MOTOR COMPETENCE, PHYSICAL ACTIVITY AND FITNESS IN CHILDHOOD

THE INTERRELATIONSHIPS AMONG MOTOR COMPETENCE, PHYSICAL ACTIVITY AND HEALTH-RELATED FITNESS IN THE EARLY YEARS

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Lay Abstract

Children with motor coordination difficulties are more likely to be unfit and inactive compared to typically developing (TD) children. However, it is still not understood when these problems emerge, or if inactivity explains why children with motor difficulties are less physically fit. This thesis examines the links between motor competence, physical activity, and fitness in two large cohorts of preschool-age children. Results show that young children with motor difficulties are less physically fit, but are not less active than TD children. In addition, children with better motor skills become more active as they get older. Despite similar physical activity levels at preschool age, these findings highlight that poor motor abilities in early childhood are a risk factor for poor physical fitness and future inactivity. Interventions targeting motor skills in the early years may help children stay active and healthy as they age.

Abstract

Motor competence is positively associated with physical activity and health-related fitness (HRF) across childhood and adolescence. Owing to their motor difficulties, children with Developmental Coordination Disorder (DCD) are found to have poorer HRF and lower activity levels compared to typically developing (TD) children. It is thought that children with DCD are less physically fit due in part to hypoactivity; however, it is still unclear how young these deficits emerge, or if physical activity explains these HRF differences. This thesis aims to fill these gaps by examining physical activity and HRF in preschool children with and without DCD, and testing mediation models linking motor competence to HRF through physical activity engagement, both cross-sectionally and over time from preschool to school age.

The first and second studies demonstrated that preschool children with DCD exhibit poorer musculoskeletal and aerobic fitness compared to TD children, however physical activity engagement was similar and did not explain these fitness deficits. The third study found that the relationship between motor competence and physical activity was not significant at preschool age, but emerged over time as children reached school age. Additionally, motor competence was a significant positive predictor of

musculoskeletal fitness across the early years, again largely independent of physical activity levels.

This thesis represents the first comprehensive series of studies that examines objectively-measured motor competence, physical activity and HRF in large samples of preschool-age children both with and without DCD. These studies highlight that poor motor competence is a risk factor for poor physical fitness, irrespective of physical activity in the early childhood period. Early motor interventions may positively influence physical fitness and may help to prevent the declines in physical activity observed as children with DCD reach middle childhood and adolescence.

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List of Abbreviations and Symbols

%ile	Percentile
AAHPERD	American Alliance for Health, Physical Education and Recreation
ADHD	Attention-Deficit/Hyperactivity Disorder
ANOVA	Analysis of Variance
ANCOVA	Analysis of Covariance
BF	Body Fat
BMI	Body Mass Index
BOT-SF	Bruininks-Oseretsky Test of Motor Proficiency 2nd Edition, Short Form
CATCH	Coordination and Activity Tracking in Children
CDC	Centers for Disease Control and Prevention
CIHR	Canadian Institutes of Health Research
CO-OP	Cognitive Orientation to daily Occupational Performance
CP	Cerebral Palsy
CPM	Counts Per Minute
CVD	Cardiovascular Disease
DCD	Developmental Coordination Disorder
DCDQ	Developmental Coordination Disorder Questionnaire
DSM-5	Diagnostic and Statistical Manual of Mental Disorders, 5th Edition
HOPP	Health Outcomes and Physical activity in Preschoolers
HRF	Health-Related Fitness
KBIT-2	Kaufman Brief Intelligence Test, 2nd Edition
LJ	Long Jump
LPA	Light Physical Activity
MABC-2	Movement Assessment Battery for Children, 2nd Edition
MP	Mean Power
MPA	Moderate Physical Activity
MVPA	Moderate-to-Vigorous Physical Activity
NTT	Neuromotor Task Training
PA	Physical Activity
PHAST	Physical Health and Activity Study Team
PP	Peak Power
rDCD	At risk for Developmental Coordination Disorder
RPM	Revolutions Per Minute
SD	Standard Deviation
SED	Sedentary
SPSS	Statistical Package for the Social Sciences
SS	Standard Score

STMP	Short-Term Muscle Power
TD	Typically Developing
TPA	Total Physical Activity
TTE	Time To Exhaustion
VO_2max	Maximal Oxygen Uptake
VPA	Vigorous Physical Activity
W	Watts
WC	Waist circumference
X^2	Chi-Square

Declaration of Academic Achievement

This thesis was prepared in the “sandwich thesis” format outlined in the McMaster University School of Graduate Studies Guide for the Preparation of Master’s and Doctoral Theses, published in December 2016. The first chapter is an introduction, which sets the context for the complete body of research. Chapters 2, 3, and 4 consist of three original research papers, prepared in journal article format for submission. A concluding chapter (Chapter 5) summarizes and discusses the main findings of this thesis and includes future research directions.

Contributions to Multi-Authored Manuscripts

Chapter 2:

Manuscript: **King-Dowling S**, Kwan MYW, Rodriguez MC, Missiuna C, Timmons BW, Cairney J. Physical activity in young children with and without motor coordination difficulties. Prepared for submission to Medicine & Science in Sport & Exercise.

Contributions: This manuscript presents preliminary baseline findings from the Coordination and Activity Tracking in CHildren (CATCH) study. SKD conceived the research question, conducted the analysis and drafted the manuscript. SKD assisted with data collection and data entry, including accelerometer processing and analysis. Critical feedback on the manuscript was provided by MYWK, MCR, BWT and JC. MCR is the research coordinator on the CATCH study and oversaw and assisted with all aspects of recruitment, data collection and data entry. CM, BWT and JC are principal investigators and MYWK is a co-investigator on the CATCH study, all of who made significant contributions to the CATCH study design. All co-authors reviewed and approved the final manuscript.

Chapter 3:

Manuscript: **King-Dowling S**, Rodriguez MC, Missiuna C, Timmons BW, Cairney J. Health-related fitness in young children with and without motor delays: role of physical activity. Prepared for submission to Medicine & Science in Sport & Exercise.

Contributions: Like Chapter 2, this manuscript presents preliminary baseline findings from CATCH study. SKD conceived the research question, analyzed the data and drafted the manuscript. SKD coordinated and assisted with data collection and data entry of all health-related fitness outcomes. Critical feedback on the manuscript was provided by MCR, BWT and JC. MCR is the research coordinator on the CATCH study and oversaw and assisted with all aspects of recruitment, data collection and data entry. JC, CM and BWT are principal investigators on the CATCH study, all of who made significant contributions to the CATCH study design. All co-authors reviewed and approved the final manuscript.

Chapter 4:

Manuscript: **King-Dowling S**, Proudfoot NA, Timmons BW, Cairney J. (In Review). Longitudinal relationships among motor proficiency, short-term muscle power and physical activity in the early years. Prepared for submission to the British Journal of Sports Medicine.

Contributions: This manuscript presents longitudinal data from the Health outcomes and Physical activity in Preschoolers (HOPP) Study. SKD assisted with recruitment, data collection and data entry and scored and interpreted all motor proficiency assessments. SKD conceived the research question, analyzed the data, and drafted the manuscript with critical feedback provided by NP, JC and BWT. NP is the research coordinator for the HOPP study and oversaw and participated in all aspects of recruitment, data collection and data management. BWT is a principal investigator and JC is a co-investigator on the HOPP study, both making significant contributions to the study design. All co-authors reviewed and approved the final manuscript.

CHAPTER 1: **INTRODUCTION**

Early childhood is an important time for the acquisition of fundamental motor skills. These motor competencies form the building blocks for children to engage in sports and active play, and provide the foundation for an active and healthy lifestyle (Clark & Metcalfe, 2002). Secular declines in motor skills have been observed in children as young as preschool-age (Roth et al., 2010). The resultant poor motor abilities in the pediatric years represent a potentially modifiable risk factor for the inactivity and declining health-related fitness trends we are observing in youth today (Dollman, Norton, & Norton, 2005; Tomkinson & Olds, 2007; Tremblay et al., 2010; Tremblay & Willms, 2000).

The hypothesized interrelationships among motor competence, physical activity and health-related fitness are described and modeled in the physical education, general child development, motor development, and childhood disability literatures (Bar-Or, 1983; Hands & Larkin, 2002; Seefeldt, 1980; Stodden et al., 2008; Wall, 2004). Although these models have been investigated largely in parallel, with little recognition of each other, they are highly complementary. Regardless of the particular framework, these models all recognize motor competence as a central component to physical activity participation and future health outcomes. They all also share a developmental focus, viewing the interrelationships among those domains as dynamic, interactive, and evolving over time. There is now strong evidence supporting the direct relationship between motor competence and both physical activity and health-related fitness in childhood (Robinson et al., 2015). However, it remains unclear when, developmentally, poor motor abilities begin affecting physical activity and health-related fitness. Additionally, how these relationships change over time and the mediating pathways among these variables is still poorly understood.

This thesis aims to expand our understanding of how motor competence is linked to physical activity and health-related fitness in the early years. First these constructs are examined in preschool children who meet diagnostic criteria for Developmental Coordination Disorder (DCD). This is followed by testing a longitudinal model linking these constructs in a large sample of typically developing children from preschool to school age. Specifically, this thesis sets out to examine if motor competence is linked to health-related fitness components through the mediating effect of physical activity in the early childhood period.

1.1 **Definitions of key terms**

Throughout this thesis the term ‘motor competence’ will be used to reflect the various terminologies that are often used interchangeably in the literature, including motor proficiency, motor coordination, motor ability, motor/movement skill. Although these terms are not strictly synonymous, in the context of this thesis motor competence will be used as a general term to describe one’s ability to execute goal-directed movement (Barnett et al., 2016; Robinson et al., 2015). Motor competence can be assessed either using process-based measures that focus on the quality of the movement or technique, or product-based measures that focus on the outcome of the movement (e.g., distance thrown, number of successful catches). The studies included in this thesis measure motor competence using two product-oriented motor assessments: the Movement Assessment Battery for Children – 2nd edition (Chapters 2 and 3) and the Bruininks-Oseretsky test of Motor Proficiency - 2nd Edition Short Form (Chapter 4).

Physical activity refers to “any body movement produced by the skeletal muscles that results in energy expenditure” (Caspersen, Powell, & Christenson, 1985, p. 126), and for the purposes of this thesis will refer primarily to estimated habitual physical activity levels (min/day or hours/week). Health-related fitness is a broad term that encompasses multiple attributes, including cardiorespiratory fitness, musculoskeletal fitness, body composition and flexibility, that relate to one’s health and ability to perform physical work (Caspersen et al., 1985). In childhood, developing good health-related fitness is related to positive health markers and outcomes across the lifespan (e.g., lower cardiovascular disease risk, adiposity, cholesterol, blood pressure and improved skeletal health) (Janz, Dawson, & Mahoney, 2002; Ortega, Ruiz, Castillo, & Sjöström, 2008; Ruiz et al., 2009). Although we recognize that both physical activity and health-related fitness are influenced by a wide variety of biological (e.g., growth and maturation) and environmental factors (e.g., culture, climate, socioeconomic status) (Malina, 2001; Sallis, Prochaska, & Taylor, 2000), this thesis focuses on the effect of motor competence on these health indices.

1.2 **Overview of Developmental Coordination Disorder**

1.2.1 Definition and prevalence

Developmental Coordination Disorder (DCD) is a prevalent neurodevelopmental condition affecting approximately 5 to 6 percent of children (Blank, Smits-Engelsman, Polatajko, & Wilson, 2012; Gibbs, Appleton, & Appleton, 2007). It is found to occur more often in boys, with estimated prevalence ratios ranging from 2:1 to as high as 7:1 (Kadesjo & Gillberg, 1999; Lingam, Hunt, Golding, Jongmans, & Emond, 2009). A hallmark feature of the disorder is that children present with motor coordination abilities well below that of their same-age peers (American Psychiatric Association, 2013); however, the presentation of motor difficulties is highly heterogeneous. Children present with a wide range of motor deficits in the fine and/or gross motor domains, which interfere significantly with daily functioning including self-care, academic or leisure-time pursuits (American Psychiatric Association, 2013).

As outlined in the most recent Diagnostic and Statistical Manual of Mental Disorders (5th ed.; DSM-5; American Psychiatric Association, 2013), the full diagnostic criteria for DCD are:

A. The acquisition and execution of coordinated motor skills is substantially below that expected given the individual’s chronologic age and opportunity for skill learning and use. Difficulties are manifested as clumsiness (e.g., dropping or bumping into objects) and well as slowness and inaccuracy of performance of motor skills (e.g., catching an object, using scissors or cutlery, handwriting, riding a bike, or participating in sports).
B. The motor skill deficit in criterion A significantly and persistently interferes with activities of daily living appropriate to the chronologic age (e.g., self-care and self-maintenance) and impacts academic/school productivity, prevocational and vocational activities, leisure, and play.
C. Onset of symptoms is in the early developmental period
D. The motor skills deficits are not better explained by intellectual disability (intellectual developmental disorder) or visual impairment and are not attributable to a neurologic condition

affecting movement (e.g., cerebral palsy, muscular dystrophy, degenerative disorder)

In order to meet criterion A, a child must score at least below the 15th percentile on a standardized norm-referenced test of motor coordination such as the Movement Assessment Battery for Children- 2nd Edition (MABC-2), with cut-offs below the 5th percentile recommended for preschool-age children (Blank et al., 2012; Henderson, Sugden, & Barnett, 2007). Secondly, as stated in criterion B, the impact of motor coordination problems must interfere with some aspect of daily functioning in order to receive a diagnosis. Unfortunately, there is currently no standardized approach or tool for assessing this criterion, however parent reports using structured interviews or questionnaires (e.g., DCD Questionnaire (B. Wilson, Kaplan, Crawford, & Roberts, 2007); MABC checklist (Henderson et al., 2007)) are typically used (Veldhuizen & Cairney, 2015). Thirdly, criterion C states that there must be evidence of motor coordination problems presenting early in development, reinforcing the idea that DCD does not emerge in later life, but is a developmental disorder (Henderson & Henderson, 2003). Lastly, a diagnosis of DCD can only be made when a proper, differential examination is conducted by a physician (criterion D). If a child, for example, meets diagnostic criteria for cerebral palsy, muscular dystrophy or another condition that impairs motor function, then a diagnosis of DCD cannot be given. It is also important to ensure that no other medical condition or cognitive deficit is present that might better explain impaired motor coordination or hinder the child's ability to perform an accurate motor assessment, such as a brain tumour, intellectual delay, or recent head trauma.

1.2.2 Etiology

The precise cause of DCD is still relatively unknown; however, the disorder is believed to have neonatal origins, likely involving an insult to the developing brain *in utero* or possibly shortly after birth (Gubbay, 1975; Holsti, Grunau, & Whitfield, 2002). Clinical risk factors associated with the DCD include very/extremely preterm or low/extremely low birth weight (Arpino et al., 2010; Holsti et al., 2002) and/or perinatal oxygen perfusion complications (Pearsall-Jones, Piek, Rigoli, Martin, & Levy, 2009). Severe neonatal hypoxia has also been linked to risk of cerebral palsy (CP), and it has been proposed that the two may share an etiological pathway (Pearsall-Jones, Piek, & Levy, 2010; Spittle & Orton, 2014). With this view, motor impairment may fall on a continuum wherein DCD represents a milder form of the more severe motor difficulties experienced by individuals with CP. In addition to clinical risk factors, there is evidence for a genetic component (Martin, Piek, & Hay, 2006). Twin studies have found higher concordance rates for DCD among monozygotic versus dizygotic twins (Lichtenstein, Carlström, Råstam, Gillberg, & Anckarsäter, 2010), and case histories have discovered similar motor difficulties in siblings and parents of the same family (Gaines et al., 2008).

DCD is at least twice as common in boys compared to girls (Kadesjo & Gillberg, 1999; Lingam et al., 2009), which suggests that male sex is a risk factor for DCD. Conversely, some general population samples (e.g., Cairney, Hay, Veldhuizen, Missiuna, & Faught, 2010; Skinner & Piek, 2001) have shown a more equal gender distribution than samples from a clinical setting. This may be due to referral bias, whereby motor difficulties are viewed as more problematic for boys, or boys receiving care for common comorbid disorders (e.g., Attention-deficit/hyperactivity disorder, ADHD (Kadesjo & Gillberg, 2001), speech/language disorders (Gaines & Missiuna, 2007)) may subsequently lead to a greater probability of their motor difficulties being identified. It is also plausible that the higher prevalence in boys is related to the poorer neurodevelopmental outcomes in premature males compared to their female counterparts (Marlow, Hennessy, Bracewell, & Wolke, 2007). Taken together, the etiology is likely multifactorial, involving a combination of both genetic and environmental factors.

Despite limited understanding of the specific etiology of DCD, significant work has examined underlying neuro-cognitive deficits such as deficits in motor planning, motor imagery and processing visuo-spatial information (Geuze, 2005; P. H. Wilson & McKenzie, 1998). One popular theory in the literature is the internal modelling deficit hypothesis (P. H. Wilson & Butson, 2007). This theory suggests that children with DCD have difficulties implementing predictive models of action (e.g., generating estimates of body position). This leads to incomplete planning of the required action and forces a reliance on slower feedback-based control strategies (Adams, Lust, Wilson, & Steenbergen, 2014). Evidence supporting this hypothesis includes impaired motor imagery (Williams, Thomas, Maruff, & Wilson, 2008) and a lesser ability to plan movements with a goal of end-state comfort (van Swieten et al., 2010) in children with DCD compared to typically developing children. Another theory is the automatization deficit hypothesis (Visser, 2003), which describes children with DCD as having difficulties automatizing movements, or performing movement with little conscious effort. This is especially apparent under conditions such as dual-task paradigms (i.e. dual motor and cognitive tasks), where the addition of a secondary, cognitive task impairs performance of the primary, motor task more notably in children with DCD relative to typically developing children. For example, Tsai and colleagues (2009) found that children with DCD showed significantly more postural control impairments during dual task paradigms involving a verbal counting task. Both of these hypotheses implicate cerebellar dysfunction as one possible neurological mechanism (Zwicker, Missiuna, & Boyd, 2009), however involvement of multiple brain areas has also been proposed. This view of more globalized ‘atypical brain development’ is especially compelling considering the reported high rates of comorbidity and shared symptomology among common neurodevelopmental disorders (i.e. disorders of motor, attention and language) (Gilger & Kaplan, 2001; Kaplan, N Wilson, Dewey, & Crawford, 1998).

Neuroimaging studies have supported both of these aforementioned views, implicating dysfunction (under activation) in cerebellar, parietal and prefrontal lobes during motor tasks (P. H. Wilson et al., 2017). Diffusion magnetic resonance imaging studies have also implicated a type of ‘dys-connectivity syndrome’ whereby altered microstructural integrity of white matter networks suggests weaker connectivity between brain regions (Debrabant et al., 2016). Taken together, research supports that children with DCD have differences in brain network structure and function (P. H. Wilson et al., 2017), and that these functional neuropathologies likely explain many of the global motor coordination deficits observed in this population (Zwicker et al., 2009).

1.2.3 Treatment and intervention approaches

Two main types of interventions for children with DCD have been described in the literature: process-oriented and task-oriented. Process-oriented approaches, also known as bottom-up approaches, target the presumed underlying sensory and motor processing deficits. For example, these approaches can involve stimulating and training the proprioceptive, kinesthetic or vestibular systems with the intent that it will transfer to a wide range of tasks and activities (e.g., sensory integration therapy (Ayres, Robbins, & McAtee, 1979); kinesthetic training (Laszlo & Bairstow, 1983).

The other main types of interventions are task-oriented, or top-down approaches, which take a more dynamic systems approach to intervention. These approaches tend to focus on child- and family-centered goals and cognitive strategies to learn functional movement skills. The Cognitive Orientation to daily Occupational Performance (CO-OP) is one such program (Polatajko et al., 2001). This program allows children to make their own goals, and they are then taken through the learning process and taught problem solving strategies that can be generalized to the learning of other motor tasks. Neuromotor task training (NTT) is another such top-down approach that targets both cognitive (e.g., fear, attention, motivation) and motor control (e.g., timing and execution) difficulties by manipulating task and/or environmental constraints (Niemeijer, Smits-Engelsman, & Schoemaker, 2007). As with CO-OP, the ultimate goal of NTT is teaching functional skills that can be transferred to performance in daily activities.

Two recent meta-analyses concluded that the effects of process-oriented approaches were weak (weighted Cohen's *d*=0.12) and had little effect on the improvement in occupational performance in children with DCD (Armstrong, 2012; Smits-Engelsman et al., 2013). Task-oriented approaches conversely, were found to produce much stronger positive effects (weighted Cohen's *d*=0.83 and *d*=0.89, respectively) (Smits-Engelsman et al., 2013). These top-down approaches are likely more effective at improving daily functioning as they focus on teaching essential activities of daily living, which encourage children to increase their participation in numerous aspects of their life, such as at school or leisure pursuits. A recent review by Offor and colleagues (2016) investigated intervention strategies in the context of physical therapy and found task-oriented approaches and also traditional physical therapy approaches (e.g., motor skill, strength and core-stability training), had the greatest benefits. In addition, although the evidence is limited, contemporary physical therapy approaches including active virtual gaming, hippotherapies and aquatic therapies have demonstrated promising results (Offor et al., 2016).

Regardless of the type of intervention, the primary focus is on improving motor abilities, whether at a deeper mechanistic level or at a more functional level. This is not surprising considering that motor coordination deficits are at the core of DCD; however, it is expected that improvement in these motor deficits will then lead to improvements in the secondary consequences of the disorder such as physical activity participation and health-related fitness. However, there is little evidence that motor-based interventions will lead to significant increases in physical activity or improvement in fitness levels either acutely or over the long term (Bremer & Cairney, 2016). It is likely that interventions will need to take a more holistic approach, targeting not only the motor deficits but also psychological constructs and environmental constraints that may also be limiting participation. For example, research has shown that school-age children with DCD not only lack competence, but often lack the confidence (i.e. self-efficacy) to engage in various types of physical activity (Cairney, Hay, Faught, Mandigo, & Flouris, 2005). These self-efficacy beliefs are a salient predictor of physical activity behaviour in childhood and adolescence (Sallis et al., 2016). Teaching and reinforcing motor competencies in environments that foster autonomy, internal motivation and self-efficacy (e.g., mastery motivational climates) at a young age may therefore have more sustained effects on physical activity engagement across the lifespan (Ames, 1992; Dunn & Watkinson, 2002; Nicholls, 1984; Robinson, 2011a).

1.3 Theoretical context linking poor motor competence to physical health through hypoactivity

In the sections that follow, the theoretical models linking motor competence to physical activity and health-related fitness in childhood from both the perspectives of those studying impaired motor functioning and those studying the normative development of fundamental motor skills will be discussed. Importantly, synergies across these literatures, which, as previously argued, have largely evolved independently of one another, will be highlighted

1.3.1 Activity deficit and skill-learning gap hypotheses

Dr. Bar-Or, a clinician-scientist and pioneer in the field of pediatric exercise medicine, identified and highlighted decreased physical activity in children with medical conditions commonly characterized by impaired gross motor functioning, such as children with arthritis, cerebral palsy and muscular dystrophy (Bar-Or, 1983). The deficits in activity compared to healthy children were attributed to the disease-related physical motoric difficulties. Since then, a number of researchers have elaborated on this '***activity deficit hypothesis***' in otherwise healthy children with motor difficulties such as those with Developmental Coordination Disorder (DCD).

Children with DCD are thought to withdraw and avoid participation in physical activities because of a history of previous failures in the motor domain. For example, unsuccessful attempts at engaging in sport-related activities often lead to a lack of confidence in their motor abilities and exclusion by their peers (Cantell, Smyth, & Ahonen, 1994; Poulsen, Ziviani, & Cuskelly, 2008; Schoemaker & Kalverboer, 1994). Bouffard and colleagues (1996) were among the first to specifically test this activity deficit hypothesis in children with poor motor coordination in the absence of a clinical diagnosis of CP or other known neurological condition that would impact motor ability. They observed that these children engaged in significantly less vigorous activity during unstructured recess time compared to children without movement difficulties. This ‘activity deficit’ has also been found in self-reported free play and organized activities (Cairney, Hay, Faught, Corna, & Flouris, 2006), and has now been replicated in numerous different contexts using varying physical activity measurement techniques to compare school-age children with DCD to children who are typically developing (Rivilis et al., 2011).

Wall (2004) furthered our understanding of the activity deficit model from a developmental context. He postulated a ***developmental skill-learning gap*** that widens over time, as the nature of child’s play becomes more demanding and complex. Children with motor learning deficits will find it increasingly difficult and less enjoyable to participate and engage in activities of increasing complexity, especially without a solid foundation of automatized fundamental motor skills. This widening gap in motor skills would therefore result in widening activity deficit over developmental time.

Hands and Larkin (2002) also built on the activity deficit model by specifically highlighting the secondary health-related consequences of the activity deficit for children with DCD. They postulate that poor motor competence will lead to increasing hypoactivity and subsequently increase the risk of poor health-related fitness, including higher risk of obesity and lower cardiorespiratory fitness as children age. They also recognized that the interrelationships among these constructs are likely bidirectional, with each influencing one another. In other words, just as poor motor competence can lead to hypoactivity that can hinder the development of health-related fitness, poor health-related fitness will make it difficult to engage in physical activity, and withdrawal from activity will limit opportunities for further development of motor skills. This is conceptualized as a continuous negative feedback loop whereby poor motor skills, physical activity, and health-related fitness will continuously decline over time. Cairney (2015a) further developed this model to highlight the developmental processes involved, with the consequences of this negative feedback loop increasing over time, leading to greater risk of poor heath outcomes (e.g., cardiovascular disease) if the core components of the model are left unchanged (Figure 1-1).

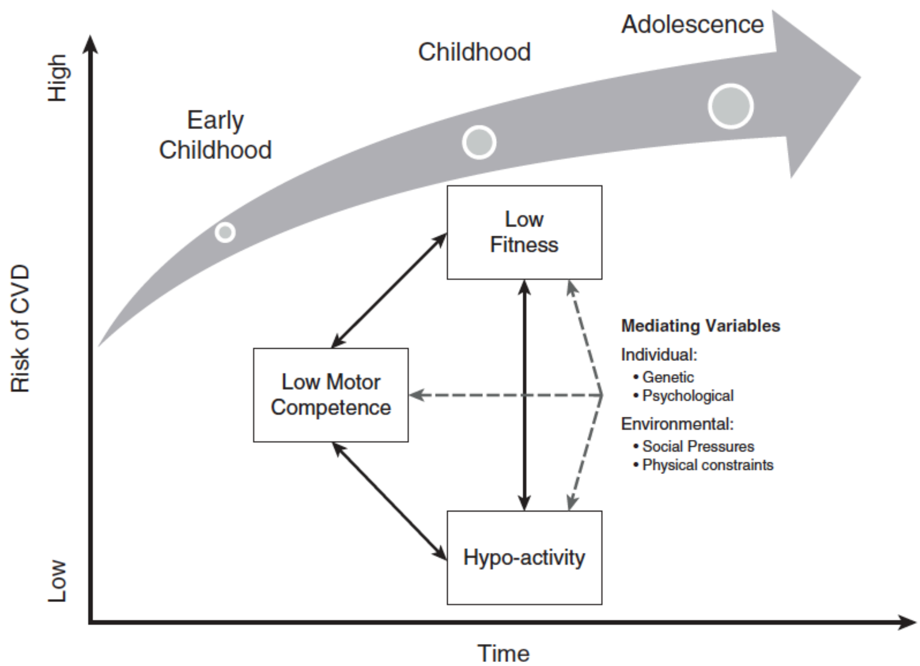

Figure 1-1 Continuous negative feedback loop over time.

Time is depicted on the x-axis and risk of poor cardiovascular heath (e.g., obesity, poor physical fitness, elevated blood cholesterol) is presented on the y-axis. The arrow that cuts across the top represents a developmental trajectory over time, with the dots distinguishing critical developmental periods in which the interactions between motor competence, physical activity and health-related fitness interact to produce health risks. The widening of both the dots and arrow over time represents the increasing relative impact of this negative feedback loop on the risk of poor cardiovascular heath across developmental time. CVD=cardiovascular disease. Reproduced from Cairney (2015a), with permission from University of Toronto Press (Appendix C).

While there are some differences between the aforementioned models, all models share the view that deficits in motor abilities, regardless of their cause, will have a negative impact on physical activity participation. Indeed, childhood disabilities and disorders will often hinder the acquisition and mastery of the necessary motor skills required to succeed and persist with physical activity. Collectively, poor motor functioning is regarded as a root cause of the ‘activity deficit’ observed in a number of clinical pediatric populations, including DCD. As it is well recognized that physical activity is associated with numerous health benefits (Janssen & LeBlanc, 2010), the aforementioned theories either imply (Wall, 2004) or explicitly link (Bar-Or, 1983; Hands & Larkin, 2002) hypoactivity in these special populations to negative health outcomes, including poor health-related fitness.

1.3.2 Stodden’s model of motor development

In 2008, Stodden and colleagues (2008) published a developmental framework, building on previous models of motor development (Clark & Metcalfe, 2002; Seefeldt, 1980). Stodden’s model highlights motor competence as a key component to positive health trajectories, specifically reduced obesity risk, through its evolving relationship with physical activity, and mediated by perceived motor competence and health-related fitness.

Although Stodden's model does not explicitly discuss DCD (or other medical conditions that directly affect motor competence), their support of Seedfeld's (1980) motor "proficiency barrier" hypothesis recognizes that a certain level of competency in fundamental motor skills (i.e. locomotor and object control skills) is necessary for children to engage in more complex sports and games. Children who are unable to reach this 'critical threshold' of motor competence will subsequently withdraw from physical activity and sport at higher rates. The exact threshold has not been quantified or empirically studied in typically developing children; however, it is plausible that many of the children who fall below this critical threshold also meet the criteria for DCD. As such, the growing body of literature on the 'activity deficit' in DCD is likely able to help identify where the critical threshold falls on the continuum of motor abilities.

Stodden and colleagues also reference and support Clark and Metcalf's (2002) model of motor development. In this model, learning to climb a mountain is used as an analogy for motor development. They describe fundamental motor skills as representative of the foundation or "base camp". In order to ascend the mountain, a strong foundation of movement skills is required. Children will follow different trajectories up the mountain (influenced by differences in abilities, environmental constraints and practice) to reach the peak, which is compared to the attainment of a skilled motor action (Clark & Metcalfe, 2002). Overall, Stodden and colleagues' (2008) developmental model recognizes that the development of proficient motor competence is a key component that will promote and sustain physical activity engagement over time.

Much like Hands and Larkin's model (2002), Stodden and colleagues (2008) theorize that the relationships between motor skill competence, physical activity and health-related fitness are bidirectional, but also incorporate a novel concept that these relationships are dynamic, and are strengthened over developmental time. They suggest that in the early years the relationship between motor competence and physical activity will be weak due to the wide variability in motor skills and physical activity, driven by differing levels of experience and environmental constraints (parental influences, socioeconomic status, physical education etc.). Over time, children with a greater skill base will be more compelled to engage in sport and active games and self-select into these activities, whereas less motorically competent children will begin to withdraw from physical activity. These compounding factors will in turn strengthen the relationship between motor competence and physical activity in the middle to late childhood period.

This dynamic relationship between motor competence and physical activity is central to Stodden's model, but the model also recognizes that this relationship will be mediated by levels of health-related fitness and perceived motor competence. These mediating relationships between motor competence and both health-related fitness and perceived motor competence are also hypothesized to become stronger over time as neuromotor and cognitive systems develop. Unlike Hands and Larkin, Stodden's main pathway suggests health-related fitness will mediate the association between motor competence and physical activity. In other words, children with more advanced motor competence will develop greater health-related fitness, which will allow them to persist and sustain physical activity engagement. Children with poor motor competence will fall into a 'negative spiral of disengagement' as poor fitness will restrict their ability to be physically active, which further limits the development of motor competencies. As Stodden and colleagues also recognize the bidirectionality among these constructs, they acknowledge, as proposed by Hands and Larkin, that children with low motor competence will not continue to be as physically active in middle childhood, and will, therefore, not be able to develop or maintain aspects of health-related fitness. Therefore, although the primary pathways are different, Stodden's 'negative spiral of disengagement' parallels the 'negative feedback loop' proposed by Hands and Larkin (2002). Overall, Stodden and colleagues

(2008) suggest that the associations and interaction among these factors will strengthen over time, leading to higher risk of obesity stemming from poor motor competence.

Although the models outlined above have been tailored to different audiences and focus on different populations, they all highlight that the relationships among motor competence, physical activity and health-related fitness are multifaceted, mutually reinforcing, and do not occur in isolation. Building on dynamic systems theory (Thelen & Smith, 1994), the models also recognize that individual (e.g., genetic, psychological), and environmental factors (e.g., social pressures, parental support) will interact to influence motor development and the strength of these relationships over time (Clark & Metcalfe, 2002; Hands & Larkin, 2002; Stodden et al., 2008). Taken together, poor motor competence is theorized to be an important determinant of hypoactivity and negative health outcomes as children age (Bar-Or, 1983; Hands & Larkin, 2002; Seefeldt, 1980; Stodden et al., 2008; Wall, 2004). Although it is presumed that early childhood is a time in which many of the secondary consequences of poor motor functioning have yet to manifest, this is largely an untested assumption. The next section highlights the current evidence supporting the aforementioned models.

1.4 **Current supporting evidence for the theoretical models**

1.4.1 Physical activity and health-related fitness in children with Developmental Coordination Disorder

Recent reviews have summarized and highlighted the emerging evidence of the negative impact of DCD on physical activity and health-related fitness in childhood (Cairney, 2015b; Cairney & Veldhuizen, 2013; Hendrix, Prins, & Dekkers, 2014; Rivilis et al., 2011). A series of studies that has dominated this area of inquiry comes from the Physical Health and Activity Study Team (PHAST) project by Cairney and his colleagues (Cairney, Hay, Veldhuizen, Missiuna, & Faught, 2010). The project set out to study the impact of DCD on physical activity, self-efficacy towards physical activity, cardiorespiratory fitness, and weight status over time in a cohort of over 2,000 children, starting in grade 4. This study was highly influential due to the large size of the cohort, the longitudinal study design, and the use of both field and laboratory-based protocols.

The results of the PHAST project confirmed that DCD was associated with lower levels of self-reported participation in both free play and organized sport and physical activity (Cairney, 2015a; Cairney, Hay, Veldhuizen, Missiuna, & Faught, 2010). They also found that children with DCD had a higher BMI, waist circumference, were at a greater risk of overweight/obesity (Cairney, Hay, Veldhuizen, Missiuna, Mahlberg, et al., 2010), and had lower levels of cardiorespiratory fitness (Cairney, Hay, Veldhuizen, & Faught, 2011b) compared to their typically developing peers. As these data were longitudinal, the investigators were able to identify that these deficits persisted, and in some cases, increased, over time from grade 4 (age 9-10) to grade 8 (age 13-14). Another interesting finding was that sex tended to moderate these associations, specifically with respect to physical activity participation. While both boys and girls with DCD reported reduced participation in free play and organized activity, the gap widened over time for girls, but not boys (Cairney, Hay, Veldhuizen, Missiuna, & Faught, 2010).

A subsample of approximately 100 children from the PHAST cohort also completed laboratory-based testing where body composition was determined via whole-body air displacement plethysmography, aerobic fitness was determined by assessing maximal oxygen uptake (VO_2max) on a cycle ergometer, and physical activity was measured objectively using accelerometers**.** This smaller sub-sample confirmed the findings from the field, in which children with DCD had higher fat mass (Cairney, Hay, Veldhuizen, & Faught, 2011a), lower VO_2max (Cairney, Hay, Veldhuizen, & Faught, 2010), and lower levels of moderate-to-vigorous physical activity (Batey et al., 2014; Kwan, King-Dowling, Hay, Faught, &

Cairney, 2016); however, when physical activity was measured objectively, the deficits appeared to be larger for boys.

Consistent with the PHAST results, other research groups have reported the same pattern of results, with children with DCD being less physically active (Beutum, Cordier, & Bundy, 2013; Cermak et al., 2015; Green et al., 2011), having higher BMI (Cantell, Crawford, & Doyle-Baker, 2008; Hands & Larkin, 2006; Lifshitz et al., 2014) and poorer aerobic fitness (Chia, Guelfi, & Licari, 2010; Gillian D. Ferguson, Aertssen, Rameckers, Jelsma, & Smits-Engelsman, 2014; Haga, 2009; Hands, 2008; Hands & Larkin, 2006; Li, Wu, Cairney, & Hsieh, 2011; Lifshitz et al., 2014; Wu, Lin, Li, Tsai, & Cairney, 2010) compared to their typically developing counterparts.

Musculoskeletal fitness and flexibility are two important dimensions of health-related fitness that were not investigated in the original PHAST project. There is accumulating evidence that children with DCD also present with poorer muscular strength and endurance when compared to typically developing children (Aertssen, Ferguson, & Smits-Engelsman, 2016; Cantell et al., 2008; Gillian D. Ferguson et al., 2014; Haga, 2009; Hands, 2008; Hands & Larkin, 2006; O’Beirne, Larkin, & Cable, 1994; Raynor, 2001; Tsiotra, Nevill, Lane, & Koutedakis, 2009). Although these studies have been largely cross sectional in nature, a limited body of longitudinal data suggests that musculoskeletal fitness (standing long jump, 50m sprint and sit-ups) deficits persist (Hands, 2008), or emerge across time in children with DCD (Li et al., 2011). In terms of flexibility, mixed findings have been reported with some studies concluding that children with DCD have poorer flexibility compared with controls (Cantell et al., 2008; Hands & Larkin, 2006; Li et al., 2011), and others showing no flexibility deficits (Gillian D. Ferguson et al., 2014; Schott, Alof, Hultsch, & Meermann, 2007; Tsiotra et al., 2009).

DCD is viewed increasingly as a fundamental determinant of physical (in)activity, and its resultant health-related consequences (Cairney & Veldhuizen, 2013). While poor motor competence may directly influence physical activity participation, it is also likely mediated by a myriad of psychosocial factors including low self-efficacy or confidence in their physical capabilities, low motivation, reduced enjoyment, and/or exclusion by peers due to previous negative physical activity experiences (Cairney, Hay, Faught, Wade, et al., 2005; Mandich, Polatajko, & Rodger, 2003; Poulsen, Ziviani, Cuskelly, & Smith, 2007). As discussed in Hands and Larkin's (2002) activity deficit model, low fitness levels in children with DCD are thought to be a consequence of this observed deficit in physical activity. To date, very few studies have tested this pathway, and have reported conflicting results. Indeed, recent work using the PHAST sample failed to find a meaningful long-term mediating effect of self-reported physical activity on cardiorespiratory fitness (Cairney, Veldhuizen, King-Dowling, Faught, & Hay, 2017), BMI or waist circumference (Joshi et al., 2015). Conversely, when examining the PHAST subsample tested in the laboratory at ages 11-12, objectively-measured physical activity was found to mediate the cross-sectional relationship between DCD and VO_2 max (Silman, Cairney, Hay, Klentrou, & Faught, 2011). These discrepancies may be due to the average age of the samples or the reliance of field versus laboratory measurement techniques.

In summary, it is clear that children with DCD are less likely to participate in physical activity, including both organized physical activity and discretionary play (Rivilis et al., 2011). They also present with lower levels of fitness and unhealthy weight, when compared to typically developing children (Hendrix et al., 2014; Rivilis et al., 2011). However, it is still unclear whether the activity deficit is an important contributor to the observed poor health-related fitness outcomes. Moreover, the majority of research in this field has examined children over the age of 7, a time in which a significant gap in physical activity and fitness is already present. Given that hypoactivity and poor health-related fitness are thought to be secondary consequences of the disorder that emerge over time, determining how young children are when these deficits appear is a critical, unanswered question in the DCD literature. Studying children with DCD at preschool-age may help to identify a critical period when deficits may be less pronounced and secondary prevention interventions may have the greatest impact.

1.4.2 Motor competence and its relationship to physical activity and health-related fitness in typically developing children

Research examining the relationship between motor competence and physical activity in typically developing children has been the subject of numerous recent reviews (Figueroa & An, 2017; Holfelder & Schott, 2014; Logan, Kipling Webster, Getchell, Pfeiffer, & Robinson, 2015; Loprinzi, Davis, & Fu, 2015; Lubans, Morgan, Cliff, Barnett, & Okely, 2010; Robinson et al., 2015). These reviews conclude that there is now strong evidence of a positive correlation between motor competence and physical activity, but the magnitude of these associations is highly variable and appears to vary based on age, sex and physical activity intensity. For example, Logan and colleagues (2015) reviewed the extant literature on the relationship between process-oriented assessments of fundamental motor skills and physical activity at different developmental ages. Although the number of included studies in their review was small (n=13), they concluded that low to moderate correlations between fundamental motor skills and physical activity existed in early childhood, whereas low to high associations were found in middle to late childhood. They also noted that object control skills tend to be more related to physical activity for boys, whereas locomotor skills may be more strongly associated with activity levels in girls. Figueroa and An (2017) reviewed 11 studies that focused solely on preschoolers (3-5 years old) and also found a significant, positive relationship between motor competence and physical activity in a large majority of the extant literature. The strength of these associations appeared to be dependent not only on age and sex, as reported by Logan and colleagues, but also on physical activity intensity, whereby associations with vigorous or moderate-to-vigorous intensity activity were more robust compared with those involving light intensity activity.

Since these reviews, studies in school-age children have further supported the positive association between motor competence and physical activity, where children with high actual and perceived motor competence present with the highest activity levels (De Meester et al., 2016, 2017; Gu, Thomas, & Chen, 2017; Khodaverdi, Bahram, Stodden, & Kazemnejad, 2016). Unlike their older counterparts, and in line with Stodden's model, the associations in preschool-age children have been relatively weak (Guo, Schenkelberg, O'Neill, Dowda, & Pate, 2017) or non-significant (Iivonen et al., 2016; Lopes, Barnett, & Rodrigues, 2016; Loprinzi & Frith, 2017).

The relationship between motor competence and health-related fitness has received more attention in recent years. Updating the review by Lubans (2010), Cattuzzo and colleagues (2016) systematically reviewed the literature and concluded that there is now strong evidence for a positive association between motor competence and both aerobic and musculoskeletal fitness, and an inverse association between motor competence and body weight status in children and adolescents. The evidence supporting an association with flexibility remains inconclusive. Unlike the physical activity findings, these associations appeared to be similar for boys and girls. Since the time of this review, these results have been further replicated in school-age children, finding moderate to high correlations between motor competence and health-related fitness (e.g., cardiorespiratory fitness, hand grip strength, body fat percentage, BMI) in both boys and girls (Gu et al., 2017; Luz, Rodrigues, Meester, & Cordovil, 2017; Marmeleira, Veiga, Cansado, & Raimundo, 2017; Milne, Leong, & Hing, 2016). In these studies, gross motor, and particularly locomotor skills, tended to show the strongest associations with health-related fitness outcomes. It is important to note that almost all of the studies investigating cardiorespiratory and musculoskeletal fitness utilized field-based fitness tests (e.g., FITNESSGRAM (Plowman & Meredith, 2013). Indeed there is currently a paucity of evidence examining these associations using reference-standard laboratory-based assessments, especially in the early years.

In 2015, Robinson and colleagues (2015) published a narrative review to update the state of the evidence for Stodden's developmental model. In keeping with the aforementioned reviews, they concluded that the direct associations between motor competence and health-related fitness, physical activity, and perceived motor competence are strongly supported in the extant literature. Conversely, evidence of the proposed mediation pathways and strengthening associations across developmental time among these constructs is still quite limited. Importantly, these conclusions are largely based on cross-sectional evidence from different age groups, which limits our ability to 1) conclude that the strength of these relationships do, in fact, increase over time, and 2) determine if motor skills predict future physical activity and fitness levels. In recent years, a growing body of longitudinal evidence examining these associations has emerged and will be the focus of the rest of this section.

The large majority of longitudinal studies have found motor competence in early to middle childhood to be predictive of future physical activity levels in later childhood, adolescence, and even into adulthood. With the exception of one study (McKenzie et al., 2002), motor competence has demonstrated positive associations with future organized physical activity and sport participation (Barnett, Van Beurden, Morgan, Brooks, & Beard, 2009; Henrique et al., 2016; Vandorpe et al., 2012), self-reported physical activity (Barnett, Morgan, Beurden, & Beard, 2008; Barnett, Van Beurden, et al., 2009; de Souza et al., 2014; Jaakkola, Yli-Piipari, Huotari, Watt, & Liukkonen, 2016; Lloyd, Saunders, Bremer, & Tremblay, 2014), and daily step counts (McIntyre, Parker, Chivers, & Hands, 2017; Venetsanou & Kambas, 2016). Furthermore, higher levels of motor competence at age 6 are linked to slower rates of decline in self-reported physical activity over 3 years compared to children with lower motor competence levels (Lopes, Rodrigues, Maia, & Malina, 2011); however, this same pattern was not observed over a shorter 9-month duration in preschool children (Bürgi et al., 2011). On the whole, it appears that childhood motor competence plays a role in future physical activity participation, and despite the sometimes low predictive values (i.e. r^2 <0.2), the largely consistent significant findings are promising given the myriad of other factors that influence physical activity participation across childhood (Sallis et al., 2000).

Relatively few longitudinal studies have examined the relationships between motor competence and future health-related fitness outcomes in typically developing children. Object control skills, in children as young as preschool age, have been found to be associated with adolescent performance on the Léger 20m shuttle run (Léger, Mercier, Gadoury, & Lambert, 1988) and AAHPERD health-related fitness test (i.e. body fat percentage, sit-ups, 1.5 mile run (American Alliance for Health, Physical Education, Recreation, and Dance, 1980)) (Barnett et al., 2008; Vlahov, Baghurst, & Mwavita, 2014). There is also evidence that gross motor competence is associated with long-term changes in BMI (D'Hondt et al., 2014; Martins et al., 2010). Fitness levels may also interact with motor competence to predict future physical activity and weight status. For example, Lopes and colleagues (2012; 2011) found that higher initial motor competence and fitness performance was associated with higher physical activity and slower accumulation of subcutaneous adiposity across childhood.

These longitudinal studies provide stronger support for a predictive link between motor competence and both future physical activity and health-related fitness. However, the majority have relied on self-reported physical activity outcomes, which are subject to recall and response biases (Sallis & Saelens, 2000), and/or field-based assessments of fitness which are typically assessed at only one point in time, therefore limiting our ability to examine how these relationships change over time. Recently, Lima and colleagues (Lima, Pfeiffer, Bugge, et al., 2017; Lima, Pfeiffer, Larsen, et al., 2017) published results from a longitudinal study using more rigorous methodologies to test some of the proposed pathways in Stodden's developmental model. They found that motor competence and objectively-measured vigorous physical activity demonstrated significant reciprocal longitudinal associations from childhood (age 6) to early adolescence (age 13), and these bidirectional relationships were mediated by cardiorespiratory fitness (VO_2max) (Lima, Pfeiffer, Larsen, et al., 2017). They also found that motor competence was directly associated with body fatness over time, and that this association was mediated by VO_2max, but not moderate-to-vigorous or vigorous physical activity (Lima, Pfeiffer, Bugge, et al., 2017). Although both analyses revealed a direct relationship between motor competence and cardiorespiratory fitness, they did not test if physical activity mediated this association. These recent studies are the first to directly test the mediation pathways in the conceptual framework proposed by Stodden and colleagues using repeated objective methods in a longitudinal design.

It is important to note that these aforementioned studies by Lima and colleagues did not assess musculoskeletal fitness, and more than half the children in this sample received an intervention that doubled the amount of time spent in physical education classes, potentially confounding the observed relationships. Additionally, these studies did not report how the associations between motor competence, physical activity, and fitness changed over time. McIntyre and colleagues (2017) recently examined motor competence and daily step counts in a cohort of children age 6- to 8- years followed over 18-months. They found an insignificant correlation between motor competence and physical activity at age 6, but a significant relationship emerged at age 7 in boys, and age 9 in girls. These findings provide initial support for a strengthening association between motor competence and physical activity across the primary school years.

The extant evidence from the aforementioned longitudinal observational studies suggests a causal relationship between motor competence and health and physical activity outcomes. If this causal relationship holds true, we would expect interventions targeting and improving motor competence to also produce positive changes in these constructs. Currently, the evidence is very limited as the majority of motor intervention studies typically focus solely on improvement to motor skills and many do not measure or report physical activity or fitness outcomes (Bremer & Cairney, 2016; Morgan et al., 2013). Of the interventions that do report these outcomes, some have led to positive changes in physical activity and health-related fitness (Cohen, Morgan, Plotnikoff, Callister, & Lubans, 2015; Johnstone, Hughes, Janssen, & Reilly, 2017; Matvienko & Ahrabi-Fard, 2010; Miller et al., 2015; Salmon, Ball, Hume, Booth, & Crawford, 2008), while others have shown no significant improvements (Barnett, van Beurden, et al., 2009; Barnett, Zask, Rose, Hughes, & Adams, 2015; Cliff et al., 2011; R. A. Jones et al., 2011; R. A. Jones, Okely, Hinkley, Batterham, & Burke, 2016).

Taken together, observational research strongly supports a positive association between motor competence and both physical activity and health-related fitness in childhood and adolescence. These relationships are hypothesized to be weaker at preschool age and proposed to strengthen over time, but a paucity of studies have empirically tested this theory using longitudinal techniques. Recent studies have begun exploring the mediating pathways among these constructs, focusing primarily on school-aged children and the proposed pathways by Stodden and colleagues (i.e. testing if heath-related fitness and/or perceived motor competence mediates the relationship between motor competence and physical activity). Few, however, have examined physical activity as a potential mediator promoting or hindering health-related fitness as proposed by Hands and Larkin. Early childhood is a critical period when interventions may have the greatest benefit. Therefore, it is essential that we understand the nature of these interrelationships across these formative years. If we can identify when poor motor competence begins to interfere with physical activity participation and health-related fitness, we can design more effective, and timely interventions that promote engagement in physical activity across the lifespan, while reducing the risk of negative health outcomes in children with motor difficulties.

1.5 **Objectives and hypotheses**

1.5.1 General objective

The general objective of this thesis is to test the core pathway in Hands and Larkin's activity deficit model by determining if physical activity levels mediate the association between motor competence and health-related fitness in the early years.

1.5.2 Specific objectives

The specific objectives of the studies in this thesis are to:

1) Explore the activity-deficit hypothesis in preschool children with and without DCD by examining the differences in the intensity and

patterns of physical activity accumulation between preschool children who meet criteria for DCD compared to typically developing children (Chapter 2).

2) Examine differences in health-related fitness (body composition, aerobic fitness, musculoskeletal fitness, and flexibility) between preschool children who meet criteria for DCD and typically developing children, and determine if these differences are mediated by daily vigorous physical activity levels (Chapter 3)

3) Determine how the strength of the association between motor competence and physical activity changes over time across early childhood, and examine if vigorous physical activity mediates the longitudinal relationship between motor competence and musculoskeletal fitness, specifically short-term muscle power, from preschool to early childhood in typically developing children (Chapter 4)

1.5.3 Specific hypotheses

The specific hypotheses of the studies included in this thesis were that:

1) Preschool-age children with DCD would have slightly lower levels of daily moderate-to-vigorous and vigorous physical activity, but the differences would be smaller than those observed in school-age children. In addition, they would accumulate their activity in shorter, less frequent bouts compared to typically developing children (Chapter 2).

2) Preschool-age children with DCD would have slightly reduced health-related fitness compared to typically developing children and this would be partially explained (mediated) by vigorous physical activity levels (Chapter 3).

3) The strength of the associations between physical activity and motor competence would increase over the study period. Motor competence would positively predict short-term muscle power over time and this would be partially mediated by vigorous physical activity engagement (Chapter 4).

1.6 **Methodological note**

Chapters 2 and 3 present findings from the same cross-sectional baseline cohort of preschool children (age 4- to 5-years) from the

Coordination and Activity Tracking in CHildren (CATCH) study (Cairney et al., 2015). This study aimed to recruit 300 typically developing children and 300 children at risk for DCD from Hamilton, Ontario and surrounding area. In order to reach the target sample size, children were screened for motor coordination difficulties either over the phone or in the laboratory. Children who were identified as having motor delays on a standardized assessment of motor coordination were automatically enrolled into the longitudinal cohort, whereas typically developing children were randomized either into or out of the cohort. Significant overlap with regards to the 1) details of the CATCH study design, 2) recruitment procedures and 3) participant characteristics is therefore evident in Chapters 2 and 3. A diagram of the flow of participants through CATCH study recruitment, which occurred from October 2013 to June 2017, is provided in Appendix A.

Chapter 4 presents findings from all 3-years of the Health Outcomes and Physical activity in Preschoolers (HOPP) study, a separate longitudinal cohort study of preschool children recruited from 2010-2012 from Hamilton, ON and surround regions (Timmons, Proudfoot, MacDonald, Bray, & Cairney, 2012). Unlike the CATCH study, this study aimed to recruit a convenience sample of healthy children 3- to 5-years of age form the community, with no randomization procedures implemented.

It is important to note that the recruitment timeline for the HOPP and CATCH studies did not overlap and represent two distinct samples of preschool children. The recruitment flow diagram and subsequent reasons for non-participation at each year of the HOPP study is presented in Appendix B.

CHAPTER 2: **Physical activity in young children with and without motor coordination difficulties**

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2.1 ABSTRACT

Purpose
Children with Developmental Coordination Disorder (DCD) are less active than typically developing (TD) children; however, little is known about when, developmentally, this activity deficit emerges. The purpose of this study was to determine if the levels and patterns of physical activity accumulation in young children with DCD are different than TD children across intensities and planes of movement.

Methods
Five hundred and ninety-two 4-to 5-year olds were recruited as part of the Coordination and Activity Tracking in Children (CATCH) study. Motor skills were assessed using the Movement Assessment Battery for Children-2; children scoring $\leq 5^{th}$ percentile comprised the DCD group (n=112), between the 6-16^{th} percentile considered to be at risk for DCD (rDCD, n=179), and children >16^{th} percentile considered TD (n=301). Physical activity was measured using Actigraph accelerometers (wGT3X) worn around the hip for 7 days. Average daily intensity of activity (sedentary, light, moderate, vigorous, and total), frequency and duration of moderate-to-vigorous PA (MVPA) bouts, and triaxial activity counts per minute (cpm) were determined. Group differences were tested using ANCOVA, with wear time, sex, and age entered as covariates.

Results
No difference in daily activity in any intensity or plane of movement was found among the 3 groups. However, young children with DCD accumulated their MVPA in slightly shorter bouts compared to TD children.

Conclusions
Differences in bout duration may have important implications for future health and fitness, especially if they become more pronounced over time. Early motor skill interventions may be able to minimize the gap in activity observed in older children with DCD.

2.2 INTRODUCTION

Affecting approximately 5-6 percent of children, Developmental Coordination Disorder (DCD) is a common neurodevelopmental disorder characterized by problems with fine and/or gross motor coordination (1). Children with DCD have lower physical activity levels compared to their typically developing (TD) peers (2). This has been found for both self-reported free play and organized activities (3) as well as objectively-measured moderate-to-vigorous physical activity (MVPA) (4). A hallmark feature of DCD is that difficulties begin early in development (1); however, DCD is typically identified after children enter school-age when motor delays begin to have a more noticeable impact on activities of daily living, including academic and leisure pursuits (e.g., dressing, writing, hygiene, sport participation). Consequently, the majority of DCD research has focused on the mid- to-late childhood period, a time in which physical activity deficits are already apparent. It is still unclear when, developmentally, this activity deficit emerges in children with motor delays.

The activity deficit hypothesis (5–7) suggests that the gap in physical activity between children with and without DCD will be minimal in early childhood but will widen over time as children with motor coordination difficulties begin to withdraw more and more from active games and/or organized sport and activity. This activity deficit is suspected to be due to a variety of physical and psychosocial factors, including increasing motor skill demands of play, reductions in aerobic fitness, and low self-efficacy towards physical activity (5, 8, 9). Since withdrawing from physical activity may have a detrimental impact on both physical and psychosocial functioning as these children age, if we are to encourage earlier identification and intervention, then it is important that we identify how young children are when movement difficulties begin to interfere with physical activity participation.

There is now strong evidence that motor competence is positively associated with physical activity in TD children and youth, and this relationship has been reported across varying assessments of motor competence (product and process orientated assessments) and both self-reported and objectively-measured physical activity (10–12). In early childhood, this relationship appears to be in the low-to-moderate range (10), and is hypothesized to strengthen over developmental time (13). Critically, emerging longitudinal evidence suggests that motor competence in early childhood positively predicts future physical activity levels (14–16).

In studies utilizing accelerometry for the assessment of physical activity in young children, relationships with motor competence tend to vary based on gender and/or intensity of activity. For example, Williams and colleagues (17) found young children in the highest motor skill tertile spent significantly more time in MVPA, but not light physical activity (LPA), a finding that replicated earlier findings by Fisher and colleagues (18) in young children. Cliff and colleagues (19) found that overall gross motor skills were positively associated with vigorous physical activity (VPA) in preschool-age boys; however, they found a negative association with moderate physical activity (MPA) in girls. Although they did not report associations with LPA, they found no association with total physical activity (TPA) as measured using average activity counts per minute (cpm) (19). In contrast, Iivonen and colleagues (20) found a significant positive relationship with both MVPA and TPA cpm with motor skills in 4-year-old children. Overall, it appears that in the early years, motor coordination is positively associated with objectively-measured physical activity in the moderate-to-vigorous range, and that this relationship may differ based on gender.

With advances in technology, accelerometers are now able to capture movement in very short sampling intervals (e.g., 1s epochs) across multiple axes. Despite these capabilities, the majority of accelerometer research in childhood has focused solely on the vertical axis, ignoring movement in the anterioposterior and mediolateral planes.

Although the vertical plane dominates in ambulatory activities, accelerations in the other two planes are likely more sensitive in capturing many non-ambulatory types of activities popular in childhood, such as climbing and jumping (21, 22), which are also likely to have benefits for health, especially muscle and bone development. In addition, recent work has found that patterns of MVPA accumulation (bout frequency and duration) may also have important implications for health outcomes in childhood, with both longer bout durations and higher bout frequency having positive influences on weight status, aerobic fitness and cardiovascular disease risk factors (23).

Given their motoric difficulties, it is possible that movement inefficiencies may cause young children with DCD to accumulate their activity differently, not only in terms of overall intensity, but also with respect to the number and duration of activity bouts as well as movement in other planes. To date, there is a paucity of research examining objectively-measured physical activity in young children with DCD. By capturing a more holistic view of movement patterns in young children, we will be able to determine if delayed motor coordination is associated with the volume and/or pattern of physical activity accumulation throughout the day. This knowledge will lay the foundation for determining how these movement patterns relate to overall health and fitness in young children with coordination difficulties and inform the development of appropriate interventions to target these health behaviours.

Therefore, the purpose of this study was to determine if there are differences in activity levels, patterns and planes among young children (age 4-5) who have DCD (≤5th percentile), children at risk for DCD (rDCD, between the 6-16th percentile) and TD children (>16th percentile). As the strength of the relationship between motor-competence and physical activity are typically low to moderate and restricted to MVPA intensities, we hypothesize that young children with DCD and rDCD will have slightly lower daily MPA and VPA, potentially due to both fewer and shorter average daily MVPA bouts compared to TD children, with no difference in LPA or TPA levels between the groups.

2.3 METHODS

Recruitment

Children 4 to 5 years old (48 to 71 months) were recruited from October 2013 to June 2017 as part of the Coordination and Activity Tracking in CHildren (CATCH) study, a prospective longitudinal cohort study of children with and without DCD in southern Ontario, Canada. The target sample size was 600 children: 300 children at risk for DCD and 300

TD children. In order to recruit a large sample of children with DCD, multiple screening procedures were undertaken and modified at different phases of recruitment in order to maximize the probability of meeting the target sample size.

In the first phase of recruitment (Oct 2013-Feb 2015), children were initially screened over the telephone using the Developmental Coordination Disorder Questionnaire (DCDQ (24)). Parents who identified potential motor difficulties on the DCDQ (score $\leq$54/75) and a random sample of children scoring in the typically developing (TD) range (score >55/75) were invited into the lab to complete a standardized motor coordination assessment (Movement Assessment Battery for Children 2^{nd} Edition, MABC-2 (25)). All children scoring $\leq 16^{th}$ percentile on the MABC-2 were considered at risk for DCD and invited to participate, whereas children scoring $>16^{th}$ percentile were considered TD and a subset was randomized into or out of the cohort. In the 2^{nd} phase of recruitment (Feb 2015-May 2015), the DCDQ telephone screening was removed and all interested participants were brought directly into the lab for a MABC-2 assessment. Children scoring $\leq 16^{th}$ percentile were again all automatically invited into the CATCH cohort study and a random sample of TD children (1 in 3) were also included. For the 3^{rd} phase of recruitment (May 2015-Aug 2016), randomization was no longer applied for the TD group; thus, all children, regardless of MABC-2 scores, were enrolled into the cohort. Once the target sample size of 300 TD children was reached (Aug 2016), only children who came into the lab and scored ≤16th percentile on the MABC-2 were enrolled. Further details of the study design and recruitment procedure can be found in a previous publication (26). Children were not eligible if they did not speak/understand English, weighed <1500g at birth, or were diagnosed with a physical disability or medical condition that affects motor coordination (e.g., cerebral palsy, muscular dystrophy). The current study is an examination of the baseline sample of children enrolled into the longitudinal cohort. Parents of all participating children provided written consent and the study was approved by the Hamilton Integrated Research Ethics Board.

Assessment of Developmental Coordination Disorder

Following both the Diagnostic and Statistical Manual of Mental Disorders 5th Edition (DSM-5)(1) and the European Academy of Childhood Disability recommendations (27), we assessed each diagnostic criterion for DCD to determine case assignment. Motor coordination was assessed using the MABC-2 (25). This assessment is a criterion standard for identifying children at risk for DCD (27). The test consists of 8 items across 3 subdomains of motor coordination (manual dexterity, aiming & catching, and static & dynamic balance). Raw scores on each item are converted into an overall standard score and corresponding percentile based on the child's chronological age. Children scoring ≤5th percentile were considered to have DCD, children scoring between the 6th-16th percentile were considered to be at risk for DCD (rDCD) and children scoring >16th percentile were considered TD. All children completed the Kaufman Brief Intelligence Test-2nd Edition (KBIT-2 (28)) to determine if motor coordination deficits may be due to an intellectual delay (intelligence quotient (IQ)<70). Parents completed a detailed medical history, and in consultation with a family physician, children were excluded if they had been diagnosed with a medical condition that would better explain their motor coordination problems. All parents also completed the DCDQ (24) and a structured interview with a research assistant to ascertain if a child's motor coordination difficulties were impacting activities of daily living including self-care, academic, and leisure pursuits.

Physical Activity

Physical activity was measured using Actigraph accelerometers (wGT3X, Pensacola Fl, USA) worn around the hip for 7 days. The participants were instructed to keep the monitor on during all waking hours, removing it only for sleep and prolonged water play. Parents completed logbooks to indicate the times the accelerometer was put on in the morning and taken off before going to bed, and any times it was removed throughout the day (e.g., for naps, swimming, or bathing). Data were analyzed in 3s epochs. Non-wear periods were defined as any times the parent indicated the monitor was not being worn and/or $\geq$ 60 min of consecutive zeros counts. A valid day was defined as ≥10 hours of wear. Only children who wore the accelerometer for ≥3 valid days were included in the analyses. Evenson cutpoints (29) were applied to determine average daily minutes in each intensity of activity (sedentary, light, moderate, vigorous, and total). Triaxial movement was determined by calculating the average daily activity cpm in each axis. Cleaning and processing of accelerometer data were conducted using Actilife software (Actigraph, Pensacola, FL)

Statistical Analysis

Differences in baseline characteristics among the DCD, rDCD and TD groups were tested using one-way ANOVA for continuous variables and chi square (X^2) for categorical variables. Differences in physical activity outcomes were tested using Analysis of Covariance (ANCOVA) with average daily minutes of wear time, age, and sex included as covariates. Significant group effects were followed by post-hoc tests using Bonferonni adjustment for multiple comparisons ($p<.017$). For each physical activity variable, linear regression analysis was conducted with the main effects of group, age, sex, and wear time, and a group by sex interaction to determine if sex moderated the relationship between DCD group and physical activity outcomes.

2.4 RESULTS

Participants

Of 1680 potential participants, 1330 were contacted and deemed eligible with 1225 (92%) providing verbal consent. Of these, 1096 participated in at least one part of the screening protocol. In total, 758 children completed motor assessments in the laboratory, with 594 invited into the longitudinal cohort based on MABC-2 scores. One participant withdrew from the study and one participant was unable to complete the baseline assessment and was excluded, leaving a total of 592 children included in the longitudinal CATCH cohort (301 TD, 179 rDCD and 112 DCD). Seven children (2 rDCD, 5 DCD) scored below 70 on the KBIT-2, indicating a potential intellectual disability. Removing these children did not significantly impact the results, and thus were included in all analyses. Children in both the rDCD and DCD groups had a higher percentage of males than the TD group, and those in the rDCD group were slightly younger than the TD group ($p<.05$). Participant characteristics of each group are presented in Table 1.

Table 2-1 Participant Characteristics

	DCD	rDCD	TD	X^2	p
n (% male)	112 (71.4)‡	179 (63.1)‡	301(48) #†	21.8	<.001
				F	p
MABC-2 (%ile)	3.2(1.9)†‡	12.7(3.5)#‡	56.2(23.4)#†	590.2	<.001
Age (months)	59.2(6.1)	58.5(7.2)‡	60.4(7.4)†	4.2	.02
Height (cm)[a]	109.5(6.1)	109.3(6.4)	110.4(6.1)	2.1	.12
Weight (kg)[b]	19.4(3.6)	19.1(3.4)	19.2(2.8)	0.2	.80
BMI %ile[c]	58.5(26.9)	57.4(27.3)	54.3(27.2)	1.3	.27

Results are presented as Mean(SD).
BMI=body mass index; DCD=Developmental Coordination Disorder; MABC-2=Movement Assessment Battery for Children – 2nd Edition; rDCD=at risk for Developmental Coordination Disorder; TD=Typically Developing; X^2= chi square. [a]Height was measured without shoes to the nearest 0.1 cm using a stadiometer (Seca 264, Chino, CA). [b]Weight was measured without shoes and in light clothing to the nearest 0.1 kg using a digital scale (SECA 869, Chino, CA). [c]BMI percentile was calculated using the Centre for Disease Control Data (30)
#Significantly different from DCD; †Significantly different from rDCD; ‡Significantly different from TD.

Physical Activity

A total of 77 (25 DCD, 28 rDCD and 24 TD) participants (13%) were excluded from the physical activity analysis. Forty-eight children were excluded due to incomplete wear time (did not meet criteria of ≥10 hours on ≥3 days) and 11 children refused to wear the accelerometer. Seventeen participants were excluded because the accelerometer was never returned (lost/unable to contact), and one parent declined the physical activity monitoring part of the study, leaving 515 (87%) participants included in the final analysis. The children who were excluded did not differ from those included on age (t=1.2, p=.24), height (t=0.2, p=0.82), weight (t=1.3, p=0.21), or sex (X^2=.3, p=.7), but did have lower total MABC-2 scores (t=3.4, p<.01). Children included in the analysis wore the accelerometer for an average of 6 valid days for 724 min (~12hr) per day. Girls were less active than boys across all measured physical activity variables (p<.001). No differences in daily minutes of sedentary time, TPA, LPA, MPA or VPA, or average daily activity counts per minute in any of the axes were found among the groups. Children in all groups engaged in similar number of daily MVPA bouts; however, children with DCD accumulated their MVPA in slightly shorter bouts compared with TD children (Table 2). Follow-up linear regression analyses indicated no significant group by sex interactions.

Table 2-2 Physical activity by group

	DCD	rDCD	TD	F	p
n	87	151	277		
SED (min/day)	443.3(44.2)	455.1(50.1)	452.7(44.4)	0.40	0.67
LPA (min/day)	201.9(28.4)	201.2(28.8)	199.4(28.6)	0.34	0.71
MPA (min/day)	40.0(9.6)	39.6(9.0)	39.8(9.3)	0.83	0.44
VPA (min/day)	31.2(10.8)	31.2(11.6)	32.3(12.2)	1.56	0.21
MVPA (min/day)	71.2(19.4)	70.9(19.1)	72.0(20.5)	1.35	0.26
TPA (min/day)	273.0(40.6)	272.1(42.1)	271.6(42.8)	0.40	0.67
MVPA bout freq (#/day)	712.5(150.6)	697.8(151.8)	705.9(159.3)	0.98	0.38
MVPA bout duration (s)	5.94(0.58)‡	6.05(0.67)	6.06(0.66)#	3.13	0.04
Axis 1 (cpm)	682.1(159.5)	681.0(185.1)	687.5(174.7)	0.92	0.40
Axis 2 (cpm)	833.2(156.7)	816.4(159.5)	788.6(151.3)	1.08	0.34
Axis 3 (cpm)	1006.6(188.4)	987.1(208.7)	979.2(193.3)	0.01	0.99

All values presented as Mean(SD).
Axis 1=vertical axis; Axis 2=mediolateral axis; Axis 3=anterioposterior axis; cpm=activity counts per minute; DCD=Developmental Coordination Disorder; LPA=light physical activity; MPA=moderate physical activity MVPA=moderate-to-vigorous physical activity; rDCD= at risk for Developmental Coordination Disorder; SED=sedentary time; TPA= total physical activity; VPA= vigorous physical activity.
#Significantly different from DCD; † Significantly different from rDCD; ‡ Significantly different from TD.

2.5 DISCUSSION

This study was the first to examine objectively-measured physical activity in a large sample of young children who met the diagnostic criteria for DCD. Overall, young children with DCD and rDCD had similar daily activity across all intensities compared to TD children. Young girls were less active than boys across all measured physical activity outcomes ($p<.001$) but no significant group by sex interactions were found, indicating that the non-significant effect of group was similar for boys and girls. Although similar MVPA levels among groups contradicts findings in older DCD samples (2), it implies that the activity deficit is not yet present in the early years, but may emerge and widen over time as children with DCD reach middle childhood and adolescence (5). The non-significant differences in VPA (and MVPA) found at this young age may be due to the fact that motoric demands of play are relatively low (8) and children with DCD are still engaging in many of the same free play and organized activities as TD children. These results are encouraging as early interventions may have the potential to help mitigate the more pronounced physical activity deficits seen as children with DCD reach adolescence (3, 4).

The lack of significant differences in MVPA and VPA appear to contradict the documented positive relationship between motor competence and MVPA found in TD preschool-age children (12). However, the literature on TD children focuses almost exclusively on fundamental motor skills (i.e. object control and locomotor abilities) as these are regarded as the building blocks for engagement in more complex skills and future engagement in physical activities (31). These are typically assessed using tools such as the Test of Gross Motor Development, which assesses the quality of movement (process) instead of the product or outcome. The association between fundamental motor skills and physical activity in the early years is generally in the low to moderate range (10, 12), and tends to vary by gender and skill domain (12). As the MABC-2 used in the current study is a product-oriented assessment that assesses both fine and gross motor skills, combined with the fact that the TD group in the current sample encompasses a wide range of motor abilities, the non-significant group differences does not mean that a relationship between gross motor abilities and physical activity is non-existent. Moreover, the relationship between motor competence and physical activity is hypothesized to get stronger over developmental time (13). Therefore, it is possible that the differences among groups will emerge as the children get older.

In terms of daily MVPA accumulation, children with more severe motor difficulties engaged in a similar number of daily MVPA bouts, but did not sustain these activity bouts for as long as TD children. The difference in average epoch duration observed between groups was quite small (0.1 s), which is unsurprising given the extremely short bursts of activity that are typical of young children (32). Reduced fitness in young children with DCD has been reported (33, 34); thus, it is possible that these fitness deficits may hinder children's ability to sustain high intensity activities as long as TD children. Conversely, it may be that the shorter bouts of MVPA are causing these fitness deficits. Future work testing these mediating pathways in the early years is necessary. As children get older and the nature of play and sport become more prolonged and complex in nature, differences in these accumulation patterns may become more pronounced, and may potentially influence future health indicators, such as aerobic fitness and weight status (23), which are common concerns for children with DCD (2).

The observed triaxial movement patterns indicate no differences among groups in any of the three planes of movement; however, an interesting pattern emerged, whereby children with DCD had higher cpm in axis 2 and 3 and lower cpm in axis 1. A follow-up exploratory analysis revealed that the relative proportion of movement in the mediolateral axis (axis 2) was higher in the DCD group, compared to the TD group ($p=.01$), which is the opposite pattern observed in the vertical axis (axis 1) ($p=.02$). This is a noteworthy finding as it demonstrates that by only focusing on the vertical axes, overall movement may be underestimated, and therefore, overall energy expenditure underestimated for children with motoric difficulties. If a higher relative movement in the mediolateral plane can be confirmed in future work, it could signify inefficient movement patterns, leading children with DCD to expend more energy than TD children in order to perform the same activities (35).

Limitations

Repeated motor assessments in the early years are recommended before a DCD diagnosis is made (27); however, as this was a cross-sectional analysis, we were not able to examine the stability of motor coordination scores. Thus, it is possible that some of these young children, especially those in the rDCD group, may not meet the criteria for DCD in future years (36). As this cohort will be followed annually, we will be able to determine how the (in)stability of motor coordination influences changes in physical activity through this early childhood period. Another limitation of the current study was that co-occurring conditions that are common in the DCD population (e.g., attention deficit hyperactivity disorder; learning disabilities) were not accounted for in these analyses as these are typically not yet diagnosed in this early age group. Hyperactivity symptomology may have confounded physical activity levels of the DCD groups (35). In addition, co-occurring behavioural difficulties may explain why a higher percentage of children in the DCD and rDCD groups did not meet the minimum accelerometer wear time criteria. Nonetheless, this is one of the largest preschool age DCD samples studied to date and the majority (82%) of children in the DCD groups were able to meet and exceed the minimum criteria of at least 10 hours per day for 3 days, with no differences in the total number of days or average daily minutes of wear between the compliant DCD and TD children.

Future Directions

Objective physical activity measurement has many benefits over parent-reported activity; however, it does not allow us to examine the type of activities or the context in which physical activity engagement occurs (37). Although overall levels of MVPA are indeed important for health in children and youth (23), the nature and type of these activities (e.g., sport, cognitively-engaging physical activity) may also serve to promote both motor and cognitive development (38). Research examining the nature of physical activity engagement and contextual factors influencing participation at this young age will help to shed light on what types of activities children with DCD may be drawn toward or avoiding, identify important areas to target for intervention, and may help to predict trajectories of motor development and future physical activity engagement. Utilizing triaxial activity counts may provide a glimpse into inefficient movement patterns and should not be ignored when measuring overall movement and/or energy expenditure in children with immature locomotor strategies (e.g., toddlers or children with motor delays). However, well-controlled observational studies combined with accelerometry are needed to examine if movement inefficiencies can be detected using this method. Following this large, longitudinal cohort will allow us to determine if and when a gap in activity emerges, how patterns of activity change, and if these influence future health-related fitness outcomes.

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CHAPTER 3: **Health-related fitness in preschoolers with and without motor delays: role of physical activity**

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3.1 ABSTRACT

Purpose
School-age children with Developmental Coordination Disorder (DCD) have poor health-related fitness (HRF), but little is known about when these deficits emerge. The purpose of this study was to determine if 4- and 5-year old children who meet the criteria for DCD exhibit poorer HRF than typically developing (TD) children, and if this relationship is mediated by vigorous physical activity (VPA) engagement.

Methods
Five-hundred and ninety-two children participated (age 5.0±0.6 years) from the Coordination and Activity Tracking in CHildren (CATCH) study. Motor skills were assessed using the Movement Assessment Battery for Children- 2nd edition (MABC-2), with groups defined as having DCD (≤5th %ile), at risk for DCD (rDCD; 6th-16th%ile), and TD (>16th percentile). Measures of body composition included body mass index, waist circumference, and body fat percentage. Musculoskeletal fitness assessments included standing long jump distance, as well as peak and mean power assessed using a 30s Wingate protocol on a pediatric cycle ergometer. Time to exhaustion on a progressive, treadmill test was used to determine aerobic fitness. Flexibility and VPA were assessed using a sit-and-reach test and 7-day accelerometry, respectively.

Results
Children in the DCD group had the poorest musculoskeletal and aerobic fitness, whereas TD children had the highest. No differences in body composition among groups were found. Daily vigorous physical activity was similar among groups and did not explain HRF differences.

Conclusions
Preschool children with DCD have decreased anaerobic and aerobic fitness compared to TD children; however, VPA and body composition appear to be less affected by DCD in the early years. Early motor interventions may be able to improve fitness and reduce the risk of hypoactivity and obesity as children with DCD get older.

3.2 INTRODUCTION

Developmental Coordination Disorder (DCD) is a neurodevelopmental disorder characterized by difficulties with fine and/or gross motor coordination that significantly impact daily functioning including leisure, academic, and self-care pursuits (1). Although it is a fairly prevalent disorder, affecting approximately 5-6% of children, it remains widely under-recognized and under-diagnosed (2). Children with DCD have lower levels of cardiorespiratory and musculoskeletal fitness (3), and higher rates of overweight/obesity (4). In longitudinal studies, the differences in cardiorespiratory fitness, body mass index (BMI), and waist circumference between children with and without DCD tends to either persist or widen over time across childhood (5–7). This is highly concerning as children with DCD are, therefore, at a greater risk of poor HRF as they age, which increases their risk of cardiovascular disease and other negative health outcomes later in life (8).

The majority of literature examining HRF in DCD has focused on children over 7 years of age, a time in which fitness and body composition differences between children with and without DCD are already present (3, 4). Therefore, it is still unclear how or when these deficits emerge. To date, only three studies that have comprehensively examined HRF in children with DCD included children under the age of 6. Aertssen and colleagues (9) found poor anaerobic capacity in young children with DCD as measured by the Muscle Power Sprint Test and Functional Strength Measures. Similarly, Schott and colleagues (10) found 4 to 6-year-old children with DCD showed deficits in anaerobic power as measured by 20 m sprint time, however no differences in aerobic fitness, upper or lower body muscular strength, flexibility, or BMI. Conversely, Hands & Larkin (11) found more global HRF deficits in young children (ages 5-8) with motor learning difficulties, reporting poorer cardiorespiratory endurance, flexibility, abdominal strength, running speed, long jump distance, and a higher BMI. These conflicting findings may be due to the different ages of the samples, whereby fitness differences may be more pronounced over age 6, or the reliance on field-based fitness assessments that may be more heavily influenced by motivational and/or environmental factors. It is, therefore, still uncertain which components of HRF are affected by motor coordination difficulties in the preschool years.

In addition to lower HRF, children with DCD are also less active compared to children with typical motor development (3). It is hypothesized that the HRF deficits observed in children with DCD are due in part to hypoactivity, whereby poor motor coordination leads to reductions in physical activity and subsequent declines in HRF (12).

Although this activity deficit is thought to explain some of the differences in HRF between children with and without DCD, there is limited evidence testing this mediation pathway and it is still uncertain if these differences in physical activity and fitness are present in the preschool years. Vigorous physical activity (VPA), in comparison to light, moderate, or moderate-to-vigorous physical activity, has been shown to be most strongly associated with health-related fitness outcomes in preschool children (13). Therefore, the purpose of this study was to examine differences in HRF (cardiorespiratory fitness, musculoskeletal fitness, body composition, and flexibility) using objective, laboratory assessments in a large sample of young children (age 4-5) with and without DCD, and to determine if HRF differences are mediated by objectively-measured VPA levels.

3.3 METHODS

Participants

Children were recruited from 2013 to 2017 as part of the Coordination and Activity Tracking in CHildren (CATCH) study, a prospective, longitudinal cohort of children with and without DCD from Southern Ontario, Canada. The target sample size was 300 TD children and 300 children at risk for DCD aged 4 and 5 years (48-71 months). In order to recruit equal samples of children with and without DCD, multiple screening procedures were undertaken and modified at different phases of recruitment.

Initially, children were screened for motor difficulties over the telephone using the DCD Questionnaire (DCDQ) and some were selected for further screening in the laboratory with the Movement Assessment Battery for Children, 2^{nd} edition (MABC-2). All children who scored $\leq 16^{th}$ percentile on the MABC-2 were invited into the longitudinal cohort, along with a random sample of TD children ($>16^{th}$ percentile). Telephone screening using the DCDQ was removed (February 2015) from the screening procedures. Random selection of TD children was also removed (May 2015) such that all children attending the lab for MABC-2 testing were invited into the longitudinal cohort. Once the TD cohort was full, only children scoring $\leq 16^{th}$ percentile on the MABC-2 were invited into the cohort (August 2016). Recruitment began in October 2013 and ended in June 2017. Details of the study design and recruitment procedure can be found in a previous publication (14). Children were eligible if they could speak/understand English, weighed over 1500g at birth, and did not have a diagnosed physical disability or medical condition (e.g., cerebral palsy or muscular dystrophy) that significantly impacted motor coordination. This study was approved by the Hamilton Integrated Research Ethics Board.

Parents of all participating children provided informed, written consent. This study is a cross-sectional examination of the baseline CATCH cohort.

Assessment of Developmental Coordination Disorder

All children completed the MABC-2 (15), the criterion standard for assessing risk of DCD in childhood (2). This assessment consists of 8 items across 3 areas of coordination: manual dexterity, aiming and catching, and balance (static and dynamic). Raw scores are converted into standard scores and an overall percentile based on the child's chronological age. Children scoring at or below the 5^{th} percentile were considered to have DCD, those in the 6^{th}-16^{th} percentile were considered to be at risk for DCD (rDCD), and children scoring above the 16^{th} percentile were considered TD. All children also completed the Kauffman Brief Intelligence Test-2^{nd} Edition (16) and parents filled out a detailed medical history in order to confirm that motor deficits were not better explained by an intellectual delay or diagnosed medical condition.

Assessment of health-related fitness (HRF)

HRF is comprised of body composition, cardiorespiratory fitness, muscular strength and endurance, and flexibility; all of which are considered the key components related to one's health and ability to engage in physical activity (17).

Body composition

Height and weight were measured in duplicate without shoes and in light clothing using a stadiometer (SECA 264, Chino, CA) and digital scale (SECA 869, Chino, CA). Measures were repeated if the two measurements were >0.1 cm or >0.1 kg apart. The average of the 2 closest measures were used to determine height and weight, and calculate body mass index (BMI, kg/m^2). BMI percentiles were then determined based on the U.S. Centers for Disease Control and Prevention growth charts (18). Presence of overweight/obesity was defined as children whose BMI was above the 85% percentile for their age and sex.

Waist circumference was measured to examine central adiposity at 2 locations: top of the iliac crest (recommended by the National Institutes of Health) and midway between the top of the iliac crest and the lowest rib (recommended by the World Health Organization) (19). Measurements were taken in duplicate and repeated if differences between measurements were >0.5 cm. Measures were taken against the skin, where possible, during normal exhalation.

Body fat percentage was determined using bioelectric impedance analysis (BIA) (RJL Quantum IV, Clinton Township, MI) while children were lying supine. Fat free mass (FFM) was first determined using an equation that has been validated for children (20). Percent body fat was then calculated as [(body weight-FFM)/body weight] x 100.

Cardiorespiratory (aerobic) fitness

Aerobic fitness was assessed using the Bruce Protocol, a progressive treadmill test that increases in speed and grade every 3 min (21). The test was initially created to assess aerobic capacity in adults but has now adapted and used extensively with children as young as preschool age (22, 23). All children started at stage 1 and were required to hold onto the handrails throughout the duration of the test to assist with balance, with a research assistant placed behind the child to ensure safety. Heart rate was measured continuously throughout the test using a heart rate monitor (Polar H7, Lachine, QC). The test was terminated when the child reached exhaustion, was no longer able to keep up with the speed of the treadmill, or refused to continue despite verbal encouragement. Time to exhaustion was used as an indicator of aerobic fitness. Only children who reached a maximum heart rate ≥180 beats per min (bpm) were included in the analyses.

Musculoskeletal Fitness

Short-term muscle power was examined using a Wingate protocol (Bar-Or 1987) on a pediatric cycle ergometer (Lode pediatric, the

Netherlands). Children were first required to sprint as fast as possible (~20 s) against the internal resistance of the ergometer only. After a short rest, the Wingate test began: children were instructed to pedal as fast as they could and a resistance relative to their body weight (0.55 Nm/kg) was applied once they reached 80% of their maximum pedaling cadence. The children then pedaled against this resistance for 30 s. Peak power (W) was determined as the highest instantaneous power achieved during the test. Mean power (W) was the average power output over the 30 s test. As young children tend to accumulate their activity in bouts shorter than 10 s (24), the mean power from the first 10 s of the test was also calculated in order to reduce the potential of confounding motivational factors on performance. This modified 10 s Wingate has been found to be reliable in preschoolers (25). Fatigue was calculated as the percentage drop off in power over the course of the first 10 s and entire 30 s test. All power outputs were calculated using the LODE Wingate software package (Lode BV, Gronigen, The Netherlands). Only children who could pedal >25 rpm were included in the analysis as this is the minimum cadence in which a resistance can be applied to the cycle ergometer.

Long jump, or standing broad jump, is a common, field-based measure of lower body muscular strength/power (26). It has been validated against peak power as measured using the modified 10 s

Wingate in preschool children (27). Children were required to stand with their feet behind a marked line and instructed to jump as far as they could and land on two feet. Distance was measured from the line to the back of the closest heel. Children were given 3 trials, with additional trials conducted only if a child fell, or did not perform a successful 2-footed take-off or landing. The best of the 3 trials was used as an indicator of lower body musculoskeletal fitness.

Flexibility

Flexibility was assessed using the sit-and-reach test. Children were instructed to keep one hand on top of the other and reach forward as far as they could using a slow and controlled motion keeping both legs fully extended. A sit-and-reach box (Novel products, Rockton, IL) was used, and measurements were taken in cm, with 23 cm corresponding to the position of the feet against the box. Trials were repeated if the child's hands came apart or their knees bent.

Physical Activity

After completion of the laboratory visit, all children were asked to wear an accelerometer (Actigraph wGT3X, Pensacola, FL) over their right hip for the following 7 days. Children were instructed to wear the accelerometer during all waking hours, only removing it for sleep and/or prolonged water activities. Parents were given a logbook to record the times the accelerometer was put on and removed. Non-wear periods were defined as any time the parent indicated the accelerometer was off and/or ≥60 min of consecutive zero counts. Only children who wore the accelerometer for at least 3 valid days (≥10 hours) were included in the analyses. Data were analyzed in 3 s epochs and Evenson cutpoints were applied to determine average daily minutes spent in VPA (28). All accelerometer data were cleaned and processed using Actilife Software (Actigraph, Pensacola, FL)

Statistical Analysis

Differences in descriptive characteristics among groups were examined using one-way ANOVA for continuous variables and chi-square for categorical variables. HRF differences were examined using Analysis of Covariance (ANCOVA) controlling for age in months, sex, height, and weight where appropriate. VPA group differences were also examined using ANCOVA and adjusted for age, sex, and daily wear time. Significant group effects were then tested using post-hoc tests with Bonferroni correction for multiple comparisons ($p<.017$). In order to examine whether sex moderated the effect of group on HRF outcomes, separate linear regressions were conducted for each HRF variable including the main effects of group and sex, and an interaction term for sex*DCD group. For

HRF outcomes in which significant group differences were found, separate mediation analyses were conducted to determine if the differences were mediated by levels of VPA. The tests for indirect (mediation) effects were conducted using the PROCESS software macro for SPSS (29), with DCD group entered as the independent (X) variable, VPA as the (M) variable, and HRF outcomes as the dependent variable (Y), with all aforementioned covariates included. As recommended by Hayes and Scharkow (29), bootstrapping was set to 10,000 samples. All analyses were conducted using SPSS v20.

3.4 RESULTS

Participants

Of the 1330 eligible families, 1225 provided verbal consent, with 594 children invited into the longitudinal cohort based on their MABC-2 scores. One participant withdrew and another was unable to complete the baseline appointment, leaving 592 children included in the final CATCH cohort (301 TD, 179 rDCD, and 112 DCD). Potential intellectual disability (IQ<70) was identified in 7 children (2 rDCD, 5 DCD); however, removing these participants did not significantly affect the results, and therefore they were included in all subsequent analyses. Participant characteristics are presented in Table 1. rDCD children were slightly younger than TD children, with a higher percentage of boys in both DCD groups compared to the TD group.

Table 3-1 Participant Characteristics

	DCD	rDCD	TD	X^2	p
n(% males)	112(71.4)‡	179(63.1)‡	301(48.2)#†	21.8	<.001
				F	p
MABC-2 (%ile)	3.2(1.9)†‡	12.7(3.5)#‡	56.2(23.4)#†	590.2	<.001
Age (months)	59.2(6.1)	58.5(7.2)‡	60.4(7.4)†	4.2	.02
Height (cm)	109.5(6.1)	109.3(6.4)	110.4(6.1)	2.1	.12
Weight (kg)	19.4(3.6)	19.1(3.4)	19.2(2.8)	0.2	.80

Results are presented as Mean(SD).
%ile = percentile; DCD=Developmental Coordination Disorder; MABC-2=Movement Assessment Battery for Children – 2nd Edition; rDCD= at risk for Developmental Coordination Disorder; TD=Typically Developing.
Significantly different from DCD; † Significantly different from at risk for DCD; ‡ Significantly different from TD

Body composition

Results of the body composition analyses are presented in Table 2. All children had valid height, weight, and waist circumference measurements. Six children were missing body fat percentage measurements due to refusal/an inability to lie still (n=5) or due to a skin rash preventing placement of the electrodes (n=1). Although there was a trend for DCD children to have higher waist circumference and absolute BMI, no statistically significant differences in any of the body composition outcomes were found among the 3 groups. This pattern of results was similar for boys and girls as no significant group*sex interactions were found.

Table 3-2 Body composition by group

	DCD	rDCD	TD	X^2	p
n overweight or obese (%)	19(16.9)	35(19.6)	47(15.6)	1.23	0.54
				F	p
WC Mid(cm)[a]	54.7(4.5)	54.1(3.8)	53.8(3.4)	2.9	0.06
WC Hip(cm)[a]	55.7(4.9)	55.2(4.2)	55.1(3.8)	2.1	0.12
BMI(kg/m^2)[a]	16.0(1.7)	15.9(1.4)	15.7(1.3)	2.7	0.07
BF(%)	22.6(4.3)	22.7(4.1)	22.8(4.3)	0.2	0.85
BMI(%ile)	58.5(26.9)	57.4(27.3)	54.3(27.2)	1.3	0.27

[a]age and sex included as covariates. Values presented as Mean(SD) unless otherwise indicated.
%ile=percentile; BF=body fat; BMI=body mass index; DCD=Developmental Coordination Disorder; rDCD=at risk for Developmental Coordination Disorder; TD=typically developing; WC hip=waist circumference at the top of the iliac crest; WC mid=waist circumference midway between the top of the iliac crest and lowest rib.

Cardiorespiratory and musculoskeletal fitness

Results of the physical fitness assessments are presented in Table 3. Overall, there was a group effect for both musculoskeletal and aerobic fitness performance; children with DCD had the greatest fitness deficits, and children at-risk for DCD performed better than the DCD group but worse than the TD group. Thirty-one children (11 DCD, 7 rDCD, 13 TD) were excluded from the aerobic fitness (time to exhaustion) analysis due to an inability to reach a HR of 180 bpm. Children in each group reached an average maximal heart rate of 198 bpm (F=0.42, p=.65); however, DCD children reached this maximal heart rate almost 1.5 min faster than did TD children. Thirteen children were excluded from all of the Wingate analyses due to an inability/refusal to pedal >25 rpm (n=7; 4 DCD, 1 rDCD, 2 TD) or equipment malfunction (n=6; 1 DCD, 2 rDCD, 3 TD); an additional 4 children with DCD were excluded from the mean power and fatigue analysis due to a refusal to continue pedaling for the entire duration of the test. Children with DCD had the lowest peak and mean muscle power on the Wingate test, and fatigued to a greater extent over both the first 10 s and the complete 30 s test compared to both rDCD and TD children. Ten children were not able to complete a long jump with two feet and were excluded from the analysis (4 DCD, 5 rDCD, 1 TD). TD children were able to jump significantly farther than the children in the rDCD and DCD groups, with children in the DCD group demonstrating the shortest long jump distance. No difference in flexibility among the 3 groups was found. The linear regression analyses found no significant group by sex interactions, suggesting that the effect of group on all physical fitness outcomes did not differ between girls and boys.

Table 3-3 Cardiorespiratory fitness, musculoskeletal fitness, and flexibility by group

	DCD	rDCD	TD	F	p
TTE (s)	542.2 (93.5)†‡	593.5 (100.6)#‡	624.7 (91.6)#†	28.9	<.001
PP (W)	89.2 (35.2) †‡	94.4 (34.2) #‡	103.5 (27.1) #†	22.8	<.001
MP30 (W)	51.7 (27.5) †‡	59.2 (28.1) #‡	70.9 (24.6) #†	38.6	<.001
MP10 (W)	71.5 (33.4) †‡	78.5 (32.6) #‡	89.2 (26.1) #†	28.4	<.001
Fatigue30 (%)	75.8 (21.8) †‡	68.1 (22.7) #‡	58.0 (20.5) #†	24.3	<.001
Fatigue10 (%)	41.0 (25.2) †‡	35.3 (23.2) #‡	25.7 (13.6) #†	22.5	<.001
Long Jump (cm)	69.2 (21.1) †‡	75.6 (20.7) #‡	86.8 (17.8) #†	47.3	<.001
Flexibility (cm)	26.9 (5.1)	27.6 (4.9)	28.0 (4.3)	1.3	0.28

Note: age, sex, height and weight are included as covariates. All values presented as M (SD).
DCD=Developmental Coordination Disorder; Fatigue30=percent fatigue over 30s; Fatigue10=percent fatigue over 10s; MP30=mean power over 30s; MP10=mean power over 10s; PP=peak power; rDCD= at risk for Developmental Coordination Disorder; TD=typically developing; TTE=time to exhaustion.
#Significantly different from DCD; †Significantly different from at risk for DCD; ‡Significantly different from TD

Role of physical activity

There were no significant differences among the three groups for daily minutes of VPA (DCD 31.2 min/day; rDCD 31.2 min/day; TD 32.3 min/day, F=1.56, p=0.21). The first mediation analysis was conducted with time to exhaustion as the dependent variable. There was a non-significant direct effect of group on VPA, 95%CI [-2.3, 0.32], p=.13, and a significant direct effect of VPA on TTE 95%CI [0.31, 1.68], p<.01. There was no significant indirect mediation effect of VPA on the DCD group to time to exhaustion relationship (95% CI [-2.9, 0.10]). A similar pattern of results was found for peak power (indirect effect 95% CI [-0.71, 0.11]), 10 s and 30 s mean power (indirect effect 95% CI [-0.80, 0.08] and [-0.75, 0.06], respectively], 10s and 30s fatigue (indirect effect 95% CI [-0.01, 0.46] and [-0.01, 0.59], respectively), and long jump distance (indirect effect 95% CI [-0.82, 0.16). Overall, although VPA had a significant direct positive effect on all aerobic and musculoskeletal fitness outcomes (p<.05), it did not explain the large differences in HRF among the three groups.

3.5 DISCUSSION

This was the first large-scale study to comprehensively examine HRF using objective laboratory measures in preschool-age children with DCD or at risk for DCD. With regard to body composition, children with DCD and rDCD, on average, did not significantly differ on measures of waist circumference, BMI, or body fat percentage, compared with TD children. While there is a trend for larger BMI and waist circumference among children in the DCD group, the relative difference in BMI and waist circumference among the groups is quite small and may have minimal clinical relevance, especially considering the overall rates of overweight/obesity in all of the groups are considerably lower than the current estimated Canadian prevalence (30). Although these findings contradict the consistent, significant body composition differences in older samples, longitudinal evidence has shown that the differences in waist circumference and BMI between children with and without DCD widen over time from middle childhood to adolescence (6). While these data are not sufficient to show causation, the minimal differences in body composition in this early childhood period is consistent with the hypothesis that overweight/obesity may be a secondary consequence of DCD that emerges in middle childhood and/or adolescence, possibly due to reduced physical activity, and that these trends may become significant as children reach school age (31).

There were no group differences in flexibility as measured by the sit-and-reach test, which may be due to the heterogeneity in flexibility profiles exhibited by children with DCD (32). Young children with DCD and rDCD demonstrated poorer aerobic and musculoskeletal fitness compared to TD children, which is consistent with the extant DCD literature in older samples of children (3). When comparing our results to previous work conducted with young children, our findings are in-line with Hands and Larkin (11), who found both aerobic and musculoskeletal fitness differences between children with and without motor learning difficulties at age 5 to 8. Although our findings confirm previous findings of poor anaerobic power in young children with DCD (9,10), Schott and colleagues (10) did not find the same deficit in cardiorespiratory fitness as found in the current study. This may be a result of the small sample size or reliance on a field-based aerobic fitness assessment, which may be more heavily influenced by motivational or environmental factors. Overall, we found young children with DCD and at-risk for DCD were not able to produce the same power outputs and fatigued faster both over an incremental, progressive aerobic endurance test and during the all-out short-term anaerobic test. These physical fitness deficits may hinder their ability to keep up with their peers in free play and organized activities, and may be a large contributor to why children with DCD begin to withdraw from physical activity as they get older.

The differences in aerobic and musculoskeletal fitness between groups was not mediated by levels of VPA. These findings are in-line with longitudinal results from the Physical Health and Activity Study Team (PHAST) study, which followed a large sample of children from ages 9 to 14. The PHAST group found that, although children with DCD had steeper rates of decline in cardiorespiratory fitness (measured using field-based tests), and greater increases in BMI and waist circumference from middle to late childhood, this widening gap in HRF was not explained by differences in self-reported physical activity (6, 7). In contrast, Silman and colleagues (33) examined a subsample of the PHAST cohort at age 12-13 using lab-based measures of cardiorespiratory fitness and found that objectively-measured physical activity significantly mediated the differences in maximal oxygen uptake (VO_2max) between children with and without DCD. These discrepancies may be due to the different methodologies used or the age of the samples, whereby the mediating effect of physical activity on cardiorespiratory fitness may not emerge until children are nearing adolescence. As described in Wall's skill gap hypothesis (34), the motoric demands of play are low in this early age group, which may be why children with DCD are not yet withdrawing from physical activity to an extent that would significantly impact fitness levels.

Since VPA levels did not explain why children with DCD had lower HRF, alternative explanations are required. Tests of fitness and motor coordination, although distinct, measure related movement components. An underlying deficit in neuromotor development, such as increased levels of co-contraction of agonist and antagonist musculature (35), may therefore hinder performance on both tests. Increased co-contraction would interfere with a child's ability to perform well-timed coordinated contractions, such as cycling at a fast cadence, propelling the body forward during jumping, and may also contribute to a reduced economy of locomotion in young children with DCD. Oxygen cost of locomotion has been studied in older children with DCD; although no significant differences were found, children with DCD had poorer quality of locomotion during treadmill running and perceived themselves as working harder (36). Impaired neuromuscular control may, therefore, also contribute to an inflated heart rate response and increased sense of perceived exertion at a given intensity compared to TD children, which may explain why children with DCD in the current study reached exhaustion on the treadmill almost 90s earlier than TD children. Psychological factors have also been linked to fitness performance in children. For example, Cairney and colleagues (37) found that generalized self-efficacy towards physical activity explained a significant proportion of the difference in performance on the Leger 20 m shuttle run between children with and without DCD. Therefore, low confidence in their physical abilities may have prevented children with DCD from performing to their true maximal potential.

Limitations

A diagnosis of DCD is not recommended for young children unless motor assessments are repeated and consistent skill delays are found (2); therefore, it is possible that not all of the children in the DCD group, especially those in the rDCD group, will meet the criteria for DCD in future years. As the CATCH study will conduct repeated annual motor assessments, we will be able to determine how potential changes in motor coordination affect changes in fitness outcomes through the early childhood period. Due to the young age of the sample, measurement of VO_2max, the gold standard in aerobic fitness testing, was not possible; however, time to exhaustion using the Bruce protocol has been found to be strongly correlated with directly measured VO_2max in children (38), and, therefore, provides a good indicator of aerobic fitness in preschool children. Furthermore, the total test duration on the Bruce protocol in the TD sample was similar to the reference values published for preschool children (23), which further support the validity of this test and our findings.

Future Directions

Future research is necessary in order to elucidate the specific causes of fitness deficits in children with DCD. Although co-activation has been studied using isometric and isokinetic movements in older children (35), this needs to be examined in younger children during more functional tasks, such as cycling and walking/running. In addition, the role of psychological factors on fitness performance has not yet been studied in preschool-age children and will be important to examine. Although the fact that these physical fitness deficits already exist at such a young age is concerning, Farhat and colleagues (39) were able to show that an 8-week motor skill intervention was able to improve motor skills, cardiorespiratory endurance, and exercise tolerance in 8-year-old children with DCD. These findings hold promise that motor interventions at an early age may help to improve fitness levels; however, longer, well-controlled randomized trials are necessary to determine if early motor interventions can lead to increases in fitness both acutely and over time in children with DCD.

Conclusions

Young children with DCD and at risk for DCD have poorer aerobic fitness and short-term muscle power compared to TD children, which is not explained by levels of daily VPA. These fitness differences may persist or widen over time, putting children with DCD at risk for poor health outcomes as they get older. Early interventions targeting perceived and actual motor competence as well as physical fitness may help to prevent the trends of unhealthy weight gain and physical inactivity observed through middle childhood and into adolescence in children with DCD.

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CHAPTER 4: **Longitudinal relationships among motor proficiency, physical activity and fitness in the early years**

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4.1 ABSTRACT

Introduction
Motor competence is associated with both physical activity and fitness levels in childhood and adolescence. The purpose of this study was to examine the strength of these associations over time and to determine if motor proficiency predicts short-term muscle power (STMP) from preschool to early childhood and whether or not this relationship is mediated by vigorous physical activity (VPA).

Methods
Participants 3 to 5 years of age were recruited and completed 3 annual assessments as part of the Health Outcomes and Physical activity in Preschoolers (HOPP) Study. Motor proficiency was assessed using the Bruininks-Oseretsky Test of Motor Proficiency 2nd Edition – Short Form. STMP, including peak power (PP) and mean power (MP), was evaluated using a modified 10-second Wingate protocol on a cycle ergometer. Physical activity was measured over 7-days using accelerometers.

Results
418 children (210 boys, age 4.5 ± 1.0 years at baseline) participated. Overall motor proficiency was a significant predictor of absolute STMP (PP estimate=0.54, p<.001; MP estimate=0.66, p<.001) and VPA (estimate=0.21, p<.001). At baseline, the relationship between motor competence and VPA was not significant, however a significant positive relationship emerged across time. Although VPA also predicted STMP (PP estimate=0.13, p=.001, MP estimate=0.16, p<.001) it did not mediate the longitudinal relationship between motor proficiency and STMP.

Conclusion
Motor proficiency is an important independent predictor of physical activity and fitness levels from preschool to early childhood. Motor proficiency may be an important target for early interventions to improve both physical activity and health-related fitness in the early years.

Keywords: Motor competence, muscle power, musculoskeletal fitness, vigorous physical activity, accelerometry

What are the new findings:

- The positive relationship between motor competence and physical activity emerges from preschool to school-age
- Motor competence is associated with musculoskeletal fitness over time in the early years
- Physical activity does not mediate the longitudinal association between motor competence and short-term muscle power in young children

4.2 INTRODUCTION

Motor competence is positively associated with physical activity and fitness levels in childhood and adolescence[1–6]. In young children, motor competence shows low to moderate correlations with physical activity[5], which is hypothesized to strengthen over time[7]. Typically, stronger associations are reported with moderate-to vigorous or vigorous intensity activities compared to total or light intensity activity[8,9]. Furthermore, it is physical activities at these higher intensities that are typically associated with positive health outcomes, with emerging evidence that vigorous activities provide additional benefits[10]. The importance of motor competency to physical activity and fitness is further reinforced as children with delayed motor development, such as those with Developmental Coordination Disorder (DCD), are repeatedly found to have lower moderate-to-vigorous physical activity levels and poorer aerobic and musculoskeletal fitness compared to their typically developing peers[11].

The theoretical underpinnings of these interrelationships have been conceptualized in the physical education literature by Stodden and colleagues[7] and in the childhood disability literature by Hands and Larkin[12]. In Stodden and colleague’s developmental model, motor skill competence is central to health trajectories as it is hypothesized to play an important emerging role in physical activity engagement and health-related fitness across developmental time[7]. They describe this as a *positive spiral of engagement* whereby motor competence promotes engagement in physical activity through the mediating effects of health-related fitness and perceived motor competence. Similarly, Hands and Larkin describe children with low motor proficiency, or DCD, to be in an ‘activity deficit’ and progressively withdraw from physical activity leading to less favourable health trajectories[12]. Although the models propose different primary mediating pathways, both highlight that the relationships among motor competence, physical activity and health-related fitness are likely reciprocal and suggest that the influence of motor competence on these health outcomes and behaviours will strengthen over time.

A recent review by Robinson and colleagues[1] concluded that the current evidence linking motor skill competence to physical activity and fitness in childhood is strong, however there are limited longitudinal studies examining how these relationships change over time and the mediating pathways among these constructs. A large majority of the existing longitudinal evidence that has examined these associations has been limited by the measurement of motor skills at only 1 point in time[13–19], reliance on self reported physical activity and field-based assessments of fitness (especially cardiorespiratory fitness)[13–22] or have focused only on the middle childhood to adolescent period[23,24]. As the early years are a time when fundamental motor skills are developed and reinforced, forming the foundation for future physical activity engagement[25], it is important we understand how longitudinal changes in motor competencies influence physical activity and subsequent health-related fitness during the transition from preschool to school-age.

Therefore the purposes of this study were to 1) examine the strength of the associations between motor proficiency and physical activity and musculoskeletal fitness over time from preschool to early childhood and 2) to determine if vigorous physical activity (VPA) levels mediate the longitudinal relationships between motor proficiency and musculoskeletal fitness (short-term muscle power, STMP) in this age group. We hypothesize that the strength of the associations between motor proficiency and VPA and STMP will increase over time. We further hypothesize that motor proficiency will be positively associated with STMP over time and this effect will be partially mediated by VPA after controlling for age at baseline, sex, height and weight.

In this study, we have annual assessments of motor skills, physical activity and musculoskeletal fitness using objective, laboratory assessments from preschool to school age and aims to fill an important gap in the literature by determining the interrelationships among these variables. By better understanding the longitudinal relationships among these constructs we will be better suited to design and implement appropriate early interventions to target inactivity and poor fitness in childhood.

4.3 METHODS

Participants

Children 3 to 5 years (36 months to 71 months) of age were recruited and completed 3 annual assessments (12 ± 1 month apart) as part of the Health Outcomes and Physical activity in Preschoolers (HOPP) Study. The HOPP study is a prospective cohort study in which participants were recruited from community-based organizations in south-central Ontario from 2010-2012. Further details on the study design are provided in a previous publication [26]. Children were ineligible if they had a diagnosed medical condition or a known physical impairment (e.g., epilepsy, cerebral palsy). Informed, written consent was given by parents/guardians of all participants. Ethical approval was obtained from the McMaster University Faculty of Health Science/Hamilton Health Sciences Research Ethics Board.

Motor proficiency

Motor proficiency was assessed using the Bruininks-Oseretsky Test of Motor Proficiency 2nd Edition – Short Form (BOT-SF[27]). This measure takes approximately 15 minutes to administer and contains 14 items covering all 8 subdomains of the full version (Fine Motor Precision, Fine Motor Integration, Manual Dexterity, Bilateral Coordination, Balance, Running Speed & Agility, Upper-limb coordination and Strength). For each item a raw score is obtained and converted into a point score. The point scores are totalled resulting in a total point score for each participant. This total point score can then be converted into a sex-combined standard score (Mean 50, SD 16) and corresponding percentile based on the child's chronological age. Evidence of reliability and validity of the test scores in studies that use similar populations have shown a strong correlation with the Peabody Developmental Motor Scales (r=0.77) and high test-retest (r=0.86) and inter-rater reliability (r=0.97)[27]. As this measure has only been validated in children ages 4 and above, motor proficiency for the 3 year old participants (n=139) was not assessed at baseline and therefore these participants were only included in the analysis for the latter 2 time points.

Physical Activity

Physical activity was measured each year over 7-days using waist worn accelerometers (Actigraph GT3X, Pensacola, FL). Actigraph accelerometers have been validated for use in young children[28] and are capable of collecting data over short sampling intervals in order to accurately capture the short intermittent nature of children's habitual physical activity[29]. Parents were instructed to have their child wear the accelerometer over the right hip during all waking time and only be taken off for water activities or sleep. They were provided a logbook in which they recorded the times the accelerometer was put on and taken off and the reason for removal. Each file was visually inspected and periods of non-wear (i.e. when parent indicated the accelerometer was removed or when there were greater than 60 min of continuous 0 activity counts) were removed. In order to determine average daily minutes of VPA, each file was then analyzed in 3-second epochs using established cutpoints for preschoolers[28]. We used a 3-second epoch in order to accurately capture physical activity in this age group[29] as it approximates the average bout duration of vigorous activity observed in young children[30]. Only children who wore the accelerometer ≥10 hours on ≥3 days were included in the analyses. All accelerometer data was cleaned and processed using Actilife software (Actigraph, Pensacola FL).

Musculoskeletal fitness

Short-term muscle power (STMP), including peak power (PP) and mean power (MP), was evaluated using a modified 10-second Wingate protocol on a pediatric cycle ergometer (LODE Corival Pediatric, The Netherlands). Max pedaling cadence was determined by having the children sprint against only the internal resistance of the ergometer for approximately 20s. Only children who could pedal >25rpm were included in the analyses. After a short rest children began the modified Wingate test where a load (0.55 Nm/kg) was applied once the children reached 80% of their maximum pedaling cadence. Children pedaled against this resistance for 10-seconds and were instructed to keep pedaling as fast as they could. PP (W) was determined as the highest instantaneous power output achieved and MP (W) was the average power output over the 10s test. PP and MP were also expressed relative to body mass (W/kg). A modified 10s test was used instead of the full 30s Wingate as children this young tend to accumulate their physical activity in bouts shorter than 10 seconds[30]. This modified Wingate has been shown to be reliable in preschoolers[31].

Statistical Analysis

Pearson's correlation coefficients between motor proficiency and outcome measures (STMP and VPA) were calculated for each year to examine the strength of these relationships at each time point. Mixed effects modeling was chosen to examine the longitudinal relationships among motor proficiency, VPA and STMP due to its ability to handle missing data as estimates are created using all available time points for each child. Separate mixed effects models including a random intercept for subject were tested for each outcome variable (PP and MP). For each outcome variable, four separate models were examined to determine if motor proficiency predicts STMP over time, controlling for age at baseline, sex, height and weight (Model 1). A sex*motor proficiency interaction term was also included to determine if these relationships differ for girls and boys. The mediating effect of VPA was then tested by first testing the direct pathways between motor proficiency and VPA (Model 2) and VPA and STMP (Model 3) over time. VPA was then entered into Model 1 (Model 4). By examining the change in the estimate between motor proficiency and STMP from Model 1 we can determine if VPA attenuates, and therefore mediates, the direct relationship between motor proficiency and STMP. All analysis was conducted using SAS University Edition.

4.4 RESULTS

Of the 691 parents contacted, 143 declined participation and 77 were deemed ineligible due to the age of the child, a diagnosed medical condition or developmental delay. The remaining 471 participants provided verbal consent and booked a baseline appointment. Of these, 43 did not attend their appointment and were unable to be rescheduled, and 6 additional children were found to be ineligible, leaving 422 children with written parental consent. Four children were unable to complete the first visit and withdrew leaving 418 children (208 girls, age 4.5 ± 0.9 years) participating at baseline. Thirty-five children were lost to follow-up due to an inability to contact or withdrew from the study due to the time commitment, travel restrictions or relocation. Total attrition over the study period was 8.4%. Descriptive statistics for the participants at each year of the HOPP study are presented in Table 1.

Table 4-1 Descriptive statistics

	Year 1	**Year 2**	**Year 3**
Total n (%male)	418 (50.2%)	400 (50.2%)	383 (49.9%)
Age (years)	4.5 ± 0.9	5.5 ± 0.9	6.5 ± 0.9
Height (cm)[a]	106.5 ± 7.7	113.5 ± 7.7	120.1 ± 7.9
Weight (kg)[b]	17.9 ± 3.2	20.3 ± 3.8	22.9 ± 4.5
BMI %ile[c]	52.4 ± 28.5	51.6 ± 28.1	50.2 ± 28.1
BOT-SF Total Point Score	32.2 ± 12.8	40.4 ± 15.5	52.3 ± 12.4
BOT-SF Standard Score	47.8 ± 7.5	48.7 ± 8.8	49.7 ± 8.5
VPA (min/day)	41.9 ± 12.6	45.0 ± 13.1	47.4 ± 14.8
PP (W)	93.9 ± 37.6	122.9 ± 37.3	149.1 ± 41.7
MP (W)	78.8 ± 34.7	106.3 ± 32.9	129.4 ± 34.7

Values are presented as Mean ± SD
[a] height was measured to the nearest 0.1cm using a calibrated stadiometer; [b] weight was measured to the nearest 0.1kg using a digital scale; [c] BMI percentile was calculated using the Centers for Disease Control and Prevention (CDC) growth charts [32]; BMI=body mass index; BOT-SF=Bruininks Oseretsky Test of Motor Proficiency – Short Form; MP=mean power; PP=peak power; VPA= vigorous physical activity.

Overall the 35 children who were lost to follow-up did not differ in age (t=.5, p=.65), height (t=1.1, p=.26) , weight (t=0.9, p-.36), BOT-SF standard score (t=.5, p=.62), VPA (t=.3, p=.78), PP (t=.8, p=.45) or MP (t=1.0, p=.32) compared to those who remained in the cohort.

A significant relationship between motor proficiency and vigorous physical activity emerged over time (Table 2). The relationship between motor proficiency and relative MP (W/kg) was moderate and significant at each year (Table 3). The same pattern was found for PP/kg (year 1, r=0.27, p<.01; year 2 r=0.29, p<.01; year 3 r=0.31, p<.01.

Table 4-2 Correlations between motor proficiency and vigorous physical activity by study year

	VPA 1	VPA 2	VPA 3
BOT SS 1	-.02 (p=0.69)		
BOT SS 2		0.13* (p=.01)	
BOT SS 3			0.21** (p<.001)

Year 1 n=250; Year 2 n=367; Year 3 n=358
BOT SS=Bruininks-Oseretsky Test of Motor Proficiency-Short Form sex-combined standard score; VPA=vigorous physical activity (min/day).

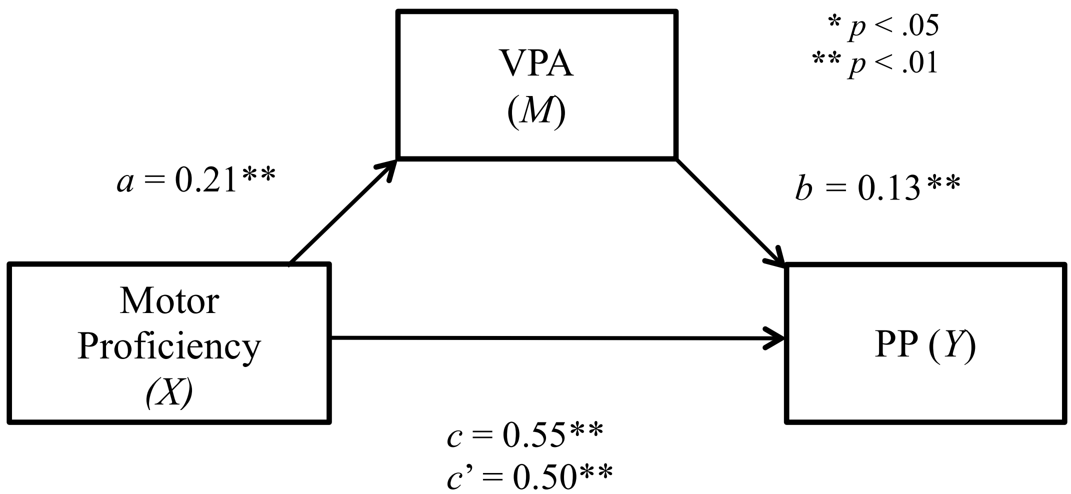
Table 4-3 Correlations between motor proficiency and relative mean power by study year

	MP/kg 1	MP/kg 2	MP/kg 3
BOT SS 1	0.32** ($p<.01$)		
BOT SS 2		0.35** ($p<.01$)	
BOT SS 3			0.39** ($p<.01$)

Year 1 n=266; Year 2 n=397; Year 3 n=383
BOT SS=Bruininks-Oseretsky Test of Motor Proficiency Short Form sex combined standard score; MP/kg=Mean power relative to body weight (W/kg)

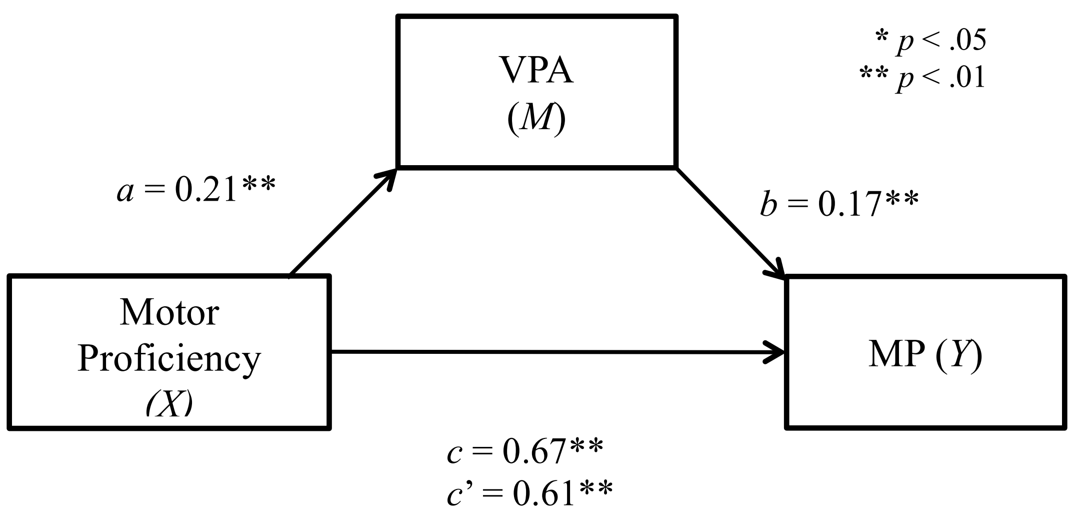
Overall, motor proficiency was a significant predictor of absolute STMP (PP estimate=0.55, $p<.001$; MP estimate=0.67, $p<.001$) and VPA (estimate=0.21, $p<.001$). There were no significant sex*motor proficiency interactions indicating that these relationships did not differ between boys and girls. Although VPA also predicted STMP (PP estimate=0.13, $p=.001$, MP estimate=0.17, $p<.001$) it did not attenuate the direct relationship between motor proficiency and STMP and therefore was not a meaningful mediator (Figure 1 and 2).

Figure 4-1 Longitudinal mediation pathways – Peak Power



a=direct effect of X on M; *b*=direct effect of M on Y; *c*=total effect of X on Y; *c'*=direct effect of X on Y; M=mediator; PP=peak power (W); VPA=vigorous physical activity (min/day); X=independent variable; Y=dependent variable

Figure 4-2 Longitudinal mediation pathways – Mean Power



a=direct effect of X on M; *b*=direct effect of M on Y; *c*=total effect of X on Y; *c'*=direct effect of X on Y; M= Mediator; MP=mean power (W); VPA=vigorous physical activity (min/day); X=independent variable; Y=dependent variable

4.5 DISCUSSION

This study examined the longitudinal relationships between motor proficiency, physical activity and musculoskeletal fitness in a large sample of young children during the transition from preschool to school age. Consistent with previous literature, we found low to moderate associations between motor proficiency and physical activity and fitness in this age group[2,5]. In line with our hypotheses, the strength of the associations between motor proficiency and physical activity emerged over the three-year study period, providing longitudinal evidence in the early years to support the emerging relationships proposed by Stodden and colleagues[7]. More motor competent children may begin to self-select or are chosen into more physically engaging activities and sports as they move from early to middle childhood and children with motor difficulties may begin to withdraw more and more from active pursuits due to a myriad of potential factors including low self-efficacy in their physical capabilities[33]. This highlights the importance of motor competence on future physical activity engagement, and suggests that the early years may be a key time to promote motor skill development in order for children to continue participation in physical activity as they transition to school age.

Motor proficiency significantly predicted positive changes in VPA and STMP over time during the early years, independent of age at baseline, sex, height and weight. VPA was associated with both motor proficiency and STMP, however, contrary to our hypothesis, VPA did not mediate the relationship between motor proficiency and STMP. These findings are in line with recent work by Cairney and colleagues[34] who found that self-reported physical activity did not explain the widening gap in cardiorespiratory fitness (predicted peak VO_2max on the 20m shuttle run) in children ages 9-14 with and without motor impairment. In contrast, Silman and colleagues[35] found objectively-measured physical activity mediated the differences in VO_2max between children with and without Developmental Coordination Disorder at age 12-13. These discrepancies may be due to different measurement techniques and study designs (e.g., self-reported vs. objectively-measured activity, predicted vs. laboratory assessed VO_2max, longitudinal vs. cross-sectional designs). Alternatively, it is possible that physical activity begins to play more of a mediating role as children transition into adolescence, physical activities become more motorically complex and physically demanding and discrepancies between children with and without motor coordination difficulties may become more pronounced. Further longitudinal research using repeated objective physical activity measurement, laboratory-based fitness assessments and standardized motor coordination tests throughout early and middle childhood and into adolescence will be needed to confirm these results.

The lack of a mediating effect of physical activity on the relationship between motor proficiency and STMP may also be explained by the coordinative demands of physical fitness tests themselves. In general, many fitness and motor coordination tasks require a high degree neuromuscular control (e.g., motor unit recruitment, firing rate, optimal co-activation), both requiring well-timed muscle contractions in order to move efficiently and economically[2,36]. It is possible that concurrent improvements in motor proficiency and musculoskeletal fitness may occur primarily through neural maturation, as a child's ability to control force in a given task improves[37], and that this may occur independently of physical activity engagement during this early developmental period. The previously reported weak associations between motor proficiency and physical activity in early childhood may also explain the lack of VPA mediation. The gap in skills between children with high and low motor competence is likely less apparent at this young age because skill demands of play are low[38]. Low skill demand, combined with the short, intermittent nature of children's' active play, will potentially increase the likelihood that children with lower motor competence will continue to engage in the same sports and activities as their more competent peers, despite their lower fitness levels. As children age and play becomes more challenging (skill-based) and sport related and physically demanding, the mediating aspect of VPA may emerge.

Limitations

Due to the young age of the sample and the time commitment for other assessments being conducted as part of the HOPP study[26], it was not feasible to conduct a full motor proficiency battery; therefore, the short-form of the BOT was used. Although this test correlates well with the full motor assessment, it does not allow for examination of the separate subdomains (e.g., fine motor vs. gross motor items). As previous research has found that object manipulation skills may be more predictive of future physical activity levels compared to other motor skills[14], it will be important for future longitudinal research to examine the relative importance of each subdomain on future health-related fitness and physical activity in this age group. As mentioned previously, we were also not able to include the children who were 3 years old at baseline testing (n=139), although we included these children in analyses at the last 2 times points, removing these children from the analysis did not significantly impact the overall results. Although continuous verbal encouragement was provided during fitness testing, we recognize that motivation and attention may have influenced test performance due to the relatively young age of our sample.

Future Directions

Since physical activity levels did not explain the relationship between motor proficiency and musculoskeletal fitness in early childhood, further research examining the underlying mechanisms (e.g., co-contraction, motor unit recruitment) linking these two constructs is needed. We will need further longitudinal studies to examine how these relationships change over time in typically developing children from preschool age through middle childhood and adolescence that include additional indicators of health-related fitness, such as aerobic fitness and body composition, in order to determine if and when physical activity begins to mediate the relationship between motor proficiency and health-related fitness. Although we do recognize that these are complex, dynamic associations influenced by many other individual and environmental factors such as growth, maturation and practice[7,25], future randomized controlled trials targeting motor proficiency would provide further evidence to support the positive influence motor competence has on long-term health outcomes.

Conclusions

Motor proficiency is an important predictor of VPA and STMP from preschool to early childhood. Although it has been hypothesized that motor proficiency is linked to fitness through physical activity engagement, we did not find evidence to support this mediation pathway. Targeting and developing appropriate motor skill interventions at an early age may help to improve both physical activity and health-related fitness in early childhood.

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Competing Interests

The authors have no competing interest to declare

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CHAPTER 5: **SUMMARY OF MAIN FINDINGS AND GENERAL DISCUSSION**

The general aim of this thesis was to enhance our understanding of the interrelationships among motor competence, physical activity and health-related fitness in the early years. Specifically, the three independent studies that comprise the body of this thesis examined: 1) differences in physical activity and health-related fitness in preschool children who meet the criteria for DCD compared to typically developing children (Chapters 2 and 3); 2) whether motor competence in typically developing children is associated with longitudinal changes in physical activity and fitness during the transition from preschool to school-age (Chapter 4); and 3) if the relationships between motor competence and health-related fitness are mediated through vigorous physical activity levels in the early years (Chapters 3 and 4). The main findings from these studies suggest that there was no differences in physical activity, body composition, or flexibility between children who met the criteria for DCD compared to typically developing children at preschool age (Chapters 2 and 3), however cardiorespiratory fitness and musculoskeletal fitness were significantly lower in young children with DCD (Chapter 3). Additionally, motor competence was positively associated with short-term muscle power both cross-sectionally (Chapter 3) and over time (Chapter 4) in the early childhood period. Physical activity was not related to motor competence at preschool-age (Chapters 2 and 4) and did not explain the differences in physical fitness between children with differing levels of motor competence (Chapters 3 and 4), however the association between motor competence and physical activity emerged as children reached school-age (Chapter 4). The following discussion will address the implications of these findings and future research directions.

5.1 Theoretical Implications

5.1.1 If not physical activity then what? Explanations beyond the activity deficit hypothesis

The primary purpose of this dissertation was to test a main tenet of the activity deficit model proposed by Hands and Larkin (2002), which suggests that low motor competence will create barriers to physical activity participation, subsequently leading to negative health-related fitness outcomes. Interestingly, the findings from this thesis did not support this mediation pathway in the early years. The significant relationships between motor competence and both cardiorespiratory and musculoskeletal fitness in young children were not explained by differences in physical activity levels. Specifically, preschool-age children who met the criteria for DCD exhausted faster on the Bruce protocol treadmill test, had lower peak and mean power on the Wingate Anaerobic test, and shorter long jump distance, but vigorous physical activity levels were similar to typically developing children and did not explain these deficits (chapter 2 and 3). Furthermore, in typically developing children, motor competence predicted positive longitudinal changes in short-term muscle power from preschool to school age, independent of significant positive changes in vigorous physical activity participation (Chapter 4).

This thesis provides strong evidence that motor competence and physical fitness outcomes are associated (Chapters 3 and 4), even in early childhood. Furthermore, the findings from Chapter 3 indicated a clear effect of the degree of motor impairment on health-related fitness outcomes. Specifically, children who had the most severe motor deficits ($\leq 5^{th}$ percentile) had the poorest cardiorespiratory and musculoskeletal fitness. However, as physical activity did not explain these associations (Chapters 3 and 4), explanations beyond the activity-deficit hypothesis are required. First, a lack of a mediating pathway may be due to the nonexistent and/or weak association between motor competence and physical activity in this early childhood period (Chapter 4). Given that preschool children with motor difficulties are engaging in similar amounts of physical activity compared to typically developing children (Chapter 2), it seems likely that another factor or factors are causing the observed differences in health-related fitness. This leads us to consider several possible alternate explanations.

First, the consistent independent link between low motor competence and low physical fitness may indicate a global delay in neuromuscular control that negatively influenced both goal-directed movement and the ability for the body to move in an efficient and economical way. For example, Raynor (2001) found higher co-activation of the vastus lateralis and biceps femoris muscles during isometric knee flexion and isokinetic knee extension, respectively, in 6- and 9-year-old children with DCD compared to controls. This manifested in lower maximal strength and power outputs in children with DCD. Similarly, during a 1-leg balance task, Gueze (2003) found increased co-activation of the tibialis anterior and peroneus muscle in children with DCD. It is therefore plausible that increased co-activation by antagonist musculature has a negative influence on both motor competence and short-term muscle power in children with DCD. It may also be that fibre type distribution or size of muscle fibres are different in children with DCD compared to typically developing children. In a study conducted by Lundberg and colleagues (1979), children with delayed walking had smaller fast twitch (type 2) muscle fibres in the vastus lateralis compared to controls. Fewer and/or smaller type 2 muscle fibres would lead to a slower rate of force development (Raynor, 1998), which may explain the deficits seen in short-term muscle power in children with DCD.

Movement inefficiencies including greater steps/min, shorter stride length, and longer stance time during running have also been observed in children with DCD when compared with typically developing children (Chia, Licari, Guelfi, & Reid, 2013). These findings likely relate to poorer postural control and lead to a reduced economy of locomotion in children with DCD, which may explain their poorer aerobic performance on the Bruce protocol. Although these mechanisms require further exploration, it is plausible that reduced movement efficiency would increase energy demands and contribute to poor fitness test performance in children with DCD, even though they may be working as hard as typically developing children.

Psychological constructs such as self-efficacy, self-control and/or perceived exertion may also contribute to the reduced fitness levels observed in children with DCD (Bandura, 1997; Barnett et al., 2008; Cairney, Hay, Faught, Léger, & Mathers, 2008; Graham, Martin Ginis, & Bray, 2017; Pageaux & Lepers, 2016; Vedul-Kjelsås, Sigmundsson, Stensdotter, & Haga, 2012). School-age children with DCD report lower perceived self-efficacy and less confidence in their physical capabilities (Cairney, Hay, Faught, Mandigo, et al., 2005; Skinner & Piek, 2001). This may translate into reduced test performance, wherein these children are less likely to persist at a task and may give up sooner, especially on endurance-based tasks.

Reduced self-efficacy may explain previously observed deficits in maximal aerobic and/or anaerobic fitness in school-age children with DCD, as well as the deficits observed in this thesis. Indeed, Cairney and colleagues (2006) found that approximately one third of the differences in 20m shuttle run performance between school-age children with and without DCD were explained by perceived (in)adequacy towards physical activity (a subscale of a generalized self-efficacy measure). As this fitness test was performed in groups, the authors suggested that the competitive and social nature of the 20m shuttle run test exacerbated the impact of perceived physical capabilities on aerobic performance (Cairney, Hay, Wade, et al., 2006). However, these findings were replicated in the laboratory setting, whereby perceived adequacy in physical activity explained a significant proportion of the variance in VO_2max between children with and without DCD (Silman et al., 2011), even after removing the confounding effect of social comparisons. This highlights that self-perceptions can influence aerobic fitness performance in school-age children with DCD even with verbal encouragement in an isolated setting, and may explain some of the performance deficits reported in the current studies (Chapter 3 and 4).

It has been demonstrated that younger children may be less able to accurately report or judge their own physical capabilities (Horn & Weiss, 1991; Rodger et al., 2003). Despite this, significant moderate correlations between perceived physical competence and actual motor competence in typically developing preschool children have been reported (Robinson, 2011b). Motor difficulties likely lead to fewer mastery experiences in the physical domain, which is one of the greatest sources of self-efficacy (Bandura, 1997). Anecdotally, it was much harder to encourage and/or motivate some of the children in the current studies who had difficulties running and/or pedaling, even during the short 30s Wingate test. Negative experiences or failures in sport or organized activity settings may therefore influence perceived self-efficacy and subsequent motivation or persistence during physical exercise tests even in children as young as preschool age.

Neurological or neuro-cognitive factors may also explain the observed relationships between motor competence and health-related fitness. Motor skills are associated with most measures of executive functioning and academic performance (Aadland et al., 2017). Interestingly, deficits in executive functions (i.e. response inhibition, set shifting, and working memory) have been observed in children with DCD (Piek, Dyck, Francis, & Conwell, 2007). Executive functions have also been linked to cognitive control, including self-regulation and self-control (Hofmann, Schmeichel, & Baddeley, 2012). As both the Bruce protocol and Wingate are maximal exercise tests that require the exertion of cognitive control to resist the urge to quit, it is possible that a deficit in self-control resulted in children with DCD not pushing themselves as hard as typically developing children on these fitness tests. It is also plausible that these tests require more cognitive effort to perform for children with low motor competence. For these children, basic movements are less automatized (running/cycling) (Wall, 2004), and therefore require more conscious effort and cognitive resources to sustain the movement. This may potentially exacerbate perceptions of physical exertion (Marcora, Staiano, & Manning, 2009), leading children with DCD to give up before true exhaustion (Chia et al., 2010). Regardless of the cognitive mechanisms involved, it is conceivable that neuro-cognitive factors may partially explain why children with poorer motor competence do not perform as well as their more motorically competent peers on fitness tests despite engaging in similar amounts of physical activity.

5.1.2 Stodden's Emerging Associations Hypothesis

The other theoretical concept that was tested in this thesis was proposed by Stodden and colleagues (2008). They postulate that the relationship between motor competence and both physical activity and fitness, will strengthen over developmental time. Although evidence is emerging to support this hypothesis (Logan et al., 2015; McIntyre et al., 2017; Robinson et al., 2015; Stodden, Gao, Goodway, & Langendorfer, 2014), the majority of extant literature relies on cross-sectional designs in combination with various definitions and measures of motor competence, fitness, and physical activity, making it difficult to make any definitive conclusions. The results from Chapter 4 provide initial support for this theory, as motor competence and physical activity were not correlated at preschool age, but a small significant correlation emerged over time as the children reached school age. We also found a strengthening association between motor competence and short-term muscle power, however this association was significant at baseline and remained in the moderate range over the three years. Results from the mixed effects models support significant, long-term positive associations between motor competence and both vigorous physical activity and musculoskeletal fitness from preschool to school age. To our knowledge these findings are the first to provide empirical support of these strengthening relationships using repeated, objective measurements of motor competence, physical activity, and fitness across early childhood.

Some underlying explanations for these emerging associations have been postulated in the literature. In Stodden and colleagues' framework, perceived motor competence is believed to be an important factor mediating the association between actual motor competence and physical activity across developmental time. In the early years, children are proposed to be less accurate at assessing their own motor (in)abilities, which may be one reason that young children with DCD and/or low motor competence are not yet withdrawing from physical activity (Chapters 2 and 4). As they age and become more cognitively mature, more accurate perceptions emerge, potentially driving the gap in physical activity participation (Cairney, Hay, Faught, Wade, et al., 2005; Horn & Weiss, 1991).

On the other hand, it is possible that preschool children with DCD do have lower self-perceptions, but that these have a stronger influence on the type of physical activity they choose to engage in (e.g., sport vs. free play), rather than the overall amount. As children get older, and the nature of physical activity becomes more sport-based (or at least, more motorically complex), a stronger association between motor competence and objectively-measured physical activity would emerge. Wall's (2004) developmental skill-learning gap hypothesis echoes this concept. In the early years the gap in skills are less noticeable as the motoric demands of play are low. Over time, activities become more motorically demanding. As the gap in skills between children with and without motor learning difficulties widens, so too does the gap in physical activity. This ultimately would translate into a stronger association between motor competence and physical activity as children age.

The relationship between motor competence and short-term muscle power also increased in strength from preschool to early childhood (Chapter 4). This is in line with findings from Stodden and colleagues (2014) who found stronger correlations between motor competence and health-related fitness in older children compared to their younger counterparts in a sample of 4- to 13-year-old children. Like physical activity, this strengthening association may be reflective of more accurate physical self-perceptions over time. That is, children with low motor competence may become more aware of their motor difficulties, leading to low self-confidence, reduced motivation, and subsequent declines in physical fitness test performance over time.

Alternatively, the strengthening association between motor competence and musculoskeletal fitness may be due to more rapid maturation of the neuromuscular system in children with more advanced motor competence. Although the quantity of activity is not lower in preschool children with low motor competence (Chapters 2 and 4), the quality of activity may not be sufficient to stimulate appropriate neuromuscular adaptations (Myer et al., 2015). The act of practicing and engaging in more ballistic and dynamic activities would hypothetically improve both motor competencies and neuromuscular efficiency, subsequently strengthening the observed associations between motor competence and physical fitness across childhood. Interestingly, a recent study by Haga and colleagues (2015) found that the correlation between motor competence and physical fitness was actually higher children age 4- to 6- years and 11- to 12-years, with lower associations found in adolescents 15- to 16-years of age. Their findings suggest that the association between motor competence and physical fitness may only continue increasing across childhood and other factors, such as significant declines in physical activity engagement (Telama, 2009) and biological maturation (M. A. Jones, Hitchen, & Stratton, 2000), may start to have a more significant impact on fitness levels as children reach adolescence.

Collectively, the studies in this dissertation provide strong evidence that motor competence across early childhood is associated with health-related fitness outcomes including cardiorespiratory and musculoskeletal fitness, with a significant association between motor competence and physical activity emerging at school age. These findings support both Hands and Larkin's and Stodden's hypotheses that motor competence is fundamentally linked to health indicators. However, contrary to Hands and Larkin's activity deficit model, the effects of motor competence on health-related fitness exist independently of physical activity engagement in the early years, replicating recent longitudinal findings in school-age children (Cairney et al., 2017; Lima, Pfeiffer, Bugge, et al., 2017). Importantly, physical fitness is predictive of future health and cardiovascular disease risk (Ortega et al., 2008; Twisk, Kemper, & van Mechelen, 2000) and both health-related fitness and motor competence show evidence of tracking through early childhood, across childhood into adolescence, and into adulthood (Caldwell et al., 2016; Janz, Dawson, & Mahoney, 2000; Malina, 1996; Twisk et al., 2000; Vandorpe et al., 2012). Therefore, these findings have significant implications for how best to intervene to reduce the risk of negative health outcomes later in life for children with low motor competence.

5.2 Practical Implications

Although this thesis set out to test one direction of the proposed activity deficit model (from motor competence to physical activity to health-related fitness), it is well recognized that these relationships are likely bidirectional, with each component influencing one another (see Fig. 1-1) (Barnett, Morgan, Van Beurden, Ball, & Lubans, 2011; Hands & Larkin, 2002; Lima, Pfeiffer, Larsen, et al., 2017; Stodden et al., 2008). Given this bidirectionality, it follows that targeting any aspect of the model should theoretically benefit the other areas. This thesis provides support that motor competence is related to physical fitness (Chapter 3), and predictive of positive changes in physical activity and musculoskeletal fitness across early childhood (Chapter 4). Therefore, it is anticipated that interventions targeting motor competence should improve both of these health-related outcomes. Despite vigorous physical activity demonstrating positive associations with markers of health-related fitness in preschool children (Chapters 3 and 4), the direct, independent associations between motor competence and both cardiorespiratory and musculoskeletal fitness suggest that solely targeting physical activity engagement, without also supporting the development of motor competence, would be less effective at improving these health-related fitness components.

Recent evidence suggests that cardiorespiratory fitness (VO_2max) mediates the long-term relationship between motor competence and physical activity from childhood to adolescence (Lima, Pfeiffer, Larsen, et al., 2017). Improving motor competence may therefore lead to a greater ability to participate and sustain physical activity engagement through its direct effect on aerobic fitness. As such, promoting both motor competence and health-related fitness would be mutually beneficial for promoting physical activity engagement. Regardless of the theoretical framework, findings from this thesis add to the mounting evidence that poor motor competence is a risk factor for poor health-related fitness and reduced physical activity participation in childhood. Although these findings at such an early age are concerning, early intervention may be able to offset the physical activity declines and cardiovascular disease risk factors, including obesity and poor cardiorespiratory fitness, that are observed in older children with poor motor competence and/or DCD (Cattuzzo et al., 2016; Hendrix et al., 2014; Rivilis et al., 2011).

It is now well recognized that motor skills do not develop spontaneously, but need to be taught and reinforced, even in typically developing children (Morgan et al., 2013). In children with DCD, the focus of intervention has been limited to improvement to motor skills, largely ignoring potential physical activity and health-related fitness benefits (Offor et al., 2016; Smits-Engelsman et al., 2013). The findings from Chapters 2 and 4 suggest that targeting motor skills may not significantly influence acute physical activity engagement at preschool age, but may help build the foundation for continuing participation as children enter school age. Specifically, preschool children who meet criteria for DCD are engaging in similar amounts of physical activity compared to their typically developing peers (Chapter 2). This therefore represents a critical time to intervene to prevent the significant deficits in activity evident when children with DCD reach middle childhood and adolescence (Rivilis et al., 2011).

Wall's skill gap model (2004) further reinforces the benefits of early intervention. He highlights that emphasizing and reinforcing fundamental motor skills in young children with motor learning difficulties will help prevent the skill and physical activity gap from emerging as these children age. This concept of teaching and reinforcing fundamental motor skills is not only restricted to children with motor difficulties. Motor-based interventions for typically developing children are also encouraged and supported throughout the physical education and motor development literature (Clark & Metcalfe, 2002; Robinson et al., 2015; Stodden et al., 2014). However, as the current evidence is mixed as to whether motor-based interventions can significantly improve physical activity and/or health-related fitness (Bremer & Cairney, 2016), carefully designed randomized controlled trials are needed to determine if they can have an impact on long-term physical activity and health outcomes in both in typically developing children and children with DCD.

Considering that in Canada, less than 10 percent of children are meeting the recommended levels of physical activity (ParticipACTION, 2016), it is unlikely that remediation or compensation of motor deficits alone would drastically change participation patterns in children with low motor competence. A holistic, physical literacy approach targeting not only motor abilities, but also affective, motivational, and environmental factors might have the greatest impact on physical activity participation for both typically developing children and children with DCD (Cairney, Bedard, Dudley, & Kriellaars, 2016; Thelen & Smith, 1994; Wade, Johnson, & Mally, 2005). Physical literacy is defined as the "motivation, confidence, physical competence, knowledge and understanding to value and take responsibility for engagement in physical activities for life" (ParticipACTION et al., 2015). This approach explicitly targets motor competence and confidence by ensuring opportunities for mastery are available while having the autonomy to be creative in a fun, enjoyable cognitively-enriching environment (Cairney et al., 2016). Using the physical literacy framework for intervention design may have positive influences on motor skills, while also building a strong foundation for children to enjoy and continue participating in physical activity for life.

The results from this thesis emphasize that cardiorespiratory and musculoskeletal fitness are already impaired in preschool-age children who meet the criteria for DCD (Chapter 3), and a moderate relationship between motor competence and musculoskeletal fitness exists throughout preschool to early childhood (Chapter 4). If, as Stodden's developmental model proposes, these fitness deficits are what lead to the physical activity declines seen later in childhood, it is imperative that fitness outcomes are measured and targeted along with motor skills. This is especially evident given the current findings that physical activity participation does not appear to meaningfully offset these fitness deficits (Chapter 3 and 4).

Exercise training or motor skill interventions may be able to improve both motor competence and fitness in children with DCD. Case reports have found that strength training has benefits not only to muscle strength but also gross motor function (Kaufman & Schilling, 2007; Menz, Hatten, & Grant-Beuttler, 2013), potentially through improvement in neuromuscular control. Recently, Farhat and colleagues (2015) found that an 8-week motor skill and agility training program was able to improve both motor and fitness outcomes including MABC-2 scores, distance on the 6 min walk test, and VO_2max, as well as reduce perceived exertion during fitness tests in children with DCD, with no changes in either the DCD or typically developing control groups. Furthermore, Ferguson and colleagues (2013) found neuromotor task training (a type of task-oriented motor-based intervention) was successful at improving cardiorespiratory fitness, functional strength, as well as motor competence in children with DCD. Interestingly, exercise interventions have not only been able to demonstrate improvements in physical fitness and motor skills, but they have also had positive effects on physical self-perceptions in both adolescent boys and girls with DCD (Bonney, Ferguson, & Smits-Engelsman, 2017; McIntyre, Chivers, Larkin, Rose, & Hands, 2015), which may increase their motivation and likelihood of participating in physical activity (Cairney, Hay, Faught, Wade, et al., 2005; Poulsen et al., 2008). Taken together, emerging evidence suggest that targeting motor competence will positively influence physical fitness and vice versa in children with low motor competence; however, whether these changes influence acute or long-term physical activity engagement remains to be determined.

The findings from this thesis, together with the existing literature, create a strong case that the development of motor competencies in children as young as preschool age are highly important for health-related fitness and future physical activity (Cattuzzo et al., 2016; Figueroa & An, 2017; Hendrix et al., 2014; Holfelder & Schott, 2014; Logan et al., 2015; Lubans et al., 2010; Rivilis et al., 2011; Robinson et al., 2015). It is therefore essential that teaching and reinforcing motor competencies are incorporated into the curriculums of early childcare centres and kindergarten physical education classes, with the ultimate goal of offsetting declines in physical activity participation and health-related fitness as children age (Kolen-Thompson, Baxter-Jones, Mirwald, & Bailey, 2003; Nader, Bradley, Houts, McRitchie, & O'Brien, 2008; Sallis, 1993). By encouraging and helping children form a strong base of fundamental motor skills, physical educators may be able to reverse the secular declines in physical activity and health-related fitness we are observing in youth today (Dollman et al., 2005; Tomkinson & Olds, 2007; Tremblay et al., 2010; Tremblay & Willms, 2000).

5.3 Future Research Directions

Although the studies included in this thesis provide a comprehensive understanding of the interrelationships among motor competence, physical activity and health-related fitness in the early years, they also serve to highlight areas for future investigation. Firstly, there are aspects of the theoretical models proposed by both Hands and Larkin and Stodden and Colleagues that were not included in this thesis that may also play important roles in explaining these interrelationships. For example, future research might examine the effects of psychological (self-efficacy, perceived physical exertion) and environmental (parental support, social pressures) factors on the associations between motor competence and both physical activity and fitness in the early years. Additionally, examining the specific domains of motor competence that are most predictive of future health-related fitness and physical activity in this young age group will further support and better inform intervention strategies.

Secondly, although the included studies were some of the first to measure cardiorespiratory and musculoskeletal fitness using objective laboratory techniques in preschool children, there is limited evidence that these outcomes in the early years will be related to future health in adulthood. We recognize that performance on these tests are largely dependent on growth (i.e. height and weight), and as such, we controlled for these growth parameters in our analyses. Still, performance on these fitness tests may be more indicative of global maturation than future health per se. Future research following children from preschool age to adolescence and into adulthood is needed to determine the predictive validity of these preschool health-related fitness assessments on future health and cardiovascular risk. Longer follow-up periods will also be needed to examine if the strength of these associations continue to increase as children transition from early childhood into adolescence. Furthermore, the underlying neuromuscular link between motor competence and physical fitness outcomes has yet to be elucidated in young children, and should be examined further in order to understand the evolving nature of the association.

Thirdly, as this thesis relied on observational longitudinal data (Chapter 4), a causal relationship between motor competence and physical activity and health-related fitness can be suggested, but not definitively concluded. As such, there is a need for well-designed randomized controlled trials targeting motor competence in the early years, with repeated objective assessments of motor competence, physical activity, and health-related fitness in both typically developing children and children with DCD. Empirical evidence of the effects of improved motor competence on acute and long-term physical activity and health-related fitness changes, will provide further support for incorporating motor-based teachings into physical education curriculums.

Finally, as rates of motor development are highly variable (Clark & Metcalfe, 2002), it is possible that some children who meet the criteria for

DCD at preschool age, specifically those in the at-risk group, may ‘catch up’, perhaps as a results of more opportunities for practice (Pless, Carlsson, Sundelin, & Persson, 2002). Following the CATCH cohort over time will allow us to determine how changes, or stability, in DCD status impact physical activity and health-related fitness as they age. As body composition and physical activity deficits were not found in this young cohort, but have previously been reported in school-age youth with DCD, this longitudinal study will elucidate when, or if, these differences become apparent over time. This will provide insight into the level or ‘critical threshold’ of motor impairment that limits physical activity participation and significantly raises the risk of obesity as children age. This work will therefore fill an important gap in both the DCD and physical education literatures.

5.4 Conclusions

This thesis makes a significant contribution to the literature as it represents the first series of studies examining objectively-measured motor competence, physical activity, and health-related fitness in two large cohorts of preschool-age children. Specifically, these results are the first to demonstrate that physical activity deficits are not yet apparent in preschool children who meet the criteria for DCD, and therefore offer promise that early identification and intervention may offset the gap in activity that emerges later in childhood (Chapter 2). Furthermore, results from Chapters 3 and 4 provide strong evidence that motor competence is associated with health-related fitness, specifically cardiorespiratory and musculoskeletal fitness, independent from physical activity engagement in young children. Importantly, results from Chapter 4 demonstrate that motor competence positively predicts trajectories of short-term muscle power and vigorous physical activity from preschool to school age, and provides empirical support for the emerging association between motor competence and physical activity across the early years. Collectively, these studies highlight that poor motor competence in early childhood is an independent risk factor for decreased cardiorespiratory and musculoskeletal fitness as well as lower levels of physical activity engagement as children age.

CHAPTER 6: **REFERENCES**

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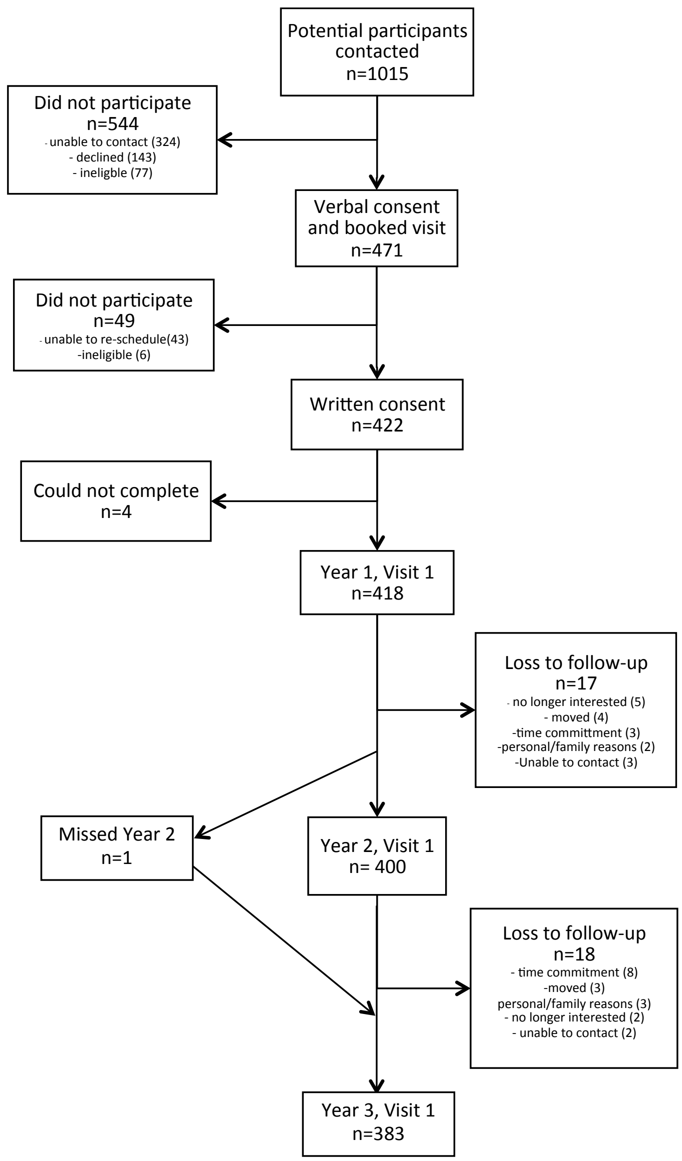
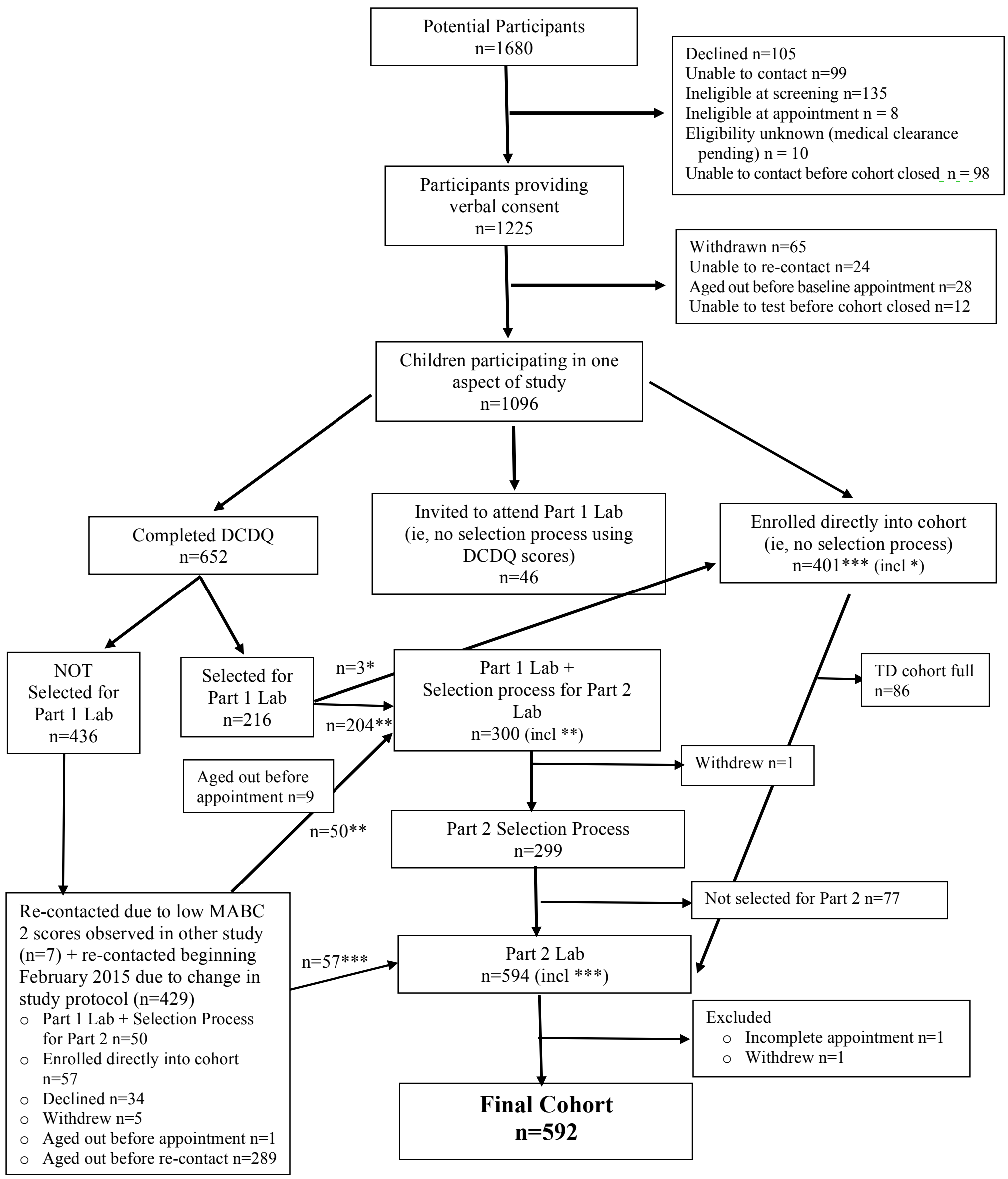
APPENDIX A: HOPP STUDY FLOW OF PARTICIPANTS



APPENDIX B: CATCH STUDY FLOW OF PARTICPANTS



Note: DCDQ=Developmental Coordination Disorder Questionnaire

APPENDIX C: COPYRIGHT PERMISSION

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INVOICE/CONTRACT

30 October 2017

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