FUNDAMENTAL MOVEMENT SKILLS IN CHILDREN WITH AND WITHOUT AUTISM SPECTRUM DISORDER, AND THE MULTI-DOMAIN EFFECTS OF AN EARLY MOTOR INTERVENTION

By

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In the United States, obesity is more likely in children with Autism than their typical peers. Despite this, research and clinical practice show a lack of consensus on how to address this issue. The current dissertation first studied the fundamental movement skills, body composition, and physical activity of children with and without Autism Spectrum Disorder (ASD). Because children with ASD showed the largest delays in fundamental movement skills, the second part of this dissertation presents a fundamental movement skill teaching program. The intervention is the first of its kind implemented in a service delivery model, within early intensive behavioral intervention preschools. Motor scores showed improvement relative to the control group. Behavior technicians implementing the study expressed differing views on the feasibility of the intervention, rating the intervention low in feasibility overall. Despite inclusion of a social play component in the intervention, the program did not influence the success rate of the children in their social play therapy targets. These results advocate for (a) increased research attention to physical development in ASD, (b) continued fundamental movement skill training for children with ASD, and (c) cross-disciplinary collaboration between the fields of clinical Psychology and Kinesiology.

ABSTRACT

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It is increasingly clear that obesity in children with Autism Spectrum Disorder (ASD) is an important public health concern. Despite this, research shows a lack of consensus on how to address this issue. The current dissertation presents data from an observational study of children with and without Autism Spectrum Disorder (ASD), investigating their fundamental movement skills, body composition, and moderate - to - vigorous intensity physical activity (MVPA). This observational study found the largest difference between diagnostic groups in fundamental movement skills (F (1,26) = 24.71, p < 0.001), followed by body composition (F (1, 26) = 6.34, p = 0.02), and no difference in MVPA (F (1, 26) = 0.11, p = 0.75). This dissertation also presents results from an Applied Behavior Analysis fundamental movement skills intervention for preschoolers with ASD. The intervention is the first of its kind implemented in early intensive behavioral intervention centers, an ecologically valid environment with wide reach. Motor scores showed improvement relative to the control group (F (1, 12) = 4.983; p = 0.016). Behavior technicians implementing the study expressed polarized views on the feasibility of the intervention, rating the intervention low in feasibility overall. Despite inclusion of a social play component in the intervention, there were no benefits observed in their existing behavioral intervention therapy progress (F (1,12) = 0.06, p = 0.81). These results can be used to advocate for (a) increased research attention to physical development in ASD, (b) continued fundamental

movement skill training for children with ASD, and (c) cross-disciplinary collaboration between the fields of clinical Psychology and Kinesiology.

This dissertation is dedicated to the families and children who participated in these research studies. Thank you for making the path a little easier for others in the future.

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KEY TO ABBREVIATIONS

DSM - V	The Diagnostic Statistical Manual – 5 th Edition
ASD	Autism Spectrum Disorder
ADHD	Attention Deficit Hyperactivity Disorder
ABA	Applied Behavior Analysis
TEACCH	Treatment and Education of Autistic and related Communication handicapped Children
PT	Physical therapy
ОТ	Occupational therapy
MABC	Movement Assessment Battery for Children
MSEL	Mullen Scales of Early Learning
VABS	Vineland Adaptive Behavior Scales
FMS	Fundamental Movement Skills
PLS	Preschool Language Scales
TGMD	Test of Gross Motor Development
PA	Physical activity
MVPA	Moderate – to – Vigorous Physical Activity
CPRT	Classroom Pivotal Response Training
BMI	Body Mass Index
MVPA	Moderate – to – Vigorous Physical Activity
CPRT	Maximum Aerobic Capacity
DST	Dynamic Systems Theory
ВОТ	Bruininks-Oseretsky Test of Motor Proficiency

СРМ	Counts per minute
CSS	Calibrated Severity Score
ADOS	Autism Diagnostic Observational Scales
MANCOVA	Multivariate Analysis of Covariance
MANOVA	Multivariate Analysis of Variance
BCBA	Board-Certified Behavior Analyst
BT	Behavior Technician
DTT	Discrete Trial Training

CHAPTER 1

Introduction

Autism Spectrum Disorder

The Diagnostic Statistical Manual – 5th Edition (DSM – V) defines Autism Spectrum Disorder (ASD) as a pervasive, functionally significant disorder in social communication and/or interaction, often including restrictive and repetitive patterns of behavior, interests, or activities (American Psychological Asociation, 2013). ASD begins early in development and can be diagnosed as a toddler (Moore & Goodson, 2003), but the average age of diagnosis in the United States is close to 3 years of age (Mandell et al., 2005). This condition differs from other developmental disabilities such as global developmental delay or intellectual disability (American Psychological Asociation, 2013). However, those with ASD show higher rates of comorbid intellectual disability, Attention Deficit Hyperactivity Disorder (ADHD), Down syndrome, Tuberous Sclerosis, Fragile X Syndrome, and other chromosomal abnormalities than the general population (Doshi-Velez et al., 2014; Muhle et al., 2004). ASD, according to the DSM – V, includes those previously diagnosed with three other conditions: Pervasive Developmental Disorder – not otherwise specified, Autism, and Asperger's Syndrome (American Psychological Asociation, 2013).

The social communication difficulties common in ASD are present across multiple environments, and can include difficulties in social-emotional reciprocity, integration of verbal and non-verbal communication, understanding and maintaining relationships, sharing in imaginative play, making friends, or showing interest in peers (American Psychological Asociation, 2013). The restrictive and repetitive components of ASD can involve motor, speech, or action stereotypies (American Psychological Asociation, 2013). Sometimes these might

involve body rocking, hand flapping, echolalia, flipping objects, walking in the same patterns, and or repeating the same phrase idiosyncratically.

In addition to the above, individuals with ASD may show inflexibility or insist on sameness in personal routines. They may show ritualized patterns, distress over small changes, difficulties with transitions, or rigid thinking patterns (American Psychological Asociation, 2013). They might demonstrate highly restricted or fixated interests, which are abnormal either in their intensity or appropriateness. Also, increased or decreased reactivity to sensory stimuli may be present, and individuals might show unusual interest in sensory aspects (American Psychological Asociation, 2013). Sometimes this can manifest as indifference to pain or temperature, excessive smelling or touching, or fascination with movement or lights.

Public health relevance of ASD prevalence trends

Prevalence estimates of ASD in the U.S. continue to rise (Newschaffer et al., 2005). This may be partly due to the combination of several previous diagnoses into an umbrella term of ASD (Shattuck, 2006), or to enhanced screening practices. However, neither of these factors completely explains the rise in ASD prevalence during the past two decades. The most cited estimates of ASD prevalence come from analyses of yearly cross-sectional samples of 8-year-old children from a disability network in the same 11 sites in the U.S. The most current data from these sites estimates ASD occurring in approximately 14.6 per 1,000 (equivalent to 1 in 63) 8-year-old children in 2012 (Christensen, 2016) and 14.7 in 2010 (Developmental Disabilities Monitoring Network Surveillance Year 2010 Principal Investigators & Centers for Disease Control and Prevention (CDC), 2014). These rose from 11.3 in 2008 (CDC, 2012), 9.0 in 2006 (CDC, 2009), 6.6 in 2002 (Autism and Developmental Disabilities Monitoring Network

Surveillance Year 2002 Principal Investigators & Centers for Disease Control and Prevention, 2007), and 6.7 in 2000 (CDC, 2007).

These escalating rates of ASD hold corresponding ramifications for public health spending. According to economic analyses, the cost of supporting an individual with an ASD in the U.S. is 2.4 million dollars over the course of their lifetime (Buescher et al., 2014). The highest costs during childhood are special education services and parental productivity loss (Buescher et al., 2014). In adulthood, the highest costs are individual productivity loss and residential support services (Buescher et al., 2014).

Care providers in any industry serving those with ASD may also require specific training in both behavioral techniques and interdisciplinary communication skills. Individual differences in ASD presentations result in wide variability in service use by individual. In addition, individuals with ASD show higher rates of co-morbid health problems than the general population (Doshi-Velez et al., 2014). During childhood, especially, families use many different services, often for short time periods (Thomas et al., 2007). This inherent heterogeneity in both severity and symptoms, and the lifelong nature of the condition further emphasize the need for interdisciplinary communication and coordinated care, as well as the need for specific training in ASD in a broad range of service professions from dentistry to youth sport league coaching.

Many behaviors exhibited by those with ASD may interfere with routine service delivery from the allied health and education industries Employees involved in the allied health and education must therefore be trained concerning ASD and techniques for management of common behaviors that might interfere with routine healthcare. In general, there is a lack of such training. Studies of students in pre-allied health programs report low representation of ASD content in

required coursework (Freedman, 2014; Price, 2013). Similarly, medical (Shah, 2001) and dental (Weil & Inglehart, 2010) school curricula include very little preparation for handling this patient population.

Etiologies of ASD

Although it was previously believed that Autism was caused by poor parenting, ASD is now understood as a neurobiological disorder of heterogenous etiology. A single predominant cause has not been identified for all ASDs, but there are well-established risk factors for the condition.

Genetic

A component of ASD risk appears to be genetic. Twin studies have contributed a great deal of knowledge to the Autism genetics field. In one study, an average of 60% of monozygotic twins both show classic Autism, and 92% of monozygotic twins share at least a Broader Autistic Phenotype including other communication disorders. In dizygomatic twins, these same rates are 0% and 10% respectively (Monaco & Bailey, 2001). In large epidemiological family studies, younger siblings of children with ASD are up to 14 times more likely to have ASD than younger siblings of unaffected children (Xie et al., 2016). The condition is more common in males than females, with ratio estimates ranging from 4:1 to 10:1 (M.-C. Lai et al., 2015). Pure x-linked heritability appears unlikely, but epigenetic factors involving the X chromosome may be involved (Marco & Skuse, 2006). Specific genes that may contribute to ASD have been identified, but none of these targets singularly accounts for all ASD cases (Monaco & Bailey, 2001). Instead, it appears that interactions between several genes may be to blame, and that the

severity of presentation could be moderated by epigenetic factors and or gene-environment interactions.

Neurological

Research investigating the neurobiological bases of ASD is emergent. There is debate concerning most findings in this area; however, it is largely agreed that brain volume differences exist in those with ASD (Hazlett et al., 2005). Many MRI studies have shown increased brain volume in those with ASD in the cerebral hemispheres, the cerebellum, and the caudate nucleus, and decreased brain volume in the corpus callosum (Stanfield et al., 2008). One review of neuroanatomical changes in the brain of those with Autism suggests that the hippocampus, amygdala, and entorhinal cortex show smaller cells and increased cell density at all ages in those with ASD, indicating halted development in these areas ("Neuroanatomic observations of the brain in autism," 2005). The authors also found that cerebellar differences vary with age, with younger patients showing plentiful and abnormally enlarged neurons, but adult patients showing small, pale neurons reduced in number ("Neuroanatomic observations of the brain in autism," 2005). Findings indicate neuroanatomical differences in the brains of those with ASD, and that these differences are perhaps developmental, with differential effects depending on age. Current and future research in this area investigates neurotransmitter processing (Chugani Diane C. et al., 2001), neuroinflammatory changes (Vargas Diana L. et al., 2004), the neurobiological interaction between intelligence quotient and ASD, and differences in task-specific brain function detected through cerebral blood flow changes (Critchley et al., 2000). In addition, an exciting area of research investigates neuroanatomical differences present in infancy that might serve as predictors of future ASD (Wolff et al., 2012). Other risk factors which confer moderate increases

in risk for ASD include infections during gestation (Brown Alan S., 2012), advanced paternal age (Hultman et al., 2011), and birth via caesarean section (Yip et al., 2017).

Despite the exciting research concerning genetic and neuroanatomical bases of ASD, the current diagnostic process is strictly behavioral. All individuals with ASD are united by the behavioral phenotype, and these behaviors are required for diagnosis. Currently, diagnosis is completed through developmental interviews with caregivers and semi-structured assessments (Lord et al., 1994, 2001). Screening for ASD has become a national priority in the recent decade, with the American Academy of Pediatrics recommending universal screening at 18 and 24 months of age (Zwaigenbaum et al., 2015).

ASD Treatments

There is no known cure for autism. Treatments that improve quality of life and reduce symptoms include various forms of therapy and pharmacologic medications. Effective treatment plans for ASD are as diverse as the individuals affected.

Treatments that address the core deficits of ASD are therapeutic in nature, rather than pharmacological (DeFilippis & Wagner, 2016). In terms of therapeutic services, families of children with ASD most commonly report using speech, language, occupational, behavioral, and recreational therapy (Green et al., 2006; Thomas et al., 2007). For children, these services are most frequently utilized within the school setting (Thomas et al., 2007).

Pharmacology is available to reduce behavioral symptoms of ASD, but no drugs are FDA approved for the core symptoms of ASD (Farmer et al., 2013). The psychoactive drugs Risperidone and Aripiprazole are FDA approved to treat irritability that is common in ASD (DeFilippis & Wagner, 2016). In addition, many individuals with ASD utilize medications for

commonly co-morbid conditions such as sleep disturbances, GI issues, and ADHD. Medication use appears to increase with age over childhood, one study reporting 36% of children under 4 years using medication, and 68% of those aged 9-11 (Thomas et al., 2007).

Behavioral Therapy

Of importance in the current dissertation are behavioral therapies that address the core symptoms of ASD: social-communication deficits and restrictive or repetitive patterns of behavior. Studies investigating the use of such services are subject to a host of issues, primarily due to the method of classifying various therapies. For example, one study uses a survey that asks caregivers about the use of "social skills training" which is not demarcated further into intervention approach (Applied Behavior Analysis, Early Start Denver Model, or Treatment and Education of Autistic and related Communication handicapped CHildren) (Thomas et al., 2007), while others classify these individual approaches into one of several other categories including "combined programs" or "standard therapies" (Green et al., 2006). This confusion often results from differing purposes of each study, namely to determine the context in which services are used (school, family, center-based) (Thomas et al., 2007), or how ASD severity and family characteristics influence service use (Green et al., 2006).

It is, however, generally accepted that the age at which behavioral therapy starts is very important. Intervening early is essential for later outcomes (Corsello, 2005), and this forms the major conceptual point motivating recent public health campaigns for earlier diagnosis of ASD. Early behavioral intervention for children with ASD shows favorable results in cost-benefit analyses, potentially saving \$187,000 – \$203,000 per year when children are 3-22 years of age (Jacobson John W. et al., 1998). Aspects of early intervention that appear to be important for

long-term outcomes include high intensity, family involvement and a focus on generalization to independence outside the immediate teaching environment (Corsello, 2005).

In addition, most studies of service use in children with ASD agree that Applied Behavior Analysis (ABA) is the most common behavioral therapy used. In meta-analyses, early and intense ABA therapy, in particular, is capable of producing large improvements in the learning rate of young children with ASD (Howard et al., 2005; Reichow et al., 2012). When compared to other early intervention approaches, it produces larger improvements on intelligence quotient tests than two other intervention approaches: TEACCH and Colorado (Smith Tristram, 2006).

ABA is based on the work of psychologist B.F. Skinner in the early 1900s (Baer et al., 1968). Skinner's work established the existence of certain laws that govern individual behavior, most notably including the principle of positive reinforcement (Cooper et al., 2007). ABA is the systematic process of tentatively applying these principles toward meaningful behavior improvement, and using data to show that the interventions employed are effective (Baer et al., 1968). In children with ASD, ABA largely focuses on the reduction of socially inappropriate problem behaviors, and the acquisition of appropriate skills in the social, language, cognitive, and adaptive areas. In early ABA intervention, typical targets might be reduction of tantrums, improvement in responding to one's name, and improvement in appropriately asking for preferred items or activities.

The Motor Domain in ASD

Motor difficulties are not a core component of an ASD diagnosis, but they commonly cooccur in this population (Fournier et al., 2010a). According to parent-reported service use surveys, 22 - 39% of children with ASDs have tried physical therapy (PT), and approximately 6 -23% were actively using PT at the time of the survey (Goin-Kochel et al., 2007). Use of occupational therapy (OT) is even higher, with 56 - 86% of children with ASD having tried OT, and 30 - 67% of children with ASD currently using OT (Goin-Kochel et al., 2007). Hypotonia is the most commonly co-occurring motor diagnosis in ASD (51% of an ASD cohort in one study) (Ming et al., 2007). The prevalence of hypotonia in those with ASD reduces with age, but it is unclear whether this is the result of intervention or aging alone. The same study indicates that clinically noted toe-walking and gross motor delay appear in 19% and 9% of children with ASD, respectively (Ming et al., 2007). Thus, motor pathologies are an important issue in children with ASD.

Children with ASD may perform more poorly on motor skill assessments than children with other developmental disabilities, even. A study of 56 children with ASD ages 21-41 months performed at a comparable level to those with developmental delay, and both those with ASD and developmental delay performed worse than those with only developmental concerns and no motor delay diagnosed (Provost et al., 2007). Another study indicated that the motor performance of children with ASD is poorer than those with ADHD (Pan et al., 2009). Motor imitation is another area of importance for children with ASD. Motor imitation is not a core deficit of ASD, but motor imitation difficulties comprise an area of socially relevant deficit in children with ASD (Edwards, 2014; Sevlever & Gillis, 2010; Stone et al., 1997).

Some component of these motor difficulties may be related to ASD severity. A sample of children ages 10-14 with childhood Autism performed more poorly on the Movement Assessment Battery for Children – 2 (MABC – 2) than those with the diagnosis of ASD (Green Dido et al., 2009). In a meta-analysis of motor impairments in ASD, individuals with Autism showed more motor deficits than those diagnosed with Asperger's syndrome or ASD (Fournier et al., 2010b). In a sample of children with high functioning ASD, TGMD – 2 object control scores predicted ASD calibrated severity (M. MacDonald et al., 2013b). Finally, in a study conducted in 149 children with ASD, Pervasive Developmental Disorder – not otherwise specified, and developmental delay from 14-49 months of age, both gross and fine motor scores on the Mullen Scales of Early Learning (MSEL) predicted daily living skills on the Vineland Adaptive Behavior Scales (VABS) (M. MacDonald et al., 2013a). Thus, it appears that children with the most severe may also have the worst motor performance.

The motor domain is also notable in children with ASD in the common occurrence of motor stereotypy (R. MacDonald et al., 2007). Stereotypies are repetitive, purposeless, and partly controllable movements. Many typically developing infants, toddlers, and adults engage in motor stereotypies such as tapping a pencil or fidgeting thumbs. Stereotypy in children with ASD are typically more frequent and may prevent engagement in appropriate activities. Common stereotypies in individuals with ASD include hand flapping, hand twisting, or body rocking. Parent-reported motor stereotypy engagement is positively associated with ASD severity. In addition, IQ shows some influence in the presence of motor stereotypies, with one study suggesting that children with ASD and low IQ engage in motor stereotypy more often than those with ASD and high IQ or those with no ASD and low IQ (Goldman Sylvie et al., 2008).

In addition, early motor behavior shows promise as an early method of ASD detection (Teitelbaum et al., 1998). In one study, infant siblings of children with ASD (and therefore infants at high risk of ASD) showed greater gross motor delays on the Alberta Infant Motor Scale at 3 and 6 months than low-risk infants (Bhat et al., 2012). In the same study, the majority of the high risk infants also showed later social communication delays (Bhat et al., 2012). Another study was able to detect abnormalities at 4-6 months of age in retrospective analysis of home movies from 17 children diagnosed with Autism (Teitelbaum et al., 1998). Particular movement abnormalities included asymmetry of arm support during prone positioning, early rolling without rotation of the trunk, asymmetrical weight distribution while sitting, and asymmetrical walking (Teitelbaum et al., 1998). Other studies have noted high hypotonia (Adrien et al., 1993), persistent asymmetrical tonic neck reflex, and lack of self-protective response when falling in infants who are later diagnosed with ASD (Baranek, 1999).

Fundamental Movement Skills

Fundamental movement skills (FMS) are basic motor abilities that provide the foundation for more advanced specialized sport skills (Gallahue & Ozmun, 2000). FMS are usually developed during early childhood (approximately ages 3 – 10). FMS include locomotor (i.e. running, hopping, and sliding), object control (i.e. kicking a soccer ball, throwing overhand, and dribbling a ball), and balance skills (Gallahue & Ozmun, 2000). FMS are an important component of physical development often emphasized in physical education. Proficiency in FMS predicts future engagement in physical activity, and particularly vigorous physical activity (Jaakkola T. et al., 2015). FMS in typically developing children are developed through an active childhood rich in play and sports.

FMS are typically assessed using standardized assessments, most notably the Test of Gross Motor Development (Ulrich, 2000). This norm-referenced, criterion-referenced assessment is process-oriented in that its scoring criteria are movement components of each skill, rather than concrete outcomes of the skill. For example, the TGMD scores whether a child approaches a soccer ball with a rapid, continuous approach, but does not score how far or how forcefully the child kicks the soccer ball.

FMS proficiency is associated with many components of developmental health. Strong FMS performance correlates with physical fitness measures of cardiorespiratory fitness (Lubans et al., 2010a). In addition, children with proficient FMS also show higher moderate-to-vigorous physical activity (Fisher et al., 2005), and lower BMI (Lubans et al., 2010a; Okely et al., 2004). Data from FMS interventions can provide benefits in cognitive abilities. In one study in disadvantaged preschoolers, participation in a low intensity motor development program improved cognitive scores compared to a control group (Draper et al., 2012). There are gender disparities in FMS, with boys performing object control skills with greater proficiency than agematched girls (Goodway et al., 2010).

Meta-analyses of existing literature agree that FMS deficits appear as a cardinal feature of ASD (Fournier et al., 2010a). School aged children with ASD are significantly impaired in both locomotor and object control skills (Berkeley et al., 2001; Pan et al., 2009; Staples & Reid, 2010), perhaps more delayed in locomotor skills than object control skills (Berkeley et al., 2001). These delays and deficits are consistent with overall motor difficulties in children with ASD, seen as early as toddlers, and progressively becoming more delayed with age (Lloyd et al., 2013; Whyatt & Craig, 2012). In a sample of children with ASD ages 9-12 showed motor proficiency equivalent to typically developing children half their chronological age, and motor skills were

more impaired than would be expected given cognitive level (Staples & Reid, 2010). In the same study, item analysis showed clear difficulties in skills that require coordination of multiple steps in a sequence, coordination of both sides of the body and multiple limbs (Staples & Reid, 2010). This finding is echoed in kinematic studies of locomotor tasks in children with ASD, which indicate difficulties in motor planning (Vernazza-Martin et al., 2005). Thus, persistent motor delays in children with ASD exist, and are functionally important.

FMS intervention is an area of active research for typically developing children (Bardid et al., 2013; Goodway et al., 2003; Goodway & Branta, 2003; S. K. Lai et al., 2014; Logan S. W. et al., 2011; Morgan et al., 2013; Vernadakis et al., 2015). FMS instructional programs may teach FMS for their own sake, or with secondary aims of long-term physical activity engagement and obesity prevention (Capio et al., 2015; S. K. Lai et al., 2014; Logan S. W. et al., 2011). This is because FMS proficiency shows important cross-sectional associations with both physical activity (Cliff et al., 2009; Fisher et al., 2005) and measures of healthy body weight (Hardy et al., 2012; Lubans et al., 2010b; Okely et al., 2004).

A 12-week (1 hour per week) FMS intervention improved the object manipulation skills and overall motor scores of 5 4-year-olds with ASD (Bremer et al., 2015). As a secondary outcome, the study also measured adaptive and social skills. There was no improvement in adaptive or social skills from baseline to post-intervention. Individual FMS improved over the course of a 12 week intervention in an early intervention classroom serving children with ASDlike characteristics from 3-7 years old (Bremer & Lloyd, 2016). Due to a small sample size, this study was not able to present group statistics concerning social skill development, but individual visual analysis indicated improvement in social skill scores due to the intervention. Finally, classroom pivotal response training (CPRT) was used to improve the locomotor and ball skills

scores of 11 children ages 4-6 in a summer-camp based motor skill intervention over 8 weeks (20 hours per week) (Ketcheson et al., 2017b). At the end of this study, children in the intervention group spent less time solitary during free play than at the beginning of the intervention. Thus, motor interventions for children with ASD report varying results regarding secondary social benefits as a result of their motor programming.

Obesity and ASD

The public health relevance of childhood obesity cannot be underestimated, as it increases the risk of adult obesity, cardio-metabolic risk, and all-cause mortality (Sahoo et al., 2015). The risk of obesity is higher for children with ASD (Broder-Fingert et al., 2014; Curtin et al., 2010, 2014; Egan et al., 2013; Hill et al., 2015; Zheng et al., 2017), often due to unique risk factors such as atypical eating habits (Emond et al., 2010), disordered sleep, and reduced engagement in social physical activity. Children with ASD may also face more harmful consequences of obesity than their typically developing peers (Hill et al., 2015). Obesity may adversely affect functional ability (Curtin et al., 2014), social marginalization (Janssen et al., 2004), and medical costs (Tremmel et al., 2017), areas already difficult for those with ASD.

Little research in children with ASD focuses on preventing obesity through the physical domain (motor skills or physical activity). There is evidence, however, that exercise may be helpful for obesity treatment in those with ASD (Pitetti et al., 2007), as well as those with intellectual disabilities (Casey et al., 2010, p.). Most obesity research in children with ASD uses body mass index (BMI) as an outcome measure. It is well understood that body fat percentage may be a more accurate measure of obesity than BMI (Frankenfield et al., 2001), particularly in individuals with unique physical characteristics. Despite this, little research in children with ASD

measures body fat percentage. Studies that do assess body fat percentage in children with ASD do so through bioelectrical impedance (Castro et al., 2017; Pan, 2014; Pan et al., 2016). Those that include a comparison group show no difference in body composition between ASD and typically developing groups (Pan, 2014; Pan et al., 2016). This contrasts with studies of BMI in children with ASD, where it is debated whether children with ASD are more obese. Several studies detect a differences in obesity prevalence based on BMI between children with ASD and typical development (de Vinck-Baroody et al., 2015; Egan et al., 2013), whereas others do not find differences in BMI between the two groups (Curtin et al., 2010; Evans et al., 2012).

Physical Activity and ASD

The evidence regarding physical activity in children with ASD is largely contradictory. A systematic review in 2017 was unable to draw meaningful conclusions because of the methodological heterogeneity and inconsistency of findings in this area (Jones et al., 2017). In a sample of children ages 2-5, children with ASD spent less time sedentary and more time in moderate-to-vigorous physical activity (MVPA) than their typically developing peers (Ketcheson et al., 2017a). In a sample of Taiwanese children ages 7-12, those with ASD did not spend different amounts of school time in MVPA from their typically developing peers (Pan, 2008). Parents of children with ASD ages 3-11 reported that their children engaged in fewer types of PA and spent less time per year participating in them than typically developing children, with age and sex adjusted. However, in the same study, accelerometer measured MVPA was similar between children with ASD and typical development (Bandini et al., 2013). A sample of school-aged youth with ASD spent more time sedentary, less time in moderate and light PA, and less time in MVPA, than typically developing peers. In addition, the group with ASD was less physically fit on a series of physical fitness assessments (Tyler et al., 2014). In secondary-school

aged children, participants with ASD showed less accelerometer-measured MVPA than typically developing youth (Pan et al., 2016). A sample of adolescents spent less time in accelerometer-assessed MVPA than age-matched typically developing peers (Stanish et al., 2017). In sum, the evidence concerning how active children with ASD are relative to their peers is complicated.

Thus, there appear to be some age-dependent differences in physical activity of children with ASD, compared to their typically developing counterparts. The inconsistency of the above findings may be due to an effect of age, but also due to differing physical activity measurement techniques, and the implicitly heterogenous character of samples of children with ASD. Research in this population typically includes small sample sizes, and the wide variability in ASD severity presents difficulties for representative sampling. There is also the possibility that physical activity quality is different in children with ASD than their typically developing peers. Motor stereotypy or hyperactivity could potentially contribute to measures of physical activity, but this physical activity would not be considered health enhancing or developmentally appropriate.

Dynamic Systems Theory

Dynamic Systems Theory (DST) (Smith & Thelen, 1993) provides a useful theoretical framework from which to understand ASD and the constructs presented in this dissertation. DST, originally developed by theoretical physicists, posits that any phenomenon can be understood as a dynamic and reciprocally connected network of sub-systems. When one sub-system changes, others react in often unpredictable ways. According to DST, these sub-systems self-organize to produce a phenomenon, much like air currents self-organize to produce a tornado. These dynamic interactions result in changes in a phenomenon that are necessarily non-linear over time. This theory has been used by developmentalists in the motor development and motor

control fields to understand the emergence of human behaviors (Clark Jane E. & Phillips Sally J., 2008; Hemami & Wyman, 1979; Thelen et al., 1991).

For example, the emergence of independent locomotion in infants can be interpreted according to DST. Other theories might assert that independent locomotion occurs at a predetermined chronological age depending on the innate neural capacity of the individual, or that independent locomotion occurs only after standing alone because there is a pre-programmed motor skill sequence. DST instead says that the emergence of independent locomotion comes from the dynamic interactions of heterarchical, overlapping, multiple, and co-existent subsystems which interact unpredictably. It could be that this skill depends upon the infant's bone, muscle, and joint structure, individual motivation and practice afforded by the parenting style and home environment. DST would assert that independent locomotion may become unstable and regress to crawling for periods of time because of some sub-systems changing, or that locomotion may be established once sub-systems remain stable.

In the same way, the constructs in this dissertation can be understood from a DST perspective. The following three manuscripts discuss the concepts of obesity, fundamental movement skills, and social play in children with ASD. Each of these concepts can be understood as behaviors emerging from the self-organization of sub-systems.

Manuscript 1 addresses obesity development in children with ASD by measuring differences in body composition, fundamental movement skills, and physical activity compared to typically developing peers. Age, gender, and ASD severity are also measured in this manuscript. DST might interpret the emergence of obesity as a product of multiple co-existent risk factors in this population, including those measured in the manuscript.

Manuscripts 2 and 3 describe the effects of an early intervention implemented within a clinical applied behavior analysis center. In Manuscript 2, the intervention's fundamental movement skill outcomes are presented. In Manuscript 3, the social play learning rate results of the intervention are presented.

The intervention discussed in Manuscripts 2 and 3 is designed incorporating the concepts of DST. It intentionally targets multiple domains of development through multiple redundant instructional methods. Discrete trial training in fundamental movement skills is an integral part of the instructional strategy. Here, a structured format with clear expectations forms a scaffold for learning FMS, imitative skills, and attentional skills. The incorporation of video modeling, live modeling, manual prompting, contingent reinforcement, and a picture task card ensures that multiple potential sub-systems are addressed, optimizing an individual's capacity to learn the task at hand. A second component of the intervention is a socially active group game incorporating FMS. Again, multiple domains are targeted to encourage socially appropriate active play: social skills, language skills, group game rules, FMS, physical activity, and following directions. And again, multiple instructional sub-systems are targeted to optimize learning: a video model, manual prompts, therapist-level discriminative stimuli, and contingent reinforcement. Considering the heterogenous presentation of children with ASD, and the concepts of DST, this multi-dimensional approach to intervention can potentially influence overall outcomes effectively.

This dissertation aims to provide evidence concerning two aspects of FMS in children with ASD. First, the concept of obesity development is addressed in relation to FMS by measuring the differences in FMS, body composition, and physical activity of children with and without ASD. Second, FMS is addressed as a target of early therapeutic intervention. Manuscript

2 presents the FMS outcomes of an early ABA therapy intervention targeting FMS and social active play. Manuscript 3 presents the social play learning rate outcomes of the same intervention.

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CHAPTER 2

Differences in Fundamental Movement Skills, Body Composition, and Moderate - to - Vigorous Physical Activity of Children with and without Autism Spectrum Disorder

Abstract

The public health relevance of obesity for children with Autism Spectrum Disorder (ASD) continues to increase. Much research has been conducted to measure the differences between children with and without ASD in both obesity and several lifestyle factors obesity-related areas. Despite this, there is a lack of consensus on the magnitude of these differences. The purpose of the current observational study investigates the fundamental movement skills, body composition, and moderate – to – vigorous intensity physical activity (MVPA) of children with and without ASD. Measurement methods of these constructs were the Test of Gross Motor Development III, air displacement plethysmography, and accelerometry at the right hip for seven days, respectively. Results showed the largest difference between diagnostic groups in fundamental movement skills (F (1,19) = 46.479, p < 0.001), followed by body composition (F (1,19) = 46.729, p < 0.001), and no difference in MVPA (F (1, 40) = 0.136, p = 0.714). Findings underscore the disparities in fundamental movement skills in children with ASD.

Introduction

Fundamental movement skills (FMS) are foundational motor skills that underlie more specialized movements in structured and unstructured physical activity (Lubans et al., 2010); children with Autism Spectrum Disorder (ASD) show delays in FMS. Evidence exists of early motor delays in children ASD that worsen with age (Lloyd et al., 2013). In a sample of children with ASD ages 6 – 8 years, 73% showed *poor* or *very poor* FMS compared to Test of Gross

Motor Development (TGMD) population norms, with larger deficits existing in object control (manipulation of sports equipment), than locomotor scores (Berkeley et al., 2001). In another study, children ages 6-10 years with ASD showed worse motor performance than both typically developing children and those with Attention Deficit Hyperactivity Disorder (ADHD), when age was statistically controlled (C.-Y. Pan et al., 2009). Children with ASD ages 9-12 demonstrated FMS on the TGMD - 2 equivalent to typically developing children half their chronological age, and performed worse than typically developing children matched for cognitive ability (Staples & Reid, 2010). A sample of adolescents with ASD showed lower scores on the Bruininks-Oseretsky Test of Motor Proficiency – 2 (BOT – 2) than age-matched peers (C.-Y. Pan, 2014). Delays in motor skills are well-established in children with ASD.

Some components of the motor difficulties experienced by children with ASD are related to ASD severity. Adolescents with Autism diagnosed in childhood performed more poorly on a motor skill assessment than those with the more heterogenous diagnosis of ASD (Green Dido et al., 2009). In a meta-analysis of motor impairments in ASD, individuals with Autism showed more motor deficits than those diagnosed with Asperger's syndrome or the more heterogenous diagnosis of ASD (Fournier et al., 2010). In a sample of children ages 6 - 15 with high functioning ASD, TGMD – 2 object control scores actually predicted ASD calibrated severity (MacDonald et al., 2013). Thus, it appears that some component of the motor delays interacts with Autism symptoms themselves.

In typically developing children, FMS show important relationships with weight status and have gained research attention for obesity prevention (Barnett et al., 2016; D'Hondt et al., 2014). It may be helpful to consider this prospect in those with ASD as well, because the risk of obesity is higher for these children (Broder-Fingert et al., 2014; Curtin et al., 2010, 2014; Egan et

al., 2013; Hill et al., 2015; Zheng et al., 2017), often due to atypical eating habits (Emond et al., 2010), disordered sleep, and reduced engagement in social physical activity. In addition to a higher obesity risk, children with ASD may also face more harmful consequences of obesity because ASD alone has risk factors that compound those of obesity. For example, children with ASD and obesity may experience bullying, functional dependence, and medical care costs more than a child with only one of these diagnoses (Hill et al., 2015). Little research in children with ASD focuses on preventing obesity through physical activity or motor skill development. There is evidence, however, that exercise may be helpful for obesity *treatment* in those with ASD (Pitetti et al., 2007), as well as those with intellectual disabilities (Casey et al., 2010, p.).

Studies investigating body composition differences between children with and without ASD generally suggest that children with ASD are more obese than their peers. However, these findings are complicated by some contrasting results and some methodological considerations. Most obesity research in children with ASD uses body mass index (BMI) as the obesity measure. However, body fat percentage may be a more accurate measure of obesity than BMI (Frankenfield et al., 2001). Despite this, little research in this population measures body fat percentage. Currently, the only identified studies measuring body fat percentage in children with ASD do this through bioelectrical impedance (Castro et al., 2017; C.-Y. Pan, 2014; C.-Y. Pan et al., 2016). When a comparison group is included, no difference in body fat percentage emerges between ASD and typically developing groups (C.-Y. Pan, 2014; C.-Y. Pan et al., 2016). This contrasts with similar studies using BMI, where this concept is debated; some detect a difference in obesity prevalence based on BMI between children with ASD and typical development (de Vinck-Baroody et al., 2015; Egan et al., 2010; Evans et al., 2012).

The evidence regarding PA and sedentary activity patterns of children with ASD is also contradictory. A systematic review conducted in 2017 could not draw meaningful conclusions because of the paucity of consistently measured evidence, and inconsistency of findings (Jones et al., 2017). In the early years (ages 2-5), children with ASD may spend less time sedentary and more time in moderate-to-vigorous physical activity (MVPA) than their typically developing peers (Ketcheson et al., 2017). In other samples, there were no differences in the amount of school time spent in MVPA between those with ASD and those without (C.-Y. Pan, 2008). Parents of children with ASD ages 3-11 years reported that their children engaged in fewer types of PA and spent less time participating in them than typically developing children. However, in the same study, accelerometer measured MVPA was similar between children with ASD and typical development (Bandini et al., 2013). A sample of school-aged youth with ASD spent more time sedentary, less time in moderate and light PA, and less time in MVPA, than typically developing peers. In addition, the group with ASD was less physically fit on a series of physical fitness assessments (Tyler et al., 2014). Similarly, in secondary-school aged children, participants with ASD showed less accelerometer-measured MVPA than typically developing youth (C.-Y. Pan et al., 2016). Another study in adolescents founds similar results (Stanish et al., 2017). Thus, the question of whether children with ASD are more or less active than their peers is still debated.

Purpose

Considering the uncertainty still present in the literature regarding body composition and physical activity of children with ASD, the purpose of this study was to examine differences in FMS, body composition, and MVPA between a sample of children with ASD and age- and sex-matched typically developing peers.

Methods

Participants

An a priori power analysis was conducted in G*Power with an α of 0.05, power of 80%, and 4 groups (2 diagnostic groups – ASD and typically developing, and 2 covariates – socioeconomic status and ASD CSS). An f² effect size of 0.16 was included, which indicates a medium difference between groups (Cohen, 1988). The medium effect size was chosen based on the dependent variable of MVPA, for which there exists the most variability in literature detecting differences between children with and without ASD. A previously published study found differences in MVPA between school children with and without ASD, but was only able to detect differences during particular periods of the day (C. Pan, 2008). The smallest significant effect size for one of these periods in which a difference in MVPA was detected between children with and without ASD was $\eta^2 = 0.15$, indicating a moderate effect size (Cohen, 1988). This power analysis suggested that 40 participants (20 with ASD, 20 typically developing) were needed to accurately detect moderate effect sizes.

Twenty - three children with ASD were recruited through community events, and local pediatrician clinics in Michigan. Children were eligible for the study if they were 3 - 10 years and their primary guardian provided verbal verification of a medical diagnosis of ASD. Age and

sex-matched typically developing children were recruited (n = 23) through Michigan State University e-mail lists. Children were included if they were the same sex and fell within 12 months of the age of one of the children with ASD, and their primary guardian provided verbal verification that they did not have ASD.

Measures

Demographics

Primary caregivers completed a demographic survey including items concerning age, race, gender, annual family income, family education, and number of children living in the home. These variables are presented as descriptive statistics. Annual family income represented socioeconomic status and is included as a covariate in statistical analysis. Although we asked for each child's parents' height and weight, we did not use them to adjust for pubertal maturation, as we limited the sample to 3 - 10 year – olds.

ASD calibrated severity score

Autism Diagnostic Observation Schedule – 2 (ADOS – 2) provided a measure of ASD calibrated severity for participants with ASD (Gotham et al., 2009). The ADOS – 2 is a standardized semi-structured interview used clinically for the diagnosis of ASD, with excellent discriminative ability by diagnosis, stability of scores over time, and inter-rater reliability (Lord et al., 1989). The ADOS – 2 was administered by a graduate student trained to reliably administer and score using the ADOS – 2 updated algorithms (Gotham et al., 2007). The calibrated severity score (CSS) is a score from 1-10. These data are provided for descriptive purposes, as well as used as a covariate in statistical analysis.

Anthropometrics

Height without shoes was measured to the nearest 0.2 cm using a Seca standing stadiometer one time. Weight while wearing the minimal clothing worn in the Bod Pod was measured to the nearest 0.2 kg using a standing scale as the average of two measurements; both were completed again if measurements were more than 0.4 cm different. Weight measurement was replicated because the weighing procedure was used as a "warm up" to the Bod Pod measurement, done in the same room and wearing the same clothing. These values were used to calculate BMI, then transformed to BMI z-scores and BMI percentiles based on Centers for Disease Control growth curves (Centers for Disease Control and Prevention, National Center for Health Statistics, 2000). In addition, Lange skinfold calipers were used to obtain skinfold measurements at four sites for a measure of site-specific subcutaneous adiposity (biceps, triceps, subscapular, suprailiac). Skinfolds were obtained twice in each site and averaged; both measurements were repeated if the first measurements were more than 2 mm different. Body fat percentage was calculated from skinfold thicknesses through the following equations: Body density (boys) = $1.1690 - 0.0788 \times \log$ (sum of skinfold thicknesses at 4 sites) (Brook, 1971), Body density (girls) = $1.2063 - 0.0999 \times \log(\text{sum of skinfold thicknesses at 4 sites})$ (Brook, 1971), and Body fat percentage = [(4.95/body density) - 4.5]*100 (Siri, 1956). These data are presented to describe the sample.

Body fat percentage

Air Displacement Plethysmography (ADP) (Bod Pod) is considered the gold standard technique to measure body fat percentage and provided a measure of body composition in this sample (Fields et al., 2002). All participants wore a swimming cap over their hair and minimal form-fitting clothing for their scans. The Bod Pod was calibrated with an iPad inside of it, and participants held and used an iPad while undergoing measurement. Bod Pod software used the Lohman model to provide an estimate body fat percentage which is used in statistical analysis.

FMS

The Test of Gross Motor Development – 3 (TGMD – 3) is a norm- and criterionreferenced test of FMS for children ages 3 to 10.9 years (Ulrich, 2013). It tests six locomotor skills: run, gallop, skip, hop, horizontal jump, slide and seven ball skills: two-hand strike of a stationary ball, one hand strike of a self-bounced ball, dribble, kick, two-handed catch, overarm throw, underarm throw. Test-retest reliability coefficients for the TGMD – 3 range from 0.95 – 0.97, and the test shows acceptable item difficulty and item discrimination ability, as well as acceptable construct validity (Webster & Ulrich, 2017). Participants are provided a live model of the skill, afforded an opportunity to practice, then asked to perform the skill twice. Assessments occurred in a gym and were administered by a graduate student who had prior training in the correct shape of each skill and had administered over fifty TGMD – 3 previously. There are 3-5 performance criteria for each skill, scored with a 0 when not achieved, and a 1 when achieved. Performance on the TGMD – 3 skills were video-recorded for later scoring, which has been shown to provide excellent reliability with live scoring (Rintala et al., 2017). Undergraduate research assistants scored video-recorded TGMD – 3 administrations. All were trained through scoring video-recorded TGMD administrations of typically developing children provided by the authors of the TGMD – 3. The first author explained any divergent items between the evaluators' answers and the provided keys. To control for potential errors in experimenter drift, two raters would together re-score a test if two scorers independently disagreed on more than 15% of the items on a TGMD – 3 assessment. The TGMD – 3 total raw score was used in statistical analysis. The interrater reliability of the two scorers was 84%. Locomotor and ball skill subtest scores are presented to characterize the sample.

Physical Activity

To measure physical activity, participants were asked to wear an Actigraph wGT3x-BT (Pensacola, FL) triaxial accelerometer at the right hip for seven days. Data were excluded if the accelerometer was worn for less than 4 days or less than 10 hours per day. Accelerometers were worn on an elastic belt around the hips. Participants who were apprehensive to wear the belt were provided social stories, and/or felt fabric covers for the belts (Hauck et al., 2016). All accelerometer data were downloaded and analyzed using ActiLife 6 software. 1-second epochs were used when collecting data in order to provide high resolution data in an effort to account for the wide age range of participants. Cut-points for children ages 5 and up were applied to demarcate sedentary (0-100 counts per minute (CPM), light (101-2295 CPM), moderate (2296-4011 CPM), and vigorous (>4012 CPM) physical activity intensity (Evenson et al., 2008). The cut-points used were developed with 15 second epochs, but ActiLife 6 can only apply cut points to files with 60 second epochs. The software therefore reintegrated the datafile to 60 second epochs and applied the Evenson cut points multiplied by 4. The cut points were applied to the data from the vertical axis only, consistent with the methods by which the Evenson cut points were created. Time spent in each category was divided by the total wear time to produce

percentage of wear time spent in each intensity category. These data are presented to describe the sample. MVPA is particularly associated with positive health and fitness outcomes, and thus percentage of wear time in MVPA is used in statistical analysis.

Procedures

The project was approved by the Human Research Protection Program at Michigan State University. Informed consent was explained to participants and primary caregivers. All primary caregivers provided informed consent for their children to participate, and participants provided verbal assent. Assessments occurred at a university laboratory in a building that also housed an available gym for TGMD – 3 administration and a separate space that housed the Bod Pod.

Statistical analysis

Descriptive statistics of both groups (ASD and typically developing) were represented by means and standard deviations. A correlation matrix among all three dependent variables (TGMD – 3 raw score, percentage of wear time in MVPA, and body fat percentage) was used to assess the appropriateness of a multivariate analysis of covariance (MANOVA). Because there was no significant correlation between percentage of wear time in MVPA and either of the other two variables (TGMD – 3: r = -0.146, p = 0.362; body fat percentage: r = -0.348, p = 0.122), three separate univariate analyses of variance (ANOVAs) were used instead. It can be noted that TGMD – 3 and body fat percentage were moderately correlated (r = -0.679, p = 0.001) (The three ANOVAs analyzed TGMD – 3 raw score, percentage of wear time in MVPA, and body fat percentage by group (ASD or typically developing). Socioeconomic status and CSS did not correlate with FMS, MVPA, and body fat percentage variables, so they were not statistically controlled in the analysis. Effect sizes of differences between groups in main outcome variables

were calculated as partial η^2 . Missing data were coded as missing and excluded from analysis, except in the case of body fat percentage. If body fat percentage measurement was not completed through the Bod Pod, this value was calculated from the skinfold measurements obtained (Brook, 1971). Although we recognize this as a limitation, post-hoc independent t-tests ensured that there was no difference between the body fat percentages of children who completed the Bod Pod and those who did not (t = - 0.498, p = 0.622). In addition, the skinfold-based body fat percentage variable and the Bod Pod-based body fat percentage variable showed a high correlation (r = 0.926, p < 0.001).

Results

Participant information is provided in Tables 2.1 and 2.2. The total sample's BMI percentile rested close to 50%, with a large difference between children with ASD and TD (74.4 \pm 30.9 % vs. 42.5 \pm 26.6 % respectively). Typically developing children were 90.9% normal weight and 9.1% overweight. Children with ASD were 50.0% normal weight, 18.2% overweight, and 31.8% obese. For between subjects effects, diagnosis illuminated a significant difference in total TGMD – 3 score (F (1,19) = 46.729, p < 0.001, $\eta^2 = 0.570$) and body fat percentage (F (1, 26) = 46.729, p < 0.001, $\eta^2 = 0.711$), but not MVPA (F (1, 40) = 0.136, p = 0.714, $\eta^2 = 0.003$). Partial η^2 effect sizes of the effect of diagnosis on FMS, body fat percentage, and MVPA, were large, large, and non-significant, respectively. Comparing the magnitude of differences in each variable, the largest difference between children with ASD and TD was in FMS (28.5 \pm 22.7 compared to 63.5 \pm 14.6), followed by the difference in body fat percentage (22.5 \pm 14.2 % compared to 13.2 \pm 4.8 %). The standard deviations of both variables in children with ASD were higher than those with TD. These results are represented visually as box and whisker plots in Figure 2.1.

Discussion

In this sample of 46 children ages 3 - 10 years (82.970 ± 21.165 months), there were differences between children with ASD and TD in FMS and body fat percentage, but not MVPA. FMS showed a greater difference between diagnostic groups than body fat percentage. As expected, effect sizes mirrored this, with diagnosis showing a large effect size on FMS, a large effect size on body fat percentage, and none on MVPA.

For FMS, our sample replicated findings of many studies that find differences between the children with and without ASD (Ketcheson et al., 2017; Staples & Reid, 2010). The replication of these findings underscores the importance of FMS intervention for children with ASD. In addition, it reinforces the need for more research regarding the causes of motor delays in children with ASD. The reasons for motor skills delays in children with ASD are still poorly understood. Considering existing knowledge that children with ASD communicate differently than others, there may be assessment techniques that better unlock the actual abilities of children with ASD, and it is unclear if presently available measures fit this description (Allen et al., 2017; Breslin & Rudisill, 2011; Wilson et al., 2018). The current study did not use picture task cards, video modeling, or any other means of making the TGMD-3 more accessible to children with ASD. Thus, it is unclear whether our measurement of FMS in this sample has true internal validity.

For body fat percentage, our study replicated findings suggesting a difference in the body composition of children with and without ASD (Hill et al., 2015). Our results contrast those of some studies that find no difference in body composition by diagnosis. Although this is a debated area of the literature, the findings of this study by no means settle the question. This study

showed large differences in body fat percentage between children with and without ASD. A potential explanation for the conflicting findings in this area of literature is the heterogeneity in studies regarding inclusion of secondary conditions, medication use, and other important covariates. Those data were not collected and therefore not statistically controlled. If they were available, it is possible our results would have been tempered.

Estimates of obesity rates in children with ASD typically fall somewhere between 18 - 35%, which is higher than the general population (Granich et al., 2016; Hill et al., 2015; Must et al., 2017). These estimates are from the most recently published evidence from studies with large representative datasets and should generally be trusted. One of these studies is an observational analysis of over 5,000 children in the Autism Speaks Autism Treatment Network (Hill et al., 2015). Another study draws from over 43,000 children with ASD (Must et al., 2017). The highest estimate of these studies (35% obese) comes from a study of 208 children ages 2 - 16. In the present study, children with ASD were 31.8% obese. It is not the intention of this study to support or contradict these large epidemiological studies. Instead, we hoped to provide a high-quality measure of body composition and study it within its context of relatedness to FMS and PA, rather than using it as the only outcome measure.

The finding suggesting a difference in the body fat percentage of children with and without ASD should be interpreted with caution because (1) there are limited comparative Bod Pod data available for children with ASD and (2) our sample of children with TD appears quite lean compared to the general population. To our knowledge, this is the first study that presents Bod Pod measurements for children with ASD. This is a strength, especially considering the high ASD severity of the sample and the relative difficulty of obtaining Bod Pod measurements compared to weight and height. In one recent study using bioelectrical impedance analysis to

quantify the body fat percentage of children with ASD, the average body fat percentage was 23%, (22.5% in the current study), but this was among a wider age range of children (4 - 16) years), and took place in Brazil (Castro et al., 2017). The BMI percentiles of our sample with ASD appear to be low compared to some data reported in another study (Heffernan et al., 2018). It is very clear, however, that our sample with TD appears to have body fat percentages much lower than published Bod Pod data from similar children (Nieman et al., 2015). Thus, although the use of the Bod Pod in an ASD population is novel, results regarding comparison to the typical population should be interpreted with caution because this sample may not be entirely representative of norms.

The seriousness of the obesity disparity in children with ASD is of enormous practical importance, considering the myriad health risks associated with obesity and the already vulnerable state of children with ASD (Hill et al., 2015). Indeed, there is compelling evidence that the disparities in obesity rates between children with and without obesity actually increase with age (Must et al., 2017). As the rates of ASD rise in the U.S., we may have a particularly difficult health crisis on our hands (Baio, 2018). Finally, with increasing age, the likelihood that children with ASD cause damage with aggressive or self-injurious behaviors increases, and increased risk of obesity with age only further exacerbates this issue. Obesity may be particularly challenging for caregivers of children with ASD to manage.

Other studies concerning obesity in children with ASD offer some potential mechanisms, but they offer such different explanations that it is challenging to make any meaningful conclusions. For example, Granich et al's result indicate that many factors did not relate to weight status in children with ASD; these included gender, age, parental education, family income, ethnicity, autism spectrum disorder severity, social functioning, psychotropic and

complementary medication use (Granich et al., 2016). Interestingly, the same study found no differences in the MVPA of children with ASD compared to those with TD. Conversely, McCoy et al found higher rates of obesity in children with ASD, and more importantly that this difference in obesity rates could be almost entirely explained by a lack of PA engagement in the children with ASD (McCoy et al., 2016). Another study finds that the higher obesity rates in children with ASD can be completely explained when adjusting for medication use and secondary health conditions (Corvey et al., 2016). Thus, the only clear conclusion within the field of obesity in ASD is that the mechanisms are very complicated.

The present study finds no difference between the MVPA of children with and without ASD. This agrees with some research studies previously published (Sandt & Frey, 2005), and also contradicts others (McCoy et al., 2016). Findings of the current study do not offer much assistance in definitively answering this research question. However, these results confirm the finding that the study of MVPA in ASD is complex. There may be unexpected factors at play in the reasons for actual differences in PA behavior in autism. In addition, there may also be significant measurement concerns specific to this population. For example, it still is not clear how much of the MVPA captured by accelerometry is health-enhancing in children with ASD. Children with ASD can engage in restrictive and repetitive behaviors such as jumping, flapping their hands, or other motor stereotypies; there is also the possibility of hyperactivity or aggression. Stereotypy, hyperactivity, and aggression might differentially contribute to PA measurements and effect results in unpredictable ways. In the future, a secondary analysis of the PA data from this sample might prove illuminating to at least offer more context to the PA behavior. Such analyses could compare step counts, daily or weekly patterns of activity, sedentary, light, and vigorous PA between diagnostic groups. In addition, future studies might

attempt a measure of stereotypy along with PA measurement, to remove the effect of these behaviors.

This sample failed to meet the guidelines for school-aged children in the U.S. for moderate – to – vigorous physical activity (Piercy et al., 2018). Both children with ASD and TD fell below 60 minutes per day of MVPA (57.900 \pm 22.064 min. vs. 54.223 \pm 12.699 min. respectively). As with FMS and body fat percentage, this sample of children with ASD showed large variability in MVPA per day and percentage of wear time in MVPA compared to the children with TD (Figure 1 offers a visual of the spread of data). It might be that the relatively high severity of ASD represented in this study (6.85 \pm 1.95 on a scale of 1 - 10) may be responsible. Although CSS did not correlate with MVPA, most of our sample showed high severity and this may play a role. Otherwise, the wide age range represented in this sample may contribute to the wide MVPA pattern distribution. Although we control for age using calendar age-matched controls, there is some debate concerning the correct use of age when controlling for PA differences in children with ASD vs. TD. Other studies might choose to use developmental age, rather than simple calendar ages.

The results of this study emphasize the need for FMS and obesity interventions for children with ASD. It is also important to note that the current study takes place before puberty. Puberty is a period of large hormonal changes that can enhance obesity risk further. With puberty also may come metabolic complications from new medications, as puberty might be the first time that a child's body is large enough to cause damage during problem behaviors. According to other evidence, the risk of motor deficiency and obesity rises with age in children with ASD (Lloyd et al., 2013; Must et al., 2017). All of this underscores the necessity for early targeted

intervention for children with ASD, given strong evidence that early intervention is effective for children with ASD (Reichow et al., 2012).

This study has several important strengths and limitations. The sample size is smaller than comparable studies that investigate differences in only one area of interest (Bandini et al., 2013). In addition, the typically developing participants included here appear to be a rather lean group, and therefore are not particularly representative of the U.S. Considering methods, this study uses an objective measure of PA and the gold standard measure for body fat percentage.

Conclusions

This study provides confirmatory evidence of differences in FMS and body composition between children with and without ASD, while controlling for each other and MVPA. This study also finds no difference in the quantity of MVPA between children with and without ASD. These results emphasize the importance of quality FMS and obesity prevention interventions in this population, and the complexity of assessing PA in children with ASD. APPENDIX

TABLES

	TD (N = 23)		ASD (N = 23)		Total ($N = 46$)	
	Mean	SD	Mean	SD	Mean	SD
Age (months)	84.32	21.42	81.62	21.32	82.97	21.17
CSS	-	-	6.85	1.95	6.85	1.95
Race	16 white		13 white		29 white	
	5 black		4 black		9 black	
	1 Asian		3 Asian		4 Asian	
			1		1	
			American		American	
			Indian or		Indian or	
			Alaska		Alaska	
			Native		Native	
			• • •		2 X C · ·	
D (1 · ·)	20		2 missing		2 Missing	
Ethnicity	20 non-		18 non-		38 non-	
	Hispanic		Hispanic		Hispanic	
	2		5 Hispanic		7 Hispanic	
a 1	Hispanic					
Gender	5 female		4 female		9 female	
	17 male		19 male		36 male	
Mother	2 SC		3 HS		3 HS	
Education	8 BS		2 SC		4 SC	
	9 MS		4 AA		4 AA	
	2 PhD		4 BS		12 BS	
	1 missing		7 MS		16 MS	
			3 missing		2 PhD	
					4 missing	
Annual	2 < \$25k		3 < \$25k		5 < \$25k	
Household	11 < \$100k	Σ.	6 < \$50k		6 < \$50k	
Income	8 > \$100k		6 < \$100k		17 < \$100k	
	1 missing		8 > \$100k		16 > \$100k	
					1 missing	

Table 2.1.Participant demographic values.

Note: TD = Typically developing; ASD = Autism Spectrum Disorder; SD = Standard deviation; CSS = ADOS – 2 calibrated severity score; HS = High school diploma; SC = Some college; AA = Associate's degree; BS = Bachelors' degree; MS = Master's degree; PhD = Doctorate or professional degree; TGMD = Test of Gross Motor Development; MVPA = Moderate – to – vigorous intensity physical activity; CPM = counts per minute; BMI = Body mass index.

•	TD (N = 23)		ASD (N = 23)		Total $(N = 46)$	
	Mean	SD	Mean	SD	Mean	SD
Locomotor	28.8	6.4	11.2	10.8	19.6	12.5
Ball skills	33.9	9.7	10.6	11.0	21.7	15.6
TGMD-3 total	63.5	14.6	28.6	22.7	49.8	24.9
Sedentary	73.62	15.77	76.27	6.87	74.94	12.09
Percentage						
Light Percentage	15.24	2.898	15.79	5.57	15.51	4.39
Moderate	3.66	0.75	3.51	1.15	3.59	0.96
Percentage						
MVPA	7.83	1.56	7.57	2.72	7.73	2.05
percentage						
Vigorous	4.16	1.00	4.57	2.22	4.36	1.71
Percentage						
Average daily	54.22	12.70	57.90	22.06	56.06	17.88
MVPA						
(minutes)						
CPM	1160.61	231.60	1196.69	354.98	1178.65	296.59
Steps per minute	9.43	2.07	10.05	2.50	9.74	2.29
Calendar days of	9.29	1.65	8.05	1.75	8.67	1.79
accelerometer						
wear						
BMI percentile	42.50	26.62	74.42	26.71	58.46	30.90
Body Fat	13.2	4.8	22.5	14.2	16.9	10.5
Percentage (Bod						
Pod)						

Participant fundamental movement skills, physical activity, and body fat percentage data.

Table 2.2.

Note: TD = Typically developing; ASD = Autism Spectrum Disorder; SD = Standard deviation; CSS = ADOS - 2 calibrated severity score; HS = High school diploma; SC = Some college; AA = Associate's degree; BS = Bachelors' degree; MS = Master's degree; PhD = Doctorate or professional degree; TGMD = Test of Gross Motor Development; MVPA = Moderate - to - vigorous intensity physical activity; CPM = counts per minute; BMI = Body mass index.

15.00[.] 100.00 ** Percent of Wear Time in MVPA TGMD – III total 80.00 10.00 ** 60.00 40.00 5.00 20.00 .00 .00 Typical Development ASD Typical Development ASD ** 60.00-Body Fat Percentage 50.00-40.00⁻ 30.00 ** 20.00 10.00 .00 ASD Typical Development

FIGURES

Figure 2.1. Box – and – whisker plots of fundamental movement skills (TGMD – III total scores), percentage of wear time in Moderate – to – Vigorous Physical Activity, and body fat percentage data separated by diagnosis (Typically developing and Autism Spectrum Disorder). Note: *p < 0.05; **p < 0.001 different by diagnosis.

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CHAPTER 3

Training the Fundamental Movement Skills of Preschoolers with Severe Autism Spectrum Disorder: Preliminary Efficacy and Feasibility in Early Intensive Behavioral Intervention Centers

Abstract

It is increasingly clear that motor deficits are intertwined with autism, but published motor interventions are delivered by motor experts. This randomized controlled trial assessed the preliminary efficacy and feasibility of motor training within early intensive behavioral intervention (EIBI) centers. Thirteen Applied Behavior Analysis behavior technicians (BTs) without a motor background and 14 children with autism participated for 20 weeks. Blinded motor assessment occurred before, after, and 4-weeks post intervention. BTs took a feasibility survey post-intervention. Motor scores improved relative to the control group with a large effect size (F (1, 12) = 4.983; p = 0.016; ES = 0.312). BTs expressed polarized views, rating the intervention low in feasibility overall. Findings hold relevance for interdisciplinary collaboration in EIBI centers.

Introduction

Fundamental movement skills (FMS), basic motor prerequisites to physical activity engagement, are increasingly prominent intervention targets for children with Autism Spectrum Disorder (ASD). Evidence exists that children with ASD show motor delays as infants and toddlers that magnify with age (Lloyd, MacDonald, & Lord, 2013), and that by ages 9 - 12years, these children perform at a level equivalent to those approximately half their chronological age (Staples & Reid, 2010). Given evidence that early intervention is helpful for children with ASD (Reichow, Barton, Boyd, & Hume, 2012), early FMS interventions are particularly

common (Bremer, Balogh, & Lloyd, 2015; Bremer & Lloyd, 2016; Ketcheson, Hauck, & Ulrich, 2017). FMS do not "naturally" develop through maturation (Clark, 2005), but must be intentionally taught and practiced (Logan S. W., Robinson L. E., Wilson A. E., & Lucas W. A., 2011). This may be particularly true for children with ASD, who likely require more practice to master FMS than their typically developing peers. Early FMS interventions are therefore garnering increased research attention.

FMS appear intertwined with social and communication skills, and FMS interventions may therefore offer benefits that cascade into domains beyond the physical. A recent motor intervention showed that social communication skills moderated motor skill improvements; children with the most severe ASD showed the biggest gains in motor skills (Bo et al., 2019). A recent FMS early intervention also achieved benefits in spontaneous social play behavior (Ketcheson et al., 2017). In addition, motor abilities are associated with social communication abilities (Hsu et al., 2004; MacDonald, Lord, & Ulrich, 2013b), adaptive behavior (MacDonald, Lord, & Ulrich, 2013a), language skills (Bedford, Pickles, & Lord, 2016; Kim, 2008), and autism severity (MacDonald, Lord, & Ulrich, 2014). Finally, motor and physical activity programs for children with ASD can provide benefits in many ways, including improved social skills (Pan et al., 2017a) reduced self-stimulation (Kern, Koegel, & Dunlap, 1984), increased rates of academic responses (Kern, Koegel, Dyer, Blew, & Fenton, 1982), more on-task time, (Kern et al., 1982) improved executive function (Pan et al., 2017b), and improved sleep quality (Wachob & Lorenzi, 2015).
Early FMS interventions for those with ASD show promising results, but remain implemented in research settings, under the close watch of conscientious researchers. For example, a 12-week FMS intervention implemented by adapted physical education (APE) researchers improved object manipulation and overall motor scores for nine 4-year-olds with ASD (Bremer et al., 2015). Some individual FMS improved from an intervention orchestrated by APE researchers and a special education teacher within an early intervention classroom with five children showing ASD-like characteristics ages 3-7 (Bremer & Lloyd, 2016). Finally, the locomotor and ball skills of 11 children ages 4-6 (9 controls) improved in a summer-camp motor skill intervention, again implemented by APE researchers (Ketcheson et al., 2017). In a systematic review of FMS interventions for typically developing young children, all studies took place in a school setting (Veldman, Jones, & Okely, 2016), underscoring a lack of research on translation of FMS interventions to novel environments. Although early FMS interventions yield positive results, it is unclear whether such results are attainable once interventions are transported to ecologically valid environments with implementors untrained in motor development.

To our knowledge, the early intensive behavioral intervention (EIBI) applied behavior analysis (ABA) center environment (hereafter, ABA therapy) has never been examined for early FMS intervention. Prior to school entrance, ABA may be the most widely used ASD therapy (Green et al., 2006). A rich legacy of evidence supports the efficacy and importance of ABA, and it is largely considered the standard-of-care treatment for ASD (Cooper, Heron, & Heward, 2007; Virués-Ortega, 2010). Unlike APE provided through school systems, ABA is largely offered at one-to-one adult-to-child ratio by staff trained specifically in ASD, rather than all disabilities. ABA is covered by most insurance companies in the United States subsequent to an

ASD diagnosis, and can therefore be offered before school systems offer APE services (Lloyd et al., 2013). In preschoolers, ABA primarily addresses cognitive, adaptive, language, academic, social, and language skills, with an ever-present focus on reducing problem behaviors. However, the underlying ABA literature provides myriad approaches that can be slightly modified for an effective FMS education program. Specifically, discrete trial training of motor imitation, video modeling, and graduated guidance are all easily modified to teach FMS (Cooper et al., 2007). Taken together, these characteristics make ABA centers attractive arenas to promote early FMS.

The early FMS intervention literature offers space for improvement in study design and several unanswered questions. Studies present several methodological issues, for example a lack of blinded assessment of outcomes and a lack of randomization to treatment groups. These studies all vary in total instructional time, frequency of sessions, and structure of the curriculum (station-based, free play-based, etc.). Very few published early FMS interventions in children with ASD present follow up data as a measure of maintenance of skills after cessation of instruction. In addition, many research-based studies use up to 160 hours of instructional time, which is likely not feasible for many environments. Thus, the early FMS intervention literature can be improved with studies of high scientific rigor, follow-up measures, and clinically feasible instructional time.

In order to consider any intervention meaningful, it is essential to ultimately translate to an ecologically valid environment (Dingfelder & Mandell, 2011). This process is difficult, considering differences between research and practice in priorities, quality control, staffing, and resource access. In order to facilitate a successful translational intervention, feasibility is an essential component of pilot studies (Bowen et al., 2009; Leon, Davis, & Kraemer, 2011). Many studies show lack of fidelity when implemented in the community, and the lack of fidelity can

affect participant outcomes (Dingfelder & Mandell, 2011; Stahmer, 2007). Interventionists must therefore always remain cognizant of feasibility concerns. In the current intervention study, feasibility concerns must be at the forefront, as this FMS intervention is implemented by adults with no motor or kinesthetic training. Their typical day involves running cognitive, social, and behavioral programs in a controlled environment where behaviors can be managed.

The purpose of this randomized controlled trial was to assess the efficacy and feasibility of FMS training when provided by two sites of an ABA center. Preliminary efficacy was assessed using the Test of Gross Motor Development across pre-intervention, post-intervention, and 4-week follow-up; feasibility was assessed using a quantitative and qualitative postintervention survey administered to behavior technician implementers (acceptability, perceived efficacy, practicality and integration) and comparison of intervention session videos to a fidelity checklist tool (fidelity of implementation). We hypothesized that children in the intervention group would show higher motor scores relative to the control group across time. In addition, we hypothesized that behavior technicians would implement the intervention with high fidelity and find the intervention feasible.

Methods

Participants

Children

Participants were fourteen full-time clients at one of two ABA centers serving children with ASD between ages two and five years. In order to be eligible to attend, children must have a physicians' recommendation for at least 25 hours per week of ABA, and therefore these centers provided service to children requiring substantial to very substantial support (one-to-one instruction, minimal language, management of severe interfering behaviors). Both EIBI centers followed the same classroom structure, activity schedules, and ABA curriculum; the sites also trained all behavior technicians together and their training was considered equivalent so that technicians could be shared across sites. Low-income families were prioritized for admittance, with approximately 75% of families relying on Medicaid for insurance coverage at the time of this study. Children were included in the study if they had a medical diagnosis of ASD, were between the ages of two and five, and were enrolled in one of the two sites approved for this study. Fifteen students met eligibility for this study; however only fourteen of these individuals' guardians provided informed consent to participate. Participants were on average 53.86 months old (SD = 6.80 months). Participants were 71.43% male, and 50% white (35.7% African American, and 14.3% Asian-American). Participants were excluded if their families had plans of relocating within the next 9 months.

Behavior Technicians

Each center aimed for a 1 child: 1 behavior technician ratio. Behavior technicians were entry-level staff holding a bachelor's degree. A Master's level Board-Certified Behavior Analyst (BCBA) was also present at each site to design and supervise therapy plans, organize staff and equipment, and provide training and administrative support. Behavior technicians were assigned randomly to work with specific children, rotating approximately every 4 weeks; however, changes in pairing were frequently made due to unique therapeutic considerations (for example if a behavior technician had minimal experience supporting clients with challenging behavior) and staffing needs at other sites. This process was orchestrated at the BCBA level without researcher input. All behavior technicians included in the current study (seventeen) were employed by the EIBI center, available to implement the intervention, and completed the anonymous feedback survey post-intervention (described below).

Study Design

This study was a randomized controlled trial designed to evaluate (1) the efficacy of the twenty-week intervention on total fundamental movement scores, and (2) the feasibility and fidelity of the intervention implementation in an ABA center environment. Fourteen participants from 2 centers were randomized at the child level to form a control (n = 6) and intervention group (n = 8). Each site had its own control and intervention group. The primary outcome was efficacy of the intervention; a treatment effect would be indicated by a significant interaction term (between time and group) in a repeated measures ANOVA on the Test of Gross Motor Development – III (TGMD – III) total raw scores. TGMD – III assessments were evaluated by blinded evaluators. The secondary outcomes of feasibility and fidelity were measured with a behavior technician feedback survey and an implementation fidelity tool. Feasibility and fidelity data were not used for hypothesis testing, but to frame motor outcomes.

Procedure

All procedures were approved by the human research protection program institutional review board at Michigan State University before data collection began and all participants' caregivers gave informed consent. Caregivers supplied descriptive characteristics on a questionnaire. Autism Severity (Autism Diagnostic Observation Scales – 2 calibrated severity score; ADOS-2), Cognitive abilities (Mullen Scales of Early Learning Age Equivalent scores; MSEL), language abilities (Preschool Language Scales – 5 expressive communication and auditory comprehension standardized scores; PLS-5), and adaptive skills (Vineland Adaptive

Behavior Scales – III composite standardized score; VABS-3) were measured by trained clinicians under the supervision of a licensed psychologist who were external to the ABA center and reported pre-intervention. Anthropometrics and FMS (Test of Gross Motor Development – III; TGMD – III) were assessed prior to intervention (pre-intervention), following twenty weeks of intervention (post-intervention), and at four weeks follow up in a pre-specified order of participants replicated at all time points. Anthropometrics were also measured pre-intervention and post-intervention. Children receiving the intervention were pulled out of their typical programming with their behavior technician once per school day, while the other participants continued standard-of-care ABA therapy. Participant characteristics are detailed in Table 3.1, along with evidence for no significant differences between the control and intervention groups in any of these parameters.

Intervention

To facilitate translation of the program to implementors with no formal motor background, the intervention was designed with attention to simplicity and clarity (Stahmer et al., 2015). In particular, we chose to use discrete trial training for gross motor imitation, video modeling, and graduated guidance, staples of ABA therapy with which these implementors were very familiar. The novelty arrives in intentional choices: the specific imitation models are thirteen gross motor skills that predict future engagement in physical activity (run, gallop, hop, skip, horizontal jump, slide, two-handed strike of stationary ball, one-handed forehand strike of a self-bounced ball, dribble, two-hand catch, kick, overhand throw, underhand throw) (Ulrich, 2013). ABA includes a rich legacy of imitation training through discrete trials, which can be easily modified to teach FMS (Cooper et al., 2007). Finally, ABA embraces the use of video models, a strategy effective for children with ASD (Charlop-Christy, Le, & Freeman, 2000), and

for teaching FMS (Yanardag, Akmanoglu, & Yilmaz, 2013). In a meta-analysis of evidencebased teaching strategies for children with autism, video modeling (but not social stories or peermediated interventions) met criteria for being evidence-based and highly effective across 38 studies (Wang & Spillane, 2009). The ABA concept of graduated guidance can also be adapted for early FMS training. When teaching a new behavior through manual prompting, graduated guidance asks the prompter to fade the intrusiveness of their prompt as the learner initiates the movements on their own. Such prompt fading allows the learner to develop independence quickly, because they are given the opportunity to practice activating the required muscles while still guided in the correct form (Cooper et al., 2007). Again, this prompt hierarchy is conducive to early FMS training, when relatively complex body positions must become independently initiated by the learner to produce a meaningful outcome.

Direct FMS instruction sessions lasted 15 minutes each and occurred 4 days per week for 20 weeks from January to July (school holidays and snow days were not counted toward the 20 weeks). The implementation schedule of each site was staggered, allowing research staff to conduct pre-intervention assessments at one site before moving onto the next site. Each session consisted of discrete trial training (Cooper et al., 2007) in one of the thirteen FMS for one individual child. Trials were implemented by one behavior technician already working with the child. One research staff member was present to answer questions and collect video.

Each trial consisted of viewing a tablet-displayed video of the parent skill, a picture task card, and an abbreviated verbal direction, which have both been shown useful for administering the TGMD with young children with ASD (Breslin & Rudisill, 2011). Following this stimulus, the participant completed one trial of the skill. The behavior technician implemented a most-toleast manual prompting hierarchy and provided immediate differential reinforcement. The intrusiveness of the prompt was faded over subsequent trials. Reinforcement was provided immediately after successful prompting or independent response. After an unsuccessful prompted or independent response, no reinforcement was provided, the verbal direction, "let's try again" was provided, and the discriminative stimulus was given again. Reinforcers were chosen according to each child's preferences, often adjusted in the moment if potent reinforcers emerged during the session. Reinforcement items and schedules were established daily prior to session beginning, by the behavior technician considering the child's preferences and motivations that day. All instruction took place in a gym or hallway with adequate space, depending on each site's facility availability.

The order of FMS taught was based on pre-intervention assessment scores, skills performed better were taught first and skills performed worse taught last. Once each of the 13 skills had been taught once, the order was repeated to ensure an equal representation of each of the skills. Effort was made to teach alternating locomotor and ball skills each day to maintain learner interest across a given week. Within each of the 13 parent skills on the TGMD – 3, scoring criteria were adapted to form target behaviors. For example, the TGMD – 3 specifies several criteria for successful kicking, one of which is a rapid, continuous approach to the ball. If participants did not demonstrate proficiency in this criterion during pre-intervention testing, it became their first target behavior under the parent skill of "kick". After the participant achieved 70% independent performance in this target behavior, the participant's target behavior shifted to a more advanced component of kicking (for example, placing the support leg close to the ball before kicking). Only criteria that could be manually prompted could be included as target behaviors. Only the target behavior itself was reinforced, but participants were prompted through

the entire parent skill each time (for example, participants were prompted through the final kick of the ball, regardless of their target behavior).

Before instruction could begin, several participants required instruction in prerequisite skills. For example, before dribble could be prompted, participants had to demonstrate holding the ball steady rather than throwing it at the ceiling or mouthing it. Before one-handed strike of a self-bounced ball could be taught, participants had to demonstrate holding the equipment in the correct hands for 3 seconds. These prerequisite skills were prompted and reinforced using the same procedures for other target behaviors, but participants could progress after 3-4 independent trials, rather than 70% independence, as these skills were often static rather than dynamic, and were deemed remedial in that they shared no meaningful relationship with the final target skills. For a minority of children, target behaviors were minorly adjusted to account for problem behaviors or aversion to manual prompting. For example, one child consistently ran in the predetermined direction, but would not stop at the finish point, and used the opportunity to elope from the intervention team. Thus, for this child, the target behavior became stopping at the finish line and walking back to start. Children showing independence on seven trials of a ten-trial block would be introduced to the next target behavior within the parent skill.

The prompt type was manual prompting from most to least intrusive, using graduated guidance to fade prompts as independence emerged. Some children were known by the therapy team to respond poorly to manual prompting, and in these cases, verbal prompts or concurrent modeling were intermittently included and noted. The prompt level and type were noted at start and finish of each 10-trial block. Instruction was adjusted according to individual learner style and success, a hallmark of successful ABA therapy. BCBAs and behavior technicians were

instrumental in optimizing successful instruction strategies. Specific prompt level descriptions are available upon request.

An additional 5 minutes per day, 4 days per week, the entire intervention group at each campus played rotating active social games to focus on social integration of FMS. These games consisted of the group watching a television-displayed video model of young children playing active social recess games (tag, duck-duck-goose, race, T-ball, etc.). Behavior technicians physically prompted and reinforced following directions and positive imitation of the videos when the group was instructed to do so.

Measures

Descriptive data

Demographics

Demographic surveys addressing family income, race, ethnicity, and education were completed by parents at the pre-intervention time point. These data are presented to describe the sample and considered as potential covariates. Anthropometrics

Height without shoes was measured once to the nearest 0.2 cm (Seca Stadiometer). Weight in light clothing was measured once to the nearest 0.1 kg (standing scale). Height and weight were used to compute body mass index (BMI), BMI Z-scores, and BMI percentiles according to Centers for Disease Control growth curves for U.S. Children (Centers for Disease Control and Prevention, National Center for Health Statistics, 2000). These data were included for descriptive purposes.

Autism calibrated severity

The Autism Diagnostic Observational Scales – 2 (ADOS – 2) provided a measure of Autism severity (Lord, Rutter, DiLavore, & Risi, 2001). The ADOS – 2 is a "gold standard" diagnostic semi-structured behavioral observation for ASD in individuals aged 12 months to adulthood, with excellent diagnostic discriminative ability, interrater reliability, and test-retest reliability (Gotham, Risi, Pickles, & Lord, 2007; Lord et al., 1989). ADOS – 2 calibrated severity score (CSS) was used to describe the sample, as this metric allows comparison of individuals across differing modules, remains stable over 12 - 24 months (Shumway et al., 2012), and may show less susceptibility to language abilities than the total raw score (Gotham, Pickles, & Lord, 2009; Shumway et al., 2012). CSS is used to qualify participants undergoing ADOS – 2 assessment as (a) non-spectrum (CSS rating 1 - 3), (b) non-Autism ASD (CSS rating 4 - 5), or (c) autism (CSS rating 6 - 10) (Gotham et al., 2007).

Developmental level

Developmental level was measured using the Mullen Scales of Early Learning (MSEL; Mullen, 1995), a standardized assessment of cognitive ability for use in children from birth to 68 months, widely used in young children with ASD (Akshoomoff, 2006). The MSEL shows good convergent validity for assessment of nonverbal and verbal intelligence quotient in children with ASD (Bishop, Guthrie, Coffing, & Lord, 2011). The four cognitive scales of the MSEL (fine motor, visual reception, receptive language, expressive language) were administered upon entry to the ABA therapy institute. We calculated the developmental quotient for all four scales combined (DQ = Age Equivalent score / chronological age x 100); this avoids potential floor or ceiling effects (Rogers et al., 2012; Vivanti et al., 2014) and provides an estimate of intelligence quotient familiar in the literature regarding young children with ASD (Lord et al., 2006; Munson et al., 2008).

Adaptive function

The Vineland Adaptive Behavior Scales – III (VABS – 3) provided a measure of adaptive function. It often provides additional information to aid in the diagnosis of intellectual and developmental disabilities (Sparrow, Cicchetti, & Saulnier, 2016). It reports scores in the domains of communication, daily living skills, socialization, and motor skills. Scores from these domains are combined and transformed to provide an adaptive behavior composite standardized score, which is used here to describe the sample.

Language ability

The Preschool Language Scales – 5 (PLS - 5) provided a measure of early language abilities (expressive communication and auditory comprehension) from birth to age 7 years, 11 months (Zimmerman, Steiner, & Pond, 2011). In newly diagnosed preschoolers with ASD, internal consistency reliability coefficients ranged from 0.66 to 0.96, and for most ages they are above 0.81 and agreement with subtests of previous versions of PLS range from 0.65 – 0.79 (Volden et al., 2011). The PLS – 5 was administered during entrance to the ABA center and reported to the study team. Raw scores are included for descriptive purposes.

Outcome Measures

Efficacy: TGMD-III

The present study used picture task cards (Breslin & Rudisill, 2011), short instructions (Breslin & Rudisill, 2011), and administration provided by a single live model (Allen, Bredero, Van Damme, Ulrich, & Simons, 2017). Participants were asked to perform one practice trial, followed by two scored trials for each of the thirteen skills on the TGMD – III. No manual prompting or physical assistance was provided during trials. Video-recorded assessments were scored by individuals blinded to group (control or intervention) and time (baseline or mid-intervention), and whom had achieved 90% reliability (Rintala, Sääkslahti, & Iivonen, 2017) using videos and scores disseminated by the assessment authors.

Feasibility: Behavior technician feedback survey

Feasibility (acceptability, perceived efficacy, practicality, integration) within the ABA center was assessed through a survey administered to the behavior technicians who implemented

the intervention. All behavior technicians working at either site for any amount of time during the intervention period were asked to complete surveys. Surveys were anonymous and completed on paper during break times on days when research staff were not present. Eleven substantive items were presented on the survey, and respondents were asked to rate on a 6 – point Likert scale to what extent they agreed with the statement (1 = Strongly disagree, 6 = Strongly agree). Items were (1) The skills targeted for teaching in this study are valuable and should be taught. (2) The intended methods (discrete trial training with graduated guidance, group video modeling with graduated guidance) are appropriate for teaching these skills. (3) The program did improve the skills it targeted. (4) The program helped my students. (5) The way that the program ended up being delivered was effective. (6) The training I was given by the research staff prepared me to do the study. (7) If I were provided training ahead of time, and feedback on my technique, I think that I and other BTs could run the sessions without research staff present. If no, what are the things that would make this difficult? (8) I would participate in a program like this again. (9) I would actively start a program like this in my own ABA center. (10) It was easy to do these sessions on a day to day basis. If not, what were the hardest parts? (11) The program fit into the early intensive ABA environment well. If not, what made it difficult from a program standpoint?

In addition to the above items, the survey asked two questions to quantify involvement, experience level, and potential of the technician to advance to the BCBA level. Questions were (12) How many months have you been a BT? (13) If you don't mind sharing, what is your next position after this job?

Finally, the survey sought qualitative feedback. For items (7), (10), and (11), the last part of each item was a question (see above), followed by open space where participants could write qualitative responses. Qualitative responses were transcribed from handwritten surveys.

Responses were analyzed with thematic analysis. The first author immersed herself in the data by reading through each transcript twice before beginning analysis. She next defined and labeled meaning units within each transcript. She then sorted these units roughly into emergent categories. The second author and the first author then met to discuss and change the placement of these units. No coding software was used to manage the data, and participants did not provide feedback on the findings. Representative participant quotations are presented to illustrate findings.

Implementation fidelity

The extent to which the intervention was administered in the way it was intended across sites was measured using an implementation fidelity tool. Fidelity assessments were completed on random selection of 10% of the videos of intervention sessions over the intervention, based on behavior coding of video recorded intervention sessions. Videos were coded by two reliable and independent evaluators (research staff) uninvolved in the instructional components of the intervention, who were blinded to the study purpose and groupings (alpha = 0.83). An 85% fidelity criterion was specified before any intervention occurred. Research staff reviewed videos each day. If the percent fidelity achieved was below 85%, a brief informal meeting was conducted during which expected criteria were demonstrated to involved behavior technicians. Items included: video model presented, activity card presented, behavior technicians provide prompting, reinforcement provided for target behaviors. Items were rated on a 3 – point Likert rating system (1 – not present; 2 = sometimes present; 3 = clearly present).

Statistical Analysis

To assess the success of randomization, t-tests were used to compare pre-intervention TGMD – III, ADOS-2, MSEL, PLS-5, and VABS-3 scores, as well as anthropometrics between groups. Intervention efficacy was assessed with a repeated measures ANOVA. We tested the significance of a time * group interaction as a predictor of total TGMD – III raw scores across 3 time points (pre-intervention, post-intervention, follow-up) between the two groups. A significant interaction term would indicate that intervention group TGMD – III scores had improved compared to the control. Partial η^2 were calculated to represent effect sizes. Box-and-whisker plots visually represented these findings. An a priori power analysis for repeated measures ANOVA was conducted with power of 0.80, a priori α of 0.05, and using an effect size (f² of 0.325) from a previously published intervention with a similar sample and design (Bremer, 2015). This analysis yielded a required sample size of 18. Survey responses to items were averaged and presented descriptively. All statistical procedures were carried out in SPSS version 25 (IBM Corp., 2017) with a pre-determined alpha of 0.05.

Results

At study entry, there were no differences between control and intervention groups in TGMD – III, ADOS-2, MSEL, PLS-5, and VABS-3 scores, or anthropometrics (Table 3.1). All participants were substantially delayed in their FMS but showed similar proficiency in locomotor (mean raw score = 4.86, SD = 4.31) and ball skills (mean raw score = 5.43, SD = 4.34) at study entry. Participants showed higher than average BMI percentiles, with a wide variability in this metric (mean BMI percentile = 69.31, SD = 32.28). In terms of autism calibrated severity, all participants fell into the highest severity "Autism" category, except for one participant with a

CSS of 5, characterized as non-Autism ASD. Five of the fourteen participants scored the highest possible CSS of 10, underscoring the relatively severe range of ASD present in this sample.

Participants were mostly male. Although the intervention group (3 females) contained more girls than the control group (1 female), there was no significant difference between groups in gender balance. Participants showed a diverse distribution of family annual income level, as well as race (50.0% White; 35.7% African American; 14.3% Asian American).

Efficacy

The repeated measures ANOVA showed a significant interaction between time (preintervention, post-intervention, and follow-up) and group for total TGMD – III raw score, with a large effect size (F (1, 12) = 4.983; p = 0.016; ES = 0.312) (Cohen, 1988). In the intervention group, locomotor skills increased by an average of about 8 points, and ball skills increased by an average of about 10 points. Table 3.2 and Figure 3.1 detail these findings.

Feasibility

Thirteen behavior technicians completed the feasibility survey. Behavior technicians were all female, and on average had worked as behavior technicians for one of the EIBI center sites for 13.58 ± 11.62 months. Scores on the feasibility survey were spread over the full range from 1 (strongly disagree) – 6 (strongly agree) within responses for every item, indicating potentially polarized opinions. Responses on average indicated that behavior technicians found the program less feasible than neutral: the average of all responses on all items was 3.11 ± 1.53 (an impartial response would have been indicated by 3.5, the center of the scale).

Item responses are presented here as mean \pm standard deviation as well as frequencies (Selected response number: number of participants selecting this response). The item receiving the lowest average score (indicating low agreement with the statement) was (10) It was easy to do these sessions on a day to day basis (2.79 \pm 2.65; 1: 7, 2: 3, 4: 1, 6: 2). This item also showed high variability; its standard deviation larger than all others except for (11) The program fit into the early intensive ABA environment well (3.57 \pm 2.67; 1: 3, 2: 4, 3: 2, 4: 2, 5: 1, 6: 2). The items with the highest average scores were (1) The skills targeted for teaching in this study are valuable and should be taught. (3.86 \pm 1.41; 1: 1, 3: 2, 4: 5, 5: 4, 6: 1) and (2) The intended methods (discrete trial training with graduated guidance, group video modeling with graduated guidance) are appropriate for teaching these skills (3.85 \pm 1.70; 1: 1, 2: 2, 3: 2, 4: 1, 5: 3, 6: 3). The remainder of the items are presented in Figure 3.2 as histograms showing the response of each behavior technician.

Results of the behavior technician feedback survey indicate that most behavior technicians were excited about the importance of FMS development for their clients but found the program difficult to implement in the EIBI environment. When qualitative responses were grouped into meaning units and categorized, the specific issues identified to limit feasibility in this environment were: (1) inadequate staffing ratios and scheduling, (2) difficulty understanding the physicality of the tasks and lack of guidance on prompt fading, (3) increased problem behaviors in the less restrictive environment, and (4) uncertainty regarding the importance of these skills for a sample with severe ASD.

First, staffing and scheduling made implementation difficult for most behavior technicians. One technician noted that the hardest parts of the study were "being off ratio and having a therapist out of the room for extended times". Another noted that the hardest parts were

"scheduling". This references the fact that the time given to researchers to implement the study was at first during behavior technicians' breaks, when only half were active, with the same number of clients in the therapy room; pulling out one technician and one child to run the study therefore reduced their ratio in the therapy room even further. This comment was repeated by several behavior technicians. One noted, "the time during which it was implemented when the staff were off-ratio". When asked for suggestions for improvement, many behavior technicians mentioned scheduling and staffing. One suggested, "coordinate with BTs better".

Second, the physical nature of the program was challenging for most behavior technicians. Behavior technicians wrote that the hardest parts of the program were "completing the physical task", "not knowing what prompt level", "lack of information from supporting staff", "not our area of expertise, using multiple prompt and don't know what to fade or when to fade", "the prompting made it difficult", "Not understanding prompt fading" "manual prompts can be aversive we are providing them at times too much because we didn't know which to prompt". These comments summarize a strong sentiment among most behavior technicians: difficulty implementing a motor-based program with only the in-the-moment training that was provided. Technicians noted that they would have liked more clear instructions regarding physical prompts, or "set prompting levels and types, set directions from implementors".

Third, most behavior technicians felt that problem behaviors were more common during intervention sessions than in the therapy room, and that this made sessions very difficult to manage. Many technicians noted that it was very difficult "handling problem behaviors" "blocking problem behavior and eloping". One said that she, "can't manage materials and block problem behaviors". Also, these problem behaviors appeared worse when the child was having a hard day, and that this session tended to exacerbate an already bad day. One behavior technician

noticed that "remaining focused" and "when the child was having a rough day" were difficult to deal with.

Finally, the severity of the participants' ASD was a common theme in the behavior technicians' responses. Many technicians believed the hardest thing about the study was "the learning level of the students" or, "the skill level of our learners". Several indicated that 15minute sessions were quite demanding, given the severity of their clients. One noted, "15 minutes is a long time", another that "too long of sessions (15 min)". Others noted that the skills being taught were perhaps too advanced for some of the children in this group, noticing that it was difficult "not having pre req skills". All behavior technicians recommended termination criteria for a given session as well as exclusion criteria for participation in the program. Others recommended following a multiple-baseline type study design instead of the randomized controlled trial design, i.e. changing the order of skills taught, "teaching one skill to mastery prior than introducing a new skill. Would allow students to know what to expect"

Implementation Fidelity

A total of 16 sessions were rated for fidelity. Fidelity assessments yielded 92.0% fidelity across the two sites. Therefore, no follow up brief meetings were required between research staff and behavior technicians. Average score was 2.95 ± 0.21 on a Likert scale from 1 - 3 on all 4 items. Item one obtained a mean score of 3.00 ± 0.00 , item two obtained 3.00 ± 0.00 , item three obtained 2.93 ± 0.25 , and item four obtained 2.93 ± 0.26 .

Discussion

Results from this pilot study support the efficacy and fidelity of a 20 – week daily FMS discrete trial training intervention and offer guidelines to enhance the feasibility of implementation in the early intensive ABA environment. Findings suggest that behavior technicians had average – to – low opinions of acceptability, feasibility, perceived efficacy, practicality, and integration, with wide variability in each. Despite these concerns, behavior technicians were able to deliver the intervention to a high level of fidelity and efficacy. Given that previous interventions in FMS have relied on expert implementors, the present study may represent an important step toward delivering physical skill instruction on a larger scale. However, several concrete steps must be taken before this is possible.

The program does improve the TGMD skills, but the improvement in skills by scale indicates a differential influence of the intervention on ball skills compared to locomotor skills. Ball skills are very different from locomotor skills in that they use equipment that offers an additional piece of instruction that converges with the verbal instruction. This may help establish stronger stimulus control over responding. This may be an advantage for young learners with ASD, and particularly for young learners with high severity ASD, of which this sample is mostly comprised. Interestingly, this study replicates the differential effectiveness of motor intervention for ball skills vs. locomotor skills found in another similar study (Bremer et al., 2015).

There are few FMS early interventions that include a follow-up measure. The corresponding results in this study indicate a dramatic drop-off in the skill level of the intervention group after only 4 weeks without training. This staggering drop-off indicates the importance of consistent and longitudinal practice and maintenance, even after participants have

mastered the skills. These results contrast those of Ketcheson et al, who found no difference between post-intervention and follow-up scores of the intervention group following a 160-hour intervention (Ketcheson et al., 2017). The dosage represented in the current study was 1/8th that of the Ketcheson et al. study, and it is not surprising that differential drop-offs of intervention effect should arise. This finding indicates the importance of continued regular instruction for maintenance of FMS proficiency in this group, particularly when intervention dose is limited, as it practically will be in any translatable setting.

There are several important action points to integrate in any future FMS intervention with a similar population. We would recommend thorough preparation and concrete practice with implementors before starting physical skill-based interventions. The current study offered no training without clients, as administrative staff indicated that on-the-job training was usually most effective with this group of behavior technicians. However, in future, an in-service of even one day could provide benefits in reducing the motor education deficits present in behavior technicians. A training manual with pictures and abbreviated directions might also help, again, introduced before implementation. To promote buy-in of behavior technicians, enhanced education concerning the cognitive and social benefits of FMS development may help to facilitate buy in from implementors. Physical activity interventions have increased the academic responding, on-task time, and sleep, and reduced the motor stereotypy of children with ASD (Neely, Rispoli, Gerow, & Ninci, 2015; Wachob & Lorenzi, 2015).

It seems likely that any behavior technicians asked to implement an FMS intervention will be out of their comfort zone; however, the results of this study offer some guidance in how to help them. Some concrete action points include straightforward manuals, more quiet and controlled environments to limit eloping and problem behaviors, shorter sessions, constant

prompting definitions, clear explanations of potential benefits of the program, and earlier and more controlled training. Similar programs might be implemented on a wide scale, after some of the major feasibility concerns are resolved. Alternatively, the results of this study could be used to advocate for wider and earlier availability of adapted physical education services and physical therapy to promote early FMS development in children with ASD, as an alternative or adjunct to physical skills being taught in ABA. Regardless, this study identifies a clinical need for direct collaboration between service providers in ABA and physical development (APE or PT, for example).

This study has several limitations. It is underpowered by 3 participants for efficacy investigations. In addition, only a subsample of the intervention sessions was reviewed for fidelity; however, the selection was random. The administrator of the main outcome measure, the TGMD – III was not blinded due to a lack of research personnel. The results of this study are not necessarily generalizable to every child, or even every preschooler with ASD, as the sample appears to be rather racially homogenous. It should be noted that demographic surveys were not returned from any of the parents at only one of the campuses, which also happened to lie in a low-income area; thus the sample's descriptive information may not be an accurate description of the true sample.

This is the first early FMS intervention to our knowledge that is designed and implemented in the ABA setting; this is the study's biggest strength. In addition, the strong randomized controlled trial design underscores the internal validity of this study. This study shows that 20 hours of reluctant and difficult implementation of FMS instruction can have a large effect on the FMS of children with severe ASD. This suggests the importance of future

investigations in this environment. In all, this study adds to a growing body of literature examining methods for impacting the motor development of children with ASD.

APPENDIX

TABLES

Table 3.1. Baseline descriptive	e informatio	n for th	e control a	nd inter	vention gro	uns		
Measure	Control		Intervention		Total		t-test	
	(n = 6)	(n = 6)		(n = 8)				
	Mean	SD	Mean	SD	Mean	SD	р	D
TGMD - III								
Pre-intervention	5.00	.20	4.75	.68	4.857	.31	.92	0.06
Locomotor								
Pre-intervention	5.17	76	5.63	.98	5.429	.35	.85	0.11
Ball skills								
Pre-intervention	10.17	7.57	10.38	.70	10.290	.93	.96	0.03
Total								
Descriptive								
information								
Height (cm)	104.70	.56	105.26	.54	105.050	.39	.10	< 0.01
Weight (kg)	17.17	.57	18.40	.66	17.94	.23	.86	0.15
BMI percentile	70.00	9.51	68.88	29.9	69.31	2.28	.96	0.04
-				0				
ADOS-2 CSS	7.50	.98	8.29	.06	7.92	.98	.50	-0.43
Gender								
Female	3		1		4			
Male	3		7		10		.15	0.90
Age (years)	53.83	7.17	53.88	.02	53.857	.80	.99	0.01
Annual								
Household								
Income								
<\$24,000	1		1		2	2		
\$25,000-	0		1		1	1		
\$49,999								
\$50,000-	2		1		3	3		
\$75,000								
>\$75,000	0		1		1	1		
Missing	3		4		7	7	.87	0.17
Race								
White	2		5		7 (50%)			
	(33.3%)		(62.5%)					
AA	3		2		5			
	(50.0%)		(25.0%)		(35.7%)			

Table 3.1 (cont'd)

Asian	1 (16.7%)		1 (12.5%)		2 (14.3%)		.43	0.50
PLS-5								
Expressive Communicat	64.75	3.15	59.00	2.44	61.30	2.35	.51	0.55
Auditory Comprehens ion	23.750	.54	18.00	.01	20.30	.54	.31	0.80
Total	125.25	3.96	117.50	6.77	120.60	8.26	.72	0.33
MSEL								
Verbal Reasoning	33.75	1.62	35.33	3.17	34.70	1.92	.83	0.16
Fine Motor	31.75	.50	27.50	.13	29.20	.12	.11	1.27
Receptive Language	28.75	7.86	25.17	2.80	26.60	4.17	.74	0.30
Expressive Language	25.50	3.77	21.80	4.76	23.44	13.56	.67	0.34
VABS Adaptive Behavior	65.67	.52	60.67	.22	62.33	.81	.40	0.68

Note: SD = Standard Deviation; p = significance; D = Cohen's D; TGMD – III = Test of Gross Motor Development – III, scores reported as raw scores; BMI = Body Mass Index; ADOS = Autism Diagnostic Observational Scales-2; CSS = Calibrated Severity Score; F = Female; M = Male; AA = African American; PLS-5 = Preschool Language Scales-5, Standard scores reported; MSEL = Mullen Scales of Early Learning, scores reported as Developmental Quotients unless otherwise noted; VABS = Vineland Adaptive Behavior Scales, Adaptive Behavior Composite-Standard score reported.

	Control		Interventio	n
	$(\mathbf{n} = 0)$		$(\mathbf{n} = \mathbf{\delta})$	
Measure	Mean	SD	Mean	SD
TGMD - III				
Pre-intervention				
Locomotor	5.000	4.195	4.750	4.683
Ball skills	5.167	3.764	5.625	4.984
Total	10.170	7.574	10.380	8.700
Post-intervention				
Locomotor	7.500	4.231	14.143	9.771
Ball skills	7.500	3.937	17.714	10.029
Total	15.000	6.870	33.286	19.094
Follow-up				
Locomotor	7.000	7.537	10.714	10.688
Ball skills	3.000	2.530	12.714	11.161
Total	10.000	8.367	23.429	20.647

Table 3.2. TGMD - 3 scores of both groups at all time points.

Note: SD = standard deviation; TGMD = Test of Gross Motor Development.



FIGURES

Figure 3.1 Box - and - whisker plots of total, locomotor, and ball skill raw scores by interventional group at baseline and mid-intervention.

The program	fit into the early in	tensive ABA enviro	onment well.		
H					
It was easy to	o do these sessions	on a day to day basi	s.		
20 1			0		*
I would activ	ely start a program	like this in my own	ABA center.		
·[
I would partic	cipate in a program	like this again.			
could run the	sessions without re	esearch staff presen	t.	ue, i think that i an	
The training I	I was given by the	research staff prepa	red me to do the st	tudy.	
The way the j	program ended up	being delivered was	effective.		٥
The program	helped my student	s.			
H					
The program	did improve the sk	ills it targeted.			
H					
The intended appropriate for	methods (discrete or teaching these sk	trial training with g cills.	raduated guidance	, video modeling) a	ure
The skills tar	geted for teaching	in this study are val	able and should b	be taught.	
0				1	
Strongly disagree		554		_	Strongly agree
1	2	3	4	5	6

Figure 3.2. Box- and – whisker plots of behavior technician responses to feasibility survey items.

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CHAPTER 4

Social Play Outcomes of an Applied Behavior Analytic Early Intervention Program Targeting Both Social and Motor Domains

Abstract

There is increasing evidence of the interrelatedness of motor skills with social skills, especially in children with Autism Spectrum Disorder (ASD). This study presents the secondary outcome results from an Applied Behavior Analysis fundamental movement skills intervention for preschoolers with ASD. The intervention is the first of its kind implemented in early intensive behavioral intervention centers, an ecologically valid environment with wide reach. Despite inclusion of a social play component in the intervention, there were no benefits observed in their existing behavioral intervention therapy progress (F (1,12) = 0.06, p = 0.81). This result suggests that the inclusion of fundamental movement skill training for 15 minutes per day over 20 weeks did not alter progress on Applied Behavior Analysis therapy.

Introduction

Autism Spectrum Disorder (ASD) is defined as a persistent impairment in social communication or interaction, often accompanied by restricted and repetitive patterns of behavior, interests, or activities (American Psychological Association, 2013). According to estimates in 2012, 1 in 68 children in the United States has a diagnosis of ASD (Christensen, 2016), and this number has increased dramatically since 2002 (1 in 151.5) (CDC, 2007). This increase has prompted research attention on this population. Early skill development programs for those with ASD primarily focus on the core deficits of ASD: social and communication skills. Effective approaches for teaching such skills are varied and can include traditional

behavioral therapy (Reichow et al., 2012), parent-training curricula (Okuno et al., 2016; Oono et al., 2013), video modeling (Charlop-Christy et al., 2000; Nikopoulos & Keenan, 2004), and peermediated interventions (Brock et al., 2017; Mason et al., 2014).

Activity-based programs may also provide benefits in social skills, particularly if social development is emphasized and instructional focus is individualized (Sowa & Meulenbroek, 2012). For example, a 12-week therapeutic horseback riding program provided benefits to 5-10 year-olds with ASD in social responsiveness and social motivation (Bass et al., 2009). Children in this program also showed improvements in sensory-seeking behavior (Bass et al., 2009). Social skill scores appeared to improve after a 12 week in-school fundamental movement skills (FMS – basic motor abilities that provide the foundation for more specific athletic abilities) intervention for children with ASD-like characteristics aged 3-7 years (Bremer & Lloyd, 2016). Also, after an intensive FMS intervention for children aged 3-6 years with ASD, children spent less free play time solitary than at baseline (Ketcheson et al., 2017). In other cases, no changes in social skills arose after physical activity interventions, often explained by low statistical power or a lack of concrete emphasis on social skills (Bremer et al., 2015; Pan, 2010). Vigorous activity also may provide cross-domain benefits in stereotypic behaviors (Kern et al., 1984; Neely et al., 2015), academic engagement (Neely et al., 2015), on-task behavior (Kern et al., 1982), and executive function (Pan et al., 2017). Thus, it appears that programs targeting both social and motor development could synergistically improve both outcomes.

Literature reinforces this assertion, as it suggests a relationship may exist between the social and motor domains (Craig et al., 2018; Garrido et al., 2017; MacDonald et al., 2013b), despite the uncommonness with which these two domains are explicitly paired in ASD early intervention. Several early social skills are in fact motor-based, including eye contact and gross

motor imitation. In addition, many commonly taught early language and social targets include a motor component, such as learner response to picture task cards or picture exchange systems, tapping individuals to request, imitating peers, and engaging in movement-based play with others. However, there are very few published interventions equally and explicitly targeting both social and motor domains.

Effective programming targeting both social and motor development could take many forms. During the early intervention age range, a natural blend of these domains is present in typical play behavior (Jahr et al., 2000; Jung & Sainato, 2013; Stahmer et al., 2003). Age appropriate play often consists of gross motor games involving other children. Such behaviors arise in typically developing children without instruction. However, children with ASD show deficits in appropriate social play (Libby et al., 1998). Applied Behavior Analysis (ABA) is widely accepted as a component of evidence-based care for individuals with ASD. Although ABA early intervention typically includes instruction in social and motor development separately, these two domains are rarely targeted together.

The purpose of this study was to measure the effect of an ABA motor and social early intervention program on ABA therapeutic progress.

Methods

Experimental Design and Setting

The current study was a randomized controlled trial conducted at two sites of an ABA early intervention institute. Children enrolled at this institute attended 30 hours per week of ABA therapy at a 1:1 ratio with behavioral therapy technicians.

Participants

Participants were included if they had a medical diagnosis of ASD and their parents provided informed consent to participate. Participants were randomized prior to baseline testing to form a treatment and a control group (sealedenvelope.com) at each site. Randomization was broken in one case because initial randomization placed a pair of twins in the treatment group. To minimize loss of instructional time to family absences, one of these twins was allocated to the control group and replaced by a participant with a comparable baseline FMS score. Participants were recruited by research staff and ABA center leadership at child pickup times. Participant characteristics are presented in Table 4.1. Characteristics include age, gender, maternal education, Autism calibrated severity score (CSS) on the Autism Diagnostic Observation Schedule - 2 (ADOS - 2), cognitive t-scores on the Mullen Scales of Early Learning (MSEL), expressive and receptive language scores on the Preschool Language Scales (PSL - 5), and adaptive behavior composite on the Vineland Adaptive Behavior Scales (VABS). Procedures

This project was approved by the Human Research Protection Program at Michigan State University and approved by the research board of the ABA institute where it was conducted. Informed consent was obtained from all participants' primary caregivers before they were enrolled. Participants ages 5 and older provided informed assent. Participants underwent baseline assessments over one week, then the treatment group participated in a twenty-week intervention integrated into their therapy plan, while the control group received treatment as usual. Both groups then participated in post-treatment assessments over one week.

Measurement

Baseline

Demographics

At baseline, primary caregivers were asked to complete a questionnaire that included items such as their child's race, ethnicity, their annual family income, and education level. Autism CSS

Autism CSS was reported to research staff by the ABA institute at baseline, from the Autism Diagnostic Observational Scales – 2 (ADOS – 2) scores administered by trained clinicians during the diagnostic process. The ADOS – 2 is a semi-structured interaction-based assessment and the gold standard diagnostic assessment for ASD. It shows high interrater reliability, score stability over time, and ability to discriminate between children with and without ASD (Lord et al., 1989, 2001; Shumway et al., 2012). The ASD CSS is a score from 0-10, where higher score indicates more severe ASD symptoms (Gotham et al., 2009). Cognitive ability

Cognitive ability was assessed with the Mullen Scales of Early Learning (MSEL), a standardized developmental assessment for children birth to 68 months (Mullen, 1995). The fine motor, visual reception, expressive communication, and receptive communication subtests were administered by ABA institute staff at entrance to therapy and reported to the research team at baseline. These subtests each produce a t-score (ranging from 20-80). This assessment shows good internal, test-retest and interrater reliability, and moderate correlations with other early childhood cognitive assessments (Bishop et al., 2011; Mullen, 1995). We calculated the developmental quotient for all four scales combined (DQ = Age Equivalent score / chronological age x 100); this avoids potential floor or ceiling effects (Rogers et al., 2012; Vivanti et al., 2014)

and provides an estimate of intelligence quotient familiar in the literature regarding young children with ASD (Lord et al., 2006; Munson et al., 2008).

Language ability

Expressive and receptive language ability were assessed using the Preschool Language Scales (PLS – 5) (Zimmerman et al., 2011). The PLS – 5 shows excellent internal consistency in newly diagnosed preschoolers with ASD (Volden et al., 2011). Agreement with subtests of previous versions of PLS range from 0.65 - 0.79 (Volden et al., 2011).

Adaptive behavior

Adaptive behavior was assessed using the Vineland Adaptive Behavior Scale (VABS), a semi-structured interview conducted with parents that provides a measure of adaptive skills in children from preschool to 18 years of age (Sparrow et al., 2016). The VABS has been used with young children with ASD (Balboni et al., 2016; Perry et al., 2009). The communication, daily living, social skills, and motor skills subtests were administered by ABA institute staff at entry, and these scores reported to research staff at baseline. These scores were combined to provide an adaptive behavior composite score.

Outcome variable

Targets mastered

The primary dependent measure was the number of therapeutic targets that participants mastered over the course of the intervention. These targets were not explicitly taught during motor and social intervention described here, but were instead the specific goals of the long-term ABA therapy plan. They were unique for each participant and selected by the ABA institute clinicians based on individual needs on a standardized placement test: the Verbal Behavior Milestones and Placement Program. This variable was calculated for all participants through retrospective client file review between the baseline and post-treatment assessment dates. Thus, this variable is a change score (number of targets mastered at post-intervention minus number of targets mastered at preintervention).

Treatment

All instruction occurred in a gym, a small treatment room, or an empty hallway. Treatment occurred for twenty weeks of instruction (4 days each), with treatment stopping for scheduled ABA institute breaks and weather-related cancellations. Daily instruction consisted of two activities: individual instruction in one of thirteen FMS through discrete trial training (DTT), and a social play group for all participants in the treatment group.

FMS DTT

Daily FMS DTT sessions lasted fifteen minutes each with one participant, one behavior therapy technician, and one research staff available for consultation. The behavior therapy technician present was assigned daily depending on clinical availability and was frequently different for each participant. The discriminative stimulus (S^D) consisted of a video model of the mature FMS displayed on a tablet, a picture task card for the FMS, a brief verbal direction (Breslin & Rudisill, 2011), and the presentation of any sports equipment used for these skills (determined from TGMD – 3 administration guidelines) (Ulrich, 2013). Manual most-to-least prompting with graduated guidance was implemented to teach the correct form of each FMS while also encouraging independence. Reinforcement was provided upon successful performance of the skill level specified, regardless of whether prompted or independent. In keeping with the procedures of the ABA institute, behavior therapist technicians used a wide variety of reinforcers and reinforcement schedules, adjusted in real-time depending on the preferences of each participant, including breaks.

Baseline performances on the TGMD - 3 informed the order of skill training, the skills with the highest baseline scores taught first. One FMS was taught each day, with an effort to alternate locomotor and ball skills. Skill order was preserved despite absences, with the next skill in order taught at the next available opportunity after an absence. Once all thirteen skills had been taught, the order of skill training was repeated. Each of the thirteen FMS skills was broken into small components that could be manually prompted to comprise target behaviors. For certain pre-requisite skills (moving along the area used for locomotor skills without eloping, holding equipment in the correct hands, not inappropriately using sport equipment), participants were advanced to the next challenge level once the skill was demonstrated. For more integral components of skills (scoring criteria on the TGMD - 3), participants had to demonstrate 70% independence on a 10-trial block of one level (approaching the soccer ball rapidly and continuously) before being prompted to perform the next challenge level (placing a preparatory foot close to the soccer ball before kicking it). Trial-level data were collected by the behavior therapy technician, with help from research staff when physically necessary, to record the success of the participant's approximation of the challenge level target behavior and whether it was prompted, any errors, and the intrusiveness and form of prompt used. Although the major focus for each trial remained the challenge level target behavior, the product of each FMS was also manually prompted when possible (participants were prompted to complete the kick of the soccer ball).

The research staff present rotated between the first author and undergraduate research assistants. Research staff daily informed the behavior therapist technicians what the challenge

level target behavior was, and ways to manually prompt it. They were reminded to use graduated guidance to fade manual prompts as soon as possible, and to prompt through the product of each FMS. They were told the S^{D} (tablet video, picture task card, verbal direction), and provided with the appropriate sport equipment. They were told to reinforce successfully prompted or independent target behaviors using the reinforcement most effective for that child.

Social Play Group

Daily social play groups occurred in six to fifteen-minute sessions with all participants in the treatment group of a given site present, an equal ratio of behavior therapy technicians, and one research staff facilitator. Participants performed ten games or activities for two weeks each. During weeks of excessive absences, the same game or activity was extended for another week. The procedures of the group session were modeled after existing imitation groups within the ABA institute, to which the participants were accustomed. This first involved watching a video model of the activity from a 40-inch television mounted on a rolling stand. During the video, a facilitator provided linguistic mapping. After this, the group was invited to try the activity, and similar linguistic mapping was provided for their actions. Behavior therapist technicians used child – level S^Ds so that individual expectations were clear. They manually prompted the corresponding target behaviors, using graduated guidance to fade when available. Behavior therapist technicians used a wide variety of contingent reinforcers and reinforcement schedules, depending on the daily preferences of each participant.

The control group did not engage in either FMS DTT or the active social play group but underwent therapy as usual in a separate room of the ABA institute.

Therapist Training

No extra training was required of behavior therapist technicians, but limited real-time formative feedback was provided by research staff. Undergraduate research staff underwent training in the FMS target behavior shapes and the protocols for DTT and social play group. Their training consisted of two training sessions with the first author, one observation hour of traditional therapy at the ABA institute, two observation days of DTT and active social play group, and one day of supervised DTT and active social play group. Feedback was provided consistently in real-time.

Statistical analysis

First, to ensure the effect of randomization, we conducted (multiple independent sample t-tests of age, sex, race, family income, maternal education, FMS, ASD CSS, average baseline mastery rate, MSEL t-scores, adaptive behavior composite, receptive language score, and expressive language score between the intervention and control groups. To adjust for multiple testing, a Bonferroni correction was used. To test the effect of the intervention on targets mastered, we used a univariate analysis of variance (ANOVA) between intervention and control groups. The dependent variable was targets mastered over the course of the intervention (the number of therapy targets mastered at post-intervention minus those mastered at pre-intervention).

Results

No differences arose between the control and treatment groups in any of the baseline characteristics tested, therefore these variables were not included as covariates in the ANOVA. Descriptive characteristics of the groups are included in Table 4.1. In the ANOVA, there was no significant difference between the targets mastered of the intervention group, compared to the

control group (F (1,12) = 0.06, p = 0.81). The R^2 value was < 0.01, corresponding to a weak effect size (Moore et al., 2013).

Discussion

The therapeutic progress of young children enrolled in an intensive ABA program did not vary by group within a motor intervention. The difference between the intervention and control groups in targets mastered did not approach significance and was of very small magnitude. This suggests that the intervention did not influence ABA learning rate.

The current study finds that a motor-social intervention yielded no effect on the behavioral therapy skill acquisition of preschoolers with ASD, most of which are social in nature. This contrasts several studies whose results showed the opposite. In 2018, Najafabadi et al found improvement on two social skill scales for school-aged children with ASD who participated in a motor skill intervention (Najafabadi et al., 2018). In this study, there was no specific social component to the intervention, but students improved in social skills anyway (Najafabadi et al., 2018). Bremer & Lloyd found improvements in social skills following a strictly motor-intervention in young children with ASD (Bremer & Lloyd, 2016). Ketcheson et al did not find social changes in many domains of the social scale used (Playground Observation of Peer Engagement) but children spent less time isolated during free play after an FMS intervention. Bremer et al, 2015 saw visual improvements in social skill scores following an FMS intervention (Bremer et al., 2015). Conversely, some motor intervention studies do not find any social benefits to their programs. An aquatic skills program did not cause any social improvements, even though aquatic skills did improve (Pan, 2010). Interestingly, this study suggests that its lack of social results could be due to low power. The current study also takes place in a small sample, and this may explain our null finding.

The most likely explanation for our results is the choice of measure to assess behavioral therapy progress. The current study's measure of social skills is more accurately a measure of *ABA treatment progress*, rather than social skills. The time spent in the current intervention (1 hour per week) was much lower than all other ABA therapy (35 hours per week). There are innumerable factors affecting the ABA progress of a child with ASD, hardly any of which were addressed in this study. This suggests that children exposed to ABA ultimately progress according to their severity level, at least more so than according to the contents of a small component of their overall therapeutic intervention.

All the studies discussed here used measures of social ability for which there is some data suggesting reliability, validity, and response to change through interventions. The scales used in the published FMS interventions in children with ASD reviewed so far are the School Social Behavior Scales (Pan, 2010), the Playground Observation of Peer Engagement (Ketcheson et al., 2017), the social interaction scale of the Gilliam Autism Rating Scale and Autism Treatment Evaluation Checklist (Najafabadi et al., 2018), and the Social Skills Improvement Scale (Bremer et al., 2015; Bremer & Lloyd, 2016). In addition, some observational studies choose to use the CSS of the ADOS – 2 as a measure of social-communication (MacDonald et al., 2013b). It is also notable that the authors of these studies all chose different scales to measure the same construct. Future work investigating connections between motor and social progress in the intervention setting might benefit from some coherency in the choice of social scale used.

When investigating cross-domain results of interventions, it is challenging to compare findings with other studies because it is often unclear which components of intervention are "motor" or "social". It is also unclear whether the benefits in social skills result from social components of the intervention, or from purely motor skill instruction. To make matters more complex, many of the interventions published do not explicitly teach specific social skills, but it is obvious that some social skill development is inherent in the activity (horseback riding, for example) (Bass et al., 2009). In other cases, the motor skill being taught is more strictly motor-based (Bremer & Lloyd, 2016; Ketcheson et al., 2017).

Another area of interest that is helpful here is the literature investigating the academic or stereotypy effects of physical activity in ASD. Vigorous activity such as running also may provide cross-domain benefits in stereotypic behaviors (Kern et al., 1984; Neely et al., 2015), academic engagement (Neely et al., 2015), on-task behavior (Kern et al., 1982), and executive function (Pan et al., 2017). In these cases, it appears that these studies are investigating a different research question: the cross-domain effects of an *acute bout of movement*. It seems more likely that the biochemical effects of vigorous exercise influence the brain in real-time here. In the current study, it is assumed that expected social benefits would occur due to the *cumulative* effect of *developing motor skills*.

Considering this, what is the nature of the connection between motor and social skills? The literature in this area is emergent, but most studies do confirm that a connection exists. In autism research, there are two areas of relevant literature: cross-sectional relationships between motor and social skills, and longitudinal studies that relate infant motor timing and future social skills. These two areas of literature are discussed below.

Cross-sectional relationships between motor and social skills exist, and their relationships may be particularly interesting in ASD. One study in over 233 children ages 5 and younger, fine motor and gross motor skills both related to adaptive skills, and gross motor skills related to daily living skills too (MacDonald et al., 2013a). The same research group found relationships

between motor ability and social-communication skills in school-aged children with ASD (MacDonald et al., 2013b). Leonard and Hill present a review of 43 studies showing relationships between motor skills and social development. They find these relationships in children with ASD, as well, but indicate that this literature is more complex (Leonard & Hill, 2014). In all, they argue that motor and social skills are particularly intertwined in early development. However, they posit that all domains of development are ultimately related and that researchers and clinicians should think *developmentally* by considering development from a dynamic systems approach (Leonard & Hill, 2014).

Motor skills during infancy have gained much attention due to their potential relevance as predictive factors of ASD diagnosis. Indeed, it appears that motor behavior can be an early indicator of developmental progress, including the social and communication domains. One study conducted in over 200 children with ASD found that the age at which children learned to walk independently was very important for later language development (Bedford et al., 2016). Another study found evidence for "cascading consequences between domains" in a sample of infants at risk of autism (LeBarton & Iverson, 2016). The age at which infants learned to sit independently correlated with the age at which they started babbling, and their prone positioning skills correlated with their gesture abilities (LeBarton & Iverson, 2016). Many other studies confirm these early motor – social connections (Leonard et al., 2015). However, one study of a very large dataset of typically developing children does not support the finding of early motor skills predicting later language development and in fact finds the reverse to be the case (Wang et al., 2014). Thus, there is ultimately some debate in this area.

This study has several strengths and limitations to acknowledge. As previously stated, the measure of mastery rate on ABA social targets has limitations as a proxy for social ability, and it

may not be particularly sensitive to the small level of intervention delivered. Alternatively, the findings of this study might be interpreted differently if mastery rate on ABA social targets were thought of as a measure of ABA treatment progress in general. Future studies in the area of motor and social cross-domain changes might benefit from sticking with a standardized scale, perhaps one that has been used in this area before. In addition, the small sample size of this study provides limitations in power. However, the effect size of the coefficient in question is so small, and the significance level so high, that it is unlikely that a larger sample would have yielded significant differences from our results.

Conclusions

This study finds that there is no effect of a 20 – week motor – social play intervention upon the rate at which students learned objectives in their ABA program. These results contrast those of similar motor interventions, most of which found cross-domain benefits in social skills. Findings hold relevance for practitioners teaching motor or social skills to young children with ASD. In the early intervention ABA setting, this motor intervention had no effect on social skill learning rate, likely because of the small intervention time relative to total therapy time or because of an insensitive measure.

APPENDIX

TABLES

Table 4.1.Baseline participant demographic characteristics.

A	Control		Intervention		Total	
	(n = 6)		(n = 8)		(n = 14)	
	Mean	SD	Mean	SD	Mean	SD
Age (months)	53.83	7.17	53.88	.02	53.86	.80
Gender						
Male	3		1		4	
Female	3		7		10	
ADOS - 2 CSS	7.50	.98	8.29	.06	7.92	.98
MSEL Total DQ	56.63	14.93	53.13	18.52	54.53	16.37
PLS-5						
Expressive	64.75	3.15	59.00	2.44	61.30	2.35
Language Score						
Receptive	23.75	.54	18.00	.01	20.30	.54
Language Score						
VABS Adaptive	65.67	.52	60.67	.22	62.33	.81
Behavior						
Composite Score						

Note: $*p \le 0.05$; $**p \le 0.001$. SD = Standard deviation; ADOS – 2 CSS = Autism Diagnostic Observation Scales – 2 Calibrated severity score; MSEL total DQ = Mullen Scales of Early Learning total Developmental Quotient; PLS – 5 = Preschool Language Scales – 5; VABS = Vineland Adaptive Behavior Scales.

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CHAPTER 5

Summary

The current dissertation addresses the early childhood development of individuals with Autism Spectrum Disorder (ASD). The specific areas studied are fundamental movement skills (FMS), physical activity, body composition, and social skills. This dissertation presents original data from both an observational study and a comprehensive early intervention. The chapters are arranged in an intentional order: the observational study (chapter 2), followed by the primary outcomes of intervention (chapter 3), and finally the cross-domain outcomes of intervention (chapter 4). This approach was chosen to illustrate the process of implementing evidence-based programs in a new setting. in that the findings of chapter 2 support the rationale for chapters 3 and 4. A summary of each chapter and a brief discussion of each chapter's relevance within the context of the whole dissertation and larger literature are presented below.

Chapter 2: Differences in Fundamental Movement Skills, Body Composition, and Moderate - to - Vigorous Physical Activity of Children with and without Autism Spectrum Disorder

The second chapter of this dissertation was a cross-sectional, observational study investigating physical and behavioral differences between children with and without ASD. This study measured the body composition, physical activity, and FMS, of 46 children ages 3 - 10years. 23 of the children had ASD, and the other 23 were typically developing children selected to be age and sex matched controls. ASD diagnoses was verified by written statement from guardians, and a score indicating "ASD" or "Autism" on the Autism Diagnostic Observation Scales - 2 (ADOS - 2), completed by the research team. All participants' guardians also reported demographic information. Participants' body composition data was collected using the Gold Standard measure of body fat percentage, Air Displacement Plethysmography with the Bod Pod (Cosmed, Concord, CA). Physical activity was measured using the objective measure of

actigraphy (Actigraph wGT3x – BT, Actigraph Corp, Pensacola, FL). Participants' FMS were measured using the Test of Gross Motor Development – III (TGMD – 3). A multivariate analysis of variance (MANOVA) investigated whether the children with ASD were significantly different from their typically developing peers in body composition, physical activity (specifically moderate – to – vigorous physical activity - MVPA), and FMS. Children with and without ASD were significantly different in body composition and FMS but showed no difference in percentage of the day spent in MVPA. Children with ASD showed higher body fat percentage, and lower FMS ability than those with typical development; the magnitude of the effect was larger for FMS than for body fat percentage.

Recent meta-analytic summaries of the literature suggest that rates of obesity in children with ASD are higher than in the general population; the results of chapter 2 replicate this finding using the gold standard measure of body fat percentage (Must et al., 2017). The results of this dissertation provide evidence of higher body fat in children with ASD, but several considerations must be noted. The current study includes a comparison group that is uncharacteristically lean (mean body fat percentage close to 14%). In addition, it is important to consider the covariates included in any study attempting to study obesity. Other studies have explained the increased obesity risk in autism by adjusting for certain characteristics, emphasizing the complex and multi-factorial process at hand (Corvey et al., 2016). Chapter 2 does not covary for any variables. However, it does match controls by age and sex, and it also includes FMS and MVPA in the MANOVA used to address the research question. In other words, this study considers body fat percentage with consideration for its context in motor behavior. Considering future research, the field of obesity and ASD might benefit from more advanced epidemiological or biostatistical examinations of complex interactions between covariates and ASD. These might clarify for future work which covariates are essential risk factors to include in every obesity study in ASD.

To our knowledge, this is the first study including Bod Pod data for children with ASD. This is likely due to lack of researcher comfort managing problem behaviors This contributes an element of depth to the existing literature on obesity and ASD, most of which is based on body mass index (BMI) measurements. Our body fat percentage data correlates closely with our participants' BMI data. This provides some initial evidence that in our sample, BMI and Air Displacement Plethysmography obtain similar information. Although this is not a validation study of BMI as a measure of body fat in children with ASD, the high correlation between these measures fortifies their measurement validity. Thus, findings can support the findings of previously published obesity and ASD studies using BMI; these studies do indeed capture the concept of body fat percentage through BMI. For example, some of these studies are quite large in sample size and have meaningful findings: higher rates of obesity, higher obesity risk with age, and more serious complications of obesity for children with ASD compared to their typically developing peers.

A large body of literature supports the existence of significant delays in motor development in children with ASD. Results of chapter 2 replicate this finding. Although not part of the diagnostic criteria for ASD, children with ASD consistently show motor delays (Staples & Reid, 2010) that begin as toddlers and get larger with age (Lloyd et al., 2013). It appears that motor skills in this population also relate to developmental areas beyond the physical (MacDonald et al., 2014), emphasizing the importance of FMS and adapted physical education (APE) services for children with ASD. This is increasingly relevant for young children, given strong evidence that early intervention is effective for children with ASD (Reichow et al., 2012).

These findings thus provide strong evidence to support the importance of Chapter 3, which is an early FMS intervention.

Existing literature is conflicted in its understanding of the physical activity behavior of children with ASD. Some literature shows that children with ASD perform less physical activity than their peers during recess at school (Pan, 2008). In addition, one study showed children with ASD participating in less MVPA than their typically developing peers during the school day (Pan, 2008). A study comparing 13 – year – olds with and without ASD in Ireland found children with ASD engaged in less MVPA and light activity, were less likely to participate in sports, and spent more time watching television than their peers (Healy et al., 2017). As is the case in children with typical development, it appears that children with ASD become less active as they age from 9 – 18 years (Macdonald et al., 2011). There are reports of adolescents with ASD engaging in much more sedentary time than their peers (Corvey et al., 2016). Conversely, a study of children with ASD aged 2-5, children with ASD spent less time sedentary (Ketcheson et al., 2017a). A study of children ages 3 - 11 found that children with ASD spent similar amounts of time in MVPA, but parent report indicated that they spent less time engaging in sports, and were involved in fewer activities than their peers (Bandini et al., 2013). These contradicting findings create a complicated body of literature that remains unsettled. The findings of chapter 2 do not do much to clear the waters but show again a lack of difference in the physical activity habits of children with ASD.

Analogous to the situation of obesity research in children with ASD, contradicting findings in the field of physical activity and autism might be due to uncertainty regarding measurement. In addition, the physical activity in autism research also tends to be comprised of small sample sizes under heterogenous conditions, making summary difficult. physical activity

measurement is complex. A recent systematic review of physical activity in children with and without ASD summarizes the state of knowledge beautifully by calling for more and better investigations (Jones et al., 2017). Specifically, the review noted that most studies in this area have a high risk of bias, small sample sizes, conflicting results, unidentified correlates of physical activity behaviors (Jones et al., 2017). Many of these study characteristics are inherent in the study of children with ASD, which is a relatively rare condition and is extremely heterogenous in presentation. In addition, parents of children with ASD report higher levels of stress than those of neurotypical children (Schieve et al., 2007), and parents under pressure likely prioritize more pressing parenting matters over research engagement.

Chapter 2 of this dissertation finds that children ages 3 - 10 in this sample have higher body fat percentages, lower FMS proficiency, and engage in the same amount of MVPA as age and sex matched controls. Findings provide support for early interventions for weight management and FMS development.

Chapter 3: Training the Fundamental Movement Skills of Preschoolers with Severe Autism Spectrum Disorder: Preliminary Efficacy and Feasibility in Early Intensive Behavioral Intervention Centers

The third chapter of this dissertation presents the efficacy, feasibility, and fidelity of an early intervention targeting FMS and social play through Applied Behavior Analysis (ABA). Participants at two sites of an early intervention ABA institute were randomized to control and intervention groups. Participants in the control group continued to receive therapy as usual. Participants in the intervention group received an individualized FMS intervention. Over 20 weeks, this randomized controlled trial aimed to teach FMS through discrete trial training and graduated guidance. Behavior technicians already providing full time therapy to the participants

were the individuals delivering the intervention. The behavior technicians prompted FMS on a one – to – one level for 15 minutes per day, four days a week with research staff present. The FMS taught were the 13 skills assessed on the TGMD – 3, intentionally chosen because of their importance in motor skill development. The intervention also included a component addressing social play skills through video modelling and group play activities (see chapter 4). This preliminary study also included measurement of intervention fidelity and implementor – rated feasibility.

Results indicate that the intervention group showed a significant improvement in FMS relative to the control group. Ball skills improved more than locomotor skills. The intervention was implemented with high fidelity according to random video review according to a fidelity checklist. Some behavior technicians agreed that FMS were important to teach, but others noted that more serious problem behaviors should be addressed first. Behavior technicians expressed discontent with the training they received, noting that the prompt levels were inconsistent and the training they received was insufficient. They also noted several significant challenges for implementing an active intervention in the early intervention ABA setting. These challenges provide support for both the delivery of APE services earlier in life to children with ASD, and the increased interdisciplinary collaboration between ABA and physical/occupational therapy experts, as the expertise of both could improve the outcomes of either.

Future directions for this intervention study include the secondary analysis of half-point data (at which point there were 10 hours of intervention delivered) as a potential minimally effective dose of instructional time. The 2018 Patricia Austin Award Presentation at the North American Federation for Adapted Physical Activity addressed the question of dose (Case, 2018). In other words, "How much instructional time is necessary to change the FMS of children with

ASD?" through a meta-analysis. Case found a substantial publication bias in this literature, wherein most published interventions showed a significant treatment effect, and very few used less than 12 instructional hours (Case, 2018). Currently, the question of minimal instructional time is unanswered, and data existing from the current study might be helpful in addressing this question.

This is the first FMS intervention to attempt integration into an early intervention ABA center. These centers have gained attention in recent times for ASD treatment in general, however. These centers are generally specified for children with severe ASD who qualify for intensive behavioral treatment before entry into kindergarten. There are typically no APE services offered in this environment. These centers typically use individualized therapy plans and a small staff: student ratio, thus they hold promise as a delivery platform for early FMS intervention services. The results of chapter 3 do not discount the early intervention ABA environment as a location for FMS intervention. In fact, they suggest that significant improvement is achieved in only 20 hours of individual intervention time. However, many serious implementation issues would need to be addressed for continued collaboration of this kind.

Social Play Outcomes of an Applied Behavior Analytic Early Intervention Program Targeting Both Social and Motor Domains

Chapter 3 of this dissertation is concerned with the cross-domain effects of the intervention described in chapter 3. The social play component of the intervention described in chapter 3 was aimed at integrating FMS into naturalistic social play environments. Thus, participants and their behavior therapists engaged in age-appropriate active games with both an FMS and a social component. Games were played in a group with the entire intervention group

at each site and their behavior therapists. Video modeling was used as the instructional method, and behavior technicians provided manual prompting and reinforcement to encourage following the rules and social customs of each game. Chapter 3 investigated whether the complete intervention resulted in significant social play improvement. In contrast to other published studies investigating cross-domain benefits of motor interventions, chapter 3 used a social play measure that was deeply ingrained in the ABA center's treatment. The social play measure was the number of mastered therapy targets in the child's program plan over the course of the intervention. The number of mastered therapy targets was not different between the control and intervention groups.

These findings contrast with several published studies (Sowa & Meulenbroek, 2012). A 12-week horseback program provided social motivation and responsiveness benefits to 5-10 year-olds with ASD (Bass et al., 2009). Social skill scores appeared to improve after a 12 week in-school FMS intervention for children with ASD-like characteristics aged 3-7 (Bremer & Lloyd, 2016). After an intensive FMS intervention for children aged 3-6 with ASD, children spent less free play time solitary than at baseline (Ketcheson et al., 2017b). In other cases, no changes in social skills arose after physical activity interventions, but authors explained these null results by low statistical power or a lack of concrete emphasis on social skills (Bremer et al., 2015; Pan, 2010).

The differences between chapter 4 and the published studies in this area are glaring. The current study had much less time available for instruction (Ketcheson et al., 2017b). In other words, the ratio of the time spent in intervention to the rest of the time in the child's day was small. It seems unlikely that such a small dosage of FMS and social play intervention could have a powerful effect. Similarly, social skills were not explicitly taught in this intervention, and it is

understood that children with ASD may have trouble generalizing learned skills to new environments. The current study's environment was also intently focused on social play targets for the rest of the day outside of intervention time, so the effect of intervention was likely washed out by the intense efforts of the rest of the day in this area. The current study's sample also had relatively severe ASD, compared to another study conducted in children with ASD-like characteristics(Bremer & Lloyd, 2016). Lastly, the current study did not use a standardized scale to measure social skills, and this may have affected the sensitivity for detection of change. Finally, there may be a publication bias present in this situation, as some motor interventions in this population likely measured social outcomes but had difficulty publishing the null results.

Implications and Future Directions

Obesity prevention and FMS are meaningful goals for children with ASD

Obesity is a relevant concern for children with ASD. Results of Chapter 2 demonstrate higher body fat percentage in a sample of children with ASD compared to their typical peers. This is the first study to use the gold standard technique of air displacement plethysmography in children with ASD. FMS remains an area in which children with ASD struggle, and early intervention is justified. Challenges of the field addressing obesity and ASD remain salient, including measurement differences, disagreements on which covariates are most important at the population level, and appropriately measuring the wide variety of presentations in various domains in ASD. Researchers understand that there are unique challenges for children with ASD, and that these can inform effective obesity interventions.

In chapter 2, we demonstrated significant deficits in FMS as well as the higher body fat percentage for children with ASD. The role of FMS in obesity prevention should not be

forgotten. FMS are early competencies that must be mastered on the path to lifelong physical activity engagement, which is a path that children with ASD may find quite difficult, especially as they age. As demonstrated in chapter 3, there are effective techniques that improve the FMS of children with ASD. In fact, some of the unique characteristics of children with ASD could conceivably predispose them toward excellence in athletics; sports could become a major strength of theirs if taught effectively. Children with ASD might gravitate toward the serious athletic environment, even perhaps more than their peers, if they were taught to understand the structure at an early age.

Measurement in ASD is complex

Chapter 2, in which body composition, physical activity, and FMS were all measured in a sample of children with ASD. As previously discussed, there is some debate in the literature regarding the differences between children with and without ASD in body composition and physical activity. As much of the literature acknowledges, most of this is due to measurement concerns perhaps relating to medications or ASD severity. Physical activity is a complicated concept to measure even outside of children with ASD, and the study of measuring physical activity specifically in children with ASD is still in its early stages. An area of interesting challenge in this field is the measurement challenge of stereotypies or other repetitive behaviors. It's unclear whether any or all these activities should be considered "health enhancing physical activity". It has not yet been established if and how much these behaviors inflate objective physical activity measurements. Future work could better validate objective physical activity measurement devices in special populations such as those with ASD.

FMS intervention is effective and interdisciplinary collaboration is encouraged

The results of the study in chapter 3 tell us that the FMS of children in an ABA early intervention center can be improved by technicians unfamiliar with FMS development. Implementation of interventions by individuals from a distinctly different profession than the intervention area can be successful with improved interdisciplinary training or integration of physical educators in delivery of services. The distracting environment, lack of instructional time, and enhanced triggering of problem behaviors were identified as concerns. However, it is noted that typical adapted physical education services are provided in similarly chaotic environments such as hallways, cafeterias, and entranceways. In addressing these concerns, it may be a more elegant solution to ensure that any instructional techniques for such situations account for these variables, or at least acknowledge their inevitability. However, within the ABA setting, if an FMS program were integrated into an existing therapy plan and completed in short bursts throughout the day, the environmental, time, and problem behavior variables might be reduced significantly. In fact, the active FMS targets could turn out to be stimulating or redirecting and could be effective tools for avoiding problem behaviors caused in other scenarios.

Cross-domain development

The study in chapter 4 found no social changes related to participation in a 20-week FMS and social play intervention, in contrast to most similar published literature of FMS interventions that also yielded social improvements. Future directions include explicitly teaching the social play skills assessed in the intervention being studied, rather than hoping that cross-domain benefits would simply emerge. In addition, more sensitive outcome measure that specifically addressed social skills in a standardized fashion would also serve this study well. Unpublished data from the same sample shows that daily total problem behaviors actually reduced in the

intervention group more than the control group over the course of the intervention. Thus, there is some evidence to support cross-domain benefits of the intervention in chapter 4. It also could be that this effect is simply due to providing a 15-minute period each day for the cathartic release of problem behavior energy, rather than truly an effect of the FMS intervention. However, it initially suggests that this FMS intervention provided some cross-domain benefit in the form of reduced problem behaviors.

Limitations

The current dissertation has several limitations that must be acknowledged. Chapter 2 presents observational data from a small sample of children that may not be representative of the general population. The sample showed relatively high severity ASD and the control group showed very low body fat percentages compared to published literature of similar children. Chapter 2 is therefore a small preliminary study intended to inform the intervention presented in chapters 3 and 4. Chapters 3 and 4 present preliminary FMS and ABA therapy effects of an FMS and social play intervention within an early intervention ABA environment. Again, the sample size in chapters 3 and 4 are small. In addition, chapters 3 and 4 were implemented in a research-friendly university-based ABA clinic, rather than a for-profit business. It is unclear whether the program could be scaled to more commercial situations, considering both the perhaps low priority of motor development in ASD treatment.

Conclusions

This dissertation suggests that children with ASD exhibit FMS delays and obesity risk, with less clear results concerning physical activity habits. Due to large FMS delays, early interventions can be designed and implemented in unconventional environments, but continued interprofessional communication and training is necessary. ABA behavior technicians with no
motor training can implement a successful FMS intervention within 20 hours of intervention time. Multi-domain interventions often have multi-domain results, but measurement of these concepts is challenging. Those working with children with ASD are encouraged to think "developmentally" with an understanding that early development takes place through dynamic interactions. APPENDIX

IRB Approval Letters

Chapter 2

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Continuing Review APPROVAL Pre-2018 Common Rule

August 12, 2019

- To: Janet L Hauck
- Re: MSU Study ID: LEGACY15-787 IRB: Biomedical and Health Institutional Review Board (BIRB) Principal Investigator: Janet L Hauck Category: Expedited 4, 7 Submission: Continuing Review CR00000818 Submission Approval Date: 8/12/2019 Effective Date: 8/12/2019 Study Expiration Date: 8/11/2020

Title: Motor Skills, Physical Activity, and Body Composition of Children Ages 1-18

This submission has been approved by the Michigan State University (MSU) BIRB. The submission was reviewed by the Institutional Review Board (IRB) through the Non-Committee Review procedure. The IRB has found that this study protects the rights and welfare of human subjects and meets the requirements of MSU's Federal Wide Assurance (FWA00004556) and the federal regulations for the protection of human subjects in research (e.g., pre-2018 45 CFR 46, 28 CFR 46, 21 CFR 50, 56, other applicable regulations).

IRB approval expired on 8-5-19. Project granted renewed approval on 8-13-19 after PI confirmed no research activities occurred during the expired period.

How to Access Final Documents

To access the study's final materials, including those approved by the IRB such as consent forms, recruitment materials, and the approved protocol, if applicable, please log into the Click™ Research Compliance System, open the study's workspace, and view the "Documents" tab. To obtain consent form(s) stamped with the IRB watermark, select the "Final" PDF version of your consent form(s) as applicable in the "Documents" tab. Please note that the consent form(s) stamped with the IRB watermark must typically be used.

Continuing Review: IRB approval is valid until the expiration date listed above. If the research continues to involve human subjects, you must submit a Continuing Review request at least one month before expiration.

Modifications: Any proposed change or modification with certain limited exceptions discussed below must be reviewed and approved by the IRB prior to implementation of the change. Please submit a Modification request to have the changes reviewed. If changes are made at the time of continuing review, please submit a Modification and Continuing Review request.

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517-355-2180 Fax: 517-432-4503 Email: irb@msu.edu www.hrpp.msu.edu New Funding: If new external funding is obtained to support this study, a Modification request must be submitted for IRB review and approval before new funds can be spent on human research activities, as the new funding source may have additional or different requirements.

Immediate Change to Eliminate a Hazard: When an immediate change in a research protocol is necessary to eliminate a hazard to subjects, the proposed change need not be reviewed by the IRB prior to its implementation. In such situations, however, investigators must report the change in protocol to the IRB immediately thereafter.

Reportable Events: Certain events require reporting to the IRB. These include:

- Potential unanticipated problems that may involve risks to subjects or others
- Potential noncompliance
- Subject complaints
- Protocol deviations or violations
- Unapproved change in protocol to eliminate a hazard to subjects
- Premature suspension or termination of research
- Audit or inspection by a federal or state agency
- · New potential conflict of interest of a study team member
- Written reports of study monitors
- Emergency use of investigational drugs or devices
- Any activities or circumstances that affect the rights and welfare of research subjects
- Any information that could increase the risk to subjects

Please report new information through the study's workspace and contact the IRB office with any urgent events. Please visit the Human Research Protection Program (HRPP) website to obtain more information, including reporting timelines.

Personnel Changes: Key study personnel must be listed on the MSU IRB application for expedited and full board studies and any changes to key study personnel must to be submitted as modifications. Although only key study personnel need to be listed on a non-exempt application, all other individuals engaged in human subject research activities must receive and maintain current human subject training, must disclose conflict of interest, and are subject to MSU HRPP requirements. It is the responsibility of the Principal Investigator (PI) to maintain oversight over all study personnel and to assure and to maintain appropriate tracking that these requirements are met (e.g. documentation of training completion, conflict of interest). When non-MSU personnel are engaged in human research, there are additional requirements. See HRPP Manual Section 4-10, Designation as Key Project Personnel on Non-Exempt IRB Projects for more information.

Prisoner Research: If a human subject involved in ongoing research becomes a prisoner during the course of the study and the relevant research proposal was not

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reviewed and approved by the IRB in accordance with the requirements for research involving prisoners under subpart C of 45 CFR part 46, the investigator must promptly notify the IRB.

Site Visits: The MSU HRPP Compliance office conducts post approval site visits for certain IRB approved studies. If the study is selected for a site visit, you will be contacted by the HRPP Compliance office to schedule the site visit.

For Studies that Involve Consent, Parental Permission, or Assent Form(s):

Use of IRB Approved Form: Investigators must use the form(s) approved by the IRB and must typically use the form with the IRB watermark.

Copy Provided to Subjects: A copy of the form(s) must be provided to the individual signing the form. In some instances, that individual must be provided with a copy of the signed form (e.g. studies following ICH-GCP E6 requirements). Assent forms should be provided as required by the IRB.

Record Retention: All records relating to the research must be appropriately managed and retained. This includes records under the investigator's control, such as the informed consent document. Investigators must retain copies of signed forms or oral consent records (e.g., logs). Investigators must retain all pages of the form, not just the signature page. Investigators may not attempt to de-identify the form; it must be retained with all original information. The PI must maintain these records for a minimum of three years after the IRB has closed the research and a longer retention period may be required by law, contract, funding agency, university requirement or other requirements for certain studies, such as those that are sponsored or FDA regulated research. See HRPP Manual Section 4-7-A, Recordkeeping for Investigators, for more information.

Closure: If the research activities no longer involve human subjects, please submit a Continuing Review request, through which study closure may be requested. Human subject research activities are complete if there is no further interactions or interventions with human subjects and/or no further analysis of identifiable private information.

For More Information: See the HRPP Manual (available at hrpp.msu.edu).

Contact Information: If we can be of further assistance or if you have questions, please contact us at 517-355-2180 or via email at IRB@msu.edu. Please visit hrpp.msu.edu to access the HRPP Manual, templates, etc.

Expedited Category. Please see the appropriate research category below for the full regulatory text.

Expedited 1. Clinical studies of drugs and medical devices only when condition (a) or (b) is met.

(a) Research on drugs for which an investigational new drug application (21 CFR Part 312) is not required. (Note: Research on marketed drugs that significantly

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increases the risks or decreases the acceptability of the risks associated with the use of the product is not eligible for expedited review.)

(b) Research on medical devices for which (i) an investigational device exemption application (21 CFR Part 812) is not required; or (ii) the medical device is cleared/approved for marketing and the medical device is being used in accordance with its cleared/approved labeling.

Expedited 2. Collection of blood samples by finger stick, heel stick, ear stick, or venipuncture as follows:

(a) from healthy, nonpregnant adults who weigh at least 110 pounds. For these subjects, the amounts drawn may not exceed 550 ml in an 8 week period and collection may not occur more frequently than 2 times per week; or (b) from other adults and children, considering the age, weight, and health of the subjects, the collection procedure, the amount of blood to be collected, and the frequency with which it will be collected. For these subjects, the amount drawn may not exceed the lesser of 50 ml or 3 ml per kg in an 8 week period and collection may not occur more frequently than 2 times per week.

Expedited 3. Prospective collection of biological specimens for research purposes by noninvasive means.

Examples: (a) hair and nail clippings in a nondisfiguring manner; (b) deciduous teeth at time of exfoliation or if routine patient care indicates a need for extraction; (c) permanent teeth if routine patient care indicates a need for extraction; (d) excreta and external secretions (including sweat); (e) uncannulated saliva collected either in an unstimulated fashion or stimulated by chewing gumbase or wax or by applying a dilute citric solution to the tongue; (f) placenta removed at delivery; (g) amniotic fluid obtained at the time of rupture of the membrane prior to or during labor; (h) supra- and subgingival dental plaque and calculus, provided the collection procedure is not more invasive than routine prophylactic scaling of the teeth and the process is accomplished in accordance with accepted prophylactic techniques; (i) mucosal and skin cells collected by buccal scraping or swab, skin swab, or mouth washings; (j) sputum collected after saline mist nebulization.

Expedited 4. Collection of data through noninvasive procedures (not involving general anesthesia or sedation) routinely employed in clinical practice, excluding procedures involving x-rays or microwaves. Where medical devices are employed, they must be cleared/approved for marketing. (Studies intended to evaluate the safety and effectiveness of the medical device are not generally eligible for expedited review, including studies of cleared medical devices for new indications.) Examples: (a) physical sensors that are applied either to the surface of the body or at a distance and do not involve input of significant amounts of energy into the subject or an invasion of the subject's privacy; (b) weighing or testing sensory acuity; (c) magnetic resonance imaging; (d) electrocardiography, electroencephalography, thermography, detection of naturally occurring radioactivity, electroretinography; (e) moderate exercise, muscular strength testing, body composition assessment, and flexibility testing where appropriate given the age, weight, and health of the individual.

Expedited 5. Research involving materials (data, documents, records, or specimens) that have been collected, or will be collected solely for nonresearch purposes (such as medical treatment or diagnosis). (NOTE: Some research in this category may be exempt from the HHS regulations for the protection of human subjects. 45 CFR 46.101(b)(4). This listing refers only to research that is not exempt.)

Expedited 6. Collection of data from voice, video, digital, or image recordings made for research purposes.

Expedited 7. Research on individual or group characteristics or behavior (including, but not limited to, research on perception, cognition, motivation, identity, language, communication, cultural beliefs or practices, and social behavior) or research employing survey, interview, oral history, focus group, program evaluation, human factors evaluation, or quality assurance methodologies. (NOTE: Some research in this category may be exempt from the HHS regulations for the protection of human subjects. 45 CFR 46.101(b)(2) and (b)(3). This listing refers only to research that is not exempt.)

Expedited 8. Continuing review of research previously approved by the convened IRB as follows:

(a) where (i) the research is permanently closed to the enrollment of new subjects;
(ii) all subjects have completed all research-related interventions; and (iii) the research remains active only for long-term follow-up of subjects; or
(b) where no subjects have been enrolled and no additional risks have been identified; or

(c) where the remaining research activities are limited to data analysis.

Expedited 9. Continuing review of research, not conducted under an investigational new drug application or investigational device exemption where categories two (2) through eight (8) do not apply but the IRB has determined and documented at a convened meeting that the research involves no greater than minimal risk and no additional risks have been identified.

Chapters 3 and 4

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Continuing Review APPROVAL

September 18, 2018

To: Janet L Hauck

Re: MSU Study ID: LEGACY17-1557 IRB: Biomedical and Health Institutional Review Board Principal Investigator: Janet L Hauck Category: Expedited 4, 6, 7 Submission: Continuing Review CR00000332 Submission Approval Date: 9/7/2018 Effective Date: 9/18/2018 Study Expiration Date: 9/6/2019

Title: Preschool-based applied behavior analytic gross motor skill instruction for children with Autism Spectrum Disorder

This submission has been approved by the Michigan State University (MSU) BIRB. The submission was reviewed by the Institutional Review Board (IRB) through the Non-Committee Review procedure. The IRB has found that this study protects the rights and welfare of human subjects and meets the requirements of MSU's Federal Wide Assurance (FWA00004556) and the federal regulations for the protection of human subjects in research (e.g., 45 CFR 46, 21 CFR 50, 56, other applicable regulations).

How to Access Final Documents

To access the study's final materials, including those approved by the IRB such as consent forms, recruitment materials, and the approved protocol, if applicable, please log into the Click™ Research Compliance System, open the study's workspace, and view the "Documents" tab. To obtain consent form(s) stamped with the IRB watermark, select the "Final" PDF version of your consent form(s) as applicable in the "Documents" tab. Please note that the consent form(s) stamped with the IRB watermark must typically be used.

Continuing Review: IRB approval is valid until the expiration date listed above. If the research continues to involve human subjects, you must submit a Continuing Review request at least one month before expiration.

Modifications: Any proposed change or modification with certain limited exceptions discussed below must be reviewed and approved by the IRB prior to implementation of the change. Please submit a Modification request to have the changes reviewed. If changes are made at the time of continuing review, please submit a Modification and Continuing Review request.

Immediate Change to Eliminate a Hazard: When an immediate change in a research protocol is necessary to eliminate a hazard to subjects, the proposed

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