordination, self-perceptions, and anxious and depressive symptomatology in adolescents.

#### Daniela Rigoli

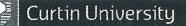
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#### An examination of the relationship between motor coordination and executive functions in adolescents. Rigoli, D., Piek, J. P., Kane, R., & Oosterlaan, J. Developmental Medicine & Child Neurology, in press.

This paper is based on data collected for my PhD thesis which involved individual assessments with each participant, for which I was the examiner. This paper investigated the relationship between motor coordination and executive functions in adolescents.

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Motor coordination, working memory, and academic achievement in a normative adolescent sample: testing a mediation model. Rigoli, D., Piek, J. P., Kane, R., & Oosterlaan, J. Archives of *Clinical Neuropsychology, in press.* 

This paper is based on data collected for my PhD thesis which involved individual assessments with each participant, for which I was the examiner. This paper investigated the relationship between motor coordination, working memory, and academic achievement in adolescents.

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#### To Whom It May Concern,

I, Daniela Rigoli, was the major contributor to the conceptualisation, coordination, and implementation of the project which resulted in the following paper:

NUMBER OF STREET, STRE

The Revised DCDQ: Is It a Suitable Screening Measure for Motor Difficulties in Adolescents? Pannekoek, L., Rigoli, D., Piek, J. P., Barrett, N. C., & Schoemaker, M. (2012). Adapted Physical Activity Quarterly, 29, 81-97.

I contributed to the conceptualisaton, drafting and proof-reading of the paper above which is based on data collected for my PhD thesis. Data collection involved individual assessments with each participant, for which I was the examiner. This paper, with Linda Pannekoek as lead author, investigated the psychometric properties of the revised Developmental Coordination Disorder Questionnaire, a parent-rated screening tool designed to assess motor difficulties in children. Linda Pannekoek was a visiting student at Curtin University who fulfilled her research requirements of her degree through my PhD project.

Daniela Rigoli

Regel.

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Linda Pannekoek

Jan Patricia Piek

Nicholas C. Barrett

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Customer name	Daniela Rigoli
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License number	2941380222659
License date	Jul 03, 2012
Licensed content publisher	Elsevier
Licensed content publication	Research in Developmental Disabilities
Licensed content title	Motor disorder and anxious and depressive symptomatology: A monozygotic co-twin control approach
Licensed content author	Jillian G. Pearsall-Jones, Jan P. Piek, Daniela Rigoli, Neilson C. Martin, Florence Levy
Licensed content date	July–August 2011
Licensed content volume number	32
Licensed content issue number	4
Number of pages	8
Start Page	1245
End Page	1252
Type of Use	reuse in a thesis/dissertation
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Expected completion date	Jul 2012
Estimated size (number of pages)	100
Elsevier VAT number	GB 494 6272 12
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Thursday 9 August, 2012

Dear Daniela,

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Piek, J. P., Rigoli, D., Pearsall-Jones, J. G., Martin, N. C., Hay, D., Bennett, K. S., & Levy, F. (2007) 'Depressive symptomatology in child and adolescent twins with attention-deficit hyperactivity disorder and/or developmental coordination disorder,' *Twin Research and Human Genetics*, *10*(4), p. 587-596.

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## APPENDIX G

Parent and young person information and consent forms



Dear Parent/Carer,

My name is Daniela Rigoli and I am writing to you on behalf of Curtin University of Technology. I am conducting a PhD research project that aims to assess movement, mood, and maths in adolescents who may or may not be experiencing difficulties in these areas. Research has found that there is a relationship between coordination and maths performance in children and adolescents. Research has also shown that children and adolescents with differing levels of math and/or movement ability experience different kinds of thoughts and feelings. Therefore, the study intends to increase our understanding of how best to help families in which a child experiences such problems. The project is being conducted with my supervisors at Curtin University, Prof. Jan Piek (Primary Supervisor), Dr. Melissa Davis, Dr. Nicholas Barrett, and Prof. Jaap Oosterlaan (Associate Supervisors).

I would like to invite you and your child to take part in the project.

### What does participation in the research project involve?

**Your child** is invited to participate in one-on-one assessment which will be broken up over one or two sessions totalling approximately 4.5 hours. This may be carried out at your family home or Curtin University, depending on what suits you.

Assessment will include a mixture of tasks such as problem solving, puzzle activities, some reading and writing, answering questions, and movement games (e.g., ball and bead activities). For example-

*Session 1:* This involves a one-on-one assessment of skills such as coordination and thinking ability (problem solving). This will take approximately 2.5 hours.

*Session 2:* Your child will be assessed in areas such as thinking ability (e.g., memory), academic achievement, and social and emotional areas. This will also take approximately 2 hours.

Please note: Unfortunately due to the purposes of this study, if your child has a physical disability or chronic illness, including hearing difficulties or a vision impairment (that doesn't simply require wearing glasses), or a medical condition that affects development (e.g. Down Syndrome), you should decline to participate on that basis and note the reason on the enclosed response form. If you have any questions about eligibility, please contact us at one of the numbers or email addresses listed below.

Also, **you are invited** to participate in the research by completing three questionnaires assessing attention and social-emotional functioning in your child. This should take no longer than half an hour.

You will also be asked to fill out the two **screening questionnaires** (assessing movement ability and medical history).

### Do I or my child have to take part?

No. Participation in this research project is entirely voluntary.

If you do not want your child to take part in the project, or your child does not wish to take part, then they simply do not. This decision should always be made completely freely, and any and all decisions are respected by members of the research team without question.

Your child has also been provided with a letter from us that we encourage you to discuss with him/her.

If you and your child agree to your child's participation, however, you do not wish do take part in filling out the parent questionnaires (on emotional functioning and attention in your child), this is also ok. Any participation is greatly appreciated and your child is able to take part without your own participation. However, if you agree to your child's participation in the project, you will be required to complete screening questionnaires, as these are important in screening the diverse medical histories and movement ability of our young participants.

### What if either of us was to change our mind?

Once a decision is made to participate, either you or your child can change your mind at any time within the minimum 5-year storage period of the research data (see below). All contributions made to the project will be destroyed unless explicitly agreed to by you.

If the project has already been published at the time you and your child decide to withdraw, your child's contribution that was used in reporting the project can not be removed from the publication.

There will be no consequences relating to a decision by you and your child to participate or not, or to participate and then withdraw, other than those already described in this letter.

# What will happen to the information collected, and is privacy and confidentiality assured?

Information that identifies anyone will be removed from the data collected. The data is then stored securely in hard and electronic copy at the School of Psychology, Curtin University and can only be accessed by the research team. The data will be stored for a minimum period of 5 years, after which it will be destroyed. This will be achieved by deleting all electronic data and shredding data which is on hard copy. The data is maintained in a way that enables us to re-identify an individual's data and destroy it if participation is withdrawn. This is done by using a system of individual codes, known only to the research team, which is used to link each individual's consent form to all data that relate to that individual.

The identity of your child will not be disclosed at any time, except in circumstances where the research team is legally required to disclose that information. Participant privacy, and the confidentiality of information disclosed by participants, is assured at all other times.

The data will be used only for this project, and will not be used in any extended or future research without first obtaining explicit written consent from you and your child.

It is intended that the findings of this study are published in a journal and/or presented at a conference. A summary of the research findings will also be made available upon completion of the project. You can access this by contacting me on the number provided, and expect it to become available in July, 2010.

### What are the benefits of this research for my child's education?

Although your child's participation may not directly benefit their education, each family will be provided with feedback on the assessment results in a brief report format. Also, parents will be informed if the scores suggest any difficulties in the areas assessed (academic, coordination, and social-emotional) and recommendations will be made for suitable services should you wish to follow up further assessment and/or treatment.

Ultimately, this project is important as it intends to increase our understanding of how best to help families in which a child experiences difficulties in the area of mood, coordination, and math ability.

#### Are there any risks associated with participation?

Although participation in the study is not anticipated to cause distress, the child- and parent rated questionnaires include reflection on social and emotional functioning. Should distress arise, you may contact me and I will provide a list of recommended child and family counselling services.

It is also possible that your child's results will suggest difficulties in the areas assessed (i.e., academic, movement, psychosocial). If the results suggest difficulties, you will be informed by letter. The letter will note the area/s that may be of concern and will include a list of recommendations which may be sought if considered appropriate by your family.

# How do I know that the people involved in this research have all the appropriate documentation to be working with children?

Under the Working with Children (Criminal Record Checking) Act 2004, people undertaking research that involves contact with children must undergo a Working with Children Check. I am also happy to provide you with copies if you have any concerns.

### Is this research approved?

The research has been approved by the Curtin University Human Research Ethics Committee (Approval Number HR 171/2007).

### Who do I contact if I wish to discuss the project further?

If you would like to discuss any aspect of this study with a member of the research team, please contact me on **9266 2286 (d.rigoli@curtin.edu.au)** or my supervisor Prof. Jan Piek on 9266 7990. If you wish to speak with an independent person about the project, please contact Linda Teasdale, Ethics Committee Secretary, by telephoning 9266 2784.

### How does my child become involved?

Please ensure that you:

- discuss what it means to take part in the project with your child before you both make a decision; and
- take up my invitation to ask any questions you may have about the project.

Once all questions have been answered to your satisfaction, and you and your child are both willing for him/her to become involved, please complete the **Consent Forms** (your child is also asked to complete the Consent Form attached to his/her letter) and return them back to me at the School of Psychology, Curtin University.

This project information letter is for you to keep.



## **Parent Consent Form for Self Participation**

- I have read this document, or have had this document explained to me in a language I understand, and I understand the aims, procedures, and risks of this project, as described within it.
- For any questions I may have had, I have taken up the invitation to ask those questions, and I am satisfied with the answers I received.
- I understand that participation in the project is entirely voluntarily.
- I am willing to become involved in the project, as described.
- I understand that I am free to withdraw that participation at any time within 5 years of project completion.
- I understand that data will be stored securely for a minimum period of 5 years, after which it will be destroyed. Also, all contributions made to the project will be destroyed unless explicitly agreed to by myself.
- I give permission for the contribution that I make to this research to be published in a journal and/or presented at a conference, provided that I or my child are not identified in any way.
- I understand that a summary of findings from the research will be made available to me and my child upon its completion.

Name of Parent/Carer (printed):					
Signature of Parent:			Date:	/	/
Contact details:					
	(Home contact number)	(Mobile)			



# **Parent Consent Form for Child's Participation**

- I have read this document, or have had this document explained to me in a language I understand, and I understand the aims, procedures, and risks of this project, as described within it.
- For any questions I may have had, I have taken up the invitation to ask those questions, and I am satisfied with the answers I received.
- I understand that participation in the project is entirely voluntarily.
- I am willing for my child to become involved in the project, as described.
- I have discussed with my child what it means to participate in this project, and he/she has explicitly indicated a willingness to take part, as indicated by his/her completion of the child consent form.
- I understand that both my child and I are free to withdraw that participation at any time within 5 years of project completion.
- I understand that data will be stored securely for a minimum period of 5 years, after which it will be destroyed. Also, all contributions made to the project will be destroyed unless explicitly agreed to by myself and my child.
- I give permission for the contribution that my child makes to this research to be published in a journal and/or presented at a conference, provided that my child or the school are not identified in any way.
- I understand that a summary of findings from the research will be made available to me and my child upon its completion.

Name of Child (printed):					
Date of Birth:					
Gender: M / F					
Name of Parent/Carer (printed):					
Signature of Parent:			Date:	/	/
Contact details:					_
	(Home contact number)	(Mobile)			



My name is Daniela Rigoli and I am from Curtin University of Technology. I would like to invite you to take part in a research project that I am doing. It is about movement (coordination), mood, and maths in adolescents who may or may not experience difficulties in these areas.

I am asking for your help with the project because I am inviting young people aged 12 to 16 to participate.

### What would I be asked to do?

If you agree to take part, you would be asked to participate in a mixture of tasks such as movement games (e.g., ball and bead activities), puzzle activities, problem solving, some reading and writing, and answering questions. This may be carried out at your family home or Curtin University and will occur in one or two sessions, depending on what suits you.

For example-

*Session 1:* This involves a one-on-one assessment of skills including, coordination and thinking ability (e.g., problem solving). This will take approximately 2.5 hours.

*Session 2:* You will be asked to participate in activities looking at thinking ability (e.g., memory), academic achievement, and social and emotional areas. This will also take approximately 2 hours.

Also, I will be inviting your parent to participate in filling out some questionnaires about your movement ability, attention, and mood.

#### **Do I have to take part?**

No. You are completely free to say yes or no. You either volunteer or you don't volunteer. If you do not want to volunteer, then simply don't write your name on the space provided on the next page. It is that easy.

I will respect your decision whichever choice you make, and I will not question it.



### What if I wanted to change my mind?

If you say yes, but then change your mind, you are free to stop participating in the project and withdraw. When you withdraw, what you have given to the project will be destroyed, unless you and your parents agree that I can use it. The period in which you can withdraw is any time within 5 years after the project takes place.

If the project has already been published at the time you decide to withdraw, your contribution that was used in that publication cannot be removed from the publication.

### What will happen to the information I give - is it private and confidential?

Information that identifies anyone will be removed from the data collected. The data is then stored securely by lock or password protection at the School of Psychology, Curtin University and can only be accessed by the research team. Data will be stored for a minimum period of 5 years. Records are destroyed immediately after this period, unless the law requires them to be held longer. This will be done by deleting all data that is in electronic form (i.e., on computers) and shredding all data that is in hard copy (i.e., on paper).

All information you provide is stored in a way that enables us to re-identify what you contributed to the project and destroy it if you withdraw your participation. This is done by using a system of individual codes, known only to the research team, which is used to link each individual's consent form to all data that relate to that individual.

After I have collected what each student has given to the project and analyse all of it, I intend to write about what I found and publish it in a journal, which is like a magazine, so that other people can read about it and I may also present the findings at a conference. But when I do this, I won't write or tell anyone your name.

A summary of the project will also be made available to you when it is completed. You can read this by contacting me about your interest. You can expect it to become available in July, 2010.

What you provide for this project will be used only for this project, and will not be used in any extended or future research without first obtaining an agreement from you and your parents/carers.

### Will you tell anyone what I say while I am contributing to the project?

In almost all cases no. If you tell me something that later I need to tell someone else because the law requires me to do so, then I will have to. I may also have to reveal something you say to me if I think that you might be being mistreated by someone. If this happens I will make sure that someone who can discuss this with you further will come to talk with you.

In all other situations, I will treat what you tell me as being private and confidential (I won't tell anyone unless you agree that I should).



### What are the benefits of this research for me?

There are no direct benefits to you, however, you and your parent/s will be provided with feedback about the assessment. This means you get to find out how you went in the areas that we looked at, for example, coordination, thinking (e.g., memory), and academic skills.

### Are there any risks associated with participation?

The questionnaires that you will be asked to complete get you to think about areas such as your mood and social support. It is possible that some people may feel upset after thinking about these things. Should distress arise, you and/or your parent may contact me and I will provide a list of recommended child and family counselling services. It is also possible that your results will suggest difficulties in the areas assessed (i.e., academic, movement, psychosocial). If the results suggest difficulties, you and your parent/s will be informed by letter. The letter will note the area/s that may be of concern and will include a list of recommendations which may be sought if considered appropriate by your family.

#### Is this research approved?

The research has been approved by the Curtin University Human Research Ethics Committee (Approval Number HR 171/2007).

#### Who do I contact if I wish to talk about the project further?

Please talk about the project with your parents first. Then, if you would like to talk with me more, please contact me on 9266 2286 (**d.rigoli@curtin.edu.au**) or my supervisor Prof. Jan Piek on 9266 7990. If, at any time, you wish to speak with a person who is not involved in the project about how something was handled, please contact Linda Teasdale, Ethics Committee Secretary, by telephoning 9266 2784.

#### OK - so how do I become involved?

You have already discussed the project and what it means to take part with at least one of your parents, and now you get to say for yourself.

If you **<u>do</u>** want to be a part of the project, then please read the next page and write your name in the space provided.

This letter is for you to keep.

# Consent Form for Young Person

- I know that I don't have to be involved in this project, but I would like to.
- I know that I will be doing activities looking at movement (coordination), academic ability, thinking, and social-emotional areas as part of the project.
- I understand that data will be stored securely for a minimum period of 5 years, after which it will be destroyed.
- I understand I am free to stop and withdraw from the project at any time within 5 years and my contribution to the project will be destroyed, unless my parents and I agree that you can use it in your reporting of the project.
- I understand that all information provided is treated as strictly confidential and will not be disclosed to anyone without my permission, except in a format that does not allow me or my family to be identified (e.g., in publications). However, I give permission for myself and my parent/s to be given feedback about the assessment results.
- I understand that I need to write my name in the space below, before I can be a part of the project.

Your name:

Today's Date: / /

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