PHYSICAL ACTIVITY FOR CHILDREN WITH AUTISM SPECTRUM DISORDER

EXPLORING THE ROLE OF PHYSICAL ACTIVITY FOR HEALTH, BEHAVIOUR, AND COGNITION IN CHILDREN WITH AUTISM SPECTRUM DISORDER

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LAY ABSTRACT

Children with autism spectrum disorder (ASD) experience a range of behavioural and physical health difficulties. Participation in physical activity may address these difficulties. However, there is limited evidence regarding the utility of physical activity for children with ASD. This dissertation explored the role of physical activity for this population through a set of studies designed to fill multiple gaps in the literature. First, the ability to use common fitness assessments for children with ASD was established. Next, this dissertation explored the importance of adaptive behaviour for the physical health of children with ASD, finding a positive relationship between motor competence and adaptive behaviour and that adaptive behaviour influences the relationship between motor competence and fitness. Lastly, this dissertation provided preliminary support for improved cognitive abilities following 20-minutes of exercise. Together, these findings highlight the role of physical activity in the health, behaviour, and cognitive functioning of children with ASD.

ABSTRACT

Children with autism spectrum disorder (ASD) experience difficulties with social communication and social interactions, in addition to the presence of a restricted or repetitive range of behaviours and interests. On top of these core symptoms, children with ASD experience numerous health deficits, as well as impairments in executive functioning. It is possible that physical activity may be an effective intervention to collectively address these deficits and behavioural challenges. Yet, to-date, literature in this area has been limited. Therefore, this dissertation explored the role of physical activity and its relationship to pertinent health, behavioural, and cognitive variables in children with ASD.

The first study in this dissertation demonstrated the feasibility and test-retest reliability of select fitness assessments in 7-12 year old children with ASD. Study two demonstrated the association between motor competence and adaptive behaviour. The third study built on this work by showing that motor competence and fitness are positively associated and that adaptive behaviour moderates this relationship. However, significant associations were not present between physical activity and health or behavioural variables. Lastly, study four demonstrated that an acute bout of exercise improves executive functions in children with ASD, with a circuit-based workout eliciting larger effects than aerobic exercise.

Collectively, this dissertation provides a comprehensive examination of the role of physical activity for children with ASD. Specifically, these studies highlight the important relationships between motor competence and behaviour, irrespective of physical activity levels, while also highlighting the acute effect of physical activity on executive functions. These findings provide important foundational knowledge that can be built upon to improve the health and behavioural well-being of children with ASD through physical activity-based programs and interventions.

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LIST OF ABBREVIATIONS AND SYMBOLS

ADHD	Attention Deficit Hyperactivity Disorder
ANOVA	Analysis of Variance
ASD	Autism Spectrum Disorder
BMI	Body Mass Index
bpm	Beats per Minute
BRIEF	Behavior Rating Inventory of Executive Functions
cm	Centimetre
CV	Variation Coefficients
DCD	Developmental Coordination Disorder
fNIRS	Functional Near-Infrared Spectroscopy
GARS-3	Gilliam Autism Rating Scale, 3rd Edition
HR	Heart Rate
ICC	Intraclass Correlation
IOTF	International Obesity Task Force
kg	Kilogram
kg/s^2	Kilogram per Second Squared
m	Metres
M	Mean
MABC-2	Movement Assessment Battery for Children, 2nd Edition
mm	Millimetres
µmol	Micromole
MPST	Muscle Power Sprint Test
MVPA	Moderate-to-Vigorous Physical Activity
m/s	Metres per Second
m/s^2	Metres per Second Squared
M6MWT	Modified 6-Minute Walk Test
Nm/kg	Newton Metre per kilogram
NSCH	National Survey of Children's Health
η_p^2	Partial Eta Squared
oxy-Hb	Oxygenated Hemoglobin
PACER	Progressive Aerobic Capacity Endurance Run
RPE	Ratings of Perceived Exertion
RPME	Ratings of Perceived Mental Exertion
RBS-R	Repetitive Behavior Scale – Revised
s	Seconds
SD	Standard Deviation
SE	Standard Error
SPSS	Statistical Package for the Social Sciences
VABS-2	Vineland Adaptive Behavior Scales, 2nd Edition
VAS	Visual Analogue Scale
W	Watts
6MWT	6-Minute Walk Test
Δ	Change

DECLARATION OF ACADEMIC ACHIEVEMENT

This thesis was prepared in the “sandwich thesis” format as outlined in the McMaster University School of Graduate Studies Guide for the Preparation of Master’s and Doctoral Theses. It includes a general introduction, four original research papers, and a general discussion. The candidate is the first author on all of the manuscripts. At the time of the thesis preparation, Chapter 2 was under peer-review (revisions submitted), Chapter 3 was published, and Chapters 4 and 5 were under peer-review.

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CHAPTER 1: INTRODUCTION

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Preamble

Regular participation in physical activity has numerous physical (Janssen & LeBlanc, 2010; Poitras et al., 2016) and mental health (Dale, Vanderloo, Moore, & Faulkner, 2018; Lubans et al., 2016) benefits for children and youth. In order to gain these benefits, it is recommended that children and youth engage in 60 minutes of moderate to vigorous physical activity (MVPA) each day (Tremblay et al., 2016). Despite these recommendations and the known benefits of regular engagement, physical activity levels remain incredibly low with only one-third (36%) of Canadian children and youth meeting current physical activity guidelines (Roberts et al., 2017). Alarmingly, children and youth with disabilities are at an even higher risk of inactivity than their typically developing peers (Rimmer & Rowland, 2008). Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by impairments in the areas of social interaction and social communication, and the presence of restricted or repetitive interests and behaviours (American Psychiatric Association, 2013). The combination of social and behavioural challenges experienced by children with ASD may place them at an even greater risk of physical inactivity; yet, this also makes them an ideal population to benefit, physically and behaviourally, from regular participation. Indeed, exercise has been recognized as 1 of 27 types of intervention to meet the criteria for evidence-based practice in individuals with ASD (Wong et al., 2015). Yet, in order to introduce feasible, effective, and sustainable physical activity interventions for this population, we must first understand how physical activity is related to health and behavioural outcomes, along with how these outcomes interact, to elicit positive behavioural change in this population.

Autism Spectrum Disorder

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by impairments in the areas of social communication and social interactions, in addition to the presence of restricted or repetitive behaviour, interests, or activities (American Psychiatric Association, 2013). Specifically, the disorder is characterized by 1) persistent deficits in social communication and social interaction across multiple contexts; and 2) restricted, repetitive patterns of behaviour, interests, or activities (American Psychiatric Association, 2013). Further, these symptoms must be present in the early developmental period; cause clinically significant impairment in social, occupational, or other important areas of current functioning; and not be better explained by intellectual disability or global developmental delay (American Psychiatric Association, 2013). Moreover, the severity level of the deficits in social communication and repetitive behaviours, respectively, are classified on a scale of 1-3, wherein a higher severity level indicates a greater amount of support is required in that area (see Table 1).

Table 1. Description of ASD severity levels from the Diagnostic and Statistical Manual of Mental Disorders, 5th Edition (American Psychiatric Association, 2013, pp. 50–59).

Severity Level	**Social Communication**	**Restricted, Repetitive Behaviours**
Level 1 “Requiring support”	Without supports in place, deficits in social communication cause noticeable impairments. Difficulty initiating social interactions, and clear examples of atypical or unsuccessful response to social overtures of others. May appear to have decreased interest in social interactions. For example, a person who is able to speak in full sentences and engages in communication but whose to- and-fro conversation with others fails, and whose attempts to	Inflexibility of behavior causes significant interference with functioning in one or more contexts. Difficulty switching between activities. Problems of organization and planning hamper independence

	make friends are odd and typically unsuccessful.	
Level 2 "Requiring substantial support"	Marked deficits in verbal and nonverbal social communication skills; social impairments apparent even with supports in place; limited initiation of social interactions; and reduced or abnormal responses to social overtures from others. For example, a person who speaks simple sentences, whose interaction is limited to narrow special interests, and who has markedly odd nonverbal communication.	Inflexibility of behavior, difficulty coping with change, or other restricted/repetitive behaviors appear frequently enough to be obvious to the casual observer and interfere with functioning in a variety of contexts. Distress and/or difficulty changing focus or action.
Level 3 "Requiring very substantial support"	Severe deficits in verbal and nonverbal social communication skills cause severe impairments in functioning, very limited initiation of social interactions, and minimal response to social overtures from others. For example, a person with few words of intelligible speech who rarely initiates interaction and, when he or she does, makes unusual approaches to meet needs only and responds to only very direct social approaches	Inflexibility of behavior, extreme difficulty coping with change, or other restricted/repetitive behaviors markedly interfere with functioning in all spheres. Great distress/difficulty changing focus or action.

As of 2015, the prevalence of ASD in Canada was 1 in 66 children between 5-17 years of age, with the prevalence being four times higher in males than females (National Autism Spectrum Disorder Surveillance System, 2018). In addition to the social, communicative, and behavioural challenges experienced by children with ASD, this is a population who also exhibits low levels of adaptive behaviour (Kanne et al., 2011) and a high incidence of medical, psychiatric, and developmental comorbidity (Matson, 2016). Indeed, the presence of both co-occurring and co-morbid conditions often represent some of the most challenging aspects of ASD, with many pointing to the management of these conditions as one of the most critical aspects of the disorder (Gillberg & Fernell, 2014; Matson, 2016).

Given that ASD is considered to be a neurodevelopmental disorder, much attention has also been given to neural connectivity and cerebral functioning in this population (Belmonte, 2004). Indeed, work in the area of brain imaging has found both functional and anatomical disturbances in neural connectivity (Anagnostou & Taylor, 2011; Belmonte, 2004; Schipul, Keller, & Just, 2011; Wass, 2011). This abnormal connectivity is generally regarded as long-distance (i.e., between region) under-connectivity and local (i.e., single region) over-connectivity (Wass, 2011). In fact, it has been proposed that regional under-connectivity may be one of the biggest contributors to both the behavioural challenges and deficits in executive functioning evident in ASD (Just, 2004; Just, Cherkassky, Keller, Kana, & Minshew, 2007). Moreover, research has found that neural disruptions, including regional under-connectivity, may be most prominent in later developing brain regions including the prefrontal cortex (Just et al., 2007; Wass, 2011). This has important implications for executive functions, the higher-order brain-based processes that are necessary for goal-directed behaviours, that are largely orchestrated through the prefrontal cortex (Anderson, 2001). Executive functions include inhibition, working memory, and cognitive flexibility, and they play an important role in virtually all aspects of life (Diamond, 2013). For instance, executive functions are needed to maintain good physical and mental health; they lead to success in academic and vocational roles and are associated with overall quality of life (Diamond, 2013). Individuals with ASD experience deficits in executive functioning (Demetriou et al., 2018; Hill, 2004b; Russo et al., 2007), which can be attributed to the widespread neural disruptions evident in the disorder. Executive functions can, however, be improved with training and practice, with individuals with the largest deficits in executive functions often showing the largest gains (Diamond, 2013). Improving executive functions may help children with ASD to better regulate their emotions, inhibit repetitive or stereotypic behaviours, and increase their focused attention on social cues. Thus, it is imperative that interventions for children with ASD address the executive dysfunction associated with the disorder.

Health Deficits

In addition to the core challenges of the disorder, individuals with ASD also experience a number of physical health deficits, which may be related to the many behavioural challenges that are present. Individuals with ASD have a 2.5-fold increased risk of all-cause mortality when compared to the general population (Hirvikoski et al., 2016). Moreover, individuals with ASD have an increased risk of specific-cause mortality across virtually all mortality categories including mental and behavioural disorders, and diseases of the nervous, circulatory, respiratory, and digestive systems (Hirvikoski et al., 2016). Not surprisingly, individuals with ASD also experience a high prevalence of co-occurring health conditions, starting in childhood and persisting through adulthood (Flygare Wallén, Ljunggren, Carlsson, Pettersson, & Wändell, 2017; Levy et al., 2018; Matson, 2016). For example, one recent review found that children and adolescents with ASD had an increased prevalence of immunological, gastrointestinal, and neurological conditions in comparison to typically developing controls (Muskens, Velders, & Staal, 2017). Levy et al. (2018) similarly found that children with ASD had significantly more gastrointestinal disorders, feeding difficulties, and sleep problems than children with developmental delays and the general population. Ultimately, individuals with ASD experience a high degree of medical, psychiatric, and developmental comorbidity (Matson, 2016), all of which can increase the complexity of treatment (and shift treatment priorities) for this population. Given this treatment complexity, it is imperative that interventions for individuals with ASD simultaneously target multiple health and behavioural outcomes. Physical activity is one modifiable health behaviour that can have positive effects on multiple areas of health, including physical, mental, and cognitive domains (Lee et al., 2012; Lubans et al., 2016; Poitras et al., 2016). Yet, the role of physical activity in the treatment of ASD is often overlooked.

Typical Treatment for ASD

Treatment for children with ASD typically focuses on the behavioural challenges associated with the disorder, through a combination of behavioural interventions (e.g., applied behaviour analysis), social skills training, speech-language therapy, and occupational therapy (Myers & Johnson, 2007; Virués-Ortega, 2010). These treatment strategies are generally effective at improving behavioural and social functioning outcomes for children with ASD, particularly if they are intensive in dose and introduced in early childhood (Corsello, 2005; Virués-Ortega, 2010). For example, long-term comprehensive applied behaviour analysis leads to positive changes in intellectual functioning, adaptive behaviour, and language development, with effects that are medium to large in magnitude (Virués-Ortega, 2010). However, these behavioural interventions can be extremely cost prohibitive for families, social services, and health care systems (Buescher, Cidav, Knapp, & Mandell, 2014; Chasson, Harris, & Neely, 2007; Horlin, Falkmer, Parsons, Albrecht, & Falkmer, 2014). Furthermore, many families face long wait times for their child’s services, often leading to missed treatment windows (Piccininni, Bisnaire, & Penner, 2017) and reduced family quality of life (Brown, MacAdam–Crisp, Wang, & Iarocci, 2006).

Physical activity is not considered a usual treatment for children with ASD, but may be a cost-effective intervention that could augment or support current practices. Physical activity has known physical (Janssen & LeBlanc, 2010; Poitras et al., 2016) and mental health (Dale et al., 2018; Lubans et al., 2016) benefits, and has shown promise in improving behaviour, social skills, and cognition in children with ASD (Bremer, Crozier, & Lloyd, 2016; Lang et al., 2010; Tan, Pooley, & Speelman, 2016). Moreover, families of children with ASD have reported that having ample opportunity to engage in leisure and recreation activities is associated with improved family quality of life (Jones, Bremer, & Lloyd, 2017). Yet, these families are not participating in leisure and recreation activities with their child with ASD to the extent that they would like to participate (Jones et al., 2017). Therefore, it is important to explore how physical activity relates to the health and behaviour of children with ASD, along with its role as a viable intervention for this population.

Physical Activity

Children and youth with ASD tend to engage in fewer minutes of physical activity (Healy & Garcia, 2018; Jones et al., 2017), along with fewer types of activities (Bandini et al., 2013), than their peers with typical development. The most recent systematic review to examine the levels and correlates of physical activity in children and adolescents with ASD included 15 studies that reported rates of physical activity participation; 5 of which compared children with ASD to those with typical development (Jones et al., 2017). Four of these studies reported that children with ASD engaged in less physical activity when compared against their typically developing peers (Jones et al., 2017). The authors of the review also identified 24 potential correlates of physical activity in children with ASD across 10 studies (Jones et al., 2017). Unfortunately, most child-level, demographic, and environmental correlates were not sufficiently investigated to draw meaningful inferences. However, age was found to be negatively associated with physical activity, while day of the week showed no association (Jones et al., 2017). A large study recently published by Healy and colleagues (2018) also found a significant, negative, association between age and physical activity in children with ASD, in addition to a positive association between normal weight status and physical activity. The authors did not, however, find an association between physical activity and any environmental factors (e.g., neighbourhood safety and amenities) (Healy, Garcia, et al., 2018).

Since Jones and colleagues' review (2017), new literature continues to provide evidence that children with ASD may be less physically active than their peers with typical development (Healy & Garcia, 2018; Healy, Haegele, Grenier, & Garcia, 2017; Stanish et al., 2017). For example, Stanish and colleagues (2017) found that adolescents, 13-21 years of age, with ASD engaged in significantly less physical activity (29 minutes of daily MVPA) than their typically developing peers (50 minutes of daily MVPA). Similar deficits are evident in samples of 9 and 13 year old children with ASD, respectively, whose engagement in both physical activity and sports was found to be significantly lower than peers without the disorder (Healy & Garcia, 2018; Healy et al., 2017). Evidence also suggests that the differences in physical activity participation also extend to low rates of participation in leisure activities, including social (e.g., involvement in clubs or organizations), skill (e.g., participation in sports teams or lessons), and chore-based activities (Ratcliff, Hong, & Hilton, 2018); thus, placing children and youth with ASD at an even greater risk of inactivity as they transition into adulthood. Indeed, adults with ASD engage in significantly less physical activity when compared to children and adolescents with the disorder (Garcia-Pastor, Salinero, Theirs, & Ruiz-Vicente, 2018), which is alarming given the already low levels of physical activity evident in childhood.

Beyond measuring physical activity levels, it is important to understand how different variables both relate to, and can be influenced by, participation in physical activity in children with ASD. This level of understanding is necessary to guide intervention planning. Yet, similar to the literature in typical populations (Metcalf, Henley, & Wilkin, 2012), there is little evidence that physical activity interventions actually increase physical activity levels in children with ASD. Moreover, it would appear that the majority of research employing physical activity interventions do not measure change in physical activity behaviours as an outcome. For instance, a recent meta-analysis of physical activity interventions for youth with ASD reported positive effects in the areas of body composition, fitness, motor skills, and social functioning (Healy, Nacario, Braithwaite, & Hopper, 2018); yet, there was no mention of the impact on physical activity levels. This may be due to a lack of effect of physical activity interventions on actual physical activity levels or may be that these interventions have not been long enough to produce a meaningful change in activity levels. Further, publication biases may result in null effects on physical activity variables not being reported in the literature. However, it is also possible that increasing physical activity levels is simply not a priority for individuals working with children with ASD. This may then speak to the need to harness physical activity as a behavioural, rather than physical health, intervention for this population.

Models of Physical Activity Participation

In order to use physical activity as an effective intervention for children and youth with ASD, it is imperative to understand the correlates and outcomes of physical activity participation, along with how these variables relate to one another. One model that is frequently used in the motor development literature is Stodden's developmental model of motor skill

(Stodden et al., 2008), which has recently been updated with substantial support for many of its pathways (Robinson et al., 2015). The model proposes positive relationships between motor competence, physical activity, fitness, and perceived motor competence; with these positive relationships also being associated with improved weight status (Robinson et al., 2015; Stodden et al., 2008). There is a great deal of support for aspects of this model in children with typical development (Robinson et al., 2015), along with some support for the model in clinical populations (Cairney, 2015; King-Dowling, Rodriguez, Missiuna, Timmons, & Cairney, 2018; Rivilis et al., 2011). However, limited research has tested these proposed pathways in children with ASD.

More recently, a model of physical activity participation was proposed by Arnell and colleagues (2017) specifically for children with ASD. This model, called the “Conditional Participation Model”, positions engagement in physical activity as conditional upon the intersection of five factors: competence and confidence, motivation, adjustment to external demands, predictability, and freedom of choice (Arnell et al., 2017). This model may be more appropriate for children with ASD as it considers some of the behavioural and social characteristics that may impact their participation; yet, it has not been empirically tested.

It is likely that aspects of both of these models need to be considered in the design of appropriate physical activity interventions for children with ASD and accordingly, aspects of both of these models can likely be improved through intervention. Therefore, it is essential to examine the state of the literature in regard to the variables that may be related to, or outcomes of, physical activity. This will help us to identify gaps in the literature and variables that may have the greatest influence on physical activity for children with ASD.

Predictors and Outcomes of Physical Activity

Physical Health

Given the potential influence of motor competence and fitness on physical activity levels in children (Robinson et al., 2015), it is important to identify the state of the literature in regard to these variables in children with ASD. This will help us to recognize the gaps in knowledge that need to be addressed in order to better understand the role of physical activity for this population. Importantly, the physical health variables discussed in this section can be considered both predictors (i.e., necessary for participation) and outcomes (i.e., influenced by participation) of physical activity.

Health-related Fitness

Health-related fitness includes cardiovascular fitness, muscular endurance, muscular strength, flexibility, and body composition (Corbin, Pangrazi, & Franks, 2000). While the fitness levels of children with ASD have generally received little empirical attention, the research that does exist suggests this is a population who consistently experiences low levels of fitness (Borremans, Rintala, & McCubbin, 2010; Bricout et al., 2018; Pan, 2014; Tyler, MacDonald, & Menear, 2014). For example, Bricout and colleagues (2018) recently investigated the aerobic capacity, flexibility, and muscular strength and power of a sample of 20 males with ASD averaging 10 years of age. The results indicated that, in comparison to their peers with typical development, the children with ASD performed significantly lower on all fitness variables (Bricout et al., 2018). Similarly, in one of the larger investigations of the fitness of adolescents (10-17 years of age) with (n=31) and without (n=31) ASD, Pan (2014) found that those with ASD scored significantly lower on measures of aerobic fitness, muscular endurance, and flexibility. Likewise, Borremans et al. (2010) reported deficits in flexibility, muscular strength, running speed, and cardiorespiratory endurance in a slightly older sample of 15-21 year old individuals with ASD (n=30), compared to their peers with typical development (n=30). In contrast, a study of the fitness of children 9-17 years of age with ASD (n=17) found that significant differences were evident only in strength, but not aerobic capacity or flexibility, when compared to children with typical development (Tyler et al., 2014).

Taken together, these findings suggest that children with ASD experience deficits across fitness domains. However, this literature is limited by a lack of consensus on how best to measure fitness in this population, which assessments to use, and whether they are valid for this population. We cannot assume that fitness assessments that were initially designed for adults, and later validated for children with typical development, will be suitable for use with children with ASD. We know that this is a population who experiences challenges in the motor domain (Fournier, Hass, Naik, Lodha, & Cauraugh, 2010), along with deficits in social communication skills and a restricted range of interests and behaviours (American Psychiatric Association, 2013). Further, children with ASD often experience difficulties with sensory processing and modulation (Ben-Sasson et al., 2009). This unique combination of challenges may make fitness testing particularly difficult for a child with ASD, particularly if it involves novel equipment (e.g., treadmill, cycle ergometer), or the need to regulate their behaviour to follow multi-step instructions in order to complete the test. To-date, none of the studies reporting on fitness outcomes in children with ASD have reported information regarding the feasibility, reliability, or validity of the assessments used. As such, one cannot discern whether the fitness tests are being completed as intended, nor whether they are actually measuring the fitness parameters that they claim to measure. As fitness is both a critical component of overall health and a common outcome of physical activity interventions, it is imperative that we establish key measurement properties of commonly used fitness assessments.

Body composition. The literature regarding body composition, an aspect of health-related fitness, has received more attention than many other physical health variables. This data overwhelmingly indicates that children with ASD are at risk of being overweight and obese (Balogun, 2016; Healy, Aigner, & Haegele, 2018; McCoy, Jakicic, & Gibbs, 2016). For example, a recent review including multiple large, population-based, studies found that children and adolescents with ASD experience an increased prevalence of obesity, ranging from 10-30% of those with ASD (Balogun, 2016). Similarly, McCoy and colleagues (2016) examined differences in body composition and physical activity behaviours in a large sample of adolescents with (n=915) and without (n=41,879) ASD through a secondary data analysis from the US National Survey of Children's Health (NSCH). They found that the adolescents with ASD were 27% more likely to be overweight and 72% more likely to be obese than their peers with typical development. Interestingly, they also found that those with ASD were 48% more likely to be underweight than their peers with typical development. However, when participation in physical activity, sports, and clubs was controlled for, only the odds of being obese remained significantly elevated in those with ASD, with an odds ratio of 1.33 (McCoy et al., 2016). More recently, Healy and colleagues (2018) examined a newer data cycle from the NSCH that included 750 adolescents (weighted n=875,963; 10-17 years of age) with ASD in comparison to 25,173 adolescents (weighted n=31,913,657) with typical development. They found that those with ASD were at close to 1.5 times greater odds of both overweight and obesity than those without. Further, adolescents with more severe ASD had a significantly higher odds of obesity (odds ratio = 3.35) than those with mild ASD. Similar findings from another large-scale study found that children with ASD (n=668) were 1.5 times more likely to be overweight or obese than general population controls (n=884), even when controlling for co-occurring medical, behavioural, developmental, and psychiatric conditions (Levy et al., 2018). Additionally, this study also found that children with ASD with moderate or severe symptoms were 1.7 times more likely to be overweight or obese than those children with mild symptoms (Levy et al., 2018). Taken together, these results demonstrate that children and adolescents with ASD are at an increased risk of an unhealthy body composition, with this risk being exacerbated as symptom severity increases.

Despite the detrimental effects of an unhealthy body composition, very few interventions (N=12) have addressed weight management in this population, with only half of these studies finding significant weight loss (Healy, Pacanowski, & Williams, 2018). There are likely many factors that contribute to the increased risk of overweight and obesity in children with ASD including metabolic side-effects from medications, sleep disturbances, and abnormal eating behaviours (Curtin, Jojic, & Bandini, 2014). However, one potentially modifiable factor (or set of factors) that may contribute to this increased risk is the motor competence – physical activity – physical fitness pathway (Robinson et al., 2015; Stodden et al., 2008), which would suggest that children who experience deficits in these areas will also have an unhealthy body composition. Yet, these pathways have not been explored in children with ASD. Understanding the relationship between body composition and physical activity in children with ASD will provide insight into whether body composition is likely to improve with physical activity, as well as the need to potentially account for differences in body composition in the design and analysis of physical activity interventions.

Motor Competence

Children with ASD experience significant delays in their motor competence, which have been noted since Kanner's (1943, 1971) first descriptions of the disorder. It is evident that these motor delays appear early in development (Lloyd, MacDonald, & Lord, 2013) and persist through childhood and into adulthood (Bhat, Landa, & Galloway, 2011). Further, motor delays persist across multiple realms of the motor domain including motor coordination and postural stability, and include deficits in planning and sensorimotor integration, in addition to the actual execution of motor tasks (Fournier et al., 2010). Motor delays may have implications across multiple domains of functioning for children with ASD as a result of limited opportunities to engage with one's environment and peers through movement-based exploration. The development of motor and social skills are indeed interrelated in children with and without neurodevelopmental disorders (Leonard & Hill, 2014), yet the implications for children with ASD may be even more far-reaching. Previous research has found associations between motor skills and measures of social communicative skills (Hirata et al., 2014; MacDonald, Lord, & Ulrich, 2013b; Sipes, Matson, & Horovitz, 2011), adaptive behaviour (MacDonald, Lord, & Ulrich, 2013a), autism severity and status (LeBarton & Landa, 2019; MacDonald, Lord, & Ulrich, 2014) and language (Bhat, Galloway, & Landa, 2012; LeBarton & Iverson, 2013; LeBarton & Landa, 2019). Given the priority of these areas for intervention in children with ASD, their relationship with the motor domain cannot be overlooked.

In terms of participation in physical activity, fundamental movement skills are considered the foundational movements necessary for regular engagement (Lubans, Morgan, Cliff, Barnett, & Okely, 2010; Payne & Isaacs, 2011). Fundamental movement skills are positively associated with physical activity, fitness, and a healthy body composition in children with typical development (Lubans et al., 2010; Robinson et al., 2015). While there is some evidence of a positive association between motor competence and fitness in children with ASD (Pan, 2014), these pathways have traditionally received little attention in this population. This is a concern given that previous research indicates that the fundamental movement skills of children with ASD are significantly delayed (Bremer, Balogh, & Lloyd, 2015; Ketcheson, Hauck, & Ulrich, 2018; Staples & Reid, 2010); thus, potentially placing them at risk of numerous health and behavioural deficits. Fundamental movement skills have been shown to improve through intervention (Bremer et al., 2015; Bremer & Lloyd, 2016; Ketcheson, Hauck, & Ulrich, 2017; Sowa & Meulenbroek, 2012). However, despite continual evidence of motor delays in this population, there has yet to be a shift to include motor skill interventions as standard care for children with ASD.

From a participation standpoint, these motor delays are concerning as they may lead to deficits in both physical activity and further skill development. Indeed, the developmental skill-learning gap hypothesis proposes that children lacking the movement skill necessary to engage in age-appropriate physical activity will withdraw from said activity as their skill level is no longer sufficient to keep-up with their peers; thus, greatly limiting any further opportunity for skill development (Wall, 2004). For children with ASD, this skill-learning gap may in fact extend to developmental areas beyond movement skill and into the social communicative skills that are also necessary for meaningful participation in play and physical activity with peers. Understanding the role of motor competence, and its association with other physical and behavioural health variables, may provide further insight into how motor competence influences (and is impacted by) physical activity in children with ASD.

Behavioural and Cognitive Well-being

There are many reasons for the high levels of physical inactivity seen in children with ASD, one of which may be that parents tend to prioritize behavioural interventions over physical activity programs (Gregor et al., 2018). Yet, physical activity may offer numerous behavioural and cognitive benefits, on top of the physical health benefits. For example, previous reviews have indicated improvements in the areas of repetitive and stereotyped behaviours, social-emotional functioning, and cognition for individuals with ASD (Bremer et al., 2016; Lang et al., 2010; Sowa & Meulenbroek, 2012; Tan et al., 2016). However, there is still little consensus on the type and dose of activity that is most beneficial for this population, along with the mechanisms that may be driving these behavioural changes. Much like the pertinent physical health variables, it is important to understand how behavioural and cognitive variables relate to participation in children with ASD. Indeed, it is possible that a certain level of behavioural or cognitive functioning is necessary for meaningful participation in physical activity. However, it is also likely that these variables can be improved through participation.

Repetitive or Stereotypic Behaviour

The majority of research that has examined the effect of physical activity on behavioural outcomes in children with ASD has focused on reductions in repetitive or stereotypic behaviours, typically through direct observation of participants immediately following a bout of activity (Bremer et al., 2016; Lang et al., 2010). For example, one of the first studies to examine this effect found an average reduction in stereotypic behaviours of approximately 30% following 8-10 minutes of jogging when compared to a typical academic session in five males with autism (Watters & Watters, 1980). Since this work, researchers have continued to find that stereotypic behaviours are reduced in children and adolescents with ASD following a brief session of jogging (e.g., Kern, Koegel, & Dunlap, 1984; Kern, Koegel, Dyer, Blew, & Fenton, 1982; Levinson & Reid, 1993; Rosenthal-Malek & Mitchell, 1997). While the results of this work are positive, the findings need to be considered in light of numerous limitations. For example, these studies included small samples, unstandardized outcome measures, and generally failed to characterize the intensity of the jogging activity. Further, participants were often chosen for these studies based on having a high level of stereotypic behaviours at baseline; thus, the effect of physical activity on children with mild stereotypies is unclear.

In contrast, Schmitz Olin and colleagues (2017) recently employed a within-subject experimental design to investigate the effect of exercise intensity and dose on reductions in stereotypic behaviours in adolescents (N=7) with ASD. The authors compared the effects of 10 and 20 minutes of both low- and high-intensity exercise on reductions in stereotypic behaviours in comparison to a sedentary control condition. They found reductions in these behaviours following all exercise conditions, with the exception of the 20-minute high-intensity condition, which resulted in an increase in stereotypic behaviours compared to the control condition. The authors also found that the 10-minute low-intensity condition resulted in the greatest reduction in behaviours across all time points (15-, 30-, 45-, and 60-minutes post-exercise) (Schmitz Olin et al., 2017).

One hypothesis for the reduction in stereotypic behaviour following physical activity has been that participants are too fatigued to engage in these negative behaviours. However, the literature would suggest that this is not the case as previous research has demonstrated improvements in positive behaviours (e.g., appropriate responding) following physical activity in children with ASD (e.g., Kern et al., 1982; Nicholson, Kehle, Bray, & Heest, 2011; Oriel, George, Peckus, & Semon, 2011; Rosenthal-Malek & Mitchell, 1997). If fatigue were causing a reduction in stereotypic behaviours, it should simultaneously cause a reduction in positive behavioural responses. Further, results from Schmitz Olin and colleagues' (2017) also help to rule out the fatigue hypothesis as they found that the highest intensity and dose of exercise actually resulted in an increase in stereotypic behaviours rather than a reduction. In all, these results suggest that physical activity can result in a decrease in stereotypic behaviours in children with ASD and that this reduction is likely not attributed to fatigue. It is therefore likely that other factors, such as cognition, play an important intermediary role in these behavioural changes.

Cognition

As noted above, physical activity has been shown to result in improved behavioural outcomes in children with ASD (Bremer et al., 2016), with these improvements possibly related to improved cognition. Indeed, previous research has also demonstrated a positive effect of physical activity on aspects of cognition, such as correct academic responding (e.g., answering questions in class) and time on-task, in this population (Bremer et al., 2016; Tan et al., 2016). Further, meta-analytic findings point to a significant, medium (r=.471), effect of exercise on cognition in individuals with ASD (Tan et al., 2016). However, the literature in this area has generally been limited by few studies (e.g., N=6 in Tan et al.'s 2016 meta-analysis), with a limited number of participants (median sample size of 10), and unstandardized outcome measures. Further, the research on physical activity and cognition to date in children with ASD has largely focused on behavioural outcomes (e.g., time on task), rather than more specific aspects or mechanisms of these cognitive effects. One aspect of cognition that warrants further attention is executive functioning, given the neural connectivity issues and subsequent executive dysfunction experienced by children with ASD (Demetriou et al., 2018; Wass, 2011).

Executive functions. Executive functions represent a number of higher-order cognitive processes including inhibition, working memory, and cognitive flexibility, that are necessary for purposeful, goal-directed behaviours (Anderson, 2001). A large body of literature has examined the effects of acute and chronic physical activity on executive functions in children and adolescents with typical development (Best, 2010; Diamond, 2015; Diamond & Ling, 2018; Gunnell et al., 2018; Hillman & Biggan, 2017; Raine et al., 2018). Research is still mixed in regard to the type(s) and duration(s) of exercise that may elicit the greatest improvements, in addition to which executive functions benefit the most; however, results generally suggest some improvement across aspects of executive functioning (Gunnell et al., 2018).

In their systematic review on the behavioural benefits of physical activity for children with ASD, Bremer and colleagues (2016) hypothesized that executive functions may be one mechanism driving these behavioural improvements. Despite this hypothesis, few studies have investigated this effect in children with ASD. Nevertheless, preliminary results are promising. For example, Anderson-Hanley and colleagues (2011) explored the effects of exergaming on aspects of executive functioning in two samples of 10-18 year old adolescents with ASD. First, in a within-subjects design, they found that participants (N=12) with ASD significantly improved in tests of working memory and inhibition, with large effect sizes, following a 20-minute bout of a dance-based exergame in comparison to a sedentary control condition (Anderson-Hanley et al., 2011). Second, they found that an additional 10 participants (8-21 years of age) significantly improved on a working memory task following a 20-minute bout of cyber-cycling when compared to the control condition from the first experiment (Anderson-Hanley et al., 2011). Although the evidence of exergaming for physical health benefits is mixed (Kari, 2014; LeBlanc et al., 2013), these findings point to improvements in executive functioning following an acute bout of activity for adolescents with ASD.

More recently, Pan et al. (2017) investigated the effect of a 12-week table tennis intervention on aspects of executive functioning (working memory and cognitive flexibility) in 6-12 year-old children with ASD (N=22). Following the intervention, participants in the experimental group (n=11) demonstrated significant improvements in their executive functions (η^2 range from 0.33-0.44) in comparison to control participants (n=11) (Pan et al., 2017). These findings suggest that improvements in executive functioning may be seen following a long-term physical activity intervention, although it is unknown whether these results could be replicated using other modes of physical activity. Lastly, Memari and colleagues (2017) examined the relationship between physical activity and cognitive flexibility in a sample of individuals 6-16 years of age with ASD (N=68). They found a significant, albeit weak, correlation between cognitive flexibility and physical activity whereby children with lower cognitive flexibility scores engaged in less physical activity (Memari et al., 2017). These findings are strengthened by a large sample size; however, they are limited by the cross-sectional design which limits our ability to make inferences regarding causality.

Taken together, the findings from these studies suggest that aspects of executive functioning may be related to physical activity in children and adolescents with ASD, and that both acute and chronic physical activity may elicit improvements to executive functions in this population.

Gaps and Limitations in the Literature

Children with ASD experience challenges in the areas of social communication and social interactions, along with the presence of restricted or repetitive behaviours and interests (American Psychiatric Association, 2013). These challenges can be attributed to widespread disruptions in neural connectivity (Just et al., 2007; Wass, 2011), which may have a particular impact on communication between brain regions with later developing regions, such as the prefrontal cortex, being the most affected (Just et al., 2007; Wass, 2011). As such, ASD is also considered a disorder of executive dysfunction (Demetriou et al., 2018; Hill, 2004b, 2004a), with deficits in executive functions attributed to the behavioural characteristics of the disorder. Yet, beyond these core challenges, children with ASD also experience multiple physical health deficits including low levels of motor competence (Fournier et al., 2010), low fitness (Bricout et al., 2018; Pan, 2014), and a high prevalence of overweight and obesity (Healy, Aigner, et al., 2018). It is plausible that all of these factors – from behavioural to physical health variables – are related to the low levels of physical activity evident in this population (Jones et al., 2017). More importantly, it is possible that we may be able to harness physical activity to elicit improvements in the behavioural functioning of children with ASD, in addition to their physical health. In order to effectively use physical activity as a behavioural intervention, we must first address many of the fundamental gaps in the literature.

First, while we know that children with ASD experience many physical and behavioural health deficits, we do not have a good understanding of how these variables (e.g., motor skills, fitness, weight status, behavioural functioning) are related to participation in physical activity in this population. While we can draw from the literature on children with typical development, using models such as Stodden's (2008), we do not know if these variables have the same effect on participation in children with ASD. Moreover, there may be other, more ASD-specific (e.g., social communicative skills), factors that influence these relationships. Therefore, it is important to understand how common correlates and outcomes of physical activity are related to one another in children with ASD, along with the role of other variables (e.g., behavioural functioning) that may be more pertinent to the population, in order to design effective physical activity interventions.

Second, in order to be confident in our assessments of the variables associated with physical activity, we need to have knowledge regarding the measurement properties of assessments and measures used in this population. While many of these variables, such as motor competence, have reliable and valid standardized assessments, the reliability of fitness assessments has not been explored for children with ASD. This is a critical gap in the literature as we cannot assume that fitness assessments used for other populations can simply be transferred to children with ASD. Moreover, beyond measurement properties we must also understand which fitness assessments, and types of physical activity, are feasible to implement in children with ASD.

Third, while there is evidence that physical activity can elicit behavioural improvements in children with ASD (Bremer et al., 2016), there is limited understanding of what is driving these effects. It is plausible that improvements to executive functions may be the key factor in these behavioural improvements; yet, the effect of physical activity on executive functions has not been sufficiently tested in children with ASD.

Finally, the extant literature has generally approached the topic of physical (in)activity in children with ASD in a silo: studies tend to focus on physical activity levels alone, or measured alongside a few other variables without testing associations. Further, while some physical activity-based interventions have measured behavioural outcomes, the studies have often been limited by a lack of controls, unstandardized measures, and poor characterization of the activity itself. It is important that we take a more comprehensive approach when exploring the role of physical activity for children with ASD to ensure that we understand how physical, behavioural, and cognitive variables both influence and change with participation.

Interdisciplinary Framework of Physical Activity

In light of the gaps and limitations in the literature and the need to take a more comprehensive approach to the study of physical activity in children with ASD, we propose here an interdisciplinary framework of physical activity correlates and outcomes (Figure 1). This framework seeks to connect work in the area of motor development and exercise psychology in order to advance our understanding of the role of physical activity for children with ASD. In brief, the framework proposes reciprocal relationships between motor competence, health-related fitness, and physical activity. Further, we propose that adaptive behaviour is an outcome of physical activity and also reciprocally related to motor competence. Lastly, we position cognition as both an outcome of physical activity and related to both adaptive behaviour and health-related fitness. While aspects of this framework have been presented before (e.g., Stodden et al., 2008), the specific inclusion of adaptive behaviour makes it relevant to the study of ASD and indeed, all children with neurodevelopmental disorders. In this sense, the framework is novel with regard to the inclusion of this domain.

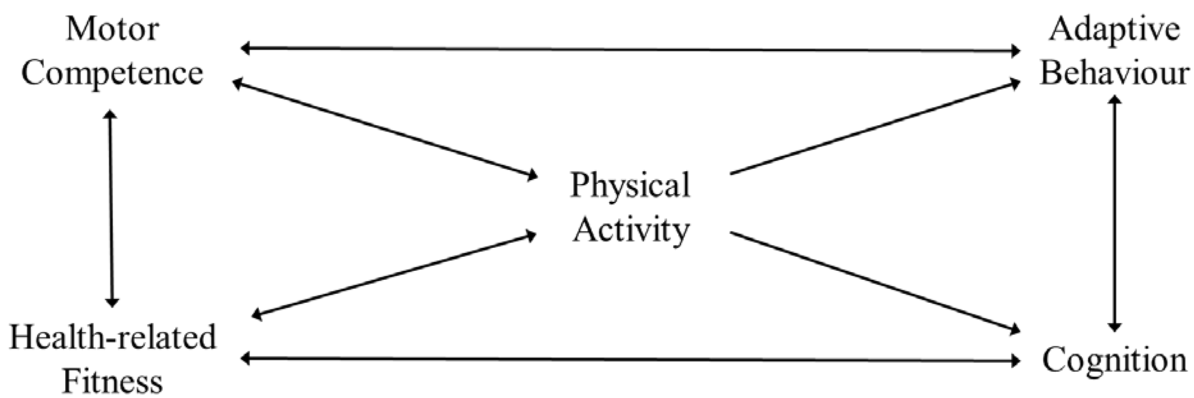

Figure 1. Proposed interdisciplinary framework of physical activity correlates and outcomes.

General Purpose of Dissertation

Guided by the literature and our proposed framework of physical activity correlates and outcomes, the objective of this dissertation was to explore the role of physical activity with the overarching intent of understanding how physical activity may be used as a behavioural intervention for children with ASD. While the studies included in this dissertation were joined by a common interest in understanding the role of physical activity for children with ASD, they were not designed as a sequential set of studies. Rather, given the general paucity of research in this area to-date, this dissertation set out to address these gaps through a set of studies designed to build the necessary foundational knowledge in this area. These studies were driven by theoretical considerations, grounded in the study of motor development, and identified through a review of correlates and outcomes of physical activity that are contextually relevant for children with ASD.

Specific Objectives

The specific objectives of the studies in this dissertation were:

1) To investigate the test-retest reliability and feasibility of select fitness assessments for use with children with ASD.
2) To examine the relationship between motor competence and adaptive behaviour in this population.
3) To examine the associations between motor competence, physical activity, and health-related fitness, and the role of adaptive behaviour in these associations in children with ASD.
4) To investigate the acute effect of exercise on executive functioning in children with ASD.

Specific Hypotheses

The specific hypotheses of the studies in this dissertation were:

1) Fitness assessments would be feasible and reliable to implement for children with ASD, with reliability being highest in assessments that were relatively short in duration and required limited instructions.
2) Motor competence and adaptive behaviour would be positively associated in children with ASD.
3) Motor competence, physical activity, and health-related fitness would be positively associated with one another, and adaptive behaviour would moderate these associations in children with ASD.
4) An acute bout of exercise would result in significant improvements to aspects of executive functioning, when compared to a control condition, in children with ASD.

References

American Psychiatric Association. (2013). *Diagnostic and Statistical Manual of Mental Disorders (DSM-5)* (5th ed.). Washington, DC: American Psychiatric Publishing.

Anagnostou, E., & Taylor, M. J. (2011). Review of neuroimaging in autism spectrum disorders: What have we learned and where we go from here. *Molecular Autism*, *2*(1), 4. https://doi.org/10.1186/2040-2392-2-4

Anderson, V. (2001). Assessing executive functions in children: Biological, psychological, and developmental considerations. *Pediatric Rehabilitation*, *4*(3), 119–136. https://doi.org/10.1080/13638490110091347

Anderson-Hanley, C., Tureck, K., & Schneiderman, R. L. (2011). Autism and exergaming: Effects on repetitive behaviors and cognition. *Psychology Research and Behavior Management*, *4*(1), 129-137. https://doi.org/10.2147/PRBM.S24016

Arnell, S., Jerlinder, K., & Lundqvist, L.-O. (2017). Perceptions of physical activity participation among adolescents with autism spectrum disorders: A conceptual model of conditional participation. *Journal of Autism and Developmental Disorders*. https://doi.org/10.1007/s10803-017-3436-2

Balogun, F. (2016). Prevalence and correlates of obesity in childhood autism spectrum disorders: A literature review. *Journal of Psychiatry*, *19*(5). https://doi.org/10.4172/2378-5756.1000385

Bandini, L. G., Gleason, J., Curtin, C., Lividini, K., Anderson, S. E., Cermak, S. A., … Must, A. (2013). Comparison of physical activity between children with autism spectrum disorders and typically developing children. *Autism*, *17*(1), 44–54. https://doi.org/10.1177/1362361312437416

Belmonte, M. K. (2004). Autism and abnormal development of brain connectivity. *Journal of Neuroscience*, *24*(42), 9228–9231. https://doi.org/10.1523/JNEUROSCI.3340-04.2004

Ben-Sasson, A., Hen, L., Fluss, R., Cermak, S. A., Engel-Yeger, B., & Gal, E. (2009). A meta-analysis of sensory modulation symptoms in individuals with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *39*(1), 1–11. https://doi.org/10.1007/s10803-008-0593-3

Best, J. R. (2010). Effects of physical activity on children’s executive function: Contributions of experimental research on aerobic exercise. *Developmental Review*, *30*(4), 331–351. https://doi.org/10.1016/j.dr.2010.08.001

Bhat, A.N., Galloway, J. C., & Landa, R. J. (2012). Relation between early motor delay and later communication delay in infants at risk for autism. *Infant Behavior and Development*, *35*(4), 838–846. https://doi.org/10.1016/j.infbeh.2012.07.019

Bhat, A.N., Landa, R. J., & Galloway, J. C. (2011). Current perspectives on motor functioning in infants, children, and adults with autism spectrum disorders. *Physical Therapy*, *91*(7), 1116-1129.

Borremans, E., Rintala, P., & McCubbin, J. A. (2010). Physical fitness and physical activity in adolescents with Asperger syndrome: A comparative study. *Adapted Physical Activity Quarterly*, *27*(4), 308–320.

Bremer, E., Balogh, R., & Lloyd, M. (2015). Effectiveness of a fundamental motor skill intervention for 4-year-old children with autism spectrum disorder: A pilot study. *Autism*, *19*(8), 980–991. https://doi.org/10.1177/1362361314557548

Bremer, E., Crozier, M., & Lloyd, M. (2016). A systematic review of the behavioural outcomes following exercise interventions for children and youth with autism spectrum disorder. *Autism*, *20*(8), 899–915. https://doi.org/10.1177/1362361315616002

Bremer, E., & Lloyd, M. (2016). School-based fundamental-motor-skill intervention for children with Autism-like characteristics: An exploratory study. *Adapted Physical Activity Quarterly*, *33*(1), 66–88. https://doi.org/10.1123/APAQ.2015-0009

Bricout, V.-A., Pace, M., Dumortier, L., Baillieul, F., Favre-Juvin, A., & Guinot, M. (2018). Reduced cardiorespiratory capacity in children with autism spectrum disorders. *Journal of Clinical Medicine*, *7*(10), 361. https://doi.org/10.3390/jcm7100361

Brown, R. I., MacAdam–Crisp, J., Wang, M., & Iarocci, G. (2006). Family quality of life when there is a child with a developmental disability. *Journal of Policy and Practice in Intellectual Disabilities*, *3*(4), 238–245. https://doi.org/10.1111/j.1741-1130.2006.00085.x

Buescher, A., Cidav, Z., Knapp, M., & Mandell, D. (2014). Costs of autism spectrum disorders in the United Kingdom and the United States. *JAMA Pediatrics*, *168*(8), 721–728. https://doi.org/10.1001/jamapediatrics.2014.210

Cairney, J. (2015). *Developmental Coordination Disorder and its Consequences*. Toronto, ON: University of Toronto Press.

Chasson, G. S., Harris, G. E., & Neely, W. J. (2007). Cost comparison of early intensive behavioral intervention and special education for children with autism. *Journal of Child and Family Studies*, *16*(3), 401–413. https://doi.org/10.1007/s10826-006-9094-1

Corbin, C. B., Pangrazi, R. P., & Franks, B. D. (2000). *Definitions: Health, Fitness, and Physical Activity*. Washington, DC: President's Council on Physical Fitness and Sports,. Retrieved from https://files.eric.ed.gov/fulltext/ED470696.pdf

Corsello, C. M. (2005). Early intervention in autism. *Infants & Young Children*, *18*(2), 74–85.

Curtin, C., Jojic, M., & Bandini, L. G. (2014). Obesity in children with autism spectrum disorder: *Harvard Review of Psychiatry*, *22*(2), 93–103. https://doi.org/10.1097/HRP.0000000000000031

Dale, L. P., Vanderloo, L., Moore, S., & Faulkner, G. (2018). Physical activity and depression, anxiety, and self-esteem in children and youth: An umbrella systematic review. *Mental Health and Physical Activity*. https://doi.org/10.1016/j.mhpa.2018.12.001

Demetriou, E. A., Lampit, A., Quintana, D. S., Naismith, S. L., Song, Y. J. C., Pye, J. E., … Guastella, A. J. (2018). Autism spectrum disorders: A meta-analysis of executive function. *Molecular Psychiatry*, *23*(5), 1198–1204. https://doi.org/10.1038/mp.2017.75

Diamond, A. (2013). Executive functions. *Annual Review of Psychology*, *64*(1), 135–168. https://doi.org/10.1146/annurev-psych-113011-143750

Diamond, A. (2015). Effects of physical exercise on executive functions: Going beyond simply moving to moving with thought. *Annals of Sports Medicine and Research*, *2*(1), 1011.

Diamond, A., & Ling, D. S. (2018). Aerobic-Exercise and resistance-training interventions have been among the least effective ways to improve executive functions of any method tried thus far. *Developmental Cognitive Neuroscience*. https://doi.org/10.1016/j.dcn.2018.05.001

Flygare Wallén, E., Ljunggren, G., Carlsson, A. C., Pettersson, D., & Wändell, P. (2017). High prevalence of diabetes mellitus, hypertension and obesity among persons with a recorded diagnosis of intellectual disability or autism spectrum disorder: Diabetes and intellectual disability or ASD. *Journal of Intellectual Disability Research*. https://doi.org/10.1111/jir.12462

Fournier, K. A., Hass, C. J., Naik, S. K., Lodha, N., & Cauraugh, J. H. (2010). Motor coordination in autism spectrum disorders: A synthesis and meta-analysis. *Journal of Autism and Developmental Disorders*, *40*(10), 1227–1240. https://doi.org/10.1007/s10803-010-0981-3

Garcia-Pastor, T., Salinero, J. J., Theirs, C. I., & Ruiz-Vicente, D. (2018). Obesity status and physical activity level in children and adults with autism spectrum disorders: A pilot study. *Journal of Autism and Developmental Disorders*. https://doi.org/10.1007/s10803-018-3692-9

Gillberg, C., & Fernell, E. (2014). Autism plus versus autism pure. *Journal of Autism and Developmental Disorders*, *44*(12), 3274–3276. https://doi.org/10.1007/s10803-014-2163-1

Gregor, S., Bruni, N., Grkinic, P., Schwartz, L., McDonald, A., Thille, P., … Jachyra, P. (2018). Parents' perspectives of physical activity participation among Canadian adolescents with autism spectrum disorder. *Research in Autism Spectrum Disorders*, *48*, 53–62. https://doi.org/10.1016/j.rasd.2018.01.007

Gunnell, K. E., Poitras, V. J., LeBlanc, A., Schibli, K., Barbeau, K., Hedayati, N., … Tremblay, M. S. (2018). Physical activity and brain structure, brain function, and cognition in children and youth: A systematic review of randomized controlled trials. *Mental Health and Physical Activity*. https://doi.org/10.1016/j.mhpa.2018.11.002

Healy, S., Aigner, C. J., & Haegele, J. A. (2018). Prevalence of overweight and obesity among US youth with autism spectrum disorder. *Autism*, 1–5. https://doi.org/10.1177/1362361318791817

Healy, S., & Garcia, J. M. (2018). Psychosocial correlates of physical activity participation and screen-time in typically developing children and children on the autism spectrum. *Journal of Developmental and Physical Disabilities*. https://doi.org/10.1007/s10882-018-9642-9

Healy, S., Garcia, J. M., & Haegele, J. A. (2018). Environmental factors associated with physical activity and screen time among children with and without autism spectrum disorder. *Journal of Autism and Developmental Disorders*. https://doi.org/10.1007/s10803-018-3818-0

Healy, S., Haegele, J. A., Grenier, M., & Garcia, J. M. (2017). Physical activity, screen-time behavior, and obesity among 13-year olds in Ireland with and without autism spectrum disorder. *Journal of Autism and Developmental Disorders*, *47*(1), 49–57. https://doi.org/10.1007/s10803-016-2920-4

Healy, S., Nacario, A., Braithwaite, R. E., & Hopper, C. (2018). The effect of physical activity interventions on youth with autism spectrum disorder: A meta-analysis. *Autism Research*, *11*(6), 818–833. https://doi.org/10.1002/aur.1955

Healy, S., Pacanowski, C. R., & Williams, E. (2018). Weight management interventions for youth with autism spectrum disorder: A systematic review. *International Journal of Obesity*. https://doi.org/10.1038/s41366-018-0233-8

Hill, E. L. (2004a). Evaluating the theory of executive dysfunction in autism. *Developmental Review*, *24*(2), 189–233. https://doi.org/10.1016/j.dr.2004.01.001

Hill, E. L. (2004b). Executive dysfunction in autism. *Trends in Cognitive Sciences*, *8*(1), 26–32.

Hillman, C. H., & Biggan, J. R. (2017). A review of childhood physical activity, brain, and cognition: Perspectives on the future. *Pediatric Exercise Science*, *29*(2), 170–176.

Hirata, S., Okuzumi, H., Kitajima, Y., Hosobuchi, T., Nakai, A., & Kokubun, M. (2014). Relationship between motor skill and social impairment in children with autism spectrum disorders. *International Journal of Developmental Disabilities*, *60*(4), 251–256. https://doi.org/10.1179/2047387713Y.0000000033

Hirvikoski, T., Mittendorfer-Rutz, E., Boman, M., Larsson, H., Lichtenstein, P., & Bölte, S. (2016). Premature mortality in autism spectrum disorder. *British Journal of Psychiatry*, *208*(03), 232–238. https://doi.org/10.1192/bjp.bp.114.160192

Horlin, C., Falkmer, M., Parsons, R., Albrecht, M. A., & Falkmer, T. (2014). The cost of autism spectrum disorders. *PLoS ONE*, *9*(9), e106552. https://doi.org/10.1371/journal.pone.0106552

Janssen, I., & LeBlanc, A. G. (2010). Systematic review of the health benefits of physical activity and fitness in school-aged children and youth. *International Journal of Behavioral Nutrition and Physical Activity*, *7*:40. https://doi.org/10.1186/1479-5868-7-40

Jones, R. A., Downing, K., Rinehart, N. J., Barnett, L. M., May, T., McGillivray, J. A., … Hinkley, T. (2017). Physical activity, sedentary behavior and their correlates in children with autism spectrum disorder: A systematic review. *PLOS ONE*, *12*(2), e0172482. https://doi.org/10.1371/journal.pone.0172482

Jones, S., Bremer, E., & Lloyd, M. (2017). Autism spectrum disorder: Family quality of life while waiting for intervention services. *Quality of Life Research*, *26*(2), 331–342. https://doi.org/10.1007/s11136-016-1382-7

Just, M. A. (2004). Cortical activation and synchronization during sentence comprehension in high-functioning autism: Evidence of underconnectivity. *Brain*, *127*(8), 1811–1821. https://doi.org/10.1093/brain/awh199

Just, M. A., Cherkassky, V. L., Keller, T. A., Kana, R. K., & Minshew, N. J. (2007). Functional and anatomical cortical underconnectivity in autism: Evidence from an fMRI study of an executive function task and corpus callosum morphometry. *Cerebral Cortex*, *17*(4), 951–961. https://doi.org/10.1093/cercor/bhl006

Kanne, S. M., Gerber, A. J., Quirmbach, L. M., Sparrow, S. S., Cicchetti, D. V., & Saulnier, C. A. (2011). The role of adaptive behavior in autism spectrum disorders: Implications for functional outcome. *Journal of Autism and Developmental Disorders*, *41*(8), 1007–1018. https://doi.org/10.1007/s10803-010-1126-4

Kanner, L. (1943). Autistic disturbances of affective contact. *Nervous Child*, *2*(3), 217–250.

Kanner, L. (1971). Follow-up study of eleven autistic children originally reported in 1943. *Journal of Autism and Childhood Schizophrenia*, *1*(2), 119–145.

Kari, T. (2014). Can exergaming promote physical fitness and physical activity? A systematic review of systematic reviews. *International Journal of Gaming and Computer-Mediated Simulations*, *6*(4). https://doi.org/10.4018/ijgcms.2014100105

Kern, L., Koegel, R. L., & Dunlap, G. (1984). The influence of vigorous versus mild exercise on autistic stereotyped behaviors. *Journal of Autism and Developmental Disorders*, *14*(1), 57–67. https://doi.org/10.1007/BF02408555

Kern, L., Koegel, R. L., Dyer, K., Blew, P. A., & Fenton, L. R. (1982). The effects of physical exercise on self-stimulation and appropriate responding in autistic children. *Journal of Autism and Developmental Disorders*, *12*(4), 399–419. https://doi.org/10.1007/BF01538327

Ketcheson, L., Hauck, J. L., & Ulrich, D. (2018). The levels of physical activity and motor skills in young children with and without autism spectrum disorder, aged 2–5 years. *Autism*, *22*(4), 414-223. https://doi.org/10.1177/1362361316683889.

Ketcheson, L., Hauck, J., & Ulrich, D. (2017). The effects of an early motor skill intervention on motor skills, levels of physical activity, and socialization in young children with autism spectrum disorder: A pilot study. *Autism*, *21*(4), 481–492. https://doi.org/10.1177/1362361316650611

King-Dowling, S., Rodriguez, C., Missiuna, C., Timmons, B. W., & Cairney, J. (2018). Health-related fitness in preschool children with and without motor delays. *Medicine & Science in Sports & Exercise*. https://doi.org/10.1249/MSS.0000000000001590

Lang, R., Koegel, L. K., Ashbaugh, K., Regester, A., Ence, W., & Smith, W. (2010). Physical exercise and individuals with autism spectrum disorders: A systematic review. *Research in Autism Spectrum Disorders*, *4*(4), 565–576. https://doi.org/10.1016/j.rasd.2010.01.006

LeBarton, E. S., & Iverson, J. M. (2013). Fine motor skill predicts expressive language in infant siblings of children with autism. *Developmental Science, 16*(6), 815-827. https://doi.org/10.1111/desc.12069

LeBarton, E. S., & Landa, R. J. (2019). Infant motor skill predicts later expressive language and autism spectrum disorder diagnosis. *Infant Behavior and Development*, *54*, 37–47. https://doi.org/10.1016/j.infbeh.2018.11.003

LeBlanc A.G., Chaput J.-P., McFarlane A., Colley R.C., Thivel D., Biddle S.J.H., et al. (2013) Active video games and health indicators in children and youth: A systematic review. *PLoS ONE, 8*(6): e65351. https://doi.org/10.1371/journal.pone.0065351

Lee, I.-M., Shiroma, E. J., Lobelo, F., Puska, P., Blair, S. N., & Katzmarzyk, P. T. (2012). Effect of physical inactivity on major non-communicable diseases worldwide: An analysis of burden of disease and life expectancy. *The Lancet*, *380*(9838), 219–229. https://doi.org/10.1016/S0140-6736(12)61031-9

Leonard, H. C., & Hill, E. L. (2014). Review: The impact of motor development on typical and atypical social cognition and language: A systematic review. *Child and Adolescent Mental Health*, *19*(3), 163-170. https://doi.org/10.1111/camh.12055

Levinson, L. J., & Reid, G. (1993). The effects of exercise intensity on the stereotypic behaviors of individuals with autism. *Adapted Physical Activity Quarterly*, *10*(3), 255–268. https://doi.org/10.1123/apaq.10.3.255

Levy, S. E., Pinto-Martin, J. A., Bradley, C. B., Chittams, J., Johnson, S. L., Pandey, J., … Kral, T. V. E. (2018). Relationship of weight outcomes, co-occurring conditions, and severity of autism spectrum disorder in the study to explore early development. *The Journal of Pediatrics*. https://doi.org/10.1016/j.jpeds.2018.09.003

Lloyd, M., MacDonald, M., & Lord, C. (2013). Motor skills of toddlers with autism spectrum disorders. *Autism*, *17*(2), 133–146. https://doi.org/10.1177/1362361311402230

Lubans, D. R., Morgan, P. J., Cliff, D. P., Barnett, L. M., & Okely, A. D. (2010). Fundamental movement skills in children and adolescents. *Sports Medicine*, *40*(12), 1019–1035.

Lubans, D. R., Richards, J., Hillman, C., Faulkner, G., Beauchamp, M., Nilsson, M., … Biddle, S. (2016). Physical activity for cognitive and mental health in youth: A systematic review of mechanisms. *PEDIATRICS*, *138*(3), e20161642–e20161642. https://doi.org/10.1542/peds.2016-1642

MacDonald, M., Lord, C., & Ulrich, D. (2013a). The relationship of motor skills and adaptive behavior skills in young children with autism spectrum disorders. *Research in Autism Spectrum Disorders*, *7*(11), 1383–1390. https://doi.org/10.1016/j.rasd.2013.07.020

MacDonald, M., Lord, C., & Ulrich, D. A. (2013b). The relationship of motor skills and social communicative skills in school-aged children with autism spectrum disorder. *Adapted Physical Activity Quarterly*, *30*(3), 271–282.

MacDonald, M., Lord, C., & Ulrich, D. A. (2014). Motor skills and calibrated autism severity in young children with autism spectrum disorder. *Adapted Physical Activity Quarterly*, *31*(2), 95–105. https://doi.org/10.1123/apaq.2013-0068

Matson, J. L. (Ed.). (2016). *Comorbid Conditions Among Children with Autism Spectrum Disorders*. Switzerland: Springer International Publishing. https://doi.org/10.1007/978-3-319-19183-6

McCoy, S. M., Jakicic, J. M., & Gibbs, B. B. (2016). Comparison of obesity, physical activity, and sedentary behaviors between adolescents with autism spectrum disorders and without. *Journal of Autism and Developmental Disorders*, *46*(7), 2317–2326. https://doi.org/10.1007/s10803-016-2762-0

Memari, A. H., Mirfazeli, F. S., Kordi, R., Shayestehfar, M., Moshayedi, P., & Mansournia, M. A. (2017). Cognitive and social functioning are connected to physical activity behavior in children with autism spectrum disorder. *Research in Autism Spectrum Disorders*, *33*, 21–28. https://doi.org/10.1016/j.rasd.2016.10.001

Metcalf, B., Henley, W., & Wilkin, T. (2012). Effectiveness of intervention on physical activity of children: Systematic review and meta-analysis of controlled trials with objectively measured outcomes. *BMJ*, *345*(sep27 1), e5888–e5888. https://doi.org/10.1136/bmj.e5888

Muskens, J. B., Velders, F. P., & Staal, W. G. (2017). Medical comorbidities in children and adolescents with autism spectrum disorders and attention deficit hyperactivity disorders: A systematic review. *European Child & Adolescent Psychiatry*. https://doi.org/10.1007/s00787-017-1020-0

Myers, S. M., & Johnson, C. P. (2007). Management of Children With Autism Spectrum Disorders. *PEDIATRICS*, *120*(5), 1162–1182. https://doi.org/10.1542/peds.2007-2362

National Autism Spectrum Disorder Surveillance System. (2018). *Autism Spectrum Disorder among Children and Youth in Canada 2018: A report of the National Autism Spectrum Disorder Surveillance System*. Ottawa, ON: Public Health Agency of Canada. Retrieved from https://www.canada.ca/content/dam/phac-aspc/documents/services/publications/diseases-conditions/autism-spectrum-disorder-children-youth-canada-2018/autism-spectrum-disorder-children-youth-canada-2018.pdf

Nicholson, H., Kehle, T. J., Bray, M. A., & Heest, J. V. (2011). The effects of antecedent physical activity on the academic engagement of children with autism spectrum disorder. *Psychology in the Schools*, *48*(2), 198–213.

Oriel, K. N., George, C. L., Peckus, R., & Semon, A. (2011). The effects of aerobic exercise on academic engagement in young children with autism spectrum disorder. *Pediatric Physical Therapy*, *23*(2), 187–193. https://doi.org/10.1097/PEP.0b013e318218f149

Pan, C.-Y. (2014). Motor proficiency and physical fitness in adolescent males with and without autism spectrum disorders. *Autism*, *18*(2), 156–165. https://doi.org/10.1177/1362361312458597

Pan, C.-Y., Chu, C.-H., Tsai, C.-L., Sung, M.-C., Huang, C.-Y., & Ma, W.-Y. (2017). The impacts of physical activity intervention on physical and cognitive outcomes in children with autism spectrum disorder. *Autism*, *21*(2), 190–202. https://doi.org/10.1177/1362361316633562

Payne, V., & Isaacs, L. (2011). *Human Motor Development: A Lifespan Approach* (8th ed.). New York, NY: McGraw-Hill.

Piccininni, C., Bisnaire, L., & Penner, M. (2017). Cost-effectiveness of wait time reduction for intensive behavioral intervention services in Ontario, Canada. *JAMA Pediatrics*, *171*(1), 23. https://doi.org/10.1001/jamapediatrics.2016.2695

Poitras, V. J., Gray, C. E., Borghese, M. M., Carson, V., Chaput, J.-P., Janssen, I., … Tremblay, M. S. (2016). Systematic review of the relationships between objectively measured physical activity and health indicators in school-aged children and youth. *Applied Physiology, Nutrition, and Metabolism*, *41*(6 (Suppl. 3)), S197–S239. https://doi.org/10.1139/apnm-2015-0663

Raine, L. B., Kao, S.-C., Pindus, D., Westfall, D. R., Shigeta, T. T., Logan, N., … Hillman, C. H. (2018). A large-scale reanalysis of childhood fitness and inhibitory control. *Journal of Cognitive Enhancement*. https://doi.org/10.1007/s41465-018-0070-7

Ratcliff, K., Hong, I., & Hilton, C. (2018). Leisure participation patterns for school age youth with autism spectrum disorders: Findings from the 2016 National Survey of Children's Health. *Journal of Autism and Developmental Disorders*. https://doi.org/10.1007/s10803-018-3643-5

Rimmer, J. A., & Rowland, J. L. (2008). Physical activity for youth with disabilities: A critical need in an underserved population. *Developmental Neurorehabilitation*, *11*(2), 141–148. https://doi.org/10.1080/17518420701688649

Rivilis, I., Hay, J., Cairney, J., Klentrou, P., Liu, J., & Faught, B. E. (2011). Physical activity and fitness in children with developmental coordination disorder: A systematic review. *Research in Developmental Disabilities*, *32*(3), 894–910. https://doi.org/10.1016/j.ridd.2011.01.017

Roberts, K. C., Yao, X., Carson, V., Chaput, J.-P., Janssen, I., & Tremblay, M. S. (2017). Meeting the Canadian 24-Hour Movement Guidelines for Children and Youth. *Health Reports*, *28*(10), 3–7.

Robinson, L. E., Stodden, D. F., Barnett, L. M., Lopes, V. P., Logan, S. W., Rodrigues, L. P., & D'Hondt, E. (2015). Motor competence and its effect on positive developmental trajectories of health. *Sports Medicine*, *45*(9), 1273–1284. https://doi.org/10.1007/s40279-015-0351-6

Rosenthal-Malek, A., & Mitchell, S. (1997). Brief report: The effects of exercise on the self-stimulatory behaviors and positive responding of adolescents with autism, *27*(2), 193–202.

Russo, N., Flanagan, T., Iarocci, G., Berringer, D., Zelazo, P. D., & Burack, J. A. (2007). Deconstructing executive deficits among persons with autism: Implications for cognitive neuroscience. *Brain and Cognition*, *65*(1), 77–86. https://doi.org/10.1016/j.bandc.2006.04.007

Schipul, S. E., Keller, T. A., & Just, M. A. (2011). Inter-regional brain communication and its disturbance in autism. *Frontiers in Systems Neuroscience, 5*:10. https://doi.org/10.3389/fnsys.2011.00010

Schmitz Olin, S., Mcfadden, B. A., Golem, D. L., Pellegrino, J. K., Walker, A. J., Sanders, D. J., & Arent, S. M. (2017). The effects of exercise dose on stereotypical behavior in children with autism: *Medicine & Science in Sports & Exercise*, *49*(5), 983–990. https://doi.org/10.1249/MSS.0000000000001197

Sipes, M., Matson, J. L., & Horovitz, M. (2011). Autism spectrum disorders and motor skills: The effect on socialization as measured by the Baby and Infant Screen for Children with aUtIsm Traits (BISCUIT). *Developmental Neurorehabilitation*, *14*(5), 290–296. https://doi.org/10.3109/17518423.2011.587838

Sowa, M., & Meulenbroek, R. (2012). Effects of physical exercise on autism spectrum disorders: A meta-analysis. *Research in Autism Spectrum Disorders*, *6*(1), 46–57. https://doi.org/10.1016/j.rasd.2011.09.001

Stanish, H. I., Curtin, C., Must, A., Phillips, S., Maslin, M., & Bandini, L. G. (2017). Physical activity levels, frequency, and type among adolescents with and without autism spectrum disorder. *Journal of Autism and Developmental Disorders*, *47*(3), 785–794. https://doi.org/10.1007/s10803-016-3001-4

Staples, K. L., & Reid, G. (2010). Fundamental movement skills and autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *40*(2), 209–217. https://doi.org/10.1007/s10803-009-0854-9

Stodden, D. F., Goodway, J. D., Langendorfer, S. J., Roberton, M. A., Rudisill, M. E., Garcia, C., & Garcia, L. E. (2008). A developmental perspective on the role of motor skill competence in physical activity: An emergent relationship. *Quest*, *60*(2), 290–306.

Tan, B. W. Z., Pooley, J. A., & Speelman, C. P. (2016). A meta-analytic review of the efficacy of physical exercise interventions on cognition in individuals with autism spectrum disorder and ADHD. *Journal of Autism and Developmental Disorders*, *46*(9), 3126–3143. https://doi.org/10.1007/s10803-016-2854-x

Tremblay, M. S., Carson, V., Chaput, J.-P., Connor Gorber, S., Dinh, T., Duggan, M., … Zehr, L. (2016). Canadian 24-hour movement guidelines for children and youth: An integration of physical activity, sedentary behaviour, and sleep. *Applied Physiology, Nutrition, and Metabolism*, *41*(6 (Suppl. 3)), S311–S327. https://doi.org/10.1139/apnm-2016-0151

Tyler, K., MacDonald, M., & Menear, K. (2014). Physical activity and physical fitness of school-aged children and youth with autism spectrum disorders. *Autism Research and Treatment*. https://doi.org/10.1155/2014/312163

Virués-Ortega, J. (2010). Applied behavior analytic intervention for autism in early childhood: Meta-analysis, meta-regression and dose–response meta-analysis of multiple outcomes. *Clinical Psychology Review*, *30*(4), 387–399. https://doi.org/10.1016/j.cpr.2010.01.008

Wall, A. E. T. (2004). The developmental skill-learning gap hypothesis: Implications for children with movement difficulties. *Adapted Physical Activity Quarterly*, *21*(3), 197–218. https://doi.org/10.1123/apaq.21.3.197

Wass, S. (2011). Distortions and disconnections: Disrupted brain connectivity in autism. *Brain and Cognition*, *75*(1), 18–28. https://doi.org/10.1016/j.bandc.2010.10.005

Watters, R. G., & Watters, W. E. (1980). Decreasing self-stimulatory behavior with physical exercise in a group of autistic boys. *Journal of Autism and Developmental Disorders*, *10*(4), 379–387. https://doi.org/10.1007/BF02414814

Wong, C., Odom, S. L., Hume, K. A., Cox, A. W., Fettig, A., Kucharczyk, S., … Schultz, T. R. (2015). Evidence-based practices for children, youth, and young adults with autism spectrum disorder: A comprehensive review. *Journal of Autism and Developmental Disorders, 45*(7), 1951–1966. https://doi.org/10.1007/s10803-014-2351-z

CHAPTER 2: Reliable and feasible fitness testing for children on the autism spectrum

Preamble

Reliable and feasible fitness testing for children on the autism spectrum is the first study in the dissertation. The study examines the test-retest reliability and feasibility of select fitness assessments for 7-12 year old children with autism spectrum disorder.

The following manuscript is currently under review (revisions submitted) at *Research Quarterly for Exercise and Sport*. The word document version of the manuscript (formatted according to *Research Quarterly for Exercise and Sport* author guidelines) is included in the dissertation.

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<u>Contribution of Study 1 to overall dissertation:</u>
Study 1 provides the first evidence of critical measurement properties of common fitness assessments for use with children with ASD. Findings from Study 1 indicate that tests that are short in duration and require minimal instruction demonstrate the greatest test-retest reliability and feasibility. These results provide the necessary foundation to move forward with assessments of health-related fitness in children with ASD.

Abstract

Purpose: This study examined the test-retest reliability and feasibility of select fitness assessments in 7-12 year old children on the autism spectrum.

Method: Participants (N=14; n=1 female; Mage = 9.5 ± 1.7 years) completed 7 fitness assessments, administered in a random order, on two occasions: Bruce protocol; Modified 6-minute walk test (M6MWT); Wingate anaerobic cycling test; muscle power sprint test (MPST); sit & reach; standing long jump; and grip strength. Intraclass correlations (two-way mixed with absolute agreement) were computed to examine test-retest reliability. Feasibility was assessed by questionnaire following the first administration of each test.

Results: The Wingate (ICC = .956), standing long jump (ICC = .925), grip strength (ICC = .913), and sit and reach (ICC = .829) tests demonstrated good- to- excellent reliability, while the Bruce protocol (ICC = .811), M6MWT (ICC = .510), and MPST (ICC = .703) demonstrated moderate- to- good reliability based on the 95% confidence intervals of the ICC. All tests demonstrated assessor-rated feasibility scores of 70/100 or higher and child-rated feasibility scores of 66/100 or higher.

Conclusion: The results demonstrate moderate- to excellent test-retest reliability for select fitness tests. Short, single-instruction (e.g., standing long jump) tests may be more reliable than lengthier assessments (e.g., M6MWT) in this population. Implications of this work include the ability of practitioners and researchers to feasibly and reliably measure the fitness of school-aged children on the autism spectrum for ongoing health and behavioural monitoring and intervention purposes.

Reliable and feasible fitness testing for children on the autism spectrum

Autism Spectrum Disorder is a common neurodevelopmental disorder that occurs in approximately 1 in 59 children (Baio et al., 2018). While individual children on the autism spectrum present a unique set of strengths and difficulties, the disorder is broadly characterized by impairments in social communication, social interactions, and the presence of restricted and repetitive behaviours (American Psychiatric Association, 2013). These core impairments are typically the focus of intervention for children on the autism spectrum; however, research suggests that this population also experiences significant disparities in their physical and mental health (Gurney, McPheeters, & Davis, 2006; Hirvikoski et al., 2016). It is plausible that physical inactivity and low fitness are partially to blame for these health disparities, yet research in this area is limited.

Existing evidence suggests that children and youth on the autism spectrum are less fit than their neurotypical peers. Specifically, significant differences have been found in measures of flexibility, muscular strength and endurance, aerobic fitness, and body composition (Borremans, Rintala, & McCubbin, 2010; Pan, 2014; Tyler, MacDonald, & Menear, 2014). The fitness of individuals on the autism spectrum should be considered an essential component of their overall well-being from both a physical and behavioural standpoint: higher fitness is associated with reductions in morbidity and mortality (Fogelholm, 2010; Nocon et al., 2008), in addition to improvements in a number of behavioural domains (Bremer, Crozier, & Lloyd, 2016). Our understanding of the role of fitness for children on the autism spectrum is however limited by our ability to reliably measure it. The aforementioned studies have included a wide range of fitness assessments including shuttle runs, sit and reach tests, grip strength, standing long jump, push ups, curl ups, and walk tests. While the standing long jump, six minute walk test, and grip strength have shown adequate reliability for children with an intellectual disability (Wouters, Evenhuis, & Hilgenkamp, 2017; Wouters, van der Zanden, Evenhuis, & Hilgenkamp, 2017), the reliability and feasibility of common fitness assessments have yet to be established for children on the autism spectrum.

We cannot assume that fitness assessments that were initially designed for adults, and later validated for children with typical development, will be suitable for use with children on the autism spectrum. We know that this is a population who experiences challenges in the motor domain (Fournier, Hass, Naik, Lodha, & Cauraugh, 2010), along with deficits in social communication skills and a restricted range of interests and behaviours (American Psychiatric Association, 2013). Further, children with ASD often experience difficulties with sensory processing and modulation (Ben-Sasson et al., 2009) and exhibit lower levels of motivation to be physically active than their peers (Pan, Tsai, Chu, & Hsieh, 2011). This unique combination of challenges may make fitness testing particularly difficult for a child with ASD, particularly if it involves novel equipment (e.g., treadmill, cycle ergometer), or the need to regulate their behaviour to follow multi-step instructions in order to complete the test.

The failure of existing research to test the measurement properties of the measures used makes the interpretation of results problematic: in the absence of estimates of reliability for example, the influence of measurement error on performance results is not known. Moreover, evidence of the reliability and feasibility of approaches used to measure fitness is essential for future research. Therefore, the purpose of this study was to examine the test-retest reliability and feasibility of common fitness assessments for children 7-12 years of age on the autism spectrum.

Method

Sample

Participants were primarily recruited through a mail-out to families waiting for services at a local children's treatment centre, in addition to advertisements placed throughout the community. Participants were between 7 years, 0 months and 12 years, 11 months of age with a diagnosis of autism spectrum disorder, confirmed by the participant's parent or guardian. Participants were precluded from participating if they could not safely engage in the exercise assessments due to a physical (e.g., unstable heart condition) or behavioural (e.g., aggression toward others) concern, as indicated by their parent or guardian.

Study Design

This reliability study was embedded in a larger cross-sectional study examining the physical health of children on the autism spectrum. Participants enrolled in the study were randomized at study entry using a random number generator (randomizer.org) to one of two study groups: Regular or Reliability. All participants in the study completed an initial four study appointments and participants in the reliability group completed an additional two appointments, for a total of six. Appointment 1 was a familiarization to the lab, research team, and exercise equipment; no data collection occurred at this appoint. Appointment two included an assessment of motor competence, along with behavioural and demographic questionnaires completed by the participants' parent for use in the larger study. Seven standardized fitness assessments were administered in a random order over Appointments 3-4. These assessments were spread over two appointments in order to minimize participant fatigue. Participants in the reliability group were then retested on all fitness assessments, in the same order as they were initially completed, during appointments 5-6. The flow of participants through the study is outlined in Figure 1.

The order of each of the seven fitness assessments was randomized, per participant, using a random number generator (randomizer.org) prior to the first fitness appointment. Randomization was completed to negate any fatigue effects that may occur with an experimenter-derived order of testing. Given that there were over 5,000 permutations of the possible order of testing, no two participants had the same randomization schedule for testing. The first four fitness assessments in the randomization schedule were completed at Appointment 3, while the remaining three fitness assessments were completed at Appointment 4. This testing order was replicated at the two reliability appointments (see Figure 1). Time of day of the fitness assessments was controlled within participants as each of the participants' four fitness appointments were completed at approximately the same time each day. However, the time of day varied between participants based on participant availability. All fitness assessments occurred indoor, in a temperature-controlled research laboratory. In all, each fitness assessment was completed on two occasions, separated by an average of 21 ± 7 days.

The study was approved by an Institutional Research Ethics Board. All parents provided informed written consent and children 8 years of age and older provided informed written assent.

Measures

Demographic information and behavioural functioning. Demographic information and basic medical history was obtained through parental completion of a participant supplemental information form. The Vineland Adaptive Behavior Scales-2 (VABS-2) Parent/Caregiver Rating Form (Sparrow, Balla, & Cicchetti, 2005) was completed to provide a comprehensive measure of adaptive behavior covering the domains of communication, daily living skills, socialization, and maladaptive behavior. The VABS-2 adaptive behavior composite standard score (mean=100, standard deviation=3) and maladaptive behavior v-scale scores (mean=15, standard deviation=3) were used to describe the overall adaptive behavior levels of the sample. The VABS-2 is commonly used for individuals on the autism spectrum and has demonstrated good psychometric properties (Sparrow et al., 2005).

Anthropometry. Height was measured using a Seca SC264 stadiometer (Chino, California) to the nearest 0.1 cm and weight was measured using a Seca SC869 digital scale (Chino, California) to the nearest 0.1 kg. BMI was calculated and classifications of weight status were made using the IOTF cut-points for children (Cole & Lobstein, 2012).

Aerobic fitness. The Bruce protocol treadmill test (Bruce, Kusumi, & Hosmer, 1973) with progressive increases in speed and incline was used to assess aerobic fitness. Participants were familiarized to the treadmill at Visit 1 through a 5-10 minute practice session that included both walking and running on the treadmill. The protocol began at a speed of 1.7 mph and a 10% gradient and progressed in speed and grade every 3 minutes. Participants were instructed to hold on to the side rails of the treadmill for support and a spotter was placed at the rear of the treadmill for safety. Standard verbal encouragement was provided to all participants throughout the test (e.g., “keep going”, “you can do it”). The test was stopped whenever the participant showed signs of exhaustion, distress, or could no longer keep pace with the speed of the belt. Heart rate (HR) was assessed continuously during the test with a HR monitor (Polar H7, Lachine, QC) worn around the chest. Participants completed one trial of the Bruce protocol and the primary outcomes of this assessment were time to exhaustion (in seconds) and peak HR. Participants’ data was only included in the analysis if their peak HR was 180 bpm or higher. Participants were provided with a 10-minute seated rest following completion of the test.

The Modified 6-Minute Walk Test (M6MWT) was administered as an additional measure of aerobic fitness. The test requires participants to walk as far as they can in 6 minutes and has been shown to be reliable and valid in children and youth with typical development (Li, 2005; Li et al., 2007). Participants were familiarized to the testing space (indoor corridor) for the M6MWT at Visit 1 and allowed to practice walking back and forth in this area. Traditionally, participants walk back-and-forth along a straight 30 metre path as many times as possible in 6 minutes (ATS Committee on Proficiency Standards for Clinical Pulmonary Function Laboratories, 2002). For the current study, the test was modified to have participants travel back and forth between two pylons placed 15 metres apart. This shorter, 15-metre, distance was chosen in order to ease the completion of the task by keeping it more contained for the participants. Similar modifications to the 6MWT have been made for other populations of children [e.g., children with intellectual disability; (Wouters, van der Zanden, et al., 2017)]. The test was self-paced and standard verbal encouragement (i.e. “good work”, “keep going”) was provided each time the participants reached the end of the 15-metre distance. Participants completed one trial of the M6MWT and the primary outcome of this assessment was distance covered (in metres) during the 6 minutes. Participants were provided with a 10-minute seated rest following completion of the test.

Anaerobic fitness. Anaerobic fitness was measured with the 30-second Wingate anaerobic cycling test (Bar-Or, 1987), which measures short-term muscle power output using a cycle ergometer (LODE Pediatric, Groningen, The Netherlands). Participants were familiarized to the cycle ergometer at Visit 1 through a 2-3 minute bout of cycling at their own pace, followed by a brief (20-30) sprint at maximum speed. On the day of assessment, participants completed a brief warm-up (1-2 minutes of light pedalling) and then pedaled as fast as they could for approximately 20 seconds to establish maximum pedaling speed. After establishing maximum pedaling speed, participants had a brief rest and then completed a second sprint against a braking force (0.55 Nm/Kg) applied once they reached 80% of their maximum pedaling speed. Standard verbal encouragement was provided to participants throughout each of the two sprints (e.g., "keep going", "don't stop pedaling", "go, go, go!"). Peak power output (highest power output) and mean power output (power output over the 30 seconds) were taken in absolute values (watts). We also examined mean power over the first 10 seconds of the test due to motivational and coordination challenges with maintaining pedaling for the full 30 seconds. The LODE Wingate software package (Lode BV, Groningen, The Netherlands) was used to calculate all power outputs. This protocol has previously been used with young children with and without motor coordination difficulties (Gabel, Obeid, Nguyen, Proudfoot, & Timmons, 2011). Participants completed one trial of the Wingate and the primary outcomes of this assessment were peak power, mean power over 30 seconds, and mean power over 10 seconds. Participants were excluded from the 10 second mean power analyses if they stopped pedaling within the first 10 seconds of the load being engaged. Participants who stopped pedaling during the final 20 seconds of the load being engaged were only excluded from the 30 second mean power analysis. Participants were provided with a 5-minute seated rest following completion of the test.

The Muscle Power Sprint Test (MPST) was administered as a second measure of anaerobic fitness. The MPST was developed and validated to test the anaerobic functioning of children with cerebral palsy who have difficulty with motor coordination and are thus better suited to an intermittent sprint test, rather than a continuous test (Verschuren, Takken, Ketelaar, Gorter, & Helders, 2007). Since then, the MPST has been validated for children 6-12 years of age with typical development (Douma-van Riet et al., 2012). Participants were familiarized to the testing space (indoor corridor) for the MPST at Visit 1 and allowed to practice running as fast as they could. For the assessment, the MPST required the child to perform a total of 6 timed 15 metre runs at their maximum pace. Each of the 6 runs were timed to the hundredth of a second and a 10-second rest period was provided between each of the 6 runs. Participants were verbally encouraged to go as fast as they could during each run (e.g., “go, go, go!”, “run fast”). The following variables were created for each of the 6 sprints: velocity (m/s) = distance/time, acceleration (m/s^2) = velocity/time, Force (kg/s^2) = body mass × acceleration, and power (watts) = force × velocity (Douma-van Riet et al., 2012; Verschuren et al., 2007). Participants completed one trial of the MPST and the primary outcomes were peak power (recorded as the highest calculated power output among the 6 sprints) and mean power (average power output of all 6 sprints). Participants were provided with a 5-minute seated rest following completion of the test.

Musculoskeletal fitness. Standing long jump was administered as a measure of lower body muscular power (Castro-Piñero et al., 2010). Participants were familiarized with the standing long jump at Visit 1, where they were able to practice jumping as far as they could for up to 5 trials. Participants were asked to jump from a standing position as far as they could using a 2-foot take-off and landing. Standard verbal encouragement (e.g., “nice jump”) was provided after each jump. A maximum of 6 trials were completed in order to obtain 3 valid trials, with the primary outcome being the furthest distance jumped (measured in cm). Participants were provided with a 2-minute seated rest following completion of the test.

A digital handgrip dynamometer (Smedley Pro Digital, Japan) was used to measure grip strength, which is an indicator of total muscle strength (Wind, Takken, Helders, & Engelbert, 2010). A familiarization to the handgrip dynamometer was provided at Visit 1, where participants were able to practice squeezing the dynamometer for up to 5 trials per hand. For the actual assessment, the dynamometer was fitted to the participants hand in line with protocols suggested by España-Romero et al. (2008). Participants were instructed to stand with their arms down at their sides, but not touching the side of their body. Participants then squeezed the dynamometer as hard as they could, while standard verbal encouragement was provided (e.g., “squeeze!”, “nice job”). Two trials were completed for each hand and the primary outcome was the highest of the trials, per hand, measured to the nearest 0.1 kg. Participants were provided with a 2-minute seated rest following completion of the test.

Flexibility was assessed using the Sit and Reach Test (Wells & Dillon, 1952). The participant was seated on the floor with their legs straight and feet held flat against the sit and reach box (Flex-Tester, Creative Health Products, USA), positioned at a distance of 23 cm. A familiarization to the sit and reach box and test was provided at Visit 1 where participants were able to practice the task for up to 5 trials. For the assessment, participants were instructed to reach forward as far as they could, with hands on top of one another, and measurements were taken to the nearest 0.5 cm. A maximum of 3 trials were completed in order to obtain 1 valid trial. Standard verbal encouragement was provided during and following the test (e.g., “reach, reach, reach”, “good job”). Participants were provided with a 2-minute seated rest following completion of the test.

Test feasibility. Test feasibility was measured by the completion of 3 questions answered by the test assessor immediately following each fitness assessment. The questions asked about the ease of test administration, perceptions on whether the child understood how to perform the test, and perceptions of the child’s effort on the assessment. Each question was answered using a 100 mm visual analogue scale (VAS), with higher scores indicating greater feasibility. When possible, participants also answered 3 similar questions on a 100 mm VAS regarding the difficulty of the test, their perceived enjoyment, and their perceived effort. Higher scores on the participant questionnaire also indicated greater feasibility. Both the assessor and participant questionnaires were modified versions of those used by Verschuren (2007) to test feasibility of the MPST in children with cerebral palsy.

Statistical Analysis

Descriptive statistics were calculated for participant demographic characteristics and the primary outcomes for each of the fitness assessments. Two-way mixed intraclass correlation (ICC) analyses with absolute agreement on single measures were used to examine the test-retest reliability of the primary outcomes of each of the fitness assessments. ICC values were interpreted as poor reliability if they were less than 0.5, moderate reliability between .5 to .75, good reliability between .75 to .9, and excellent reliability if the value was greater than .9 (Koo & Li, 2016). In addition, paired samples t-tests and effect sizes (Cohen's *d*) were used to examine the differences between Measure 1 and 2 on each fitness assessment. In order to assess differential dropout on the fitness assessments, independent samples t-tests were computed for age, BMI, adaptive behavior, and feasibility scores between participants who were excluded on any fitness outcome due to behavioural difficulties and the remainder of the sample. Mean scores were calculated for each of the child- and participant-rated feasibility questions for each fitness assessment. Two total feasibility scores, assessor and participant, were also computed for each fitness assessment by taking the mean score of each of the three questions. All analyses were completed using SPSS 25 (IBM Corporation, 2017). A sample size of 10 participants was required to detect an ICC value of .7 with an alpha of .05 and 80% power (Bujang & Baharum, 2017).

Results

Participants (n=14) were predominately (93%) male and demonstrated behavioural profiles consistent with children on the autism spectrum, scoring on average one standard deviation below the mean on adaptive behavior and one standard deviation above the mean on maladaptive behavior. Demographic characteristics of the sample are presented in Table 1.

Test-retest Reliability

Results from the paired t-tests demonstrated that there were no significant differences (*p*-values > .05) between the two measurements on any of the fitness outcomes. The ICC values indicated that the test-retest reliability ranged from moderate- to- excellent. However, the 95% CIs of the ICCs of some outcomes were large and suggested reliability may be poorer for select outcomes. Specifically, the Bruce protocol, M6MWT, and MPST had 95% CIs of their ICCs that ranged from poor- to- good or poor- to-excellent. In contrast, grip strength (dominant hand), the sit and reach test, and mean power over 10 seconds on the Wingate had 95% CIs of their ICCs that ranged from moderate- to- excellent. Lastly, peak power and mean power over 30 seconds on the Wingate, standing long jump, and grip strength (non-dominant hand) had 95% CIs of their ICCs that ranged from good- to- excellent. Complete test-retest reliability results are presented in Table 2.

Five participants were excluded from the Bruce protocol analyses for two reasons: not reaching a peak HR of 180 bpm (n=4) and for refusing to wear the HR monitor (n=1). There were no differences in age, BMI, adaptive behavior, maladaptive behavior, or feasibility ratings between those participants who were excluded from the Bruce protocol analyses and the rest of the sample (all *p*-values >.05). One participant was excluded from the analyses of mean power over 10 and 30 seconds on the Wingate for stopping pedaling within the first 10 seconds of the test. Four additional participants were excluded from the analysis of mean power over 30 seconds on the Wingate as they stopped pedaling in the final 20 seconds of the test. No differences in age, BMI, adaptive behavior, or maladaptive behavior were present between those participants who were excluded from the Wingate analyses and the rest of the sample (all *p*-values >.05). There was however a significant difference (t=2.77, p<.05) on assessor-rated feasibility of the Wingate with those participants who were excluded from the analyses having a lower feasibility score than the included participants. Lastly, two participants could not complete the sit and reach test so were excluded from that analysis. The two excluded participants had a lower adaptive behavior score (t=2.41, p<.05), higher BMI (t=-2.56, p<.05), and lower feasibility score on the sit and reach test (t=5.00, p<.01) when compared to those participants who were included in the analysis.

Test Feasibility

Only 7 participants completed the child feasibility questionnaire, with the remaining 7 not understanding how to answer using the VAS scale. There were however no differences (all *p*-values >.05) in age, BMI, adaptive behavior, maladaptive behavior, or assessor feasibility ratings between those participants who did (n=7) and did not (n=7) complete the child feasibility questionnaire. Assessor-rated feasibility scores were, on average, high (>70/100) for all fitness assessments with grip strength demonstrating the highest feasibility (86.15/100) and the sit and reach test demonstrating the lowest feasibility (72.31/100). Child-rated feasibility scores were, on average, high and comparable to the assessor-rated feasibility. The assessment with the highest child-rated feasibility was standing long jump (82.55/100), whereas the lowest feasibility rating was on the sit and reach test (66.24/100). Complete results from both the assessor- and child-rated feasibility questionnaires are presented in Table 3.

Discussion

The purpose of this study was to examine the test-retest reliability and feasibility of lab- and field-based fitness assessments in school-aged children on the autism spectrum. The results demonstrate moderate to excellent test-retest reliability suggesting some tests are clearly better than others when it comes to this aspect of measurement. Specifically, the Wingate anaerobic cycling test, standing long jump, grip strength, and sit and reach test all demonstrated good to excellent reliability, with 95% confidence intervals in the moderate to excellent range. In contrast, the Bruce protocol, M6MWT, and MPST demonstrated moderate to good reliability; however, the large confidence intervals on those tests suggest that caution should be used when interpreting their test-retest reliability. These results generally align with a recent systematic review of the feasibility and reliability of fitness tests for children and youth with intellectual disabilities, where moderate reliability was demonstrated for measures of muscular strength but, evidence of the reliability of fixed time walking tests and shuttle run tests were limited (Wouters, Evenhuis, et al., 2017).

There are a number of factors that may contribute to the poorer reliability seen on the M6MWT and MPST. For example, both tests rely on a high degree of self-regulation to stay on task and motivation to complete the activity. These tests may also be more prone to distractions by the participants as they occur in an open space in contrast to the more fixed or constrained environments of the Wingate, standing long jump, grip strength, and sit and reach tests. Children on the autism spectrum also lack proficiency in motor coordination (Fournier et al., 2010), exhibit challenges with inhibition and attention (Hill, 2004), and may have difficulties with task understanding (Breslin & Liu, 2015; National Research Council, 2001); all of which may help to explain differences in reliability on the various fitness assessments. For example, it may be easier for children on the autism spectrum to complete a cycling test than a running or walking test due to difficulties with maintaining a proficient gait (Fournier et al., 2010). Likewise, tests that are shorter in duration or require only a single instruction (e.g., Wingate, grip strength) may be more suitable to the attention and task understanding abilities of children on the autism spectrum than longer (e.g., M6MWT) or multi-step (e.g. MPST) assessments.

The role of motivation and encouragement should also be further explored in the completion of fitness assessments for children on the autism spectrum. For example, the current study provided encouragement (e.g. “good work”, “keep going”) to participants at set intervals during the M6MWT, as well as throughout each of the other tests; however, the direct impact of this encouragement was not measured. One recent study found better performance on the 6MWT in 6-12 year old children with typical development when encouragement was provided (Morales Mestre, Audag, Caty, & Reychler, 2018), yet the effect for children on the autism spectrum is unknown. While the provision of encouragement during each test likely helped to improve the participants’ performance, it is important to note that from a feasibility perspective, providing encouragement was often necessary in order for the assessments to be completed. Moreover, encouragement was similar across participants and assessments.

There are no established thresholds for interpreting the results of the feasibility questionnaire; however, scores closer to 100 represent a higher degree of feasibility. Moreover, the feasibility questionnaire used in this study was based on that used by Verschuren (2007), who interpreted their mean feasibility scores ranging from 8.8 to 9.2 out of 10 (comparable to 88 to 92 out of 100 on our scores) as being “very high” feasibility. Considering this interpretation, our results would suggest that all tests in the current study are feasible to implement from the assessor’s perspective. The participant feasibility results are more challenging to interpret as only half of the children were able to respond to the questions. However, those children who did respond reported a high degree of perceived effort on all tests and overall feasibility ratings that appeared acceptable. Standing long jump, grip strength, and the Wingate had the highest child-rated total feasibility scores, which when paired with the high reliability coefficients on these assessments further speaks to their acceptable use in this population.

A limitation of the current study is that assessments were performed over 4 days (two days for each of the initial and reliability assessments). However, given the attention and behavioral profiles of children on the autism spectrum we deemed it necessary to split the assessments into multiple sessions; we further randomized the assessment order to limit any fatigue or motivational effects over the course of the appointments. Second, our test-retest window for the reliability assessments ranged from two to four weeks, which is longer than the more commonly used one- to two-week window employed for many reliability studies of fitness tests (Artero et al., 2011). Given the nature of the study design and family burden of completing six study appointments, some families could not complete weekly appointments and thus it was necessary for their reliability appointments to be outside of the two-week window. We would not however expect the participants' fitness to change over a two to four- week period due to growth or maturation, and no participants were involved in physiotherapy or other training programs to improve their fitness during the duration of the study. Third, it is possible that the low level of reliability found on the M6MWT was due to the modification of the test to a 15-metre corridor as opposed to the usual 30-metre distance. This modification may have increased participant fatigue due to the increased number of turns. As such, it may be beneficial for future research to explore the test-retest reliability of the original 6MWT for this population. Fourth, this study included a smaller sample of participants than is often included in the study of the reliability of fitness assessments [e.g., N=37 in Wouters et al. (2017) study of children with moderate to severe intellectual disabilities]. We were, however, adequately powered to detect ICC values of .70 and higher (Bujang & Baharum, 2017). Further, the fact that we found ICC values in the good- to-excellent range with small confidence intervals for some outcomes suggests that our sample size was sufficient to detect meaningful results. Finally, our sample only included one female, which means that generalizability to both sexes may be limited. However, given our smaller sample and lack of basis to exclude participants based on sex in this study, we opted to include the single female in our analyses. Future work should aim to further explore the reliability and feasibility of fitness testing for children on the autism spectrum, with a particular focus on increasing number of female participants in this work.

Establishing the reliability and feasibility of fitness assessments for school-aged children on the autism spectrum is an important step in being able to accurately measure, track, and intervene on the fitness of this population. While previous research has suggested that children on the autism spectrum exhibit low fitness levels (Borremans et al., 2010; Pan, 2014; Tyler et al., 2014), the reliability of the assessments used had not previously been established. Based on our findings, we recommend the use of the Wingate anaerobic cycling test, standing long jump, grip strength, and sit and reach test to examine the fitness of 7-12 year old children on the autism spectrum. In contrast, the Bruce protocol, M6MWT, and MPST only demonstrate moderate test-retest reliability in this population. Despite differences in reliability, it appears that all tests can be implemented with adequate levels of feasibility. We advise that researchers and clinicians take these findings into account when selecting fitness assessments for use with school-aged children on the autism spectrum, particularly when assessing changes in fitness over time or following intervention. Moreover, future research should explore the reliability and feasibility of other aerobic fitness assessments (e.g., PACER, intermittent treadmill tests), in addition to other measurement properties, for use in this population.

References

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (DSM-5)* (5th ed.). Washington, DC: American Psychiatric Publishing.

Artero, E. G., España-Romero, V., Castro-Piñero, J., Ortega, F. B., Suni, J., Castillo-Garzon, M. J., & Ruiz, J. R. (2011). Reliability of Field-Based Fitness Tests in Youth. *International Journal of Sports Medicine*, *32*(03), 159–169. https://doi.org/10.1055/s-0030-1268488

ATS Committee on Proficiency Standards for Clinical Pulmonary Function Laboratories. (2002). ATS Statement: Guidelines for the six-minute walk test. *American Journal of Respiratory and Critical Care Medicine*, *166*(1), 111–117.

Baio, J., Wiggins, L., Christensen, D. L., Maenner, M. J., Daniels, J., Warren, Z., … Dowling, N. F. (2018). Prevalence of Autism Spectrum Disorder among children aged 8 years — Autism and Developmental Disabilities Monitoring Network, 11 Sites, United States, 2014. *MMWR. Surveillance Summaries*, *67*(No. SS-6), 1–23. https://doi.org/10.15585/mmwr.ss6706a1

Bar-Or, O. (1987). The Wingate anaerobic test an update on methodology, reliability and validity. *Sports Medicine*, *4*(6), 381–394.

Borremans, E., Rintala, P., & McCubbin, J. A. (2010). Physical fitness and physical activity in adolescents with Asperger syndrome: A comparative study. *Adapted Physical Activity Quarterly*, *27*(4), 308–320.

Bremer, E., Crozier, M., & Lloyd, M. (2016). A systematic review of the behavioural outcomes following exercise interventions for children and youth with autism spectrum disorder. *Autism*, *20*(8), 899–915. https://doi.org/10.1177/1362361315616002

Breslin, C. M., & Liu, T. (2015). Do You Know What I’m Saying? Strategies to Assess Motor Skills for Children with Autism Spectrum Disorder. *Journal of Physical Education, Recreation & Dance*, *86*(1), 10–15. https://doi.org/10.1080/07303084.2014.978419

Bruce, R. A., Kusumi, F., & Hosmer, D. (1973). Maximal oxygen intake and nomographic assessment of functional aerobic impairment in cardiovascular disease. *American Heart Journal*, *85*(4), 546–562.

Bujang, M. A., & Baharum, N. (2017). A simplified guide to determination of sample size requirements for estimating the value of intraclass correlation coefficient: A review. *Archives of Orofacial Sciences*, *12*(1), 1–11.

Castro-Piñero, J., Ortega, F. B., Artero, E. G., Girela-Rejón, M. J., Mora, J., Sjöström, M., & Ruiz, J. R. (2010). Assessing muscular strength in youth: Usefulness of standing long jump as a general index of muscular fitness. *Journal of Strength and Conditioning Research*, *24*(7), 1810–1817. https://doi.org/10.1519/JSC.0b013e3181ddb03d

Cole, T. J., & Lobstein, T. (2012). Extended international (IOTF) body mass index cut-offs for thinness, overweight and obesity: Extended international BMI cut-offs. *Pediatric Obesity*, *7*(4), 284–294. https://doi.org/10.1111/j.2047-6310.2012.00064.x

Douma-van Riet, D., Verschuren, O., Jelsma, D., Kruitwagen, C., Smits-Engelsman, B., & Takken, T. (2012). Reference Values for the Muscle Power Sprint Test in 6- to 12-Year-Old Children. *Pediatric Physical Therapy*, *24*(4), 327–332. https://doi.org/10.1097/PEP.0b013e3182694a4c

España-Romero, V., Artero, E. G., Santaliestra-Pasias, A. M., Gutierrez, A., Castillo, M. J., & Ruiz, J. R. (2008). Hand Span Influences Optimal Grip Span in Boys and Girls Aged 6 to

12 Years. *The Journal of Hand Surgery*, *33*(3), 378–384. https://doi.org/10.1016/j.jhsa.2007.11.013

Fogelholm, M. (2010). Physical activity, fitness and fatness: relations to mortality, morbidity and disease risk factors. A systematic review. *Obesity Reviews*, *11*(3), 202–221. https://doi.org/10.1111/j.1467-789X.2009.00653.x

Fournier, K. A., Hass, C. J., Naik, S. K., Lodha, N., & Cauraugh, J. H. (2010). Motor Coordination in Autism Spectrum Disorders: A Synthesis and Meta-Analysis. *Journal of Autism and Developmental Disorders*, *40*(10), 1227–1240. https://doi.org/10.1007/s10803-010-0981-3

Gabel, L., Obeid, J., Nguyen, T., Proudfoot, N. A., & Timmons, B. W. (2011). Short-term muscle power and speed in preschoolers exhibit stronger tracking than physical activity. *Applied Physiology, Nutrition, and Metabolism*, *36*(6), 939–945. https://doi.org/10.1139/h11-118

Gurney, J. G., McPheeters, M. L., & Davis, M. M. (2006). Parental report of health conditions and health care use among children with and without autism: National Survey of Children's Health. *Archives of Pediatrics & Adolescent Medicine*, *160*(8), 825–830.

Hill, E. L. (2004). Executive dysfunction in autism. *Trends in Cognitive Sciences*, *8*(1), 26–32.

Hirvikoski, T., Mittendorfer-Rutz, E., Boman, M., Larsson, H., Lichtenstein, P., & Bölte, S. (2016). Premature mortality in autism spectrum disorder. *British Journal of Psychiatry*, *208*(03), 232–238. https://doi.org/10.1192/bjp.bp.114.160192

IBM Corporation. (2017). IBM SPSS statistics for windows (Version 25). Armonk, NY: IBM Corporation.

Koo, T. K., & Li, M. Y. (2016). A Guideline of Selecting and Reporting Intraclass Correlation Coefficients for Reliability Research. *Journal of Chiropractic Medicine*, *15*(2), 155–163. https://doi.org/10.1016/j.jcm.2016.02.012

Li, A. M. (2005). The six-minute walk test in healthy children: reliability and validity. *European Respiratory Journal*, *25*(6), 1057–1060. https://doi.org/10.1183/09031936.05.00134904

Li, A. M., Yin, J., Au, J. T., So, H. K., Tsang, T., Wong, E., … Ng, P. C. (2007). Standard Reference for the Six-Minute-Walk Test in Healthy Children Aged 7 to 16 Years. *American Journal of Respiratory and Critical Care Medicine*, *176*(2), 174–180. https://doi.org/10.1164/rccm.200607-883OC

Morales Mestre, N., Audag, N., Caty, G., & Reychler, G. (2018). Learning and Encouragement Effects on Six-Minute Walking Test in Children. *The Journal of Pediatrics*. https://doi.org/10.1016/j.jpeds.2018.02.073

National Research Council. (2001). *Educating children with autism*. Washington, DC: National Academy Press. Retrieved from http://public.eblib.com/choice/publicfullrecord.aspx?p=3375258

Nocon, M., Hiemann, T., Müller-Riemenschneider, F., Thalau, F., Roll, S., & Willich, S. N. (2008). Association of physical activity with all-cause and cardiovascular mortality: a systematic review and meta-analysis. *European Journal of Cardiovascular Prevention & Rehabilitation*, *15*(3), 239–246. https://doi.org/10.1097/HJR.0b013e3282f55e09

Pan, C.-Y. (2014). Motor proficiency and physical fitness in adolescent males with and without autism spectrum disorders. *Autism*, *18*(2), 156–165. https://doi.org/10.1177/1362361312458597

Pan, C.-Y., Tsai, C.-L., Chu, C.-H., & Hsieh, K.-W. (2011). Physical activity and self-determined motivation of adolescents with and without autism spectrum disorders in inclusive physical education. *Research in Autism Spectrum Disorders*, *5*(2), 733–741. https://doi.org/10.1016/j.rasd.2010.08.007

Sparrow, S. S., Balla, D. A., & Cicchetti, D. V. (2005). *Vineland Adaptive Behavior Scales, Second Edition*. Bloomington, MN: PsychCorp.

Tyler, K., MacDonald, M., & Menear, K. (2014). Physical Activity and Physical Fitness of School-Aged Children and Youth with Autism Spectrum Disorders. *Autism Research and Treatment*, *2014*, 1–6. https://doi.org/10.1155/2014/312163

Verschuren, O., Takken, T., Ketelaar, M., Gorter, J. W., & Helders, P. J. M. (2007). Reliability for Running Tests for Measuring Agility and Anaerobic Muscle Power in Children and Adolescents with Cerebal Palsy. *Pediatric Physical Therapy*, *19*(2), 108–115. https://doi.org/10.1097/pep.0b013e318036bfce

Wells, K. F., & Dillon, E. K. (1952). The sit and reach—a test of back and leg flexibility. *Research Quarterly. American Association for Health, Physical Education and Recreation*, *23*(1), 115–118.

Wind, A. E., Takken, T., Helders, P. J. M., & Engelbert, R. H. H. (2010). Is grip strength a predictor for total muscle strength in healthy children, adolescents, and young adults? *European Journal of Pediatrics*, *169*(3), 281–287. https://doi.org/10.1007/s00431-009-1010-4

Wouters, M., Evenhuis, H. M., & Hilgenkamp, T. I. M. (2017). Systematic review of field-based physical fitness tests for children and adolescents with intellectual disabilities. *Research in Developmental Disabilities*, *61*, 77–94. https://doi.org/10.1016/j.ridd.2016.12.016

Wouters, M., van der Zanden, A. M., Evenhuis, H. M., & Hilgenkamp, T. I. M. (2017). Feasibility and Reliability of Tests Measuring Health-Related Physical Fitness in Children With Moderate to Severe Levels of Intellectual Disability. *American Journal on Intellectual and Developmental Disabilities*, *122*(5), 422–438. https://doi.org/10.1352/1944-7558-122.5.422

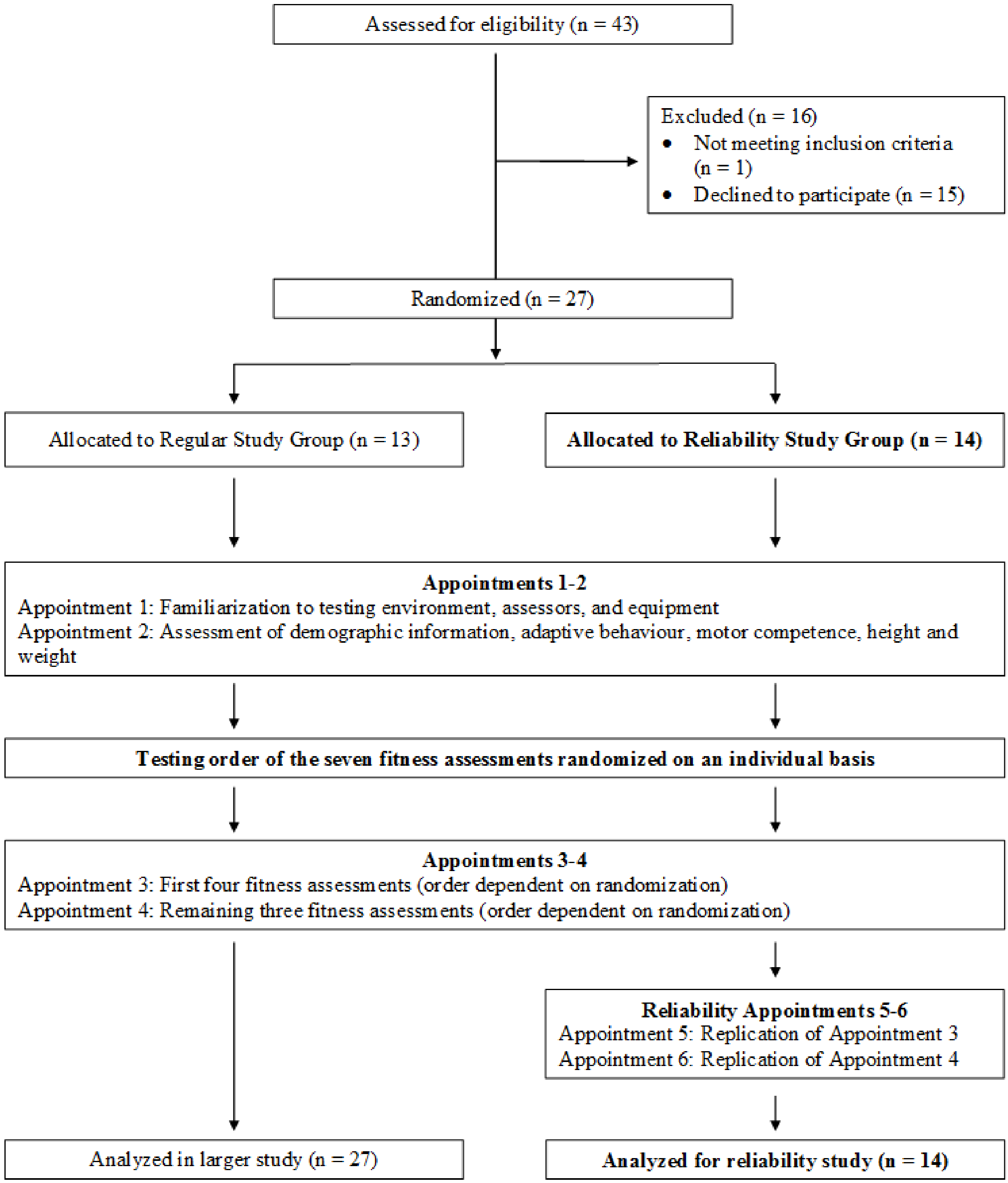

Figure 1. Flow diagram of study design.

Table 1. Participant demographic characteristics (n=14).

Demographic Variable	**Statistic**
	Mean (SD)
Age (years)	9.5 (1.7)
VABS-2 Standard and v-scale Scores	
Adaptive Behavior Composite	81.2 (13.1)
Internalizing Behavior	20.3 (1.7)
Externalizing Behavior	18.4 (2.4)
Maladaptive Behavior	20.1 (1.8)
BMI	18.8 (4.1)
	N (%)
BMI Category	
Underweight	*1 (7%)*
Normal weight	*7 (50%)*
Overweight/Obese	*6 (43%)*
Ethnicity	
Caucasian	*10 (71%)*
Other	*4 (29%)*

VABS-2: Vineland Adaptive Behavior Scales, 2nd Edition; BMI: Body Mass Index.

Table 2. Test-retest reliability of each fitness assessment.

Fitness Outcome	**Participants**	**Measure 1** mean (SD)	**Measure 2** mean (SD)	***p*-value**	**Effect Size** (*d*)	**ICC**	**95% CI**
Bruce Protocol							
Time to exhaustion (s)	9	601.9 (132.2)	638.1 (158.9)	.250	0.41	.811	.406-.953
Peak HR (bpm)	9	194.4 (6.1)	196.3 (4.0)	.237	0.43	.618	.038-.896
M6MWT							
Distance (m)	14	474.8 (88.2)	441.4 (98.2)	.193	0.37	.510	.030-.807
Wingate							
Peak Power (W)	14	229.9 (79.4)	242.3 (84.2)	.052	0.57	.956	.845-.986
Mean Power 30sec (W)	9	142.8 (49.5)	137.3 (58.7)	.399	0.30	.943	.783-.987
Mean Power 10sec (W)	13	175.4 (68.5)	185.6 (74.0)	.356	0.27	.855	.605-.953
MPST							
Peak Power (W)	14	95.0 (50.9)	99.1 (51.5)	.751	0.09	.595	.101-.851

Mean Power (W)	14	74.3 (39.7)	75.9 (36.8)	.845	0.05	.703	.284-.895
Standing Long Jump							
Distance (cm)	14	105.5 (32.9)	107.4 (32.1)	.592	0.15	.925	.787-.975
Grip Strength							
Dominant hand (kg)	14	11.0 (2.9)	11.1 (3.5)	.754	0.09	.871	.646-.957
Non-dominant hand (kg)	14	11.2 (2.9)	11.1 (3.7)	.797	0.07	.913	.752-.971
Sit and Reach							
Distance (cm)	12	18.5 (5.8)	19.2 (5.8)	.517	0.19	.829	.518-.947

HR: Heart Rate; M6MWT: Modified Six-Minute Walk Test; MPST: Muscle Power Sprint Test.

Table 3. Assessor- and child-rated feasibility of each fitness assessment.

	Bruce Protocol Mean (SD)	**M6MWT** Mean (SD)	**Wingate** Mean (SD)	**MPST** Mean (SD)	**Long Jump** Mean (SD)	**Grip Strength** Mean (SD)	**Sit and Reach** Mean (SD)
Assessor-rated							
Ease of Administration	76.29 (21.71)	82.64 (17.59)	85.00 (10.02)	82.89 (17.75)	83.96 (21.36)	88.82 (13.73)	70.96 (29.67)
Participant Understanding	79.36 (17.04)	83.18 (18.69)	83.75 (11.85)	83.71 (16.97)	85.61 (22.10)	86.64 (17.24)	74.25 (25.18)
Participant Performance	83.61 (14.44)	71.18 (22.80)	77.64 (17.12)	74.07 (24.68)	82.11 (22.30)	83.00 (17.15)	71.71 (25.65)

Total Score	79.75	79.00	82.13	80.23	83.89	86.15	72.31
	(16.89)	(18.92)	(11.23)	(18.50)	(21.65)	(15.63)	(26.49)
Child-rated (n=7)							
Ease of Task	45.86	54.07	51.07	55.07	69.14	81.29	58.00
	(37.36	(35.51)	(36.71)	(34.83)	(33.01)	(23.97)	(30.31)
Task Enjoyment	76.43	68.57	82.50	68.57	88.21	65.93	59.64
	(30.11)	(22.26)	(15.23)	(22.26)	(15.87)	(39.84)	(24.28)
Perceived Effort	86.14	86.86	88.64	88.00	90.29	92.57	81.07
	(22.9)	(21.71)	(12.43)	(16.43)	(12.22)	(8.98)	(19.38)
Total Score	69.48	69.79	74.07	70.55	82.55	79.93	66.24
	(26.92)	(20.48)	(16.94)	(13.60)	(14.99)	(17.70)	(17.07)

M6MWT: Modified Six-Minute Walk Test; MPST: Muscle Power Sprint Test.

CHAPTER 3: The interrelationship between motor coordination and adaptive behavior in children with autism spectrum disorder

Preamble

The interrelationship between motor coordination and adaptive behavior in children with autism spectrum disorder is the second study in the dissertation. The study examines the relationship between motor coordination and adaptive behaviour in a sample of 7-12 year old children with ASD.

The following manuscript is published in *Frontiers in Psychology*. The published version of the manuscript is included in the dissertation.

<u>Contribution of Study 2 to overall dissertation:</u>
Study 2 provides evidence of a positive association between motor coordination and adaptive behaviour in children with ASD. These findings underscore the need to consider the role of motor competence for participation in daily activities for this population. Overall, study 2 provides support for the motor competence – adaptive behaviour pathway proposed in the Interdisciplinary Framework presented in Chapter 1.

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The Interrelationship Between Motor Coordination and Adaptive Behavior in Children With Autism Spectrum Disorder

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Objective: Children with autism spectrum disorder (ASD) experience significant challenges with their motor coordination. It is not, however, well understood how motor coordination may impact the behavioral functioning of children with ASD. Therefore the purpose of this study was to explore the relationships between motor coordination and adaptive behavior in 7–12-year-old children with ASD.

Methods: Motor coordination was assessed using the Movement Assessment Battery for Children, 2nd Edition (MABC-2) and adaptive behavior was assessed by parental report using the Vineland Adaptive Behavior Scales, 2nd Edition (VABS-2) as part of a larger cross-sectional study. Descriptive characteristics were calculated for MABC-2 and VABS-2 scores and Spearman's rank order correlation analyses were used to examine the relationship between motor coordination and adaptive behavior.

Results: On average, the participants ($n = 26$) exhibited significant challenges in regard to their motor coordination with all but two participants classified as having significant motor impairments by scoring at or below the 16th percentile on the MABC-2. Results from the correlation analyses indicated that manual dexterity was positively related to daily living skills ($\rho = 0.58$, $p < 0.003$), and overall motor coordination was positively related to daily living skills ($\rho = 0.60$, $p < 0.003$) and overall adaptive behavior ($\rho = 0.57$, $p < 0.003$). In all instances, better motor coordination was related to more adaptive behaviors.

Conclusion: These results highlight the profound motor coordination challenges that children with ASD experience and also suggest that these challenges, particularly with manual dexterity, are related to the daily behavior of children with ASD. The interrelatedness of motor and adaptive behavior suggests the need to further explore the impact of motor-based interventions for this population, as well as conduct longitudinal studies to disentangle these relationships.

Keywords: motor development, neurodevelopmental disability, middle childhood, developmental trajectories, daily living skills

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INTRODUCTION

Autism spectrum disorder (ASD) is a neurodevelopmental disorder defined by challenges in communication, social skills, and the presence of restricted and repetitive behaviors (American Psychiatric Association, 2013). Children with ASD are heterogeneous in their abilities and difficulties, with varying levels of symptom severity, behavioral profiles, and relative level of functioning for each individual child with ASD (Geschwind, 2009; Wiggins et al., 2012; Georgiades et al., 2013a,b). Adaptive behavior is a common term used to describe and classify the degree to which an individual performs daily activities for social and personal sufficiency and for school-aged children this can include tasks such as household chores, completing school projects, and getting along with peers (Sparrow et al., 2005). Children with ASD are most commonly described as having adaptive behavior profiles with relative strengths in daily living skills, but with weaknesses in socialization and communication (Kanne et al., 2011). Adaptive behavior is consistently associated with positive outcome in individuals with ASD, regardless of symptom severity (Kanne et al., 2011), further emphasizing the importance of adaptive behavior in enabling individuals with ASD to reach a level of independence needed for personal and social sufficiency.

One area of development that is often ignored when intervening on the adaptive behavior of school-aged children with ASD is their motor coordination. Motor coordination is the organization of one's body parts to create both gross and fine movements (Payne and Isaacs, 2011; Haywood and Getchell, 2014). Motor coordination is involved in virtually all aspects of daily living including, fine motor tasks like dressing, feeding, and writing and gross motor tasks including locomotion and active play. Beyond basic motor tasks, motor coordination is inherently linked to social communicative skills through gesturing, imitation, and joint attention (Schmidt et al., 2011); skills that are typically a challenge for children with ASD. Motor delays are present early on for toddlers with ASD and this delay may become more pronounced with age (Lloyd et al., 2013). Importantly, these motor delays are not limited to infants and toddlers, but persist through childhood into adulthood (Ozonoff et al., 2008; Fournier et al., 2010; Bhat et al., 2011). For example, the most recent meta-analysis of 51 studies showed that individuals with ASD had significantly poorer motor coordination across a range of domains (e.g., balance, posture, gait, reaction time, aiming) when compared to typically developing peers (Fournier et al., 2010).

Until relatively recently, motor coordination problems in children, characteristic of a disorder known as developmental coordination disorder (DCD) (Cairney, 2015), was considered a separate, non-overlapping, condition from ASD. Indeed, in the case of DCD, until the 5th edition of the Diagnostic and Statistical Manual of Mental Disorders (American Psychiatric Association, 2013), children who met diagnostic criteria for ASD were excluded altogether from a diagnosis of DCD. This reflects a pervasive underlying concern about evaluating a child's motor ability independent of their cognitive ability. Since communicative disorders are an inherent feature of ASD (American Psychiatric Association, 2013), poor performance on motor tasks may be related to failure to comprehend or comply with verbal instructions, rather than reflecting a true deficit in motor ability. At the same time, it has been observed clinically for some time that children with neurodevelopmental disorders such as ADHD/ADD or ASD, typically present with motor impairments (Cairney and King-Dowling, 2016). Evidence from a recent systematic review suggests that behaviorally DCD and ASD are separate, but possibly co-occurring, conditions with differences in many behavioral domains (Caçola et al., 2017); however, these findings are limited by the lack of studies reporting on children with co-occurring diagnoses of ASD and DCD. Indeed, the removal of ASD as an exclusion from DCD has opened the door for more serious inquiry into co-occurring, motor, cognitive, communicative and social impairments in children who present with neurodevelopmental disorders such as ASD. Examination of these co-occurring conditions helps us to better understand the heterogeneity of ASD, while also providing opportunities to discern appropriate treatment strategies for different subgroups of the spectrum. This is important given the range of behavioral profiles exhibited by children with ASD, along with the notion that co-occurring conditions may actually be the most prominent difficulties in the daily life of a child with ASD (Gillberg and Fernell, 2014).

Despite the evidence that motor impairments are common in children with ASD, there is still relatively little evidence on how these delays may be related to the core deficits of the disorder and the ensuing implications on their adaptive behavior. Previous research has demonstrated that significant relationships exist between motor development, social skills, and communication in infants developing both typically and atypically (Leonard and Hill, 2014). However, the importance of these relationships is often neglected beyond the developmental period in children with ASD. Existing research in school-aged children is limited but suggests that fundamental movement skills (i.e., the foundational skills needed for participation in physical activity) are related to social communicative skills in this population (MacDonald et al., 2013). The relationships between fundamental movement skills and social skills inherently make sense for children with ASD: proficient movement skills are necessary to engage in active play, which in turn is a very important social activity for children. The current literature is limited in that it has yet to explore whether motor coordination, including both gross and fine motor skill, is related to outcomes beyond just social communicative skills in children with ASD, which may have important implications for independence and positive outcomes. Thus, the purpose of this study is to examine the relationship between motor coordination and adaptive behavior in school-aged children with ASD.

MATERIALS AND METHODS

Design and Procedure

A cross-sectional design was employed to assess the relationship between motor coordination and adaptive behavior in

7–12-year-old children with ASD. The study consisted of one appointment, which represented the second visit in a larger study of the physical health of children with ASD. The first study appointment was a lab familiarization session for the participant and their parent; no data collection occurred at this appointment. Participants were recruited on an on-going basis and assessments took place in a university research lab between March 2016 and December 2017. All assessments were conducted by a graduate research assistant with formal training in the assessment of motor development and coordination in children with ASD. Ethical approval for the study was provided by the university's Institutional Research Ethics Board. Informed written consent was obtained from the participants' parent or guardian at the first study appointment and participants 8 years of age and older provided informed written assent prior to their participation.

Participants

A convenience sample of participants were recruited through advertisements at local community centers, in addition to a recruitment mail out to families of children with ASD on a waitlist for government-funded applied behavior analysis services in the geographic region of the study. Children were eligible to participate if they were between the ages of 7 years, 0 months, to 12 years, 11 months with confirmation of their ASD diagnosis provided by their parent or guardian. In Ontario, Canada, where this study took place, a diagnosis of ASD can be made by a family physician, pediatrician, psychiatrist, psychologist, or a psychological associate.

Measures

Demographic and Medical History

The participants' diagnostic and treatment history was reported by parental completion of a supplemental information form. This form asked parents to answer basic demographic questions, state when (and by whom) their child was diagnosed, and asked a range of questions regarding their child's motor and behavioral treatment history.

Motor Coordination

Children were administered the Movement Assessment Battery for Children-2 (MABC-2) by a trained research assistant, which is an individually administered standardized test used to identify motor impairment in children aged 3–16 years (Henderson et al., 2007). The MABC-2 consists of eight motor tasks grouped under three domains: manual dexterity; aiming & catching; and balance. Domain and total test scores for the assessment are converted into age-normed percentile scores, with a score below the 16th percentile indicating the presence of significant motor impairments. Test re-test reliability and standard error of measurement for the total test scores have been reported to be 0.80 and 1.34, respectively (Henderson et al., 2007).

Adaptive Behavior

Parents completed the Vineland Adaptive Behavior Scales-2 (VABS-2) Parent/Caregiver Rating Form (Sparrow et al., 2005). The VABS-2 assesses adaptive behavior in the following domains: communication, daily living skills, socialization, and maladaptive behavior, and helps to provide a broad overview of each child's level of functioning. Standard scores are reported for the domains of communication, daily living skills, and socialization, in addition to the adaptive behavior composite. VABS-2 standard scores range from 20 to 160, have a mean of 100, and a standard deviation of 15. Maladaptive behaviors are reported for the domains of externalizing and internalizing, in addition to a composite maladaptive behavior index. Each of the maladaptive behavior scores are reported as *v*-scale scores, which range from 1–24 with a mean of 15, and a standard deviation of 3. Internal-consistency reliabilities of the domain and composite scores of the VABS-2 range from 0.80 to 0.97 and the test–retest reliability correlations range from 0.75 to 0.93 for 7–12-year-old children (Sparrow et al., 2005).

Statistical Analyses

Descriptive statistics (mean, standard deviations) were calculated for demographic and diagnostic variables, along with domain and total scores for the MABC-2 and VABS-2. Tests of normality revealed that the MABC-2 scores were not normally distributed, therefore nonparametric analyses were employed. Spearman's rank order correlations were used to examine the relationships between MABC-2 and VABS-2 domain and total scores. Given the multiple correlations being tested, a Bonferroni-adjusted p-value of 0.003 (0.05/16) was used as the threshold for statistical significance. All analyses were conducted using SPSS 25 (IBM Corporation, 2017).

RESULTS

Demographic and diagnostic characteristics of the sample ($n = 26$) are presented in **Table 1**. On average, the participants' exhibited impaired motor coordination with an average MABC-2 total test percentile score in the 6th percentile. In other words, participants in this sample were scoring, on average, better than only 6% of their age- and sex-matched peers. Moreover, all but two participants were classified as having significant motor impairments by scoring at or below the 16th percentile on the MABC-2 (**Table 2**). As anticipated, the participants' also exhibited challenges with their adaptive behavior. For example, the VABS-2 adaptive behavior composite score was, on average, almost two standard deviations below what would be expected for the participants' age. Likewise, on average, the maladaptive behavior scores ranged from one to almost two standard deviations above what would be expected for the participants' age, indicating an increased level of maladaptive behavior (**Table 2**).

Results from the correlation analyses (**Table 3**) indicate that the manual dexterity domain of the MABC-2 was significantly correlated with the VABS-2 daily living skill domain. Further, the MABC-2 total test score was significantly correlated with the VABS-2 daily living skill domain and the adaptive behavior composite: in all cases, children who performed better on the motor tasks had more adaptive behaviors in these domains. No other significant correlations were present.

TABLE 1 | Descriptive characteristics of the sample (n = 26).

Variable		Statistics Mean (SD)
Age (years)		9.9 (1.7)
Age at diagnosis (months)		60.6 (28.2)
		N (%)
Sex (male, female)		23 male, 3 female
Ethnicity		
	Caucasian	19 (73)
	Other	7 (27)
Diagnosis provider		
	Pediatrician	15 (58)
	Psychiatrist	3 (12)
	Psychologist	7 (27)
	Psychological associate	1 (3)
Previously received treatment		
	Behavioral (ABA or IBI)	19 (73)
	Speech-Language	19 (73)
	Motor (PT or OT)	15 (58)
Currently receiving treatment		
	Behavioral (ABA or IBI)	10 (38)
	Speech-language	4 (15)
	Motor (PT or OT)	8 (31)
Number of co-occurring diagnoses		
	0	14 (53.8)
	1	3 (11.5)
	2	6 (23.1)
	3	1 (3.8)
	4	2 (7.7)
Type of co-occurring diagnoses		
	ADD/ADHD	9 (34.6)
	Developmental coordination disorder	2 (7.7)
	Developmental delay	6 (23.1)
	Giftedness	1 (3.8)
	Intellectual disability	2 (7.7)
	Learning disability	3 (11.5)
	Selective mutism	1 (3.8)
	Tourette syndrome	2 (7.7)

ABA, Applied Behavior Analysis; IBI, Intensive Behavioral Intervention; PT, Physiotherapy; OT, Occupational Therapy; ADD/ADHD, Attention Deficit Disorder/Attention Deficit Hyperactivity Disorder.

TABLE 2 | Motor coordination and adaptive behavior scores.

Variable		Mean (SD)
MABC-2 percentile scores		
	Manual dexterity	3.8 (5.6)
	Aiming & catching	19.4 (27.4)
	Balance	13.5 (22.2)
	Total test	6.3 (13.9)
VABS-2 adaptive behavior standard scores		
	Communication	79.6 (16.2)
	Daily living skills	83.8 (16.4)
	Socialization	77.9 (14.9)
	Adaptive behavior composite	78.9 (13.4)
VABS-2 maladaptive behavior v-scale scores		
	Internalizing	20.5 (2.2)
	Externalizing	18.4 (2.4)
	Maladaptive behavior index	20.1 (1.9)

MABC-2, Movement Assessment Battery for Children, 2nd Edition; VABS-2, Vineland Adaptive Behavior Scales, 2nd Edition.

TABLE 3 | Spearman's rank order correlations between MABC-2 and VABS-2 scores.

VABS-2	MABC-2			
	Manual dexterity	Aiming & catching	Balance	Total score
Communication	0.34	0.31	0.44	0.42
Daily living skills	0.58*	0.29	0.47	0.60*
Socialization	0.34	0.17	0.34	0.45
Adaptive behavior composite	0.53	0.25	0.45	0.57*

**Correlation significant at $p < 0.003$. MABC-2, Movement Assessment Battery for Children, 2nd Edition; VABS-2, Vineland Adaptive Behavior Scales, 2nd Edition.*

DISCUSSION

The results of this study confirm the presence of significant delays in motor coordination and adaptive behavior experienced by school-aged children with ASD and add to our understanding of how these behavioral domains are related to one another. Specifically, we found that manual dexterity, or fine motor coordination, is strongly related to better functioning in daily living skills and better overall motor coordination was related to better daily living skills and overall adaptive behavior. These findings support previous work that has found positive associations between fundamental movement skills and social communicative skills (MacDonald et al., 2013), and better manual dexterity and overall motor coordination related to less social impairment in children with ASD (Hirata et al., 2014). However, the importance of manual dexterity for daily living skills and more generalized adaptive behavior was previously underexplored in school-aged children with ASD.

Motor development is so closely entwined with a child's adaptive behavior that fine and gross motor skills are included in the VABS-2 up until the age of 6 years (Sparrow et al., 2005). It is clear however, that the motor domain does not cease to influence adaptive behavior beyond the developmental period, and in fact it may be even more important for school-aged children with ASD as they also exhibit significant motor delays. Hirata et al. (2014) postulate several potential mechanisms linking motor coordination to other areas of behavior and function (specifically social impairments). First, the association may reflect an underlying neurocognitive deficit affecting regions of the brain that govern both motor control, social behavior and cognitive function. Indeed, in the area of DCD, researchers have postulated a pervasive, minimal brain dysfunction as a possible mechanism to explain the high rates of co-morbidity in children with motor coordination problems, particularly in relation to attention and executive functioning (Kaplan et al., 1998; Dewey et al., 2002). The correlation observed here may simply reflect a general disruption in brain function that affects multiple domains simultaneously. For example, both functional and anatomical disturbances in neural connectivity have been implicated in the behavioral characteristics of individuals with ASD (Belmonte, 2004; Schipul et al., 2011), with abnormal cerebellar activity being one common indicator of dysfunction (Belmonte, 2004; Mostofsky et al., 2009). The cerebellum plays an important role in motor development and coordination, and increasing focus is being placed on the interrelationships between the cerebellum and brain regions responsible for cognitive tasks, such as the prefrontal cortex (Diamond, 2000, 2007; Leisman et al., 2016). One could therefore expect that cerebellar dysfunction is not only implicated in the motor impairments associated with ASD but, also in the cognitive and executive dysfunctions associated with the disorder. Moreover, the cerebellum is also responsible for automatization processes, or the ability to complete a task with little conscious attention. The automatization deficit hypothesis, wherein individuals have difficulty automatizing their motor and cognitive behaviors, was initially used as a paradigm for children with developmental dyslexia (Fawcett and Nicolson, 1992), but has more recently been applied to children with DCD (Visser, 2003; Schott et al., 2016). These authors speculate that challenges in automatization resulting from cerebellar dysfunction are part of the neuropathology of DCD and provide rationale for the frequency of co-occurring developmental concerns in areas such as attention and learning (Visser, 2003). It is likely then, that an automatization deficit may also help to explain the motor impairments experienced by children with ASD, in addition to the co-occurrence of social and attentional difficulties.

Another explanation concerns the relationship between manual dexterity specifically and other domains of function (Hirata et al., 2014). It seems reasonable that a child who performs better in manual tasks will also score higher on self-care behaviors and other aspects of daily living, given the importance of these motoric skills for everyday tasks (e.g., combing hair, dressing, eating). With regard to communication, manual dexterity is important for school-based activities (printing, art work), so children with better skills in this domain may also have greater opportunity for developing interpersonal and communicative skills, interacting with teachers and other peers. Of course, it will be necessary to track the longitudinal development of both manual dexterity and adaptive behaviors in children with ASD to untangle possible causal linkages.

It is also important to consider the heterogeneity in ASD and how this may affect movement skill. Previous research has shown mixed results when it comes, for example, to the association between manual dexterity, vs. other domains of motor coordination, and other developmental outcomes (MacDonald et al., 2013; Hirata et al., 2014). Hirata et al. (2014) hypothesize that this in turn may be related to social and/or cultural factors. For example, in some countries, focusing on the development of manual skills through practice may be considered more important by parents than other skills. It is also plausible that the mixed results seen in previous research is simply due to the heterogeneity of the disorder itself. ASD is complex, characterized by behavior, and is often accompanied by co-occurring diagnoses (Gillberg and Fernell, 2014). It is therefore likely that the motor domain will have a varying impact, behaviorally, on children with ASD depending on the remainder of their needs and attributes. Interestingly, we found much greater variability in the motor scores of our participants than in the adaptive behavior scores. This may be reflective of a larger variability in motor functioning across children with ASD, compared to the generally pervasive deficits in adaptive behavior, or may be an artifact of the measures used. For example, the motor coordination test was a direct assessment, however, adaptive behavior was parent-reported. We generally expect to see greater variability in direct assessments as they are dependent on the child's performance that day, their understanding of how to perform the tasks, and their behavior. In contrast, parents are rating their child's adaptive behavior based on multiple experiences over time, thus providing a more stable rating that is reflective of on-going challenges in the behavioral domain. Regardless, the variability in motor functioning of individuals with ASD, and how it relates to different behavioral domains, is an important area of inquiry in need of further research.

Future research should continue to disentangle the positive relationship between motor coordination and adaptive behavior found in this study, particularly given the importance of adaptive behavior for obtaining positive outcomes in children with ASD (Kanne et al., 2011). We also propose that the efficacy of motor-based interventions for improving adaptive behavior in this population be explored. Traditional behavioral and social-communicative interventions for children with ASD are often time-intensive and expensive, particularly when implemented in a community or private setting (Chasson et al., 2007). In contrast, school-based interventions may be optimal for many children with ASD as they may provide greater ecological validity; however, limited effects have been shown (Bellini et al., 2007). School-based motor skill interventions may have the potential to improve areas of functioning beyond just the motor domain. However, it is likely that the impact of motor interventions will have a varying effect across behavioral profiles of ASD. Research into the design, delivery, and outcomes of such interventions is warranted, along with an examination of the behavioral characterization of responders and non-responders to this type of intervention.

Limitations of the current study include the small sample, which limits our ability to conduct more sophisticated statistical analyses. Another limitation is that the VABS-2 is parent-reported; ideally, we would have both subjective and objective measures of the participants' adaptive behaviors. Further, we are limited by our lack of measure of IQ, which may influence the relationship between the MABC-2 and VABS-2 found in our study. Lastly, the cross-sectional design of this study does not allow us to infer causation or disentangle the relationships between motor coordination and adaptive behavior. Future studies on this topic should employ prospective longitudinal designs and include a heterogeneous sample of participants with ASD. Strengths of the design include administration of a standardized test of motor coordination, which is also the criterion measure used to assess for DCD and other motor impairments. The multiple domains of the MABC-2 allowed us to examine interrelationships across subdomains of both motor and adaptive behaviors.

CONCLUSION

The results of this study highlight the profound challenges with motor coordination and adaptive behavior experienced by school-aged children with ASD, and suggest that these challenges are interrelated. It is possible that the interrelatedness of these domains is due to the broader neurocognitive deficits present in children with ASD, however, longitudinal neuroimaging and behavioral studies are needed to disentangle these relationships.

ETHICS STATEMENT

This study was carried out in accordance with the recommendations of Hamilton Integrated Research Ethics Board with written informed consent from all parents of participating children. All parents of participating children gave written informed consent in accordance with the Declaration of Helsinki. All child participants 8 years of age and older gave written informed assent.

AUTHOR CONTRIBUTIONS

EB designed the study, coordinated recruitment and data collection, carried out the data analyses, and drafted the initial manuscript. JC supervised the design and execution of all phases of the study and revised and approved the final manuscript as submitted. Both authors approved the final manuscript as submitted and agreed to be accountable for all aspects of the work.

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REFERENCES

American Psychiatric Association. (2013). *Diagnostic and Statistical Manual of Mental Disorders (DSM-5)*, 5th Edn. Washington, DC: American Psychiatric Publishing. doi: 10.1176/appi.books.9780890425596

Bellini, S., Peters, J. K., Benner, L., and Hopf, A. (2007). A meta-analysis of school-based social skills interventions for children with autism spectrum disorders. *Remedial Spec. Educ.* 28, 153–162. doi: 10.1007/s10803-015-2373-1

Belmonte, M. K. (2004). Autism and abnormal development of brain connectivity. *J. Neurosci.* 24, 9228–9231. doi: 10.1523/JNEUROSCI.3340-04.2004

Bhat, A. N., Landa, R. J., and Galloway, J. C. C. (2011). Current perspectives on motor functioning in infants, children, and adults with autism spectrum disorders. *Phys. Ther.* 91:1116. doi: 10.2522/ptj.20100294

Caçola, P., Miller, H. L., and Williamson, P. O. (2017). Behavioral comparisons in autism spectrum disorder and developmental coordination disorder: a systematic literature review. *Res. Autism Spectr. Disord.* 38, 6–18. doi: 10.1016/j.rasd.2017.03.004

Cairney, J. (2015). *Developmental Coordination Disorder and its Consequences.* Toronto, ON: University of Toronto Press.

Cairney, J., and King-Dowling, S. (2016). "Developmental Coordination Disorder," in *Comorbid Conditions Among Children with Autism Spectrum Disorders*, ed. J. L. Matson (Cham: Springer International Publishing), 303–322. doi: 10.1007/978-3-319-19183-6_13

Chasson, G. S., Harris, G. E., and Neely, W. J. (2007). Cost comparison of early intensive behavioral intervention and special education for children with autism. *J. Child Fam. Stud.* 16, 401–413. doi: 10.1007/s10826-006-9094-1

Dewey, D., Kaplan, B. J., Crawford, S. G., and Wilson, B. N. (2002). Developmental coordination disorder: associated problems in attention, learning, and psychosocial adjustment. *Hum. Mov. Sci.* 21, 905–918. doi: 10.1016/S0167-9457(02)00163-X

Diamond, A. (2000). Close interrelation of motor development and cognitive development and of the cerebellum and prefrontal cortex. *Child Dev.* 71, 44–56. doi: 10.1111/1467-8624.00117

Diamond, A. (2007). Interrelated and interdependent. *Dev. Sci.* 10, 152–158. doi: 10.1111/j.1467-7687.2007.00578.x

Fawcett, A. J., and Nicolson, R. I. (1992). Automatisation deficits in balance for dyslexic children. *Percept. Mot. Skills* 75, 507–529. doi: 10.2466/pms.1992.75.2.507

Fournier, K. A., Hass, C. J., Naik, S. K., Lodha, N., and Cauraugh, J. H. (2010). Motor coordination in autism spectrum disorders: a synthesis and meta-analysis. *J. Autism Dev. Disord.* 40, 1227–1240. doi: 10.1007/s10803-010-0981-3

Georgiades, S., Szatmari, P., and Boyle, M. (2013a). Importance of studying heterogeneity in autism. *Neuropsychiatry* 3, 123–125. doi: 10.2217/npy.13.8

Georgiades, S., Szatmari, P., Boyle, M., Hanna, S., Duku, E., Zwaigenbaum, L., et al. (2013b). Investigating phenotypic heterogeneity in children with autism spectrum disorder: a factor mixture modeling approach: ASD factor mixture model. *J. Child Psychol. Psychiatry* 54, 206–215. doi: 10.1111/j.1469-7610.2012.02588.x

Geschwind, D. H. (2009). Advances in autism. *Annu. Rev. Med.* 60, 367–380. doi: 10.1146/annurev.med.60.053107.121225

Gillberg, C., and Fernell, E. (2014). Autism plus versus autism pure. *J. Autism Dev. Disord.* 44, 3274–3276. doi: 10.1007/s10803-014-2163-1

Haywood, K., and Getchell, N. (2014). *Life Span Motor Development*, 6th Edn. Champaign, IL: Human Kinetics.

Henderson, S., Sugden, D., and Barnett, A. (2007). *Movement Assessment Battery for Children*, 2nd Edn. London: Pearson Education.

Hirata, S., Okuzumi, H., Kitajima, Y., Hosobuchi, T., Nakai, A., and Kokubun, M. (2014). Relationship between motor skill and social impairment in children with autism spectrum disorders. *Int. J. Dev. Disabil.* 60, 251–256. doi: 10.1179/2047387713Y.0000000033

IBM Corporation (2017). *IBM SPSS statistics for windows*. Armonk, NY: IBM Corporation.

Kanne, S. M., Gerber, A. J., Quirmbach, L. M., Sparrow, S. S., Cicchetti, D. V., and Saulnier, C. A. (2011). The role of adaptive behavior in autism spectrum disorders: implications for functional outcome. *J. Autism Dev. Disord.* 41, 1007–1018. doi: 10.1007/s10803-010-1126-4

Kaplan, B. J., Wilson, B. N., Dewey, D., and Crawford, S. G. (1998). DCD may not be a discrete disorder. *Hum. Mov. Sci.* 17, 471–490. doi: 10.1016/S0167-9457(98)00010-4

Leisman, G., Moustafa, A., and Shafir, T. (2016). Thinking, walking, talking: integratory motor and cognitive brain function. *Front. Public Health* 4:94. doi: 10.3389/fpubh.2016.00094

Leonard, H. C., and Hill, E. L. (2014). Review: the impact of motor development on typical and atypical social cognition and language: a systematic review. *Child Adolesc. Ment. Health* 19, 163–170. doi: 10.1111/camh.12055

Lloyd, M., MacDonald, M., and Lord, C. (2013). Motor skills of toddlers with autism spectrum disorders. *Autism* 17, 133–146. doi: 10.1177/1362361311402230

MacDonald, M., Lord, C., and Ulrich, D. A. (2013). The relationship of motor skills and social communicative skills in school-aged children with autism spectrum disorder. *Adapt. Phys. Act. Q.* 30, 271–282. doi: 10.1123/apaq.30.3.271

Mostofsky, S. H., Powell, S. K., Simmonds, D. J., Goldberg, M. C., Caffo, B., and Pekar, J. J. (2009). Decreased connectivity and cerebellar activity in autism during motor task performance. *Brain* 132, 2413–2425. doi: 10.1093/brain/awp088

Ozonoff, S., Young, G. S., Goldring, S., Greiss-Hess, L., Herrera, A. M., Steele, J., et al. (2008). Gross motor development, movement abnormalities, and early identification of autism. *J. Autism Dev. Disord.* 38, 644–656. doi: 10.1007/s10803-007-0430-0

Payne, V., and Isaacs, L. (2011). *Human Motor Development: A Lifespan Approach*, 8th Edn. New York, NY: McGraw-Hill.

Schipul, S. E., Keller, T. A., and Just, M. A. (2011). Inter-regional brain communication and its disturbance in autism. *Front. Syst. Neurosci.* 5:10. doi: 10.3389/fnsys.2011.00010

Schmidt, R. C., Fitzpatrick, P., Caron, R., and Mergeche, J. (2011). Understanding social motor coordination. *Hum. Mov. Sci.* 30, 834–845. doi: 10.1016/j.humov.2010.05.014

Schott, N., El-Rajab, I., and Klotzbier, T. (2016). Cognitive-motor interference during fine and gross motor tasks in children with Developmental Coordination Disorder (DCD). *Res. Dev. Disabil.* 57, 136–148. doi: 10.1016/j.ridd.2016.07.003

Sparrow, S. S., Balla, D. A., and Cicchetti, D. V. (2005). *Vineland Adaptive Behavior Scales*, 2nd Edn. Bloomington, MN: PsychCorp.

Visser, J. (2003). Developmental coordination disorder: a review of research on subtypes and comorbidities. *Hum. Mov. Sci.* 22, 479–493. doi: 10.1016/j.humov.2003.09.005

Wiggins, L. D., Robins, D. L., Adamson, L. B., Bakeman, R., and Henrich, C. C. (2012). Support for a Dimensional view of autism spectrum disorders in toddlers. *J. Autism Dev. Disord.* 42, 191–200. doi: 10.1007/s10803-011-1230-0

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CHAPTER 4: The influence of adaptive behavior on the pathways connecting motor competence and health-related fitness in children with autism spectrum disorder

Preamble

The influence of adaptive behavior on the pathways connecting motor competence and health-related fitness in children with autism spectrum disorder is the third study in the dissertation. The study examines the associations between motor competence, physical activity, and health-related fitness by mapping on to a common model of motor competence for children with typical development. Further, this study proposes a novel modification to this model, wherein adaptive behaviour moderates the relationships between motor competence and physical activity and health-related fitness, respectively.

The manuscript is currently under review for publication in the *Journal of Autism and Developmental Disabilities*. The word document version of the manuscript (formatted according to the *Journal of Autism and Developmental Disabilities* author guidelines) is included in the dissertation.

Contribution of Study 3 to the overall dissertation

Study 3 found a positive association between motor competence and health-related fitness, with adaptive behaviour moderating this relationship in children with ASD. However, no significant associations with physical activity were present. These findings emphasize the importance of the relationship between motor competence and adaptive behaviour for the physical health of this population. Further, they underscore the need to explore the role of physical activity for other areas of functioning in children with ASD. Overall, study 3 provides support for two of the pathways (motor competence – health-related fitness and motor competence – adaptive behaviour) proposed in the Interdisciplinary Framework presented in Chapter 1.

Abstract

The purpose of this study was to examine the pathways connecting motor competence, physical activity, and health-related fitness, in addition to the influence of adaptive behavior on these pathways, in a sample of 7-12 year old children with ASD (n=27). Results indicated that motor competence and health-related fitness were positively related ($r=.420$, $p<.05$); however, no other significant pathways were present. Moreover, the relationship between motor competence and health-related fitness was moderated by adaptive behavior, wherein the significant relationship was only present for those participants scoring approximately one standard deviation below the mean on adaptive behavior. Implications of these associations and directions for future research are discussed.

The development of proficient motor skills has important implications for the physical and mental health of children and youth (Lubans, Morgan, Cliff, Barnett, & Okely, 2010). For example, motor skills are positively associated with participation in physical activity, physical fitness, perceived competence, a healthy body composition, and mental health outcomes (Lubans et al., 2010). In 2008, Stodden and colleagues proposed a developmental model of motor skill wherein proficient motor skills led to a positive spiral of engagement in physical activity (Stodden et al., 2008). This model was recently updated to reflect the current state of the evidence regarding the strength of the proposed associations (Robinson et al., 2015). The updated model provides extensive evidence for a positive relationship between motor competence and physical activity, and between motor competence and health-related fitness. There is also extensive evidence of an inverse relationship between motor competence and weight status (Robinson et al., 2015), in addition to a positive relationship between perceived motor competence and physical activity and fitness, respectively. Lastly, the authors propose, albeit without extensive testing, that the relationship between motor competence and physical activity is mediated by health-related fitness (Robinson et al., 2015). Taken together, the evidence for the updated model suggests that intervening in the motor domain will have a positive influence on each of the other variables in the model.

While this model has been very influential in the study of children and youth with typical development, the application of this model to clinical populations, such as children with neurodevelopmental disorders, has been limited (King-Dowling, Rodriguez, Missiuna, Timmons, & Cairney, 2018). Neurodevelopmental disorders include autism spectrum disorder (ASD), developmental coordination disorder (DCD), and attention deficit hyperactivity disorder (ADHD), among others (American Psychiatric Association, 2013). Many of these disorders are either associated with, or characterized by, poor motor performance (American Psychiatric Association, 2013; Cairney, 2015; Fournier, Hass, Naik, Lodha, & Cauraugh, 2010; Kaiser, Schoemaker, Albaret, & Geuze, 2015). For example, in the case of DCD, the diagnosis is provided to individuals who experience deficits in motor skill that significantly and persistently interfere with activities of daily living; the onset of these deficits must appear in the early developmental period and cannot be better explained by an intellectual disability, visual impairment, or neurological condition (American Psychiatric Association, 2013). In the case of ADHD, a disorder not defined by motor deficits, we also see pervasive delays in gross and fine motor skill when compared to typically developing peers (Kaiser et al., 2015). Moreover, many researchers estimate that upwards of 50% of children with ADHD could also be classified as DCD (Kaiser et al., 2015). Research in the area of DCD does support some of the predictions in the developmental model of motor skill. For example, DCD is associated with low levels of physical activity and physical fitness (Rivilis et al., 2011), with significant discrepancies in fitness between children with DCD and typical development appearing as young as preschool (King-Dowling et al., 2018). Moreover, children with DCD have lower self-perceptions (Cairney, 2015) and have been found to have a higher body mass index (BMI) and waist circumference than their typically developing peers (Cairney et al., 2010). Taken together, research into DCD provides support for much of the developmental model of motor skill, wherein low motor competence is related to lower levels of perceived competence, physical activity and fitness, and higher weight status in this population.

Unlike DCD, less work has explored these associations in children with ASD, yet ASD is a prevalent childhood disorder with significant impacts to health and quality of life (Jones, Bremer, & Lloyd, 2017; Lee, Harrington, Louie, & Newschaffer, 2008). Research suggests that children with ASD are less active and less fit (Tyler, MacDonald, & Menear, 2014), and exhibit a higher incidence of overweight and obesity (Healy, Aigner, & Haegele, 2018), when compared to their peers with typical development. Given the significant motor delays experienced by this population (Fournier et al., 2010), it is natural to hypothesize that intervening in the motor domain will positively influence physical activity, fitness, and body composition in this population. Indeed, these relationships may be even more important for a child with ASD as they have some of the greatest room for improvement in these areas. One previous study found a positive relationship between motor coordination and fitness in 10-17 year olds with ASD (Pan, 2014). These authors also found a negative relationship between percent body fat (but not BMI) and strength and agility in this same sample (Pan, 2014); thus, providing partial support for the developmental model of motor skill. Yet, although many of the variables included in the developmental model are occasionally measured in the same study, we are unaware of any further research that has examined the associations between these variables in a sample of children with ASD.

Beyond the motor delays experienced by children with ASD, this is a population who also experiences significant challenges with their daily behaviours, inherent to their core challenges in the social communicative domain (Kanne et al., 2011). The ability of a child with ASD to functionally participate in daily activities can be represented by their level of adaptive behavior. Adaptive behavior is positively related to motor skill in children with ASD (Bremer & Cairney, 2018). As such, we hypothesize that the individual relationships between motor skill and physical activity, and motor skill and physical fitness will be moderated by adaptive behavior in children with ASD. That is, the strength of the relationship will be predicated on the presence of a certain level of adaptive behavior that is required for meaningful participation in the motor domain to result in a benefit to physical activity and fitness. If, for example, a child with

ASD does not have a prerequisite level of adaptive behavior, their participation in motor skill-based activities may not be meaningful, or deliberate enough, to elicit an association with physical activity or fitness. We are therefore proposing a modification to the developmental model of motor skill (Robinson et al., 2015; Stodden et al., 2008) where adaptive behavior will moderate the relationships between motor competence and fitness, and motor competence and physical activity, respectively (Figure 1). We hypothesize that this modification will make the model more relevant for children with ASD, as it will take into account their behavioral functioning.

Therefore, the purpose of this study is twofold: 1) to examine the associations proposed in the developmental model of motor skill (Robinson et al., 2015; Stodden et al., 2008) in a sample of children with ASD; and 2) to test a modification to the model wherein adaptive behavior moderates the relationship between motor competence and physical activity, and motor competence and fitness, respectively, in this population.

Method

Participants and Procedure

A cross-sectional design was used to explore the associations between motor competence, physical fitness, physical activity, and weight status, in addition to the moderating role of adaptive behaviour, in a sample of 7-12 year old children with ASD. Participants were primarily recruited through a mail out to families of children with ASD on a wait list for services at a government-funded children's treatment centre, in addition to advertisements placed at local community centres and on social media. Inclusion criteria required that participants be between 7 and 12 years of age with a verbal confirmation of the child's diagnosis provided by their parent or guardian. In the province of Ontario, Canada where this study took place, a diagnosis of ASD can be made by a family physician, pediatrician, psychiatrist, psychologist, or a psychological associate. Participants were excluded from participating in the study if they had a co-occurring physical disability, or could not safely engage in the assessments due to physical or behavioral concerns. All participants attended four appointments at the researcher's lab, with data collected during the last three visits. Visit 1 was a familiarization to the lab, exercise equipment, and research team. Visit 2 assessed motor competence, along with the completion of behavioral questionnaires completed by the participant's parent. Visits 3 and 4 assessed weight status and fitness. The data in this study was collected as part of a larger study that also examined the feasibility and reliability of fitness assessments in children with ASD (Bremer & Cairney, Under review). Accordingly, approximately 50% of the participants participated in an additional two study appointments (Visits 5-6) for further fitness testing, from which the data is reported elsewhere (Bremer & Cairney, Under review). This study was approved by the Institutional Research Ethics Board, all parents provided written informed consent, and participants 8 years of age and older provided informed assent prior to their participation in the study.

Measures

Demographic information and adaptive behavior

Parents provided pertinent demographic information for their child, including age of diagnosis and diagnosis provider. Parents also completed the Vineland Adaptive Behavior Scales-2 (VABS-2) Parent/Caregiver Report Form (Sparrow, Balla, & Cicchetti, 2005) .The adaptive behavior composite score was used to describe the sample and in the moderation analyses. The composite score incorporates the domains of communication, daily living skills, and socialization. The score is age-normed, has a mean of 100, and a standard deviation of 15 (Sparrow et al., 2005).

Motor competence

Participants' motor competence was assessed with the Movement Assessment Battery for Children-2 (MABC-2) (Henderson, Sugden, & Barnett, 2007). The MABC-2 is commonly used to identify movement difficulties in children, and provides scores in the domains of manual dexterity, aiming and catching, and balance, in addition to a total test score. The MABC-2 consists of three age bands (3-6 years, 7-10 years, and 11-16 years), therefore, the two older age bands were used in this study. The total test score is converted into a percentile based on the child's age and sex, with a percentile score at or below the

16th percentile indicating the presence of significant movement difficulties (Henderson et al., 2007).

Health-related fitness

Health-related fitness was assessed using tests of anaerobic power, muscular power, muscular strength, and flexibility. The Wingate anaerobic cycling test (Bar-Or, 1987) provided a measure of anaerobic fitness and was assessed using a pediatric cycle ergometer (LODE Pediatric, Groningen, The Netherlands). Participants established maximum pedaling speed through completion of a brief (i.e., 20-second) sprint. After a brief rest, they completed a second sprint with a braking force (0.55 Nm/kg) applied once 80% of their maximum pedaling speed was reached. Peak power output (in watts) was obtained from the LODE Wingate software package (LODE BV, Groningen, The Netherlands) and was considered the primary outcome from the assessment.

Standing long jump provided a measure of lower body muscular power (Castro-Piñero et al., 2010) and required the participant to jump as far as they could using a two-footed take-off and landing. The furthest distance jumped (in cm) was considered the primary outcome.

Grip strength was measured using a digital handgrip dynamometer (Smedley Pro Digital, Japan), and was considered an indicator of total muscular power (Wind, Takken, Helders, & Engelbert, 2010). Participants completed two trials per hand, with the primary outcome being the highest value recorded (in kg) for the dominant hand.

Lastly, the sit and reach test (Wells & Dillon, 1952) was completed as a measure of flexibility. Participants sat on the floor with their legs straight and feet flat against the sit and reach box (Flex-Tester, Creative Health Products, USA); with hands together, they reached forward as far as they could, with the distance reached (in cm) recorded as the primary outcome. Each of the included fitness assessments demonstrated good- to- excellent test-retest reliability and a high degree of test feasibility in our sample (Bremer & Cairney, Under review).

Physical Activity

Physical activity was assessed using an Actigraph accelerometer (wGT3X, Pensacola, Florida) provided to the participants at Visit 2. Participants were instructed to wear the accelerometer on their right hip during all waking hours for 7 consecutive days, only removing the device for water activities (e.g., swimming, bathing) and to sleep. Parents completed a logbook to record the times at which the device was put on and taken off, including whether it was removed at all during the day. Data was analysed in 3 second epochs and non-wear periods were defined as any time the parent indicated the monitor was not worn and/or 60 minutes of consecutive zero counts. At least 10 hours of wear time per day on at least 3 days was considered sufficient for inclusion in the analyses. Evenson cut-points were applied to the data to determine the average daily minutes in sedentary, light, moderate, and vigorous physical activity (Evenson, Catellier, Gill, Ondrak, & McMurray, 2008; Trost, Loprinzi, Moore, & Pfeiffer, 2011). Average daily minutes spent in moderate-to-vigorous physical activity (MVPA) was considered the primary outcome from the accelerometer data. All cleaning and processing of the accelerometer data was completed using Actilife software (Actigraph, Pensacola, Florida).

Body composition

Height was measured to the nearest 0.1 cm using a Seca SC264 stadiometer (Chino, California). Weight was measured to the nearest 0.1 kg using a Seca SC869 digital scale (Chino, California). BMI was calculated and BMI classifications were made using the IOTF cut-points for children (Cole & Lobstein, 2012).

Statistical Analysis

Missing data was imputed using single mean imputation for individual fitness, motor, and physical activity variables. The primary outcome from each of the four fitness assessments was converted to a T-score (with a mean of 50 and a standard deviation of 10) and summed to create a composite health-related fitness score. Descriptive statistics were calculated for demographic characteristics and the primary outcomes. The first analysis consisted of Pearson product correlations to assess the relationship between motor competence and fitness, physical activity, and BMI, respectively. Second, we tested whether fitness mediated the association between motor competence and physical activity. Third, we tested whether adaptive behaviour moderated the relationship between motor competence and fitness, and motor competence and physical activity, respectively. All statistical analyses were completed in SPSS version 25 (IBM Corporation, 2017). The PROCESS macro for SPSS was used to run the mediation and moderation analyses (Hayes & Rockwood, 2017). The mediation employed a bootstrapping procedure with 5,000 samples to estimate the 95% confidence intervals of the indirect effect. Moderation analyses with an interaction term that was significant at $p<.10$ were probed using the Johnson-Neyman technique to explore the conditional effect of the focal predictor at different values of the moderator. With a sample size of 27, our regression models were deemed sufficient, considering the recommendations of Austin and Steyerberg (2015) who found that a minimum of two subjects per variable was sufficient for the adequate estimation of regression coefficients, standard error, and confidence intervals.

Results

Participants (n=27) were 9.9 (SD=1.7) years of age and predominately (88.9%) male. Close to half of the participants (48.1%) had at least one co-occurring diagnosis, with the most common diagnosis being ADHD, which was diagnosed in 9 participants. Just over half (51.9%) of the sample was classified as normal weight, with the remaining participants classified as underweight (3.7%), overweight (29.6%), and obese (14.8%).

Overall, missing variables of health-related fitness, physical activity, and motor competence scores accounted for less than 10% of all data. The most common missing variable was physical activity, with 18.5% (5/27 participants) missing data on this variable. The primary outcome of each measure is presented in Table 1. On average, the results indicate the presence of significant challenges in the domains of motor skill and adaptive behaviour, along with physical activity levels below the recommended 60 minutes of MVPA per day (Tremblay et al., 2016).

In regard to our first research question, we found that motor competence was positively related to our composite score of health-related fitness. No other significant correlations were present between our primary outcomes (Table 2). Next, we tested whether fitness mediated the association between motor competence and physical activity and found that the indirect effect was small and non-significant [effect (SE) = -0.09 (0.17), 95% CI = -0.53 to 0.15].

To test our second research question, we ran two separate moderation analyses to examine the effect of adaptive behaviour on the relationship between motor competence and health-related fitness, and motor competence and physical activity, respectively. Results indicate that adaptive behavior moderates the association between motor competence and fitness, although this effect was not statistically significant at $p<.05$ [coefficient (SE) = -0.077 (0.041), 95% CI = -0.162 to 0.009, $t = -1.857$, $p = .076$]. However, we did observe a conditional effect of the focal predictor indicating a region of significance ($p<.05$) was present at a VABS-2 score of 89.1 and below. Specifically, we see that motor competence is related to health-related fitness only among participants scoring relatively low in adaptive behavior (i.e., one standard deviation or more below the standardized mean; $\Theta_{X \rightarrow Y|W=85} = .0965$, $p<.05$). There is however no statistically significant association between motor competence and health-related fitness for participants scoring at or above the mean in adaptive behavior ($\Theta_{X \rightarrow Y|W=100} = -.0183$, $p>.05$). Lastly, adaptive behavior did not moderate the association between motor competence and physical activity [coefficient (SE) = -0.046 (0.032), 95% CI = -0.112 to 0.021, t = -1.428, p =.167].

Discussion

Our results provide partial support for the application of the developmental model of motor skill for school-aged children with ASD. Specifically, we found that motor competence is associated with health-related fitness in this population. However, contrary to the model, we did not find associations between motor competence and BMI, or motor competence and physical activity, nor did health-related fitness mediate this relationship. Novel to this study, we proposed a modification to the model with adaptive behaviour moderating the associations between motor competence and fitness, and motor competence and physical activity, respectively (see Figure 1). Findings from this study partially support our hypothesis in that adaptive behaviour was found to moderate the relationship between motor competence and fitness; however, it did not moderate an association between motor competence and physical activity. Interestingly, the point of significance of this moderation was at an adaptive behaviour score of 89.1 (i.e., just over one standard deviation below the mean), indicating that the association between motor competence and fitness is significant at a relatively low level of behavioral functioning. Moreover, the majority (77.8%) of participants in our sample had an adaptive behavior score below this threshold. The implications of this association are far from trivial: fitness is a significant independent predictor of morbidity and mortality (Fogelholm, 2010; Janssen & LeBlanc, 2010) and should be considered an important aspect of overall health. In fact, individuals with ASD are at a 2.5-fold increased risk of all-cause mortality in comparison to their peers with typical development, including an elevated risk of mortality from both circulatory and respiratory conditions (Hirvikoski et al., 2016). As such, we would suggest that improving the fitness of individuals with ASD should be a top priority for their health. Moreover, the presence of a positive relationship between fitness and motor competence, particularly at low levels of adaptive behaviour, would suggest that we may be able to intervene in these domains, through for example a motor skill intervention, to produce meaningful health benefits for children with ASD.

That we did not find an association between motor competence and physical activity may be due to the general lack of variability in our physical activity data, as most participants were considered physically inactive with an average of 48.7 minutes of MVPA per day. Interestingly, 6 participants (22.2%) did meet the recommended 60 minutes of MVPA per day in our study, suggesting that the sample was not exclusively inactive. However, it is possible that these minutes of physical activity would not represent quality participation, rather excess movement due to hyperactivity or stereotyped behaviors (i.e., behaviors that do not require a high degree of motor competence), which may help to explain the lack of association in our data between physical activity and motor competence or fitness. It is also likely that there are other, unmeasured, variables accounting for participation in physical activity. For instance, participants' enjoyment of physical activity, their motivation, and their self-efficacy to engage in physical activity may be even more important for the age and social stages of our participants. Moreover, many physical activities for school-aged children are social; therefore, an additional challenge is added for children with ASD who, inherent to their diagnosis, experience challenges with social interactions. Previous qualitative research has suggested a number of individual and environmental factors related to participation in physical activity, including competence and confidence, motivation, adjustment to external demands, predictability, and freedom of choice (Arnell, Jerlinder, & Lundqvist, 2017). It is important that future work consider these factors, and how they relate to motor competence, fitness, and behavior in children with ASD. From an intervention standpoint, this means we likely need to focus our attention on improving more than just motor skills in order to get children with ASD more physically active. Tailoring programs to the individual interests of children with ASD and building their self-efficacy to engage in various activities is likely necessary. Physical literacy may therefore provide an appropriate model for physical activity programming for children with ASD. Such programming would need to intentionally target the competence, confidence, motivation, and knowledge and understanding of participants in order to shift physical activity levels in a positive direction (Dudley, Cairney, Wainwright, Kriellaars, & Mitchell, 2017). However, more research is needed, in the form of both prospective longitudinal studies and randomized trials, in order to better understand how we can increase physical activity levels in this population.

One limitation to the current study is that our composite fitness score did not include a measure of aerobic fitness. Results from our reliability study (Bremer & Cairney, Under review) indicated that neither of our aerobic fitness measures (a progressive treadmill test and six minute walk test) demonstrated acceptable test-retest reliability in our sample, therefore those measures were excluded from this study. A second limitation is that we did not measure perceived competence, which is another variable in the Stodden model (Robinson et al., 2015; Stodden et al., 2008). However, we are unaware of any validated measures of perceived competence for children with ASD. Lastly, given our study population and challenges with participant recruitment, our sample size was small. Previous research on the accuracy and stability of estimated regression coefficients in OLS regression models, such as those used in our moderation analyses, suggests that a minimum of at least two subjects per variable produces unbiased estimates of coefficients and confidence intervals (Austin & Steyerberg, 2015). Our models included three predictors, therefore suggesting a minimum total N of at least 6 cases, which was exceeded by our sample of N=27. Further, our sample size generally aligns with other studies that include direct assessments of physical activity and fitness in children with ASD, in which we see sample sizes in the range of 15 to 30 participants (Pan, 2011, 2014; Tyler et al., 2014). Our study is strengthened by the inclusion of direct measures of motor competence, health-related fitness, physical activity, and weight status within the same study. We are unaware of previous studies that have included all of these measures within a single study for children with ASD. Moreover, our findings are further strengthened by the inclusion of fitness tests shown to be reliable and feasible in our study population (Bremer & Cairney, Under review).

In conclusion, our findings demonstrate a positive association between motor competence and health-related fitness in children with ASD. Further, our results support one of our proposed modifications to Stodden and colleagues' model

(Robinson et al., 2015; Stodden et al., 2008), with adaptive behavior moderating the association between motor competence and fitness, but not motor competence and physical activity. In contrast to work with children with typical development, we did not find associations between motor competence and physical activity or BMI. Future research should use prospective longitudinal cohorts and experimental trials to further disentangle these variables in children and youth with ASD. Moreover, employing mixed-methods designs may further add to our understanding of physical activity participation in this population, which may support the development of appropriate strategies to increase physical activity levels.

References

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (DSM-5)* (5th ed.). Washington, DC: American Psychiatric Publishing.

Arnell, S., Jerlinder, K., & Lundqvist, L.-O. (2017). Perceptions of Physical Activity Participation Among Adolescents with Autism Spectrum Disorders: A Conceptual Model of Conditional Participation. *Journal of Autism and Developmental Disorders*. https://doi.org/10.1007/s10803-017-3436-2

Austin, P. C., & Steyerberg, E. W. (2015). The number of subjects per variable required in linear regression analyses. *Journal of Clinical Epidemiology*, *68*(6), 627–636. https://doi.org/10.1016/j.jclinepi.2014.12.014

Bar-Or, O. (1987). The Wingate Anaerobic Test: An Update on Methodology, Reliability and Validity. *Sports Medicine*, *4*(6), 381–394. https://doi.org/10.2165/00007256-198704060-00001

Bremer, E., & Cairney, J. (Under review). Reliable and feasible fitness testing for children on the autism spectrum. *Research Quarterly for Exercise and Sport*.

Bremer, E., & Cairney, J. (2018). The interrelationship between motor coordination and adaptive behavior in children with autism spectrum disorder. *Frontiers in Psychology*. https://doi.org/10.3389/fpsyg.2018.02350

Cairney, J. (2015). *Developmental coordination disorder and its consequences*. Toronto, ON: University of Toronto Press.

Cairney, J., Hay, J., Veldhuizen, S., Missiuna, C., Mahlberg, N., & Faught, B. E. (2010). Trajectories of relative weight and waist circumference among children with and without developmental coordination disorder. *Canadian Medical Association Journal*, *182*(11), 1167–1172. https://doi.org/10.1503/cmaj.091454

Castro-Piñero, J., Ortega, F. B., Artero, E. G., Girela-Rejón, M. J., Mora, J., Sjöström, M., & Ruiz, J. R. (2010). Assessing muscular strength in youth: Usefulness of standing long jump as a general index of muscular fitness. *Journal of Strength and Conditioning Research*, *24*(7), 1810–1817. https://doi.org/10.1519/JSC.0b013e3181ddb03d

Cole, T. J., & Lobstein, T. (2012). Extended international (IOTF) body mass index cut-offs for thinness, overweight and obesity: Extended international BMI cut-offs. *Pediatric Obesity*, *7*(4), 284–294. https://doi.org/10.1111/j.2047-6310.2012.00064.x

Dudley, D., Cairney, J., Wainwright, N., Kriellaars, D., & Mitchell, D. (2017). Critical Considerations for Physical Literacy Policy in Public Health, Recreation, Sport, and Education Agencies. *Quest*, 1–17. https://doi.org/10.1080/00336297.2016.1268967

Evenson, K. R., Catellier, D. J., Gill, K., Ondrak, K. S., & McMurray, R. G. (2008). Calibration of two objective measures of physical activity for children. *Journal of Sports Sciences*, *26*(14), 1557–1565. https://doi.org/10.1080/02640410802334196

Fogelholm, M. (2010). Physical activity, fitness and fatness: relations to mortality, morbidity and disease risk factors. A systematic review. *Obesity Reviews*, *11*(3), 202–221. https://doi.org/10.1111/j.1467-789X.2009.00653.x

Fournier, K. A., Hass, C. J., Naik, S. K., Lodha, N., & Cauraugh, J. H. (2010). Motor Coordination in Autism Spectrum Disorders: A Synthesis and Meta-Analysis. *Journal of Autism and Developmental Disorders*, *40*(10), 1227–1240. https://doi.org/10.1007/s10803-010-0981-3

Hayes, A. F., & Rockwood, N. J. (2017). Regression-based statistical mediation and moderation analysis in clinical research: Observations, recommendations, and implementation. *Behaviour Research and Therapy*, *98*, 39–57. https://doi.org/10.1016/j.brat.2016.11.001

Healy, S., Aigner, C. J., & Haegele, J. A. (2018). Prevalence of overweight and obesity among US youth with autism spectrum disorder. *Autism*, 1–5. https://doi.org/10.1177/1362361318791817

Henderson, S., Sugden, D., & Barnett, A. (2007). *Movement Assessment Battery for Children* (2nd ed). London, UK: Pearson Education.

Hirvikoski, T., Mittendorfer-Rutz, E., Boman, M., Larsson, H., Lichtenstein, P., & Bölte, S. (2016). Premature mortality in autism spectrum disorder. *British Journal of Psychiatry*, *208*(03), 232–238. https://doi.org/10.1192/bjp.bp.114.160192

IBM Corporation. (2017). IBM SPSS statistics for windows (Version 25). Armonk, NY: IBM Corporation.

Janssen, I., & LeBlanc, A. G. (2010). Systematic review of the health benefits of physical activity and fitness in school-aged children and youth. *International Journal of Behavioral Nutrition and Physical Activity*, *7*(1), 1.

Jones, S., Bremer, E., & Lloyd, M. (2017). Autism spectrum disorder: family quality of life while waiting for intervention services. *Quality of Life Research*, *26*(2), 331–342. https://doi.org/10.1007/s11136-016-1382-7

Kaiser, M.-L., Schoemaker, M. M., Albaret, J.-M., & Geuze, R. H. (2015). What is the evidence of impaired motor skills and motor control among children with attention deficit hyperactivity disorder (ADHD)? Systematic review of the literature. *Research in Developmental Disabilities*, *36*, 338–357. https://doi.org/10.1016/j.ridd.2014.09.023

Kanne, S. M., Gerber, A. J., Quirmbach, L. M., Sparrow, S. S., Cicchetti, D. V., & Saulnier, C. A. (2011). The Role of Adaptive Behavior in Autism Spectrum Disorders: Implications for Functional Outcome. *Journal of Autism and Developmental Disorders*, *41*(8), 1007–1018. https://doi.org/10.1007/s10803-010-1126-4

King-Dowling, S., Rodriguez, C., Missiuna, C., Timmons, B. W., & Cairney, J. (2018). Health-related Fitness in Preschool Children with and without Motor Delays. *Medicine & Science in Sports & Exercise*, 1. https://doi.org/10.1249/MSS.0000000000001590

Lee, L.-C., Harrington, R. A., Louie, B. B., & Newschaffer, C. J. (2008). Children with Autism: Quality of Life and Parental Concerns. *Journal of Autism and Developmental Disorders*, *38*(6), 1147–1160. https://doi.org/10.1007/s10803-007-0491-0

Lubans, D. R., Morgan, P. J., Cliff, D. P., Barnett, L. M., & Okely, A. D. (2010). Fundamental movement skills in children and adolescents. *Sports Medicine*, *40*(12), 1019–1035.

Pan, C.-Y. (2011). The efficacy of an aquatic program on physical fitness and aquatic skills in children with and without autism spectrum disorders. *Research in Autism Spectrum Disorders*, *5*(1), 657–665. https://doi.org/10.1016/j.rasd.2010.08.001

Pan, C.-Y. (2014). Motor proficiency and physical fitness in adolescent males with and without autism spectrum disorders. *Autism*, *18*(2), 156–165. https://doi.org/10.1177/1362361312458597

Rivilis, I., Hay, J., Cairney, J., Klentrou, P., Liu, J., & Faught, B. E. (2011). Physical activity and fitness in children with developmental coordination disorder: A systematic review. *Research in Developmental Disabilities*, *32*(3), 894–910. https://doi.org/10.1016/j.ridd.2011.01.017

Robinson, L. E., Stodden, D. F., Barnett, L. M., Lopes, V. P., Logan, S. W., Rodrigues, L. P., & D'Hondt, E. (2015). Motor Competence and its Effect on Positive Developmental Trajectories of Health. *Sports Medicine*, *45*(9), 1273–1284. https://doi.org/10.1007/s40279-015-0351-6

Sparrow, S. S., Balla, D. A., & Cicchetti, D. V. (2005). *Vineland Adaptive Behavior Scales, Second Edition*. Bloomington, MN: PsychCorp.

Stodden, D. F., Goodway, J. D., Langendorfer, S. J., Roberton, M. A., Rudisill, M. E., Garcia, C., & Garcia, L. E. (2008). A developmental perspective on the role of motor skill competence in physical activity: An emergent relationship. *Quest*, *60*(2), 290–306.

Tremblay, M. S., Carson, V., Chaput, J.-P., Connor Gorber, S., Dinh, T., Duggan, M., … Zehr, L. (2016). Canadian 24-Hour Movement Guidelines for Children and Youth: An Integration of Physical Activity, Sedentary

Behaviour, and Sleep. *Applied Physiology, Nutrition, and Metabolism, 41*(6 (Suppl. 3)), S311–S327. https://doi.org/10.1139/apnm-2016-0151

Trost, S. G., Loprinzi, P. D., Moore, R., & Pfeiffer, K. A. (2011). Comparison of accelerometer cut points for predicting activity intensity in youth. *Medicine & Science in Sports & Exercise, 43*(7), 1360–1368. https://doi.org/10.1249/MSS.0b013e318206476e

Tyler, K., MacDonald, M., & Menear, K. (2014). Physical Activity and Physical Fitness of School-Aged Children and Youth with Autism Spectrum Disorders. *Autism Research and Treatment, 2014*, 1–6. https://doi.org/10.1155/2014/312163

Wells, K. F., & Dillon, E. K. (1952). The sit and reach—a test of back and leg flexibility. *Research Quarterly. American Association for Health, Physical Education and Recreation, 23*(1), 115–118.

Wind, A. E., Takken, T., Helders, P. J. M., & Engelbert, R. H. H. (2010). Is grip strength a predictor for total muscle strength in healthy children, adolescents, and young adults? *European Journal of Pediatrics, 169*(3), 281–287. https://doi.org/10.1007/s00431-009-1010-4

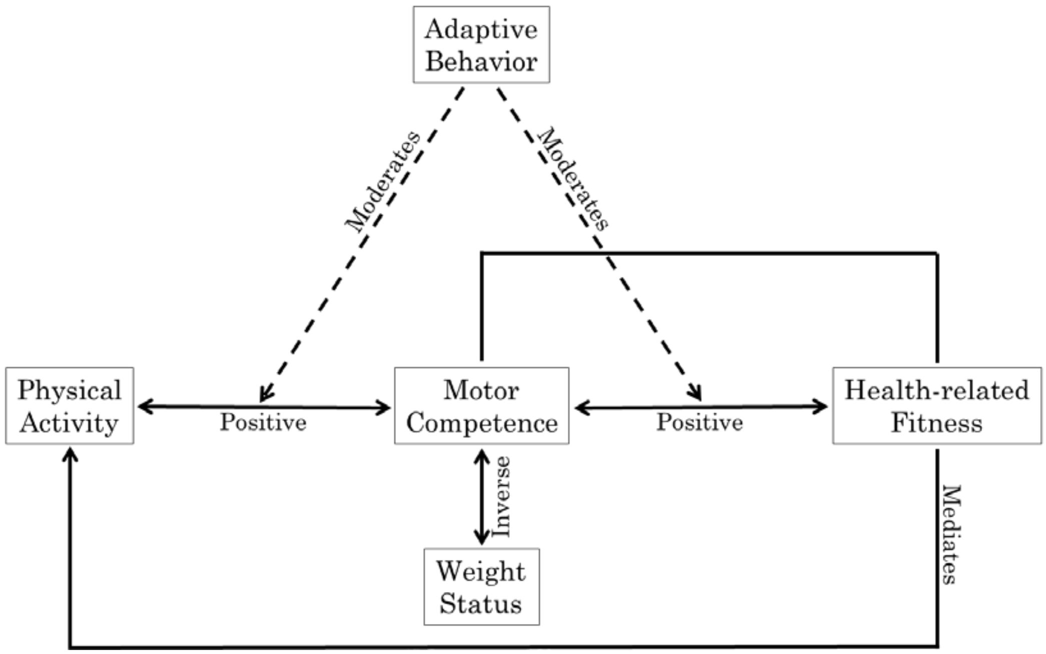

Figure 1. Effect modification model of the influence of adaptive behavior on the pathways connecting motor competence to physical activity and health-related fitness.

Note: Solid lines represent associations present in Stodden and colleagues' model (Robinson et al., 2015; Stodden et al., 2008), while the dashed lines represent our proposed modification to the model.

Table 1. Descriptive statistics of the primary outcomes.

Outcome	Statistic Mean (SD)
MABC-2 total test score (percentile)	6.3 (13.7)
Physical activity (minutes MVPA/day)	48.7 (17.1)
VABS-2 adaptive behavior composite (standard score)	78.0 (13.8)
BMI (kg/m^2)	19.1 (3.7)
Wingate (peak power)	228.9 (71.9)
Standing long jump (cm)	104.9 (32.1)
Grip strength (kg)	11.8 (3.1)
Sit and reach (cm)	18.3 (5.2)

Note: MABC-2=Movement Assessment Battery for Children, 2nd Edition; MVPA=Moderate-to-Vigorous Physical Activity; VABS-2=Vineland Adaptive Behavior Scales, 2nd Edition; BMI=Body Mass Index.

Table 2. Pearson's correlations between the primary outcomes.

	1	2	3
1. Motor competence			
2. Health-related fitness	**.420**		
3. Physical activity	.079	- .109	
4. BMI	- .125	.045	- .027

Note: Bold font indicates the correlation is significant at p<.05; BMI=Body Mass Index.

CHAPTER 5: The acute effect of exercise on executive functioning and cerebral blood flow regulation in children with autism spectrum disorder: An exploratory study

Preamble

The acute effect of exercise on executive functioning and cerebral blood flow regulation in children with autism spectrum disorder: An exploratory study is the fourth study in the dissertation. The study explores the effects of two different types of exercise, compared to a sedentary control, on executive functions and cerebral blood flow regulation in children with ASD.

The manuscript is currently under review for publication in the *International Journal of Psychophysiology*. The word document version of the manuscript (formatted according to the *International Journal of Psychophysiology* author guidelines) is included in the dissertation.

<u>Contribution of Study 4 to the overall dissertation</u>
Study 4 provided preliminary support for the positive effect of an acute bout of exercise on executive functioning and cerebral blood flow regulation in children with ASD. Findings also suggest that a circuit-based interval exercise session may elicit greater improvements to these areas than more traditional aerobic exercise. These results provide support for the physical activity – cognition pathway proposed in the Interdisciplinary Framework presented in Chapter 1.

Abstract

The purpose of this study was to examine the acute effect of exercise on executive functions and cerebral blood flow regulation in children with ASD. A secondary question was to assess potential intermediary physiological and psychological mechanisms for change. Participants (N=12) were 8-12 years of age with a diagnosis of ASD. A within-subject crossover design was employed. Participants completed three 20-minute conditions on separate days: circuit-based workout, treadmill walking, and sedentary control. Pre- and post- each condition participants completed a cancellation task as a measure of inhibitory control. Changes in cerebral blood flow regulation were assessed using functional near-infrared spectroscopy (fNIRS) during the task and measures of heart rate, affect, perceived exertion, motivation, and self-efficacy were taken throughout the experiment. A series of repeated measures ANOVAs were conducted to examine intervention effects. Results demonstrated a non-significant interaction effect for executive functions (p = .226, η_p^2 = .086) and cerebral blood flow regulation (p = .252, η_p^2 = .109). Post-hoc analyses revealed that the circuit condition elicited the largest changes in both executive functions (d = 0.75, p = .080) and cerebral blood flow regulation (d = 0.66, p = .171). Effects on the psychological outcomes were small and non-significant. Although more research is needed, these findings suggest that exercise may be a feasible intervention for enhancing executive functioning and cerebral blood flow regulation in children with ASD.

1.1 Introduction

Autism spectrum disorder (ASD) is a neurodevelopmental disorder that results in challenges in the areas of social communication and social interactions, along with the presence of restricted or repetitive behaviours (American Psychiatric Association, 2013). While the etiology of ASD is complex and includes both genetic and environmental factors (Currenti, 2010), much work has also focused on the widespread disrupted neural connectivity that is present in the disorder (Belmonte, 2004; Schipul, Keller, & Just, 2011; Wass, 2011). Brain imaging research, for example, points to connectivity issues between the frontal cortex and other brain regions, which is thought to contribute to the behavioural characteristics of ASD (Vissers, Cohen, & Geurts, 2012; Wass, 2011). Further, the prefrontal cortex, which continues to develop through childhood, is thought to be particularly impacted by these connectivity issues (Just, Cherkassky, Keller, Kana, & Minshew, 2007).

Parallel to the core features of ASD, and in line with the widespread neural disruptions, previous research has also demonstrated significant impairments in executive functioning among this population (Demetriou et al., 2018). Executive functions are higher-order cognitive processes, originating in the prefrontal cortex, that enable us to control and direct our behaviour (Diamond, 2013). Executive functions include three core processes including inhibition (i.e., self-control or interference control), working memory, and cognitive flexibility (i.e., set shifting or mental flexibility) (Lehto, Juujärvi, Kooistra, & Pulkkinen, 2003; Miyake et al., 2000). These skills are essential for virtually all aspects of life including physical and mental health, academic and vocational success, and social development (Diamond, 2013). Thus, impairments in executive functioning can have a debilitating impact across multiple aspects of functioning.

Interventions for children with ASD typically focus on addressing the behavioural challenges associated with the disorder through, for example, intensive behavioural interventions employing applied behaviour analysis strategies (e.g., positive reinforcement, antecedent-behaviour-consequence techniques). However, there has been a limited amount of interventions that directly address the deficits in executive functioning, rather than the behaviours associated with these deficits. One example of a successful intervention targeting executive functions for children with ASD employed cognitive behavioural strategies in a school-based program and found significant gains in the areas of problem solving, flexibility, and planning (Kenworthy et al., 2014). Yet, the potential mechanisms behind these changes remain unknown.

In contrast, exercise is a form of intervention that has been gaining traction as a way to improve aspects of executive functioning in children. While the results of recent reviews are mixed (Diamond & Ling, 2018; Gunnell et al., 2018; Hillman & Biggan, 2017), the guiding principal is that both acute and chronic exercise results in a number of physiological and psychological changes that, in turn, elicit improvements in brain-based processes (Lambourne & Tomporowski, 2010; Stimpson, Davison, & Javadi, 2018). While traditional forms of aerobic exercise (e.g., running) has been the focus of much of this research (Gunnell et al., 2018; Pontifex et al., 2019), the effects of exercise on executive functions may not be equal across exercise types. For example, previous research has suggested that cognitively engaging activities (i.e., physical activity paired with cognitive/mental effort) may elicit greater improvements in executive functioning than aerobic exercise alone (Best, 2010; Diamond & Ling, 2018; Schmidt, Jäger, Egger, Roebers, & Conzelmann, 2015). Further, resistance training has received a lot less attention in the literature, but has demonstrated positive results on measures of inhibition (Soga, Masaki, Gerber, & Ludyga, 2018). There are a number of potential mechanisms driving the effects of exercise on cognition. For example, increased arousal which leads to a cascade of factors including activation of the reticular activating system and the release of catecholamines and neurotrophic factors (McMorris, 2016; Pontifex et al., 2019; Voss, Vivar, Kramer, & van Praag, 2013). Further, exercise may result in an increase in cerebral blood flow regulation, resulting in improved cognitive processing through an increase in resource availability (i.e., availability of oxygenated hemoglobin) and waste clearing (i.e., removal of deoxygenated hemoglobin) (Ogoh & Ainslie, 2009). There are also numerous intermediary psychological mechanisms that may be driving changes in executive functioning following exercise. For example, exercise can lead to improved affect (Ekkekakis, Parfitt, & Petruzzello, 2011), which may then impact performance on later cognitive tasks. Further, self-efficacy to perform cognitive tasks may also be improved through changes in both affect and physiological states following exercise (Bandura, 1997).

Despite the impairments in executive functioning experienced by children with ASD, and the potential for exercise to reduce these impairments, little work in this area has focused specifically on children with ASD. There is evidence of behavioural improvements in the areas of social-emotional functioning and a reduction in repetitive behaviours following exercise in children with ASD, with these improvements hypothesized to be related to improvements in executive functioning (Bremer, Crozier, & Lloyd, 2016). Further, a recent meta-analysis of the effect of exercise interventions on cognition reported a large effect for improvements in specific aspects of cognition (e.g., time on task) for children with ASD (Tan, Pooley, & Speelman, 2016). Our interpretation of this effect is, however, limited as it was derived from only 6 studies and only 1 of these studies (Anderson-Hanley, Tureck, & Schneiderman, 2011) actually included measures of executive functioning as an outcome. Since this review, two additional studies have shown positive changes in executive functions in children with ASD following 12-week table tennis and basketball interventions, respectively (Pan et al., 2017; Tse et al., 2019). Additionally, Memari and colleagues (Memari et al., 2017) found a significant positive correlation between cognitive flexibility and physical activity in children with ASD; although, the cross-sectional design of the study limits our understanding of causality in this relationship. Taken together, these results suggest there is potential for exercise to improve executive functions in children with ASD. The current literature, however, has failed to test potential intermediary variables that may account for changes in executive functions, as well as the differential effects of varying types of exercise on executive functions in this population.

This exploratory study examined the effect of two different types of acute exercise (treadmill walking and a circuit-based interval workout), in comparison to a sedentary control condition, on executive functioning and cerebral blood flow regulation in children with ASD. We hypothesized that both exercise conditions would result in greater improvements to both variables than the sedentary control condition. As a secondary research question, we also explored changes in potential intermediary psychological (self-efficacy, motivation, affect, and perceived exertion) and physiological (heart rate) variables.

2.1 Method

2.2 Participants and Design

Participants were 12 males with ASD between 8-12 years of age (M_{age} = 11.1, SD = 1.3 years). While recruitment was open to both sexes, only male participants enlisted in the study. Eligibility criteria included no co-occurring physical health conditions (e.g., unstable heart condition) that would preclude participants from safely engaging in physical activity and the ability to follow dual-step instructions, as reported by the participants' parent. The study utilized a within-subject randomized experimental design with one independent variable (group, consisting of three levels – treadmill walking, circuit-based interval workout, and sedentary control) and two primary dependent measures (change in executive function task performance and change in cerebral blood flow regulation). The study was approved by the Hamilton Integrated Research Ethics Board, parents provided informed written consent, and participants provided informed written assent prior to participation in the study.

2.3 Experimental Manipulation

Participants completed three experimental conditions, in a random order, separated by a minimum of 48 hours. Randomization was determined using a random number generator (randomizer.org) prior to study entry. The manipulations included two exercise conditions (steady-state and interval) and one sedentary control condition. Each condition was 20 minutes in length. Heart rate was recorded continuously during each condition using a Polar V800 heart rate monitor and chest strap, with a target heart rate for the exercise conditions of 120-160 beats per minute [bpm; approximately 60-80% of predicted maximal heart rate (Gelbart, Ziv-Baran, Williams, Yarom, & Dubnov-Raz, 2017)].

2.3.1 Treadmill walking. Treadmill walking was chosen as a steady-state, aerobic exercise as this type of exercise is commonly employed in the acute exercise-cognition literature (Pontifex et al., 2019). The condition consisted of walking on a treadmill with the speed and incline starting at 1.7 mph and 10%, respectively, in order to meet target heart rate zones. The speed and incline were adjusted, if needed, to maintain the target heart rate.

2.3.2 Circuit-based interval workout. The circuit-based interval exercise condition included five exercises, completed in a circuit, for a total of 3 sets. This condition was chosen to represent a more ecologically valid type of workout, and included exercises that could be completed at home or in school or clinical settings. The exercises included jumping jacks, medicine ball (2 lb) chest press, squat jumps, seated row with a light resistance band, and alternating step-ups on an exercise step. Each exercise was completed for 45 seconds, followed by a 20 second rest and transition to the next exercise. A 2-minute rest was provided between each set. The timing of the exercises and transitions between activities were guided through a commercially available iPad application (Seconds Pro Interval Timer, Runloop Ltd.). Participants were verbally encouraged to maintain a steady pace throughout the exercises in order to stay in the target heart rate zone.

2.3.3 Sedentary control. The sedentary control condition consisted of watching an age-appropriate children's movie while seated in a chair in the exercise room. Considered a cognitive engagement control, this is the most commonly used control condition in the acute exercise cognition literature

(Pontifex et al., 2019). Further, this was considered a typical activity in which a child with ASD may engage in their free time (Jones et al., 2017).

2.4 Primary Outcome Measures

2.4.1 Executive functioning. Executive functioning was assessed with the Attention Sustained subtest of the Leiter International Performance Scale, 3rd Edition. The Leiter-3 is an individually administered standardized test used to assess aspects of intellectual ability and executive functioning in individuals aged 3-75 years, and has been validated for individuals with ASD (Roid et al., 2013). The Attention Sustained subtest measures inhibitory control through a cancellation task that requires the participant to cross out as many target items on a page as they can in a given period of time. The subtest includes four trials, with the first two trials lasting 30 seconds and the second two trials lasting 60 seconds. Each of the four trials is completed on a separate page and has a different target item for the participant to find. Each page is full of items similar to the target and the participant needs to cross out only the target items while inhibiting the desire to cross out similar, non-target, items. Further, the four trials progress in difficulty by including previous target items in subsequent trials, increasing the complexity of the target items, and moving from a linear to random arrangement of the items on the page. Prior to each of the four trials, participants are given a practice trial on a miniature version of the task to ensure task understanding. Scores are calculated as the total number of target items correctly crossed out, minus the total number of commission errors. The total number of target items for each of the four trials is 32, 64, 69, and 52, respectively, with the highest total score (sum of trials 1-4) being 217 (Roid, Miller, Pomplun, & Koch, 2013). The subtest was completed twice (pre- and post-experimental manipulation) per study appointment. The change in total score from pre- to post-test was considered the primary outcome of the study.

2.4.2 Cerebral blood flow regulation. A 20-channel, continuous wave, functional near-infrared spectroscopy (fNIRS) system (NIRSport, NIRx Medical Technologies) was used to measure the relative change in oxygenated hemoglobin (oxy-Hb) within the prefrontal cortex during the executive functioning task. The system is comprised of 8 sources and 8 detectors, resulting in 20 channels with a sampling frequency of 7.81 Hz. The optodes were placed in the cap based on the standard 10-20 international system (Jasper, 1958) with the goal of acquiring the hemodynamic measures from the prefrontal cortex. After placing the optodes in the cap, a thick black over-cap was used to cover the optodes in order to avoid interference from environmental light.

The raw fNIRS data was recorded continuously during the executive functioning task using the NIRStar acquisition software. The raw data was then processed offline with nirsLAB. First, the task onsets and rest periods were defined. Signal-to-noise performance was evaluated using variation coefficients (CV), which is the standard deviation divided by the mean from all of the raw data in the measurement time series, by channel, and expressed as a percentage (Kenville, Maudrich, Carius, & Ragert, 2017; Piper et al., 2014) . In line with previous literature, channels were removed when the CV exceeded 15% (Kenville et al., 2017; Piper et al., 2014). A digital band-pass filter with a low cut-off frequency of 0.0033 Hz and a high cut-off frequency of 0.02 Hz was applied to the raw data to remove high-frequency noise and cardiovascular artifacts. Hemodynamic states were then computed and the data was exported to Microsoft Excel for post-processing analyses. The mean oxy-Hb across all channels was calculated for each of the four blocks of the executive functioning task. Change from pre- to post-test in the average oxy-Hb across all four blocks (in µmol) was considered the primary outcome from this variable.

2.5 Secondary Outcome Measures and Manipulation Checks

2.5.1 Affect. The Feeling Scale (Hardy & Rejeski, 1989) was used to measure affective states at baseline, following each of the four executive functioning trials and at four-minute intervals throughout the experimental manipulation. The Feeling Scale is an 11-point bipolar single-item scale that ranges from -5 (*very bad*) to +5 (*very good*) along a displeasure-pleasure continuum. The average of the four affect ratings following the executive function trials was considered as a potential psychological mechanism of change, while the average of the five ratings during the experimental manipulation was considered a manipulation check.

2.5.2 Ratings of perceived mental exertion. Participants rated their perceived mental exertion (RPME) using an adapted version of Borg's CR-10 scale (Borg, 1998) following each of the four executive functioning trials and at four minute intervals throughout the experimental manipulation. Participants were instructed to rate their perceived mental exertion from 0 (no exertion at all) to 12 (absolute maximum), with 10 (extremely strong) representing the highest mental exertion they had ever experienced. The average of the four RPME ratings following the executive function trials was considered as a potential psychological mechanism of change, while the average of the five ratings during the experimental manipulation was used as a manipulation check.

2.5.3 Ratings of perceived physical exertion. Participants rated their perceived physical exertion (RPE) using Borg's CR-10 scale (Borg, 1998), at four minute intervals throughout the experimental manipulation in order to determine the extent to which they were physically exerting themselves during each of the three conditions. Participants were instructed to rate their perceived physical exertion from 0 (no exertion at all) to 12 (absolute maximum), with 10 (extremely strong) representing the highest physical exertion they had ever experienced. The average of the five ratings during the experimental manipulation was used as a manipulation check.

2.5.4. Intrinsic motivation. Motivation for performing the executive function task was assessed at pre- and post-test using the effort and importance subscale from the Intrinsic Motivation Inventory (Ryan, 1982). The subscale consists of five items that are rated on a Likert scale ranging from 1 (*not at all true*) to 7 (*very true*). An example item is *"I am going to put a lot of effort into these brain games"*. A total motivation score was calculated as the mean of the five items and change from pre- to post- test was considered a potential psychological mechanism of change.

2.5.5 Task self-efficacy. Self-efficacy to perform the executive functioning task was assessed once (i.e., prior to performing the post-test) per appointment, using a four-item, 11-point, scale adhering to recommendations by Bandura (Bandura, 1997, 2006) for measuring self-efficacy. Each item was prefaced with the stem *"For the brain games I'm about to do, I am confident I can perform*". The individual items represented gradations of performance that were relative to the participant's performance on the first executive function task (blocks 1-4). Using the 11-point scale, 0 (*not at all confident*) to 10 (*totally confident*), participants were asked to rate their confidence to perform (1) "*almost as good as the last time*", (2) "*as good as the last time*", (3) "*a little better than the last time*", and (4) "*a lot better than the last time*" when compared to their pre-test. The scores from each of these four items were averaged to create a task self-efficacy score ranging from 0 to 10.

2.5.6 Heart rate. Heart rate was measured continuously during the experimental conditions using a Polar H1 chest strap synced to a Polar V800 watch (Polar Canada, Lachine, Quebec). Average heart rate, in beats per minute, over the 20-minute condition was used as a manipulation check of exercise intensity.

2.6 Sample Descriptive Characteristics

2.6.1 ASD severity. Parents completed the Gilliam Autism Rating Scale – Third Edition (GARS-3) (Gilliam, 2014). The GARS-3 consists of 56 items and takes 5-10 minutes to complete. It yields standard scores, percentile ranks, severity level, and probability of Autism and has been shown to have good test-retest reliability (>0.80) and able to accurately discriminate children with ASD from those without (sensitivity = 0.97, specificity = 0.97).

2.6.2 Trait executive functioning. Parents completed the Behaviour Rating Inventory of Executive Functions (BRIEF) (Gioia, Isquith, Guy, & Kenworthy, 2000). The questionnaire is appropriate for children aged 5 to 18 years and took approximately 10-15 minutes to administer. The instrument was parent-reported and assessed the following domains of executive functioning: inhibition, shifting, emotional control, working memory, and planning/organizing. Scores were totalled to give a Global Executive Composite. Parent test-retest reliability scores from the normative sample of 1,419 parents of children aged 5 to 18 is 0.82. Scores from the BRIEF were used to describe the sample and as a potential covariate in the analysis.

2.6.3 Repetitive behaviours. Parents completed the Repetitive Behavior Scale – Revised (RBS-R; Bodfish, Symons, & Lewis, 1999) at the first study appointment. The questionnaire is appropriate for children with ASD and has been independently validated in this population, showing high internal consistency and good interrater reliability (ICC = .70) (Lam & Aman, 2007). The instrument was parent-reported, took approximately 15 minutes to administer, and assesses the following domains of repetitive behaviours: Ritualistic/Sameness, Stereotypic, Self-injurious, Compulsive, and Restricted Interests. Scores from the RBS-R were used to describe the sample and as a potential covariate in the analysis.

2.6.4 Body composition. Height was measured to the nearest 0.1 cm using a Seca SC264 stadiometer (Chino, California) and weight was measured to the nearest 0.1 kg using a Seca SC869 digital scale (Chino, California). Body mass index (BMI) was calculated and participants' weight status was classified using the IOTF cut-points (Cole & Lobstein, 2012).

2.7 Procedure

Participants attended three appointments at the research lab, with each appointment lasting approximately 1 hour. At the beginning of the first study appointment, participants' parents provided informed written consent and participants provided informed assent. Parents then completed a suite of surveys including the demographic information form, GARS-3, BRIEF, and RBS-R.

Participants' height and weight were measured at the first study appointment. All remaining procedures were identical between the three study appointments.

Upon entering the lab, participants were fitted with a heart rate monitor and familiarized to the feeling scale and perceived exertion scales. Participants rated their affect and answered 5 questions regarding their intrinsic motivation to perform the executive functioning task. Participants were then fitted with an fNIRS cap, which was then calibrated for signal quality. Following calibration, baseline fNIRS and heart rate recordings (1 minute) were simultaneously taken prior to the start of the executive function task. Four trials of the executive function task were then completed with trials 1-2 each lasting 30 seconds and trials 3-4 each lasting 60 seconds. A brief (approximately 15 second) practice was provided prior to each of the four trials. Following each trial, participants provided ratings of affect and perceived mental exertion. fNIRS was recorded throughout all four trials, with the start and end points of each trial manually triggered in the NIRS program for later analysis.

Following the executive function task, participants completed one of the three 20-minute experimental conditions. Heart rate was recorded continuously throughout the condition, and ratings of affect, perceived mental exertion, and perceived physical exertion were provided at 4-minute intervals.

After the experimental manipulation, participants rated their task self-efficacy to perform the executive function task followed by ratings of intrinsic motivation and affect, as in the pre-test. The remainder of the protocol was then completed as described in the pre-test.

The remaining two appointments were completed following the same procedures as described above, with the experimental manipulation changing according to each participant's randomization schedule.

2.8 Statistical Analyses

All statistical analyses were conducted using SPSS 25 (IBM Corporation, 2017). Descriptive statistics were computed for all study variables. Separate repeated measures ANOVA models were computed to assess differences in executive functioning, cerebral oxygenation, affect, RPME, and motivation from pre- to post-test by condition. Separate one-way ANOVA models were used to assess differences in means between conditions for self-efficacy and the manipulation checks (heart rate, RPE, RPME, and affect during the experimental manipulation). The level of significance for the ANOVAs was $p = .05$, and planned post-hoc comparisons were conducted using a Bonferroni corrected *p*-value of .017 for each analyses (i.e., .05 divided by 3 follow-up t-tests per analysis).

Effect sizes for the repeated measure ANOVAs are reported as partial eta squared (η_p^2) and the values for small, medium, and large are 0.01, 0.06, and 0.14, respectively (Cohen, 1992). Effect sizes for the post-hoc analyses are reported as

Cohen's *d* and the values for small, medium, and large are 0.2, 0.5, and 0.8, respectively (Cohen, 1992).

2.8.1 Sensitivity analysis. Due to challenges with recruitment, our sample size was limited to 12 participants. Therefore, a post-hoc sensitivity analysis was conducted to determine the size of effect needed to reach statistical significance (G*Power version 3.1.9.2; Faul, Erdfelder, Buchner, & Lang, 2009). Based on our sample size of N = 12, with power = 0.80, and alpha = .05, a large effect of η_p^2 = 0.219 was necessary to achieve statistical significance.

3.1 Results

Demographic characteristics of the participants are presented in Table 1. The majority of participants (75%) had an autism severity rating of level 2 (i.e., requiring substantial support), while one participant was considered level 1 (i.e., requiring support) and two participants were considered level 3 (i.e., requiring very substantial support), respectively.

Table 1. Demographic characteristics of the participants.

Variable	Mean (SD)
Age	11.1 (1.3)
BMI	18.2 (2.5)
GARS Autism Index	90.0 (17.0)
BRIEF Global Executive Composite	71.1 (9.4)
RBS Total Score	29.4 (15.3)

3.2 Primary Analyses

Two repeated measures ANOVAs were conducted to examine the change in executive functioning and cerebral oxygenation, respectively, from pre- to post-test by experimental condition. In regard to executive functioning, results indicated a significant main effect for time [$F(1, 33) = 11.76$, $p = .002$, $\eta_p^2 = .263$] and a non-significant interaction effect [$F(2, 33) = 1.56$, $p = .226$, $\eta_p^2 = .086$]. All post-hoc analyses were non-significant at a *p*-value of .017. Effect sizes were, however, in the hypothesized direction and medium for the difference in change between the circuit and sedentary conditions ($d = 0.75$, $p = .080$); small between the treadmill and sedentary conditions ($d = 0.39$, $p = .345$); and small between the circuit and treadmill conditions ($d = 0.33$, $p= .431$). Change scores on the executive functioning task are presented in Figure 1.

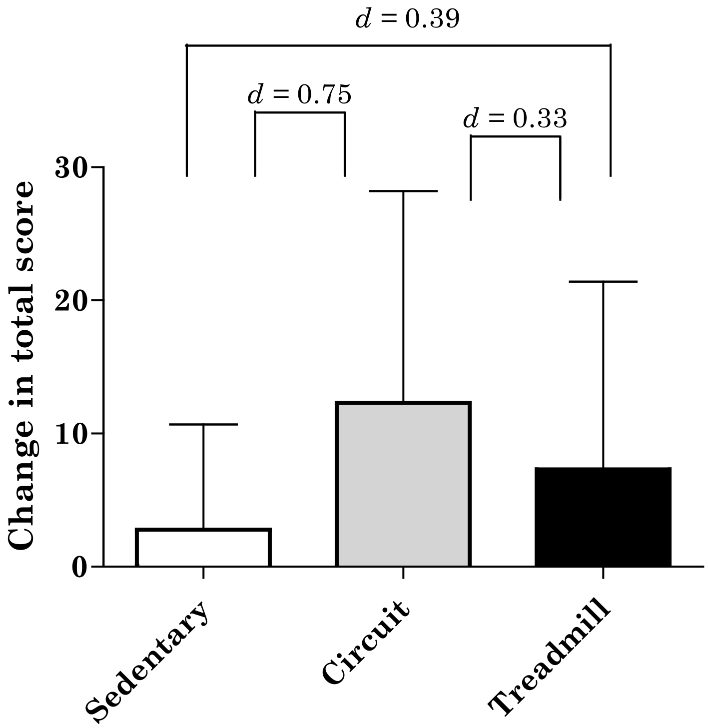

Figure 1. Change in executive functioning by condition.

In regard to changes in cerebral blood flow regulation, results indicated a non-significant main effect for time [$F(1, 24) = 0.55$, $p = .465$, $\eta_p^2 = .022$] and a non-significant interaction effect [$F(2, 24) = 1.46$, $p = .252$, $\eta_p^2 = .109$]. All post-hoc analyses were non-significant at a *p*-value of .017. Effect sizes for the difference in the change in oxy-Hb were in the hypothesized direction and medium between the circuit and sedentary conditions ($d = 0.66$, $p = .171$); small between the treadmill and sedentary conditions ($d = 0.05$, $p = .922$); and medium between the circuit and treadmill conditions (*d* = 0.71, *p* = .156). Change from pre- to post-test in oxy-Hb are presented in Figure 2.

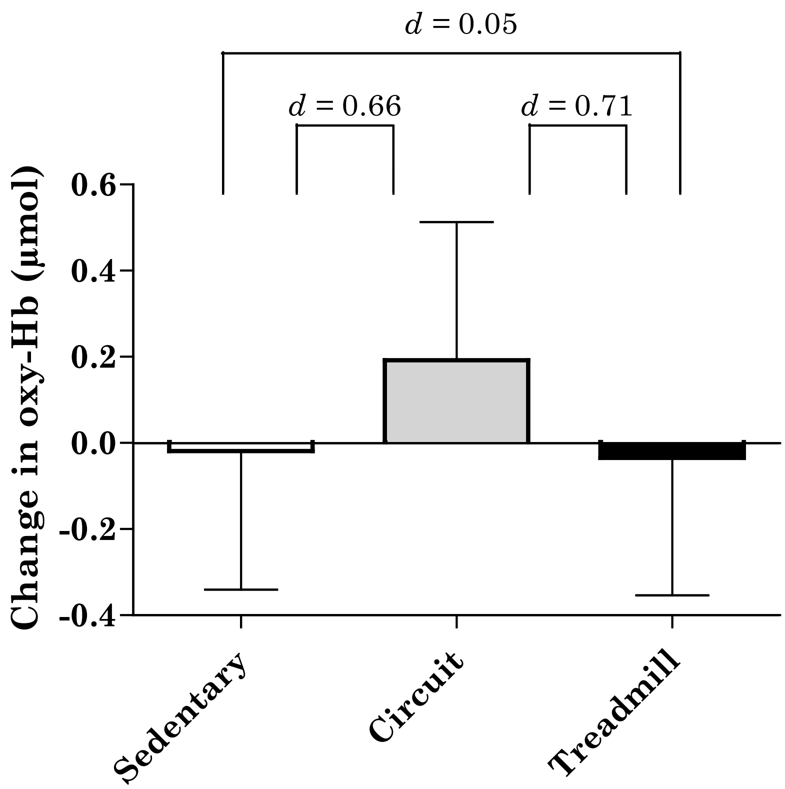

Figure 2. Change in cerebral oxygenation by condition.

3.3 Secondary Analyses

Descriptive statistics summarizing affect, perceived mental exertion, motivation and task self-efficacy scores are shown, by group, in Table 2. Three separate repeated measures ANOVAs were conducted to examine the change in affect and perceived mental exertion during the executive function task, along with changes in motivation to complete the executive functioning task from the pre- to post- test by condition. Results indicated that no significant differences between the conditions were present for affect, perceived mental exertion, or motivation. Finally, results from the one-way ANOVA to examine differences in self-efficacy to perform the executive function task between the three conditions indicated no significant differences between conditions. Given that these analyses were secondary, non-significant effects were not further explored through post-hoc analyses.

3.4 Manipulation Checks

Four separate one-way ANOVAs were conducted on average heart rate, affect, perceived physical exertion, and perceived mental exertion, respectively, during each of the experimental manipulations. Results are presented in Table 3 and indicate significant differences in heart rate and perceived physical exertion between the two exercise conditions and the control condition, respectively.

Table 2. Time by condition interaction effect for psychological outcomes.

Variable	**Sedentary** Mean (SD)	**Circuit** Mean (SD)	**Treadmill** Mean (SD)	**F**	***p*-value**
Task Affect (Δ pre- to- post- test)	0.9 (0.8)	0.1 (0.6)	1.0 (1.2)	2.77	.08
Task RPME (Δ pre- to- post- test)	-0.03 (1.6)	-0.6 (2.1)	0.7 (1.1)	1.72	.20
Task Motivation (Δ pre- to- post- test)	0.1 (0.9)	0.2 (1.2)	-0.1 (1.1)	1.00	.38
Task Self-efficacy	5.4 (2.1)	6.7 (2.9)	6.8 (3.0)	0.98	.39

Table 3. Experimental condition manipulation checks.

Variable	**Sedentary**	**Circuit**	**Treadmill**	**F**	***p*-value**
Heart Rate	89.2 (9.8)	131.5 (8.7)**	128.0 (10.2)**	63.98	<.001
Affect	2.4 (2.4)	3.5 (1.5)	3.2 (1.8)	0.87	.43
RPE	3.2 (3.5)	6.5 (2.4)*	5.8 (2.9)	3.66	.04
RPME	3.7 (3.6)	5.4 (3.1)	5.0 (3.2)	0.75	.48

* Significantly different from the sedentary condition at $p<.05$

** Significantly different from the sedentary condition at $p<.001$

4.1 Discussion

Overall, findings from this exploratory study suggest that exercise may be a promising modality to elicit changes in cerebral blood flow regulation and executive functioning in children with ASD, with effect sizes that were medium to large in size. While both exercise conditions elicited greater improvements than the control condition, the results also indicate that the circuit condition elicited some of the greatest changes in executive functioning and oxy-Hb, suggesting that not all exercise modalities are equal. While these results were not statistically significant, the magnitude of our effect sizes suggests that these changes are of practical significance (Ellis & Steyn, 2003; Sullivan & Feinn, 2012).

This is the first study to examine the acute effect of two different types of exercise on executive functioning, when compared to a sedentary control condition, in children with ASD. Importantly, the study utilized numerous manipulation checks in order to further discern the effect of exercise on our primary outcomes. Results from the manipulation checks indicated that exercise intensity was similar between both exercise conditions, with no differences in the participants' heart rate or perceived exertion; thus, it is likely that differences in executive functioning and cerebral blood flow regulation between the conditions may be due to mechanisms other than exercise intensity.

Previous research has demonstrated greater improvements in executive functioning following cognitively engaging physical activity when compared to traditional aerobic activity (Best, 2010; Diamond & Ling, 2018; Schmidt et al., 2015), which may be one possible reason for the differential effects between the circuit and treadmill conditions. Even though perceived mental exertion was similar between conditions, it is possible that the circuit condition required a greater degree of cognitive demand than the treadmill condition due to the need to switch between the different exercises and the more dynamic nature of each exercise in comparison to treadmill walking. Indeed, the exercises included in the circuit condition required a higher degree of motor coordination, given the full-body, dynamic nature of each of the five exercises. There is a high degree of co-activation between the cerebellum, responsible for motor coordination, and the prefrontal cortex, responsible for executive functions (Diamond, 2000). As such, tasks that require both motor and cognitive demands should inherently lead to greater co-activation of brain regions. It has further been proposed that the benefits of cognitively engaging physical activity can be attributed to a theory of "pre-activation" (Budde, Voelcker-Rehage, Pietraßyk-Kendziorra, Ribeiro, & Tidow, 2008; Schmidt, Benzing, & Kamer, 2016) , whereby the cognitive engagement of the physical activity primes the same cognitive processes needed for the later performance of cognitive tasks. This theory of pre-activation is supported by the fact that we saw greater improvements in executive functioning following the circuit exercise, which likely required a higher degree of cognitive engagement, in comparison to the treadmill exercise, which relied more on motor processes alone. Further, we saw an increase in oxy-Hb following the circuit condition, yet virtually no change in this outcome following the treadmill condition; thus, suggesting that this pre-activation effect may only be present when a certain degree of cognitive engagement occurs alongside physical activity.

Contrary to our initial hypotheses, we did not find differences between the conditions in our psychological outcomes. It is possible that physical activity does not have the same effect on these psychological mechanisms including affect, motivation, and self-efficacy among children with ASD compared to children with typical development (e.g., Schmidt et al., 2016). However, these results may actually mean that children with ASD can make improvements to their executive functions following exercise, regardless of their motivation or self-efficacy toward the cognitive task. This is a potentially promising finding that should be further explored in future intervention work as providing a bout of exercise prior to cognitive tasks, such as behavioural treatments or school activities, may be able to elicit improved outcomes on these activities, regardless of the child’s feelings toward the activity.

Although not a primary outcome, another important finding from this study was that all participants were able to complete both exercise conditions, with 100% adherence, while staying in the target heart rate zone. This finding suggests that exercise is a feasible intervention for children with ASD, which is important for the implementation and uptake of exercise-based programs for this population. Beyond replication of these study findings, an important next step in this work will also be to explore the role of exercise on executive functions outside of the lab setting. To this extent, circuit-based exercise may provide a more ecologically valid workout for school-age children with ASD who may not have access to specialized exercise equipment, such as a treadmill. While basic equipment (e.g., resistance band, medicine ball) was used for the circuit, it could also be completed without any equipment as a complete body-weight exercise session. Thus, the circuit workout could reasonably be completed in a home, school, or clinic environment with little space or equipment needed. Further, the larger effect on executive functioning and cerebral blood flow regulation evident following the circuit condition suggests that this type of exercise may also elicit the greatest improvements on various health and behavioural outcomes (both acutely and overtime) in children with ASD.

Finally, while the exercise sessions were 20-minutes in length, it is important that future research also explore the effects of shorter bouts of activity (e.g., Howie, Schatz, & Pate, 2015; Schmitz Olin et al., 2017) to further improve the feasibility of implementing exercise breaks into multiple settings. Given that current behavioural treatment practices for this population are both time- and cost-intensive (Buescher, Cidav, Knapp, & Mandell, 2014; Virués-Ortega, 2010), exercise may provide a relatively low-cost alternative to augment current practices. For example, the provision of a brief exercise bout either immediately before or during traditional behavioural therapy may help to improve participants' attention and inhibition, essentially boosting the benefits of the session. Future research should build on this work by exploring the effect of exercise breaks on traditional treatment outcomes for children with ASD.

One limitation to this study is the relatively small sample that was employed. While we were limited to 12 participants, our post-hoc sensitivity analysis revealed we were powered to only detect large effects. Recruitment of participants with ASD in a community setting is a challenge due to low prevalence rates and lack of interest in exercise-based studies among parents of children with ASD. Nonetheless, our sample size is not that different from previous experimental exercise-based studies in this population (Anderson-Hanley et al., 2011; Schmitz Olin et al., 2017). An additional limitation is that we only included one measure of executive functioning in the study. The cancellation task we employed was considered a measure of inhibitory control; however, most studies of this aspect of executive functioning use a Flanker task to assess this outcome (Pontifex et al., 2019; Raine et al., 2018). Pilot testing in our lab suggested that the completion of a Flanker task would not be feasible for our participants. Future work should attempt to recruit larger samples and include multiple measures of executive functioning.

Our study is strengthened by employing a within-subject pre-test post-test experimental design, which is considered the gold-standard for testing the acute effects of exercise on executive functioning (Pontifex et al., 2019). Another strength to this study is the inclusion of direct measures of executive functioning, changes in cerebral blood flow regulation, and psychological outcomes within the same study; something that has not previously been done in a sample of children with ASD. Further strengths are the inclusion of two different types of exercise, in comparison to a sedentary control condition, along with numerous manipulation checks.

Overall, results from this exploratory study suggest that an acute bout of exercise may elicit improvements in executive functioning and cerebral blood flow regulation in children with ASD, with exercises that are more cognitively and motorically demanding potentially having the greatest benefits. However, the failure to find a significant interaction effect warrants the need for more work in this area, with larger sample sizes, to truly establish the acute effect of exercise on executive functioning, and its intermediary variables, in children with ASD.

References

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (DSM-5)* (5th ed.). Washington, DC: American Psychiatric Publishing.

Anderson-Hanley, C., Tureck, K., & Schneiderman, R. L. (2011). Autism and exergaming: Effects on repetitive behaviors and cognition. *Psychology Research and Behavior Management*. https://doi.org/10.2147/PRBM.S24016

Bandura, A. (1997). *Self-efficacy: The exercise of control*. New York, NY: Freeman.

Bandura, A. (2006). Guide for constructing self-efficacy scales. *Self-Efficacy Beliefs of Adolescents*, *5*(307–337).

Belmonte, M. K. (2004). Autism and Abnormal Development of Brain Connectivity. *Journal of Neuroscience*, *24*(42), 9228–9231. https://doi.org/10.1523/JNEUROSCI.3340-04.2004

Best, J. R. (2010). Effects of physical activity on children's executive function: Contributions of experimental research on aerobic exercise. *Developmental Review*, *30*(4), 331–351. https://doi.org/10.1016/j.dr.2010.08.001

Borg, G. (1998). *Borg's perceived exertion and pain scales.* Champaign, IL: Human Kinetics.

Bremer, E., Crozier, M., & Lloyd, M. (2016). A systematic review of the behavioural outcomes following exercise interventions for children and youth with autism spectrum disorder. *Autism*, *20*(8), 899–915. https://doi.org/10.1177/1362361315616002

Budde, H., Voelcker-Rehage, C., Pietraßyk-Kendziorra, S., Ribeiro, P., & Tidow, G. (2008). Acute coordinative exercise improves attentional performance in adolescents. *Neuroscience Letters*, *441*(2), 219–223. https://doi.org/10.1016/j.neulet.2008.06.024

Buescher, A., Cidav, Z., Knapp, M., & Mandell, D. (2014). Costs of autism spectrum disorders in the United Kingdom and the United States. *JAMA Pediatrics*, *168*(8), 721–728. https://doi.org/10.1001/jamapediatrics.2014.210

Cohen, J. (1992). A power primer. *Psychological Bulletin*, *112*(1), 155.

Cole, T. J., & Lobstein, T. (2012). Extended international (IOTF) body mass index cut-offs for thinness, overweight and obesity: Extended international BMI cut-offs. *Pediatric Obesity*, *7*(4), 284–294. https://doi.org/10.1111/j.2047-6310.2012.00064.x

Currenti, S. A. (2010). Understanding and Determining the Etiology of Autism. *Cellular and Molecular Neurobiology*, *30*(2), 161–171. https://doi.org/10.1007/s10571-009-9453-8

Demetriou, E. A., Lampit, A., Quintana, D. S., Naismith, S. L., Song, Y. J. C., Pye, J. E., … Guastella, A. J. (2018). Autism spectrum disorders: a meta-analysis of executive function. *Molecular Psychiatry*, *23*(5), 1198–1204. https://doi.org/10.1038/mp.2017.75

Diamond, A. (2000). Close interrelation of motor development and cognitive development and of the cerebellum and prefrontal cortex. *Child Development*, *71*(1), 44–56. https://doi.org/10.1111/1467-8624.00117

Diamond, A. (2013). Executive Functions. *Annual Review of Psychology*, *64*(1), 135–168. https://doi.org/10.1146/annurev-psych-113011-143750

Diamond, A., & Ling, D. S. (2018). Aerobic-Exercise and resistance-training interventions have been among the least effective ways to improve executive functions of any method tried thus far. *Developmental Cognitive Neuroscience*. https://doi.org/10.1016/j.dcn.2018.05.001

Ekkekakis, P., Parfitt, G., & Petruzzello, S. J. (2011). The Pleasure and Displeasure People Feel When they Exercise at Different Intensities. *Sports Medicine*, *41*(8), 641–671. https://doi.org/10.2165/11590680-000000000-00000

Ellis, S. M., & Steyn, H. S. (2003). Practical significance (effect sizes) versus or in combination with statistical significance (p-values). *Management Dynamics*, *12*(4), 51–53.

Faul, F., Erdfelder, E., Buchner, A., & Lang, A.-G. (2009). Statistical power analyses using G*Power 3.1: Tests for correlation and regression analyses. *Behavior Research Methods*, *41*(4), 1149–1160. https://doi.org/10.3758/BRM.41.4.1149

Gelbart, M., Ziv-Baran, T., Williams, C. A., Yarom, Y., & Dubnov-Raz, G. (2017). Prediction of maximal heart rate in children and adolescents. *Clinical Journal of Sport Medicine*, *27*(2), 139–144. https://doi.org/10.1097/JSM.0000000000000315

Gilliam, J. C. (2014). *GARS-3: Gilliam Autism Rating Scale–Third Edition*. Austin, TX: Pro-Ed, Inc.

Gunnell, K. E., Poitras, V. J., LeBlanc, A., Schibli, K., Barbeau, K., Hedayati, N., … Tremblay, M. S. (2018). Physical activity and brain structure, brain function, and cognition in children and youth: A systematic review of randomized controlled trials. *Mental Health and Physical Activity*. https://doi.org/10.1016/j.mhpa.2018.11.002

Hillman, C. H., & Biggan, J. R. (2017). A review of childhood physical activity, brain, and cognition: perspectives on the future. *Pediatric Exercise Science*, *29*(2), 170–176.

Howie, E. K., Schatz, J., & Pate, R. R. (2015). Acute Effects of Classroom Exercise Breaks on Executive Function and Math Performance: A Dose–Response Study. *Research Quarterly for Exercise and Sport*, *86*(3), 217–224. https://doi.org/10.1080/02701367.2015.1039892

IBM Corporation. (2017). IBM SPSS statistics for windows (Version 25). Armonk, NY: IBM Corporation.

Jasper, H. H. (1958). The ten-twenty electrode system of the International Federation. *Electroencephalogr. Clin. Neurophysiol.*, *10*, 370–375.

Jones, R. A., Downing, K., Rinehart, N. J., Barnett, L. M., May, T., McGillivray, J. A., … Hinkley, T. (2017). Physical activity, sedentary behavior and their correlates in children with Autism Spectrum Disorder: A systematic review. *PLOS ONE*, *12*(2), e0172482. https://doi.org/10.1371/journal.pone.0172482

Just, M. A., Cherkassky, V. L., Keller, T. A., Kana, R. K., & Minshew, N. J. (2007). Functional and Anatomical Cortical Underconnectivity in Autism: Evidence from an fMRI Study of an Executive Function Task and Corpus Callosum Morphometry. *Cerebral Cortex*, *17*(4), 951–961. https://doi.org/10.1093/cercor/bhl006

Kenville, R., Maudrich, T., Carius, D., & Ragert, P. (2017). Hemodynamic Response Alterations in Sensorimotor Areas as a Function of Barbell Load Levels during Squatting: An fNIRS Study. *Frontiers in Human Neuroscience*, *11*. https://doi.org/10.3389/fnhum.2017.00241

Kenworthy, L., Anthony, L. G., Naiman, D. Q., Cannon, L., Wills, M. C., Luong-Tran, C., … Wallace, G. L. (2014). Randomized controlled effectiveness trial of executive function intervention for children on the autism spectrum. *Journal of Child Psychology and Psychiatry*, *55*(4), 374–383. https://doi.org/10.1111/jcpp.12161

Lambourne, K., & Tomporowski, P. (2010). The effect of exercise-induced arousal on cognitive task performance: A meta-regression analysis. *Brain Research*, *1341*, 12–24. https://doi.org/10.1016/j.brainres.2010.03.091

Lehto, J. E., Juujärvi, P., Kooistra, L., & Pulkkinen, L. (2003). Dimensions of executive functioning: Evidence from children. *British Journal of Developmental Psychology*, *21*(1), 59–80. https://doi.org/10.1348/026151003321164627

McMorris, T. (2016). Developing the catecholamines hypothesis for the acute exercise-cognition interaction in humans: Lessons from animal studies. *Physiology & Behavior*, *165*, 291–299. https://doi.org/10.1016/j.physbeh.2016.08.011

Memari, A. H., Mirfazeli, F. S., Kordi, R., Shayestehfar, M., Moshayedi, P., & Mansournia, M. A. (2017). Cognitive and social functioning are connected to physical activity behavior in children with autism spectrum disorder.

Research in Autism Spectrum Disorders, *33*, 21–28. https://doi.org/10.1016/j.rasd.2016.10.001

Miyake, A., Friedman, N. P., Emerson, M. J., Witzki, A. H., Howerter, A., & Wager, T. D. (2000). The Unity and Diversity of Executive Functions and Their Contributions to Complex “Frontal Lobe” Tasks: A Latent Variable Analysis. *Cognitive Psychology*, *41*(1), 49–100. https://doi.org/10.1006/cogp.1999.0734

Ogoh, S., & Ainslie, P. N. (2009). Cerebral blood flow during exercise: mechanisms of regulation. *Journal of Applied Physiology*, *107*(5), 1370–1380. https://doi.org/10.1152/japplphysiol.00573.2009

Pan, C.-Y., Chu, C.-H., Tsai, C.-L., Sung, M.-C., Huang, C.-Y., & Ma, W.-Y. (2017). The impacts of physical activity intervention on physical and cognitive outcomes in children with autism spectrum disorder. *Autism*, *21*(2), 190–202. https://doi.org/10.1177/1362361316633562

Piper, S. K., Krueger, A., Koch, S. P., Mehnert, J., Habermehl, C., Steinbrink, J., … Schmitz, C. H. (2014). A wearable multi-channel fNIRS system for brain imaging in freely moving subjects. *NeuroImage*, *85*, 64–71. https://doi.org/10.1016/j.neuroimage.2013.06.062

Pontifex, M. B., McGowan, A. L., Chandler, M. C., Gwizdala, K. L., Parks, A. C., Fenn, K., & Kamijo, K. (2019). A primer on investigating the after effects of acute bouts of physical activity on cognition. *Psychology of Sport and Exercise*, *40*, 1–22. https://doi.org/10.1016/j.psychsport.2018.08.015

Raine, L. B., Kao, S.-C., Pindus, D., Westfall, D. R., Shigeta, T. T., Logan, N., … Hillman, C. H. (2018). A Large-Scale Reanalysis of Childhood Fitness and Inhibitory Control. *Journal of Cognitive Enhancement*. https://doi.org/10.1007/s41465-018-0070-7

Roid, G. H., Miller, L. J., Pomplun, M., & Koch, C. (2013). Leiter international performance scale,(Leiter-3). *Los Angeles: Western Psychological Services*.

Schipul, S. E., Keller, T. A., & Just, M. A. (2011). Inter-Regional Brain Communication and Its Disturbance in Autism. *Frontiers in Systems Neuroscience*, *5*. https://doi.org/10.3389/fnsys.2011.00010

Schmidt, M., Benzing, V., & Kamer, M. (2016). Classroom-Based Physical Activity Breaks and Children’s Attention: Cognitive Engagement Works! *Frontiers in Psychology*, *7*. https://doi.org/10.3389/fpsyg.2016.01474

Schmidt, M., Jäger, K., Egger, F., Roebers, C. M., & Conzelmann, A. (2015). Cognitively Engaging Chronic Physical Activity, but Not Aerobic Exercise, Affects Executive Functions in Primary School Children: A Group-Randomized Controlled Trial. *Journal of Sport and Exercise Psychology*, *37*(6), 575–591. https://doi.org/10.1123/jsep.2015-0069

Schmitz Olin, S., Mcfadden, B. A., Golem, D. L., Pellegrino, J. K., Walker, A. J., Sanders, D. J., & Arent, S. M. (2017). The Effects of Exercise Dose on Stereotypical Behavior in Children with Autism: *Medicine & Science in Sports & Exercise*, *49*(5), 983–990. https://doi.org/10.1249/MSS.0000000000001197

Soga, K., Masaki, H., Gerber, M., & Ludyga, S. (2018). Acute and Long-term Effects of Resistance Training on Executive Function. *Journal of Cognitive Enhancement*. https://doi.org/10.1007/s41465-018-0079-y

Stimpson, N. J., Davison, G., & Javadi, A.-H. (2018). Joggin' the Noggin: Towards a Physiological Understanding of Exercise-Induced Cognitive Benefits. *Neuroscience & Biobehavioral Reviews*, *88*, 177–186. https://doi.org/10.1016/j.neubiorev.2018.03.018

Sullivan, G. M., & Feinn, R. (2012). Using Effect Size—or Why the P-Value Is Not Enough. *Journal of Graduate Medical Education*, *4*(3), 279–282. https://doi.org/10.4300/JGME-D-12-00156.1

Tan, B. W. Z., Pooley, J. A., & Speelman, C. P. (2016). A Meta-Analytic Review of the Efficacy of Physical Exercise Interventions on Cognition in Individuals with Autism Spectrum Disorder and ADHD. *Journal of Autism and Developmental Disorders*, *46*(9), 3126–3143. https://doi.org/10.1007/s10803-016-2854-x

Tse, C. Y. A., Lee, H. P., Chan, K. S. K., Edgar, B. V., Wilkinson-Smith, A., & Lai, W. H. E. (2019). Examining the impact of physical activity on sleep quality and executive functions in children with autism spectrum disorder: A randomized controlled trial. *Autism*, 1–12. https://doi.org/10.1177/1362361318823910

Virués-Ortega, J. (2010). Applied behavior analytic intervention for autism in early childhood: Meta-analysis, meta-regression and dose–response meta-analysis of multiple outcomes. *Clinical Psychology Review*, *30*(4), 387–399. https://doi.org/10.1016/j.cpr.2010.01.008

Vissers, M. E., X Cohen, M., & Geurts, H. M. (2012). Brain connectivity and high functioning autism: A promising path of research that needs refined models, methodological convergence, and stronger behavioral links. *Neuroscience & Biobehavioral Reviews*, *36*(1), 604–625. https://doi.org/10.1016/j.neubiorev.2011.09.003

Voss, M. W., Vivar, C., Kramer, A. F., & van Praag, H. (2013). Bridging animal and human models of exercise-induced brain plasticity. *Trends in Cognitive Sciences*, *17*(10), 525–544.

Wass, S. (2011). Distortions and disconnections: Disrupted brain connectivity in autism. *Brain and Cognition*, *75*(1), 18–28. https://doi.org/10.1016/j.bandc.2010.10.005

CHAPTER 6: GENERAL DISCUSSION

The overarching objective of this dissertation was to explore the role of physical activity for the health, behaviour, and cognition of children with ASD. Ultimately, the long-term goal of this work is to understand how physical activity may be used as a behavioural intervention for this population. Given the paucity of literature in this area to-date, this broad objective was explored through a set of studies that sought to establish foundational knowledge regarding common correlates and outcomes of physical activity. Importantly, the studies included in this dissertation were not designed to be sequential, but rather to fill a number of gaps in the literature. This work was guided through an interdisciplinary framework (Figure 1) of physical activity correlates and outcomes, developed for this dissertation. Specifically, this framework helped to guide our study of the role of physical activity for children with ASD by taking a comprehensive approach to understanding the physical, behavioural, and cognitive variables that may influence and change with participation in physical activity.

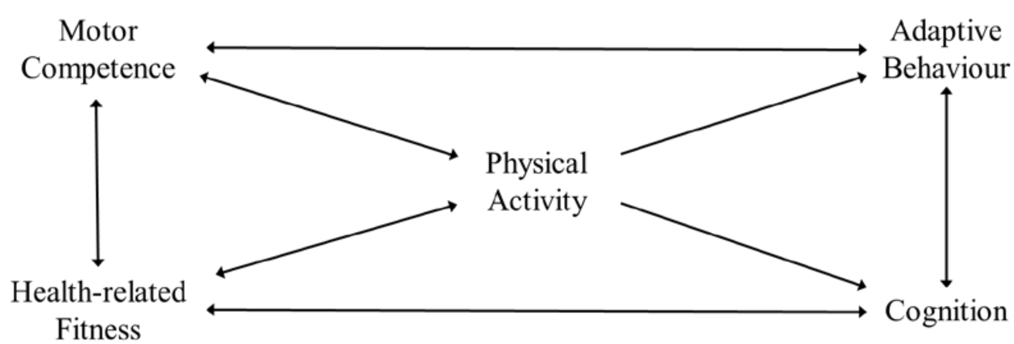

Figure 1. Interdisciplinary framework of physical activity correlates and outcomes.

Filling the Gaps

Overall, the findings from this dissertation have extended existing knowledge in several important ways. First, it is imperative that studies of the health and physical activity levels of children with ASD include measures that are both feasible and reliable for this population. Previous studies reporting on fitness outcomes in children with ASD have failed to provide evidence of these critical measurement properties. Fitness is an important independent indicator of overall health (Anderssen et al., 2007; Ortega, Ruiz, Castillo, & Sjöström, 2008; Ruiz et al., 2009). As such, it is also considered an outcome of many physical activity interventions (e.g., Kriemler et al., 2011), and is positively associated with cognitive functioning (Fedewa & Ahn, 2011; Raine et al., 2018). However, if we cannot reliably measure fitness, then we cannot assess its role as a correlate or outcome of physical activity. Therefore, an important first step in this dissertation was to establish the feasibility and reliability of select fitness assessments for use in children with ASD. Findings from Chapter 2 demonstrated that fitness tests can be completed with a high degree of feasibility in children with ASD; however, differences in reliability are present. Specifically, assessments requiring a limited number of instructions and short completion time (e.g., standing long jump, Wingate, grip strength, sit and reach) demonstrated good- to- excellent test-retest reliability. In contrast, assessments that were longer in duration or required a higher-degree of self-regulation (e.g., modified six-minute walk test, Bruce protocol, and Muscle Power Sprint Test) did not demonstrate adequate levels of reliability. Researchers and practitioners should consider these findings when choosing fitness assessments for children with ASD. Further, future work should continue to explore other measurement properties, such as the validity of these assessments, specifically for children with ASD.

Second, we know that children with ASD experience a number of physical health deficits, including low levels of motor competence (Fournier, Hass, Naik, Lodha, & Cauraugh, 2010), health-related fitness (Borremans, Rintala, & McCubbin, 2010; Pan, 2014), and high rates of obesity (Healy, Aigner, & Haegele, 2018). These variables may all be related to participation in physical activity in children (Stodden et al., 2008), yet we do not have a good understanding of how these variables interact specifically for children with ASD. Understanding these relationships, in the context of the behavioural profiles of ASD, may provide important insights necessary in the design and implementation of physical activity programs for this population. Indeed, guided by our interdisciplinary framework, this dissertation provided support for the role of adaptive behaviour in the physical health of children with ASD. Specifically, we found that adaptive behaviour is positively related to motor coordination in children with ASD (Chapter 3). Moreover, in Chapter 4 we tested an extension of Stodden's model to consider the role of adaptive behaviour in this model. Findings showed that adaptive behaviour moderates the relationship between motor coordination and health-related fitness (Chapter 4). Contrary to our hypotheses though, we did not find significant relationships between physical activity and motor coordination or health-related fitness (Chapter 4). Taken together, findings from Chapters 3 and 4 add to our understanding of the importance of motor competence in the daily lives and behaviour of children with ASD, but also emphasize the importance of the relationship between motor competence and adaptive behaviour. Moreover, that we did not find significant relationships with physical activity may mean that traditional models of participation used for children with typical development may not be as relevant for children with ASD. These findings underscore the need to explore the role of physical activity on other outcomes (e.g., behaviour and cognition) rather than only physical health outcomes, in children with ASD.

Third, changes in cognitive outcomes following physical activity may be of particular importance for children with ASD given the widespread executive dysfunction that is evident in the disorder (Demetriou et al., 2018). Research in children with typical development generally points to improvements in executive functioning following both acute (Ludyga, Gerber, Brand, Holsboer-Trachsler, & Puhse, 2016; Verburgh, Königs, Scherder, & Oosterlaan, 2014) and chronic (Xue, Yang, & Huang, 2019) physical activity, with varying effects based on the type of activity. However, very little research has explored these effects in children with ASD. In Chapter 1, we proposed that cognition may be a critical outcome of physical activity in children with ASD. Indeed, findings from Chapter 5 in this dissertation provided preliminary support for the positive effect of an acute bout of exercise on executive functioning (an aspect of cognition) in this population. Further, findings suggest that a circuit-based interval exercise session may elicit greater improvements in executive functioning than more traditional aerobic exercise.

Overall, findings from this dissertation have filled multiple gaps in the literature regarding the role of physical activity for children with ASD. Further, this dissertation provides preliminary support for three key pathways proposed in our interdisciplinary framework (outlined by the dashed boxes in Figure 2). Specifically, we found associations between motor competence, health-related fitness, and adaptive behaviour. We also provided evidence for the acute effect of physical activity on cognition in children with ASD. The implications and future directions of these findings will be discussed in the subsequent sections.

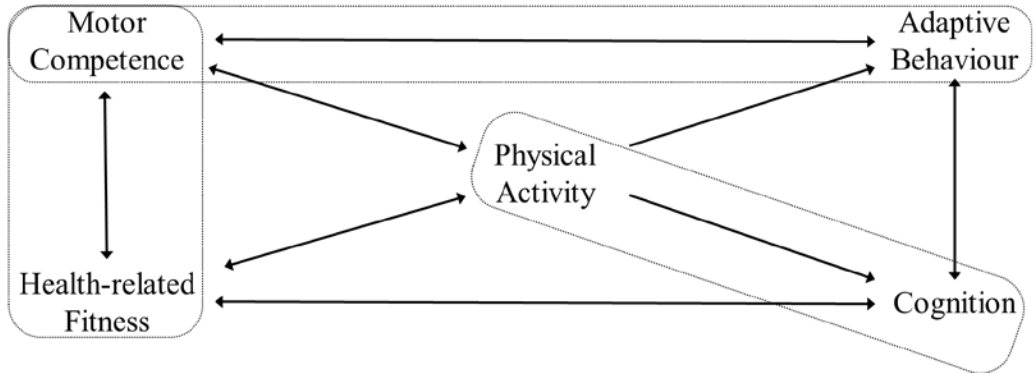

Figure 2. Evidence-supported pathways of the interdisciplinary framework of physical activity.

Implications

Theoretical Implications

Modified model of motor competence for physical activity. One of the objectives of this dissertation was to understand the relationships between physical activity and physical and behavioural health variables in children with ASD. Through this objective, an important contribution was made to the motor development literature by applying a well-established model of motor competence (Stodden et al., 2008), developed for typical populations, to children with ASD. This model positions motor competence as essential for participation in physical activity, along with improved fitness, high perceived motor competence, and a healthy weight status. While aspects of this model have been extensively tested (Robinson et al., 2015), little work had examined these pathways in children with

ASD. This dissertation provided some of the first information to suggest that many of these pathways, with the exception of the relationship between motor competence and fitness, may not be significant for children with ASD. Beyond simply testing these existing relationships, this dissertation also proposed and tested a novel modification to Stodden's model (Figure 1 in Chapter 4), wherein adaptive behaviour moderates the relationship between motor competence and fitness and physical activity, respectively. Our results provided partial support for this modified model, with adaptive behaviour only moderating the association between motor competence and fitness. Our effect modification model of the influence of adaptive behaviour on the pathways connecting motor competence to physical activity and health-related fitness provides an important basis from which to move forward in the study of physical activity and motor competence for children with ASD. Further, future research should continue to explore these relationships through a comprehensive test of the effect modification model with a larger sample, as well as examining the effect modification model in the context of our interdisciplinary framework of physical activity.

Interestingly, we did not find any significant associations between our outcomes measured in Chapter 4 and physical activity. It is possible that this is due to limited variability in our physical activity data or because participants, on average, were not meeting physical activity guidelines (i.e., 48.7 ± 17.1 minutes of MVPA/day reported in Chapter 4). Further, despite a great deal of evidence, the strength of the relationship between motor competence and physical activity in children with typical development is generally weak to begin with (Holfelder & Schott, 2014). Thus, it is also possible that we simply were not powered to detect an association between motor competence and physical activity. Conversely, the lack of association may simply point to the need to take into account other variables that were not measured. For example, although it was unavailable at the time this dissertation was initiated, Arnell and colleagues (2017) recently proposed a conditional model of participation in physical activity for children with ASD. This conditional model is composed of five interconnected domains including competence and confidence, motivation, adjustment to external demands, predictability, and freedom of choice. These factors are likely important for all children, not just those with ASD. However, it is plausible that the weighting of each factor, particularly the need for predictability and adjustment to external demands, may play a larger role in the ability of children with ASD to participate in physical activity. Moreover, in reference to our interdisciplinary framework initially presented in Chapter 1 (Figure 1), it is possible that the factors proposed by Arnell (2017) actually have varying effects on each pathway. Future work should continue to explore how these factors (e.g., adjustment to external demands, predictability, etc.) influence the physical activity pathways for children with ASD, while taking into account the associations found in this dissertation.

Physical activity and cognition. Much like the extant literature on physical activity in children with ASD, the findings discussed in the previous section were largely grounded in the study of motor development. Yet, we also provided evidence for an interaction between adaptive behaviour, motor competence, and fitness in children with ASD. Moving beyond the motor domain, physical activity has the potential to impact behaviour and cognition in children with ASD and it has been hypothesized that these improvements are due to improved executive functions (Bremer, Crozier, & Lloyd, 2016; Tan, Pooley, & Speelman, 2016). Given the widespread executive dysfunction experienced by children with ASD (Demetriou et al., 2018) and the role of executive functions in virtually all aspects of behaviour (Diamond, 2013), we specifically sought to examine if executive functions could be improved through physical activity in this population.

Chapter 5 was an exploratory investigation into the acute effect of exercise on executive functions and cerebral blood flow regulation in children with ASD. In line with previous research, we hypothesized that an acute bout of exercise would lead to increases in these variables. Indeed, findings from this study provided preliminary support for an acute effect of exercise on executive functions in children with ASD. Results also provided initial support for the role of cerebral blood flow regulation as a potential factor that may contribute to changes in executive functioning. Findings from this study also provided support for a circuit-based exercise session to result in greater improvements to these areas when compared to treadmill walking. Although we did not find significant interaction effects in this study, the effect sizes were medium to large in size, suggestive of practical significance for this population.

There are a few possible mechanisms that could be driving these changes in executive functions. One is that the activation of the cerebellum during exercise led to co-activation of the prefrontal cortex, essentially “pre-activating” the brain regions needed for the executive functioning task (Budde, Voelcker-Rehage, Pietraßyk-Kendziorra, Ribeiro, & Tidow, 2008; Schmidt, Benzing, & Kamer, 2016). This pre-activation theory is supported by the fact that we saw greater improvements in executive functioning following the circuit exercise, which may have required a higher degree of cognitive engagement, in comparison to the treadmill exercise, which may have relied more on motor processes alone. This hypothesis is further supported by the fact that we saw an increase in oxygenated hemoglobin in the prefrontal cortex following the circuit condition, yet virtually no change in this outcome following the treadmill condition; suggesting that this pre-activation effect may only be present when a certain degree of cognitive engagement is required alongside physical activity. While we were underpowered to investigate the indirect effect of cerebral blood flow regulation on changes in executive functioning, we recommend that future work explore whether this physiological change mediates changes in executive function.

Taken together, this dissertation provides preliminary evidence that executive functioning can be improved with an acute bout of physical activity in children with ASD and may occur alongside an increase in cerebral blood flow regulation. Future work should continue to explore these mechanisms along with the effect of cognition on behavioural outcomes in this population.

Practical Implications

Feasibility. In addition to the theoretical implications of this dissertation, there are many practical implications that may help to advance our understanding of the role of physical activity for children with ASD. The high rates of physical inactivity evident in children with ASD may beg the question of whether physical activity is actually a feasible activity for this population. In other words, are children with ASD not engaging in physical activity because they cannot; or is the inactivity owing to other factors? Therefore, one practical implication of this dissertation was the finding that physical activity is feasible.

Specifically, Chapter 2 demonstrated that various fitness assessments can be completed with children with ASD with a high degree of feasibility. This is important for future longitudinal studies of the health of children with ASD, and intervention studies measuring the effect of physical activity interventions on health outcomes. Further, Chapter 5 demonstrated a very high degree of feasibility for the completion of 20-minute exercise sessions. Both the treadmill and circuit-based exercise sessions were completed with 100% adherence by all participants, with the manipulation checks indicating that the exercise was completed as intended. It is particularly relevant that all children could complete the circuit-based workout, as this was the more ecologically valid activity of the two. Indeed, the circuit included exercises that could be implemented in home, school, and clinical settings. Further, these are exercises (e.g., jumping jacks and squat jumps) that are commonly included in school daily physical activity breaks as they can be completed within the classroom. Collectively, this dissertation demonstrated a high degree of feasibility for different types of physical activity (e.g., fitness assessments, aerobic activity, and interval-based activity) for children with ASD, suggesting that these activities are suitable for the population.

Motor competence and behaviour are interrelated. In addition to high rates of physical inactivity (Jones et al., 2017), children with ASD also experience low levels of motor competence (Fournier et al., 2010) . Yet, to-date we have had little information on how these variables are related to one another. The presence and direction of the relationships between these variables may impact the planning and delivery of physical activity interventions. In addition, it is also critical to explore how behavioural functioning may impact these associations, as behavioural characteristics of ASD are typically the primary focus of intervention. Therefore, another practical implication of this dissertation was establishing the importance of motor competence for the behavioural and physical health of children with ASD. We found that motor and adaptive behaviour were positively associated and that adaptive behaviour moderated the association between motor competence and fitness. Further, this moderation was apparent at a relatively low level of adaptive behaviour (i.e., over one standard deviation below the mean; Chapter 4), suggesting the importance of motor competence for virtually all children with ASD, regardless of level of behavioural functioning.

This relationship may have many practical implications for children with ASD. One is that it points to the importance of specifically targeting motor competence, through intervention, as this may provide children with the physical skills needed to engage in a number of positive, prosocial, daily behaviours. Developing competence in motor skills may help to ease many of the social and behavioural difficulties experienced by children with ASD. Improving fine motor abilities can assist with dressing, feeding, and academic activities. Developing competence in gross motor skills can provide an opportunity for children to participate in active play and recreational activities. Further, a general degree of motor coordination is necessary for children to simply navigate their environment in a sufficient manner. It is imperative that children, particularly those of school age, have the motor competence to engage in these activities at an age-appropriate level. Children who are performing at a level below their peers may be more likely to drop out of activities; thus, limiting any future opportunities for skill development (Wall, 2004). For a child with ASD who already has difficulty with social interactions this potential for further withdrawal could have drastic implications for their social development.

Previous research has demonstrated that motor skills can be improved through physical activity-based interventions in children with ASD (Colombo-Dougovito & Block, 2019). Moreover, targeted motor skill interventions have also shown positive improvements to aspects of adaptive behaviour and social skills, in addition to improvements in motor skills (Bremer, Balogh, & Lloyd, 2015; Bremer & Lloyd, 2016; Ketcheson, Hauck, & Ulrich, 2017). In contrast, there is no evidence that behavioural interventions result in simultaneous improvements to the motor domain. As such, providing increased access to motor skill interventions for children with ASD may provide benefits beyond the motor domain.

Intervening through physical activity. In addition to improved physical health outcomes, participation in physical activity may also provide additional benefits for children with ASD. For example, physical activity can lead to improved cognition, including executive functions and measures of academic achievement, in children with typical development (Álvarez-Bueno, Pesce, Cavero-Redondo, Sánchez-López, Garrido-Miguel, et al., 2017; Álvarez-Bueno, Pesce, Cavero-Redondo, Sánchez-López, Martínez-Hortelano, et al., 2017).

Previous research in children with ASD has also provided preliminary support for these effects (Bremer et al., 2016; Tan et al., 2016); however, these findings have been limited. Developing an understanding of how physical activity can be used to address the behavioural challenges experienced by children with ASD may help to provide additional options to augment current treatment practices. Yet, in order to influence future treatment practices, we must first understand how physical activity may elicit change in this population.

Chapter 5 provided preliminary support that an acute circuit-based exercise session may result in greater benefits than more traditional aerobic exercise in regard to improvements in executive functioning. This finding may have many practical implications for intervention planning moving forward. The circuit-based workout was considered more ecologically valid as it included activities that could be completed in a home, school, or clinical setting. As such, these activities may be able to be implemented as part of a more traditional treatment program for children with ASD. For example, providing a brief exercise bout to children with ASD prior to a traditional behavioural therapy session may improve aspects of their executive functions (e.g., inhibitory control) during the session and could lead to improved treatment outcomes. Likewise, providing exercise breaks in the middle of treatment sessions, or throughout the school day, may also have similar beneficial effects on treatment and academic outcomes. However, much more research is needed regarding the acute effects of exercise on executive functions in children with ASD, in addition to how these improvements may relate to behavioural outcomes including social and communicative skills.

Limitations

Guided by an interdisciplinary framework, this dissertation addressed multiple gaps in the literature and built on our understanding of the importance of physical activity for children with ASD. Nonetheless, there are still limitations to this body of work. One limitation is the relatively small sample of children from which these findings were based. For example, Chapters 2-4 were derived from one larger study of the same participants. Recruitment of children with ASD for physical activity-based studies is challenging due to generally low prevalence of the condition and a lack of interest in physical activity for this population. This larger study was, however, designed with the intent of assessing multiple outcomes (i.e., reliability of fitness assessments along with associations between health and behavioural outcomes). Further, that the studies included in this dissertation still found both statistically significant and clinically meaningful findings, despite small sample sizes, adds further strength to the importance of physical activity-based research and programming for children with ASD.

A second limitation stems from our inability to measure all potential correlates and outcomes of physical activity. For instance, Stodden's model includes perceived motor competence as another important factor related to both physical activity and actual motor competence (Stodden et al., 2008). We are, however, unaware of any valid measures of perceived motor competence for children with ASD. Similarly, we did not measure external demands (e.g., environmental and social factors) that may influence physical activity in our participants (Arnell et al., 2017). This was simply beyond the scope of resources available at the onset of this dissertation given the myriad of variables that could be correlates and/or outcomes of physical activity. These results are however strengthened by the inclusion of valid measures of physical activity, fitness, motor competence, and behavioural functioning, within the same set of studies.

Third, each of the studies were completed in a lab setting, potentially limiting their ecological validity. While this may not be as much of an issue for Chapters 2 and 3, the ecological validity of our findings may be more relevant for Chapters 4 and 5. In particular, it will be important that future work continue to examine the associations between physical activity, health, behaviour, and cognition by employing prospective longitudinal cohort studies, in order to see how these variables interact with each other over time and in the context of environmental constraints. Results from Chapter 5 provided preliminary evidence of the acute effect of different types of exercise on executive functioning in children with ASD. It was important that this work be completed in a lab setting in order to determine the efficacy of this work. Moving forward, it is critical that these results not only be replicated in lab-based studies, but are built upon in other environments, including home, school, and clinical settings. This study was strengthened by the inclusion of two different types of exercise, with the circuit-based exercise likely representing a much more ecologically valid type of activity than the treadmill walking. Moreover, the one-on-one nature of the exercise session was similar to the format of typical behavioural therapy for children with ASD, which may speak to the transferability of this work into a clinical setting.

Future Directions

Future research should build on the findings from this dissertation by comprehensively testing all of the pathways presented in our interdisciplinary framework (Figure 1). In order to further our understanding of these pathways, researchers should prospectively track the associations found in Chapters 3 and 4 over time in a large cohort of children with ASD. This will help to build on our understanding of how these physical health and behavioural variables interact in this population. Further, a large sample will help to either confirm our finding that there was no relationship between motor competence, fitness, and physical activity; or will be able to provide support for this relationship. The relationships between these predictor and outcome variables with physical activity should also be explored through experimental studies. For example, physical activity-based motor skill interventions for children with ASD should measure outcomes beyond the motor domain including physical activity, fitness, adaptive behaviour, and cognition, among others. Similarly, interventions for children with ASD should be intentional in their design, deliberately targeting multiple domains of health, behaviour, and cognition through a single intervention.

Another future direction of this work is to replicate the findings from the acute exercise study, using a larger sample of participants and multiple measures of executive functioning. A larger sample would enable researchers to investigate some of the potential mediating pathways (e.g., cerebral blood flow regulation) that may lead to changes in executive functioning. Including measures of working memory and cognitive flexibility, in addition to inhibitory control, would further our understanding of these effects. In addition to the replication of these acute findings, it is also important that future work begin to examine both the acute and chronic effects of physical activity on executive functions in clinical, community, and school settings. A logical next step would then be to explore how changes in executive function mediate change in behavioural outcomes (e.g., social communicative skills) for children with ASD.

Lastly, future work should explore how physical activity can be effectively embedded into regular treatment strategies for children with ASD. Physical activity programs are relatively inexpensive, do not require intensive work with expert providers, and may be completed in home, community, and clinical settings. When used in combination with other services, physical activity may prove complementary and the risk of adverse events is low. Moreover, even if executive functions and behaviour do not improve, the principal side effect may be improved physical health. Therefore, it may prove beneficial to provide children with ASD access to physical activity-based programming, both while on the waitlist for other services and to augment on-going treatment. However, the ability to provide such services and the effect on treatment outcomes needs to be examined.

Conclusion

This dissertation employed a series of studies, guided by an interdisciplinary framework, to establish foundational knowledge regarding the correlates and outcomes of physical activity for children with ASD, with the overarching goal of understanding how we can use physical activity as an intervention for this population. Collectively, these studies advanced our knowledge in a number of important ways. First, the feasibility and reliability of common fitness assessments was established as our ability to measure changes in fitness relies on our ability to be able to reliably assess it. Next, this dissertation established the importance of adaptive behaviour for the physical health of children with ASD, finding a positive relationship between motor competence and adaptive behaviour, as well as a moderating role of adaptive behaviour in the relationship between motor competence and fitness. Lastly, we provided preliminary support for improved executive functioning and cerebral blood flow regulation following an acute exercise session in children with ASD, with greater effects following a circuit-based exercise session. Together, these findings highlight the role of physical activity in the health, behaviour, and cognitive functioning of children with ASD.

References

Álvarez-Bueno, C., Pesce, C., Cavero-Redondo, I., Sánchez-López, M., Garrido-Miguel, M., & Martínez-Vizcaíno, V. (2017). Academic achievement and physical activity: A meta-analysis. *Pediatrics*, *140*(6), e20171498. https://doi.org/10.1542/peds.2017-1498

Álvarez-Bueno, C., Pesce, C., Cavero-Redondo, I., Sánchez-López, M., Martínez-Hortelano, J. A., & Martínez-Vizcaíno, V. (2017). The effect of physical activity interventions on children's cognition and metacognition: A systematic review and meta-analysis. *Journal of the American Academy of Child & Adolescent Psychiatry*, *56*(9), 729–738. http://dx.doi.org/10.1016/j.jaac.2017.06.012

Anderssen, S. A., Cooper, A. R., Riddoch, C., Sardinha, L. B., Harro, M., Brage, S., & Andersen, L. B. (2007). Low cardiorespiratory fitness is a strong predictor for clustering of cardiovascular disease risk factors in children independent of country, age and sex. *European Journal of Cardiovascular Prevention & Rehabilitation*, *14*(4), 526–531.

Arnell, S., Jerlinder, K., & Lundqvist, L.-O. (2017). Perceptions of physical activity participation among adolescents with autism spectrum disorders: A conceptual model of conditional participation. *Journal of Autism and Developmental Disorders*. https://doi.org/10.1007/s10803-017-3436-2

Borremans, E., Rintala, P., & McCubbin, J. A. (2010). Physical fitness and physical activity in adolescents with Asperger syndrome: A comparative study. *Adapted Physical Activity Quarterly*, *27*(4), 308–320.

Bremer, E., Balogh, R., & Lloyd, M. (2015). Effectiveness of a fundamental motor skill intervention for 4-year-old children with autism spectrum disorder: A pilot study. *Autism*, *19*(8), 980–991. https://doi.org/10.1177/1362361314557548

Bremer, E., Crozier, M., & Lloyd, M. (2016). A systematic review of the behavioural outcomes following exercise interventions for children and youth with autism spectrum disorder. *Autism*, *20*(8), 899–915. https://doi.org/10.1177/1362361315616002

Bremer, E., & Lloyd, M. (2016). School-based fundamental-motor-skill intervention for children with autism-like characteristics: An exploratory study. *Adapted Physical Activity Quarterly*, *33*(1), 66–88. https://doi.org/10.1123/APAQ.2015-0009

Budde, H., Voelcker-Rehage, C., Pietraßyk-Kendziorra, S., Ribeiro, P., & Tidow, G. (2008). Acute coordinative exercise improves attentional performance in adolescents. *Neuroscience Letters*, *441*(2), 219–223. https://doi.org/10.1016/j.neulet.2008.06.024

Colombo-Dougovito, A. M., & Block, M. E. (2019). Fundamental motor skill interventions for children and adolescents on the autism spectrum: A literature review. *Review Journal of Autism and Developmental Disorders*. https://doi.org/10.1007/s40489-019-00161-2

Demetriou, E. A., Lampit, A., Quintana, D. S., Naismith, S. L., Song, Y. J. C., Pye, J. E., … Guastella, A. J. (2018). Autism spectrum disorders: A meta-analysis of executive function. *Molecular Psychiatry*, *23*(5), 1198–1204. https://doi.org/10.1038/mp.2017.75

Diamond, A. (2013). Executive functions. *Annual Review of Psychology*, *64*(1), 135–168. https://doi.org/10.1146/annurev-psych-113011-143750

Fedewa, A. L., & Ahn, S. (2011). The effects of physical activity and physical fitness on children's achievement and cognitive outcomes: A meta-analysis. *Research Quarterly for Exercise and Sport*, *82*(3), 521–535. https://doi.org/10.5641/027013611X13275191444107

Fournier, K. A., Hass, C. J., Naik, S. K., Lodha, N., & Cauraugh, J. H. (2010). Motor coordination in autism spectrum disorders: A synthesis and meta-analysis. *Journal of Autism and Developmental Disorders*, *40*(10), 1227–1240. https://doi.org/10.1007/s10803-010-0981-3

Healy, S., Aigner, C. J., & Haegele, J. A. (2018). Prevalence of overweight and obesity among US youth with autism spectrum disorder. *Autism*. https://doi.org/10.1177/1362361318791817

Holfelder, B., & Schott, N. (2014). Relationship of fundamental movement skills and physical activity in children and adolescents: A systematic review. *Psychology of Sport and Exercise*, *15*(4), 382–391. https://doi.org/10.1016/j.psychsport.2014.03.005

Jones, R. A., Downing, K., Rinehart, N. J., Barnett, L. M., May, T., McGillivray, J. A., … Hinkley, T. (2017). Physical activity, sedentary behavior and their correlates in children with Autism Spectrum Disorder: A systematic review. *PLOS ONE*, *12*(2), e0172482. https://doi.org/10.1371/journal.pone.0172482

Ketcheson, L., Hauck, J., & Ulrich, D. (2017). The effects of an early motor skill intervention on motor skills, levels of physical activity, and socialization in young children with autism spectrum disorder: A pilot study. *Autism*, *21*(4), 481–492. https://doi.org/10.1177/1362361316650611

Kriemler, S., Meyer, U., Martin, E., van Sluijs, E. M. F., Andersen, L. B., & Martin, B. W. (2011). Effect of school-based interventions on physical activity and fitness in children and adolescents: A review of reviews and systematic update. *British Journal of Sports Medicine*, *45*(11), 923–930. https://doi.org/10.1136/bjsports-2011-090186

Ludyga, S., Gerber, M., Brand, S., Holsboer-Trachsler, E., & Puhse, U. (2016). Acute effects of moderate aerobic exercise on specific aspects of executive function in different age and fitness groups: A meta-analysis. *Psychophysiology*, *53*(11), 1611–1626. https://doi.org/10.1111/psyp.12736

Ortega, F. B., Ruiz, J. R., Castillo, M. J., & Sjöström, M. (2008). Physical fitness in childhood and adolescence: A powerful marker of health. *International Journal of Obesity*, *32*(1), 1–11.

Pan, C.-Y. (2014). Motor proficiency and physical fitness in adolescent males with and without autism spectrum disorders. *Autism*, *18*(2), 156–165. https://doi.org/10.1177/1362361312458597

Raine, L. B., Kao, S.-C., Pindus, D., Westfall, D. R., Shigeta, T. T., Logan, N., … Hillman, C. H. (2018). A large-scale reanalysis of childhood fitness and inhibitory control. *Journal of Cognitive Enhancement*. https://doi.org/10.1007/s41465-018-0070-7

Robinson, L. E., Stodden, D. F., Barnett, L. M., Lopes, V. P., Logan, S. W., Rodrigues, L. P., & D'Hondt, E. (2015). Motor competence and its effect on positive developmental trajectories of health. *Sports Medicine*, *45*(9), 1273–1284. https://doi.org/10.1007/s40279-015-0351-6

Ruiz, J. R., Castro-Pinero, J., Artero, E. G., Ortega, F. B., Sjostrom, M., Suni, J., & Castillo, M. J. (2009). Predictive validity of health-related fitness in youth: A systematic review. *British Journal of Sports Medicine*, *43*(12), 909–923. https://doi.org/10.1136/bjsm.2008.056499

Schmidt, M., Benzing, V., & Kamer, M. (2016). Classroom-based physical activity breaks and children's attention: Cognitive engagement works! *Frontiers in Psychology*. https://doi.org/10.3389/fpsyg.2016.01474

Stodden, D. F., Goodway, J. D., Langendorfer, S. J., Roberton, M. A., Rudisill, M. E., Garcia, C., & Garcia, L. E. (2008). A developmental perspective on the role of motor skill competence in physical activity: An emergent relationship. *Quest*, *60*(2), 290–306.

Tan, B. W. Z., Pooley, J. A., & Speelman, C. P. (2016). A meta-analytic review of the efficacy of physical exercise interventions on cognition in individuals with autism spectrum disorder and ADHD. *Journal of Autism and Developmental Disorders*, *46*(9), 3126–3143. https://doi.org/10.1007/s10803-016-2854-x

Verburgh, L., Königs, M., Scherder, E. J. A., & Oosterlaan, J. (2014). Physical exercise and executive functions in preadolescent children, adolescents and young adults: A meta-analysis. *British Journal of Sports Medicine*, *48*(12), 973–979. https://doi.org/10.1136/bjsports-2012-091441

Wall, A. E. T. (2004). The developmental skill-learning gap hypothesis: Implications for children with movement difficulties. *Adapted Physical Activity Quarterly*, *21*(3), 197–218. https://doi.org/10.1123/apaq.21.3.197

Xue, Y., Yang, Y., & Huang, T. (2019). Effects of chronic exercise interventions on executive function among children and adolescents: A systematic review with meta-analysis. *British Journal of Sports Medicine*, bjsports-2018-099825. https://doi.org/10.1136/bjsports-2018-099825

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(Chapter 3)

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