Examination of factors related to motor performance and physical activity in school-aged children with and without Down syndrome

by

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This dissertation is dedicated to Alyssa, Max, and Quinn. Without your love, support, and inspiration, this would not have been possible.

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ABSTRACT

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Due to widespread genetic constraints, children with Down syndrome (DS) are believed to experience deficits in several developmental domains. However, much of the research in this area is dated or inconclusive. In addition, the inter-relatedness between developmental domains is relatively unexplored in children with DS. Therefore, the primary objectives of the current studies were to: 1) describe the physical, motor, and activity profiles of school-aged children with and without DS; 2) explore physical- and activity-based correlates to gross motor skills; and 3) investigate socio-emotional characteristics of children with and without DS and determine if they were related to gross motor skills, physical activity, and participation. To address these questions, forty children with DS (N=20, mean age = 7.9 years) and with typical development (TD) (N=20, mean age = 7.9 years) were assessed on their physical, motor, activity, and socio-emotional characteristics. Results indicated that children with DS had increased range of motion, decreased leg strength, poorer gross motor skills, and less diversity in their recreational activities compared to the TD group. On average, the two groups were not

different in terms of their generalized physical activity levels, however, which may be due to hyperactivity in a subsample of the children with DS. Findings also revealed that physical characteristics (e.g. leg strength), generalized activity, and participation all significantly predicted gross motor skill performance in the DS group whereas only physical characteristics significantly contributed to models of gross motor skills for the children with TD. With respect to socio-emotional development, children with DS had more social and behavioral problems than their peers with TD. Increased gross motor skill ability and participation were associated with higher social competence in both groups. For children with DS, generalized physical activity seemed to be associated with behavioral problems curvilinearly such that there may be an optimal range of activity to promote socio-emotional well-being in this population. Collectively, results from these studies demonstrate the high level of inter-relatedness between developmental domains in school-aged children with DS. Future research should consider motor skill and physical activity interventions to promote overall development and well-being in this population.

CHAPTER I

Introduction

Overview

The main objectives of this dissertation are to provide comprehensive descriptions of the motor, physical, and socio-emotional characteristics of children with and without Down syndrome (DS) and to explore relationships between these domains. For the purpose of this dissertation, the "motor" domain refers to gross motor skills and physical activity whereas the "physical" domain consists of body composition, range of motion, and leg strength. A variety of social, emotional, and behavioral factors, such as social awareness and aggression, comprise the "socio-emotional" domain. Study 1 is largely descriptive, where the motor and physical characteristics of the two groups (DS vs. non-DS) are presented and compared. Study 2 investigates relationships between motor and physical abilities in addition to associations within the motor domain (i.e. motor skills vs. physical activity). Finally, Study 3 describes the socio-emotional characteristics of children with and without Down syndrome and examines whether group differences in these variables exist. Study 3 also explores relationships between the motor and socio-emotional domains. The ensuing pages provide a review of the literature pertinent to these three studies.

Review of Literature

A. Down syndrome: Multi-domain deficits

Down syndrome (DS), first described by Dr. John Langdon Down in 1866, is both the oldest and most common known cause of intellectual disability. Recent estimates place the rate of incidence of DS at 1 in every 733 to 1 in every 850 live births in the United States (Canfield, Ramadhani, Yuskiv, & Davidoff, 2006; Shin, Besser, Kucik, Lu, Siffel, &Correa, 2009). DS is caused by the presence of all or part of an extra 21st chromosome. Due to this chromosomal abnormality, every cell in the body is affected in most cases. Such widespread genetic abnormality may explain why individuals with DS demonstrate developmental problems within multiple domains. Though DS is most commonly associated with intellectual disability (ID), equally important deficiencies are experienced within the physical, motor, and socio-emotional domains. Our current understanding of these three domains (physical, motor, socio-emotional) with respect to DS will be described in the following sections.

1) Physical Domain

For the purpose of this dissertation, the "physical domain" includes body composition, range of motion, and muscle strength. From a dynamic systems perspective (DSP), these factors may be viewed as potential *control parameters* for motor behavior. DSP proposes that several subsystems (i.e. musculo-skeletal, sensory, cardiovascular, neural, etc.) self-organize to influence behavior within a given task (Kamm, Thelen, & Jensen, 1990). Control parameters are variables that, when scaled up or down beyond a critical point, cause a shift in behavior. For example, when water

reaches a certain temperature it begins to boil. Therefore, temperature can be considered a control parameter in this context because once it reaches a critical level there is a qualitative change in the molecular action of the water. Accordingly, it is plausible to suggest that body composition, muscle strength, or range of motion might serve as control parameters for motor performance in school-aged children with and without DS. In fact, infant studies have implicated several of these factors in early locomotor behavior (Ulrich, Ulrich, Angulo-Kinzler, & Yoon, 2001; Thelen, Fisher, Ridley-Johnson, & Griffin, 1982). Moreover, it appears that individuals with DS experience a variety of physical challenges not only in infancy but throughout the lifespan.

Of the physical factors impacted in individuals with DS, perhaps the most visibly obvious are those related to body composition. There is substantial research to indicate a high prevalence of obesity in individuals with DS. In fact, several experts believe that DS obesity rates should be considered a major public health concern (Rubin, Rimmer, Chicoine, Braddock, & McGuire, 1998). Several studies illustrate the heightened obesity levels in DS. For example, Prasher (1995) examined two-hundred individuals with DS in the United Kingdom. Findings revealed that 31% of males and 22% of females were overweight (BMI = 25-29) while 48% of males and 47% of females were categorized as obese (BMI = 30-34). In a similar study in the U.K, Bell & Bhate (1992) observed 58% of males and 83% of females with DS to be overweight or obese. These percentages were much higher than those observed in a population without intellectual disability, who demonstrated rates of 40% and 32% for males and females, respectively. Rubin and colleagues (1998) reaffirmed the previous findings within the United States by

demonstrating that individuals with DS had overweight rates 12-20% higher than the general population.

Studies on obesity in children with DS are not as conclusive as the adult literature. For example, Sharav and Bowman (1992) examined thirty pairs of siblings between the ages of 2 and 14 years, each pair consisting of one child with DS. The findings showed no differences in BMI between the children with and without DS. In contrast, Whitt-Glover and colleagues found that children with DS had higher BMI values compared to their unaffected siblings (Whitt-Glover, O'Neill, and Stettler, 2006). In yet another study, fat-free mass values measured using three different methods (bioelectrical impedance, skinfold thickness, and deuterium dilution) were not found to be different between prepubescent children with and without DS (Luke, Sutton, Schoeller, & Roizen, 1996). Therefore, inconsistent findings exist with respect to body composition differences between children with and without DS.

Unlike body composition, there is a strong consensus on the musculo-skeletal abnormalities present in children with DS. Documented problems include hypotonia, joint laxity, and hypermobility. While conducting orthopedic evaluations, Concolino and colleagues found that 30% of children with DS had at least moderate hypotonia whereas only slight hypotonia was observed in 5% of the children without DS (Concolino, Pasquzzi, Capalbo, Sinopoli, & Strisciuglio, 2006). Concolino's results are consistent with earlier work by Morris, Vaughan, & Vaccaro (1982), who also found decreased muscle tone in children with DS compared to control subjects.

Concolino's work revealed additional musculo-skeletal challenges for children with DS, namely increased range of motion and joint laxity. Specifically, they observed that all fifty children with DS had joint laxity, with 20% demonstrating "severe" laxity. In contrast, 80% of the children without DS demonstrated no laxity at all. Excessive range of motion, particularly at the hip, has been widely documented in other studies as well. Angelopoulou and colleagues (1999) examined range of motion in children with DS, other forms of ID, and no ID (Angelopoulou, Tsimaris, Chirstoulas, & Mandroukas, 1999). The children with DS had significantly higher ranges of motion for hip flexion compared to the group with non-DS intellectual disability. In addition, significantly higher ranges of motion for hip abduction were observed for the children with DS relative to the other two groups. Difficulties with range of motion in DS are not limited to the hip, however. Research has shown that in addition to hip dysplasia, individuals with DS are also susceptible to patellar instability and disorders of the ankle, such as pes planus (Mik, Gholve, Scher, Widmann, & Green, 2008).

The presence of musculo-skeletal abnormalities may contribute to observed strength deficiencies in individuals with DS. Croce (1996) found that adults with DS demonstrated significantly lower isokinetic hamstring and quadriceps peak torque levels relative to sedentary controls. Isokinetic strength in adults with DS has also been compared to adults with other forms of intellectual disability (Croce, Pitotti, Horvat, & Miller (1996). Results from Pitetti et al. (1992) showed that isokinetic arm (elbow flexion/extension) and leg (knee flexion/extension) strength were lower in DS and non-DS ID groups in relation to the adults without ID (Pitetti, Climstein, Mays, & Barrett,

1992). Additionally, the DS group exhibited decreased leg strength compared to the non-DS ID group, suggesting that factors outside of cognition contribute to strength deficiencies in DS.

Decreased muscle strength in DS has not been limited to adult populations however. Deficiencies in knee strength have been documented in children and adolescents with DS as well. Specifically, Cioni et al. (1994) examined isokinetic knee extensor strength in children and adolescents with DS, with non-specific ID, and with typical development (Cioni, Cocilovo, Di Pasquale, Araujo, Siqueira, & Bianco, 1994). Results showed that both children and adolescents with DS had significantly lower leg strength when compared with the other two groups. Moreover, by the age of 14 years, individuals with DS failed to demonstrate gains in muscle strength that typically accompany adolescence. Strength deficits in children with DS have also been observed by Mercer & Lewis (2001).

Collectively, the previously mentioned studies illustrate that children with DS demonstrate a variety of physical difficulties. Specifically, individuals with DS face constraints related to range of motion and muscle strength. Body composition and obesity appear to be a concern for adults with DS as well. However, there are inconsistent findings regarding body composition in children with DS. Nevertheless, children with DS face significant challenges within the physical domain. Difficulties in the areas of motor performance and physical activity also exist and will be the subject of the next section.

2) *Motor Domain*

In this section, descriptions of motor skill development and physical activity in Down syndrome are presented. Relationships between motor skill development and physical activity will also be addressed. Associations between these factors and those from the physical or socio-emotional domain will be discussed in subsequent sections of the literature review.

The health benefits of physical activity in children have been well-documented. They include, but are not limited to, increased HDL cholesterol, lower blood pressure, and healthier body composition (for review see Strong, 1990). Unfortunately, individuals with DS display less than optimal physical activity profiles from an early age. As evidence, McKay and Angulo-Barroso (2006) observed that the infants with DS spend more time in lower intensity activities compared to infants with typical development. Early deficiencies in physical activity may lay a foundation for ongoing patterns of inactivity in children with DS. For example, a recent pilot study found that less than half of the children with DS accumulate recommended values of daily physical activity (Shields, Dodd, and Abblitt, 2009). Sibling studies, which allow for comparisons of DS vs. non-DS activity levels, also suggest that children with DS are relatively inactive. Sharav & Bowman (1992) found that children with DS were less active and showed a preference for indoor activities when compared to their unaffected siblings. In another sibling study, Whitt-Glover and colleagues (2006) demonstrated that children with DS (ages 3-10 years) accumulated less vigorous activity than their siblings and performed these

activities for shorter durations. Whitt-Glover's study showed that children with and without DS spent similar times in moderate and lower intensity activities, however.

Research has also been conducted to determine if activity levels are impacted by setting in children with DS. For example, Faison-Hodge and Porretta (2004) compared the physical activity levels of school-age students with mild intellectual disability, including DS, during physical education and recess. Results showed a significant difference in moderate to vigorous activity between settings, with more favorable activity levels during recess. The authors concluded that physical education settings should focus on maximizing activity through games that incorporate a variety of locomotor skills. This recommendation seems warranted given the deficits in gross motor skills in children with DS and other forms of intellectual disability.

From an early age, individuals with DS demonstrate significant challenges with respect to their motor skill development. In a study of general movements in infancy, Mazzone, Mugno, & Mazzone (2004) found that infants with DS showed decreased movement speed, more abrupt beginnings and endings of movements, and fewer concurrent gross movements compared to infants with TD. More specifically, atypical patterns of leg movements in infants with DS have also been observed. For instance, Ulrich and Ulrich (1995) demonstrated that infants with DS performed less functional leg movements than their peers with TD. Low levels of functional leg movements and decreased activity may partially explain delayed walking onset in children with DS (Ulrich & Ulrich, 1995; McKay & Angulo-Barroso, 2006).

There is an abundance of research to suggest that children with DS begin to walk, on average, a year later than children with TD (Henderson, 1986; Ulrich & Ulrich, 1995; Ulrich, Ulrich, Angulo-Kinzler, & Yun, 2001). Based on growth-motor curves, Palisano and colleagues (2001) determined that children with DS have only a 40% probability of attaining independent walking by 2 years of age. Studies also demonstrate delays in crawling in children with DS, but not to the extent suggested by Palisano. Ulrich and Ulrich (1995) found that infants with DS crawl, on average, at 13.7 months. More recently, it has been observed that 75% of infants with DS crawl between 10 and 14 months (Ulrich, Lloyd, Tiernan, Looper, and Angulo-Barroso, 2008). Of note, children in Ulrich's 2008 study received treadmill intervention, which may have facilitated crawling onset. Regardless, the collective results of these studies illustrate the presence of motor skill delays in infants with DS. In addition to delays, locomotor abnormalities have been found once the skill is attained. Specifically, toddlers with DS display a wider-based gait (Lydic & Steele, 1979) and more variable gait patterns compared to toddlers with TD (Parker, Bronks, & Snyder, 1986; Looper, Wu, Angulo-Barroso, Ulrich, & Ulrich, 2006).

Motor difficulties in early life seem to persist into childhood, although they are less understood. Research has shown that children with DS score lower than their non-affected peers on a variety of functional motor tasks, including running speed, balance, strength, and visuo-motor control (Connolly & Michael, 1986). Jobling (1998), using the Bruininsk Oseretsky Test of Motor Proficiency (BOTMP), found that children with DS were highly variable in their performances depending on the motor skill that was tested.

For example, skills with a high balance component and items involving upper-limb coordination illustrated substantial problems for children with DS. On the other hand, running speed and agility performance were observed to be age-appropriate, which contradicted results from Connolly & Michael (1986). Problems with object control skills in children with DS have also been documented using Movement ABC instrument (Spano et al., 1999).

Many of the skills assessed in the preceding studies can be classified as fundamental movement skills (FMS). Researchers have argued that FMS represent requisite behavioral competencies for physical activity participation (Okely, Booth, & Chey, 2004). Stodden and colleagues (2008) go further to suggest that motor skill competence is a primary underlying mechanism that promotes engagement in physical activity (Stodden et al., 2008). Findings are not always in support of a motor skill-physical activity relationship, however. For example, McKenzie and colleagues examined whether childhood movement skills at 4, 5, and 6 years of age predicted activity in Anglo-American and Mexican-American children at age 12 (McKenzie et al., 2002). The following movement skills were measured: agility and locomotion (lateral jumping); eye-hand coordination and manipulation (catching a ball); and stability (balancing on one foot). A 7-day recall parent interview, the Physical Activity Recall (PAR), was used to determine physical activity at age 12. Results failed to show that movement skills in children predicted later physical activity levels. However, the researchers acknowledged that their movement skill measures may have been inappropriate or insufficient in number. Despite such limitations, other researchers

have also been unsuccessful in establishing a strong association between physical activity and motor skills. Specifically, Reed, Metzker, and Phillips (2004) examined the PA-motor skill relationship in middle-school students using pedometers to estimate physical activity and three skills (passing, balance, and agility) to characterize motor skills. They also concluded that motor skills were not strongly correlated with physical activity.

Despite the findings by McKenzie and Reed, there is some evidence to suggest that a motor skill-physical activity relationship may exist, particularly when accelerometry is utilized. Wrotniak and colleagues measured physical activity using accelerometers and motor proficiency with the BOTMP in sixty-five children (Wrotniak, Epstein, Dorn, Jones, & Kondilis, 2006). Results showed that motor skill ability was positively correlated with activity counts and percentage of time spent in moderate and moderate-to-vigorous intensity physical activity. Also using accelerometers but employing the Movement Assessment Battery, Fisher and colleagues (2005) found a relationship between motor skills and activity as well (Fisher et al., 2005). In addition, Fisher suggested that the motor skill-physical activity relationship is contingent on sufficient levels of physical activity. Specifically, their results demonstrated that total physical activity and percent time spent in higher intensity activities were significantly correlated with total movement skill score, while time spent during light activity was not related to movement skills. Williams and colleagues (2008) also established a relationship between motor skills and activity intensity in children using accelerometers (Williams et al., 2008).

The preceding findings suggest that level of physical activity and motor skill ability may be related. However, there is limited data on whether this relationship exists in children with DS. Recently, we found that children with DS who learned to ride a bicycle spent more time in moderate and vigorous activity than children with DS who were unable to ride a bicycle (Ulrich, Burghardt, Lloyd, Tiernan, & Hornyak, 2008). It remains unclear whether relationships between fundamental motor skills and physical activity exist in children with DS, though. Future studies addressing this issue should measure physical activity using multiple techniques. Recently, researchers have suggested employing more than one method when assessing physical activity in children with disabilities (Cervantes & Porretta, 2010). In addition, combined accelerometer and heart rate monitors have been developed and validated in children (Corder, Brage, Warehand, & Ekelund, 2005). Such monitors are believed to provide more accurate predictions of physical activity intensity than accelerometers alone. Whether such devices are feasible and effective in assessing physical activity in children with DS should also be explored. Further insight into the activity and motor skill profiles may contribute to a better understanding of the well-documented emotional difficulties in individuals with DS.

3) Socio-emotional Domain

Proper attachment formation is an important factor in a child's willingness to explore his or her environments (Bowlby, 1969). Unfortunately, there is considerable research showing that infants and young children with DS show high rates of

disorganized and insecure attachment patterns (Ganiban, Barnett, & Cicchetti, 2000; Vaughn, Lefever, Seifer, & Barlow, 1989). For instance, van Ijzendoorn, Goldberg, Kroonenberg, and Frankel (1992) found that rates for disorganized attachment in young children with DS appear to be nearly twice that of non-DS children. Similarly, research has also indicated that children with low cognitive functioning are less frequently classified as having secure attachments compared to those that demonstrate higher cognition (Atkinson et al., 1999).

In addition to cognitive explanations, motor deficits have also been implicated to understand attachment patterns in DS. For example, Ganiban and colleagues (2000) proposed that the developmental delays resulting from motor problems and neurological dysfunction may influence early parent-child interactions and, over time, negatively affect attachment relationships (Ganiban, Barnett, and Cicchetti, 2000). Such motor-attachment relationships had been suggested prior to Ganiban's work as well. Bridges and Cicchetti (1982) argued that muted affect or ambiguous affective expression due to poor muscle tone might make it difficult for parents to accurately evaluate cues of infants with DS. In turn, they suggested, these motor difficulties could impact the establishment of synchronous, reciprocal interactions between caregiver and child and ultimately result in less desirable attachment patterns. Several studies have shown that infants with DS are less communicative and smile for shorter durations during social exchanges compared to infants with TD (Slonims & McConachie, 2006; Carvajal & Iglesias, 1997).

Regardless of the cause of attachment problems, it appears that socio-emotional difficulties in infancy continue into the toddler years for individuals with DS. In a social referencing study, Knieps, Walden, and Baxter, (1994) found significant differences between toddlers with and without DS. While toddlers without DS tended to match their parent's expressions, those with DS did not. Instead, the latter group responded with positive affect even following a fearful communication by the parent. Similar findings have been replicated in slightly older children as well. For example, Kasari and colleagues (1995) observed that children with DS engaged in significantly fewer social referencing looks than their unaffected peers. Research has also suggested that children with DS may have problems with emotion recognition. According to Kasari, Freeman, & Hughes (2001), by a developmental age of 4, children with DS perform worse than controls on tasks designed to tap their knowledge regarding simple emotions. The results of the preceding studies suggest that toddlers and young children with DS look to their caregivers less in novel or ambiguous situations. Further, they fail to use the conveyed emotions to interpret situations and shape their behaviors during the times when they do reference their caregivers.

Findings from infant and toddler studies of attachment and social communication may provide insight into subsequent behavioral problems in children with DS. Approximately one-fourth to one-third of school-aged children with DS have been identified as having significant emotional and conduct problems, with non-compliance, aggression, and hyperactivity being most prevalent (Cuskelly & Dadds, 1992; Menolascino, 1967; Myers & Pueschel, 1991). Those rates are consistent with a

parental report from Coe and colleagues (1999), which demonstrated that 1 in 3 children with DS have behavioral problems. The prevalence cited by Coe exceeded that of controls by nearly a 3 to 1 margin. The most prevalent behavioral problems reported Coe's study were conduct disorders, social withdrawal, attention disorder, and psychotic disorders (e.g., repetitive speech and preoccupation of thought). Further, it has also been suggested that children with DS may overuse social behaviors throughout the school years, such as seeking help and comfort unnecessarily(Kasari & Freeman, 2001).

Based on the above discussion, it is clear that individuals with Down syndrome exhibit a wide spectrum of socio-emotional deficits that are apparent within the first few months of life. Early deficits in areas such as attachment formation and social referencing manifest into school-aged issues with attention, aggression, and social competence. Given the problems in socio-emotional behavior coupled with issues related to motor and physical abilities, it is plausible to expect multi-domain relationships in this population.

B. Multi-domain relationships

1) Motor vs. physical domains

The focus of this section is on the relationships between the motor and physical domains. Many of the physical variables described earlier (e.g., leg strength, body composition, range of motion) have been associated with motor skills and physical activity. As described previously, Ulrich and colleagues have suggested that leg strength plays an important role in locomotor development (Ulrich, Ulrich, Angulo-Kinzler, & Yun,

2001; Ulrich, Lloyd, Tiernan, Looper, & Angulo-Barroso, 2008). Strength is also believed to be related to more advanced motor skills and athletic performance. In a review of strength training and adolescents, Webb (1990) concluded that increased strength can enhance children's performance in athletic activities where strength, power, or speed is required. For example, a variety of studies show improvements in both long jump and vertical jump following strengthening (Falk & Mor, 1996; Hetzler et al., 1995; Weltman et al., 1986). Similarly, improved sprint speed and agility have been documented after strength interventions (Lillegard, Brown, Wilson, Henderson, & Lewis, 1997; Williams, 1991).

Motor skill improvements following strengthening do not appear to be limited to children with typical development. In 2003, Blundell conducted a pilot study involving group circuit training for children ages 4-8 years with cerebral palsy (Blundell, Shepherd, Dean, & Adams, 2003). Twice per week, the children performed functionally based exercises targeting lower extremity strength, including treadmill walking, step-ups, sit-to-stands, and leg presses. Not only did the children increase muscle strength, they also displayed gains in functional performance, such as increased speed and longer strides during gait. With respect to children with DS, a case study exploring the effects exercise training was recently conducted (Lewis & Fragala-Pinkham, 2005). In the case study, a 10.5-year-old girl with DS alternated strength training and aerobic exercises five to six days per week for six weeks. Her score on the Gross Motor Scale of the BOTMP increased from 2 to 19, demonstrating improved balance, coordination, and power in gross motor tasks. Although limited, evidence appears to suggest a relationship

between motor skills and strength in children with disabilities. An association between motor skills and hypermobility is less clear, though.

There are limited and inconsistent findings to support a relationship between joint range of motion and motor skills. In a study of children 12 and under, no association was found between the amount of generalized joint hypermobility and delay in motor skills (Engelbert et al., 2005). In contrast, studies focusing on children with DS seem to support some role of joint laxity in motor skill performance. As evidence, Martin (2004) explored the effects of a flexible supramalleolar orthosis (SMO) on postural stability and motor skills in children with DS. Martin's rationale was that the SMO would control pronation secondary to joint laxity. Children in the study made significant improvements in balance, walking, running, and jumping after using the orthotic. Whether relationships exist between fundamental motor skills and hypermobility at the hip or knee in children with or without DS remains relatively unexplored. More substantial effort has been dedicated to associations between body composition and the motor domains.

There is an abundance of research examining body composition and physical activity in adults and children with TD. It is well-documented that exercise increases resting metabolic rate and provides healthy body composition changes in adults (e.g., Pratley et al., 1994). With respect to children, significant reductions in body fat in preadolescents following exercise training have been observed (Faigenbaum et al., 1993). However, Reilly (2006) failed to find a decrease in the BMI of young children

following a physical activity intervention (Reilly et al, 2006). Studies of associations between body composition and activity in populations with DS or other forms of ID are less robust but seem to suggest an important role for activity on body composition. Ordonez and Rosety-Rodriguez (2006) found that a twelve-week exercise program in adolescents with DS resulted in a reduction of fat cells. Similarly, in a recent intervention study in children and adolescents with DS, Ulrich suggested that bicycle training may positively impact BMI and body fat (Ulrich et al., 2011). However, in a large study of youth with mild ID, Frey and Chow (2006) determined that BMI did not impact motor skills. To date, there are very few studies where relationships between fundamental motor skills and body composition in children with DS have been directly examined, however.

2) Motor vs. socio-emotional domains

Despite the mixed results regarding associations between various physical factors and the motor domain, there appears to be more consensus regarding emotional benefits of physical activity. The majority of these studies have been limited to non-DS populations, though. For example, a study by O'dea (2003) found that children and adolescents with TD reported an array of social and psychological benefits from physical activity. In addition, exercise treatments have been shown to increase positive mood and decrease negative mood in 9- and 10-year-old children with TD (Williamson, Dewey, & Steinberg, 2001). Past research also demonstrates that adolescents who engage in regular physical activity and participate in sports have lower

anxiety-depression scores, display less social inhibition, and illustrate more positive emotional and behavioral attributes (Kickcaldy, Shephard, & Siefen, 2002; Donaldson & Ronan, 2006).

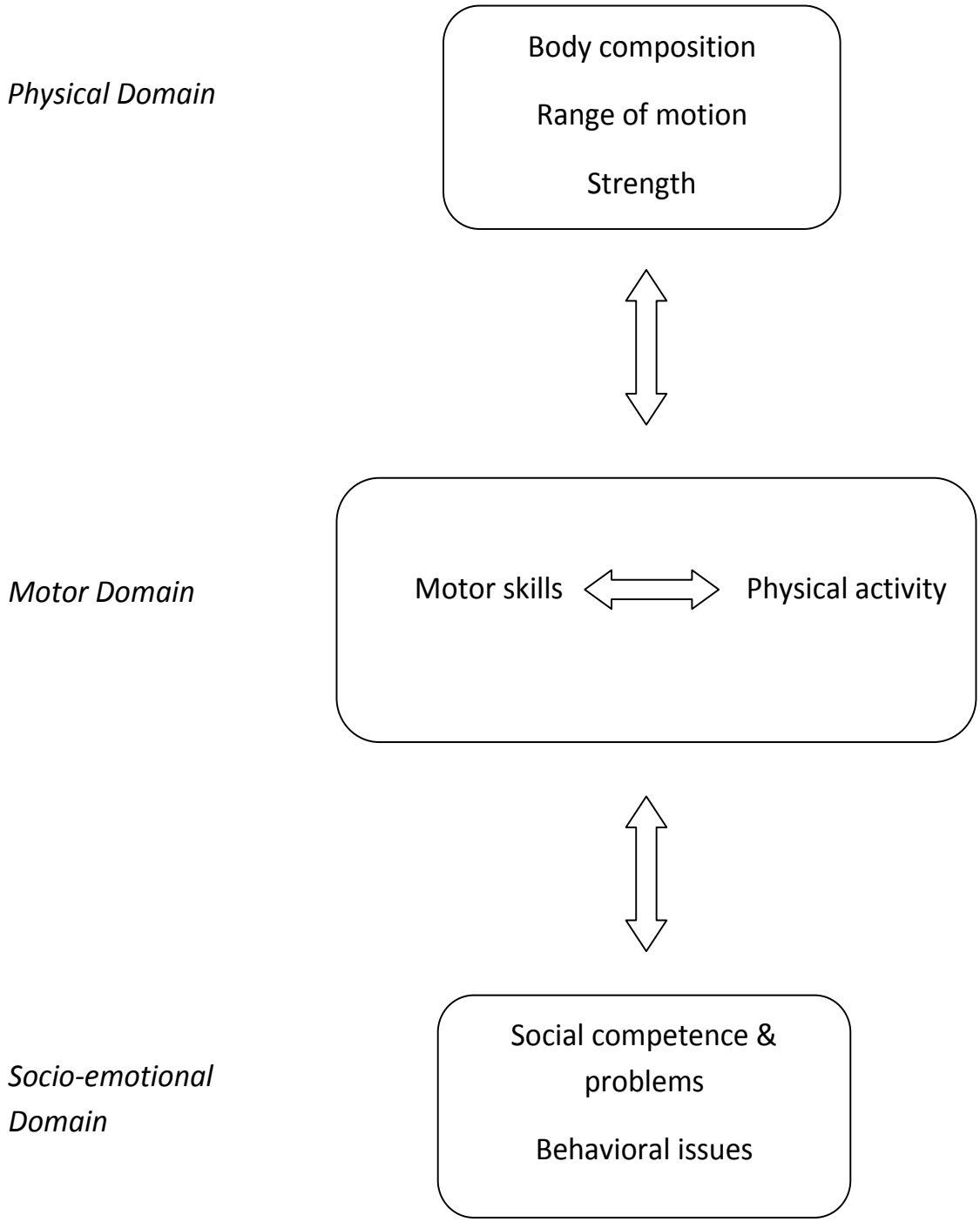
With respect to socio-emotional and motor relationships in DS, attitudinal and psychosocial outcomes of a fitness and health education program for adults have been examined (Heller, Hsieh, & Rimmer, 2004). A training group participated in a 12-week, 3 days per week, exercise and health education program. Compared to controls, the training group showed significant changes in attitudes towards exercise, including increased exercise self-efficacy, more positive expected outcomes, improved life satisfaction, and marginally lower depression. While the data in this area is limited for children with DS and other forms of intellectual disability, there appears to be some emotional gains from exercise and sport participation, namely in terms of reducing maladaptive behavior (Gencoz, 1997; Sanyer, 2006). Again, the majority of these studies focus on physical activity and participation in sport and exercise, as opposed to motor skill proficiency. Future studies should examine potential links between motor skill ability and socio-emotional health in children with and without disabilities. Namely, links between motor skills and some of the more problematic areas (e.g., social competence, aggression) for children with DS require investigation.

Purpose

The preceding review of literature described our current understanding of deficits and relationships with respect to the physical, motor, and socio-emotional domains of children with DS. While the literature strongly suggests that children with DS experience multi-domain deficits, the relationships between motor, physical, and socio-emotional characteristics remain unclear in this population. In addition, many individuals with disabilities, including those with DS, have made significant progress in recent years due to an increased public awareness of their challenges and the corresponding services now available to assist them. Further, contemporary methodological considerations may provide increased knowledge about the performance of children with and without DS, particularly in the area of physical activity. However, the utilization of such methods in special populations warrants additional exploration. Given these factors, the existence and extent of multi-domain deficits in children with DS should be reexamined comprehensively. Studies should also focus on delineating the relationships between motor ability, physical characteristics, and socio-emotional profiles. This dissertation aims to address such issues. The current studies will provide comprehensive descriptive information surrounding motor skills, physical activity, physical characteristics, and socio-emotional attributes of school-aged children with and without DS. Further, relationships between these areas will be investigated. The feasibility of using a combined heart rate monitor and accelerometer to assess physical activity in children with DS will also be explored. This dissertation will provide additional insight into the motor, physical, and socio-emotional development of

school-aged children with DS. Results will serve as a foundation for future activity-based interventions. Figure 1.1 provides a conceptual model for the studies.

Figure 1.1. Conceptual model for inter-domain relationships



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Chapter II

Examining group differences in physical and motor abilities in school-aged children with and without Down syndrome

Introduction

Down syndrome (DS) is both the oldest and most common form of intellectual disability. Recent estimates place the rate of incidence of DS at 1 in every 733 to 1 in every 850 live births in the United States (Canfield, Ramadhani, Yuskiv, & Davidoff, 2006; Shin et al., 2009). Though DS is often associated with intellectual disability (ID), developmental areas outside of cognition have also been studied. For example, significant effort has been made in the past to describe motor development in children with DS and the factors that may influence it, such as body composition, range of motion, and muscle strength (e.g. Concolino, Pasquzzi, Capalbo, Sinopoli, & Strisciuglio, 2006; Cioni et al., 1994; Connolly & Michael, 1986; Prasher, 1995; Ulrich & Ulrich, 1995). However, many of these studies are dated or have produced inconsistent findings.

Body composition in individuals with DS has received significant attention over the last twenty years. There is substantial research to indicate a high prevalence of obesity in individuals with DS (Prasher, 1995). In fact, several experts in the past have proposed that DS obesity rates should be considered a major public health concern (Rubin, Rimmer, Chicoine, Braddock, & McGuire, 1998). The majority of studies in this

area concentrate on adults with DS, however. Research related to obesity in children with DS is less conclusive. For example, Sharav and Bowman (1992) examined thirty pairs of siblings between the ages of 2 and 14 years, each pair consisting of one child with DS. Results revealed no differences in BMI between the children with and without DS. The findings were inconsistent with those from a more recent sibling study on DS (Whitt-Glover, O'Neill, and Stettler, 2006) but in agreement with Luke (1996) who observed no differences in fat-free mass (FFM) values for children with and without DS (Luke, Sutton, Schoeller, & Roizen, 1996). Therefore, it appears that the literature is currently divided with regard to body composition and obesity in children with DS.

Unlike body composition, there is a strong consensus surrounding musculo-skeletal abnormalities present in children with DS. As evidence, Concolino (2006) performed orthopedic evaluations of children aged 4-10 years with and without DS (Concolino, Pasquzzi, Capalbo, Sinopoli, & Strisciuglio, 2006). Results showed that all fifty children with DS showed joint laxity, with 20% demonstrating severe laxity. In contrast, 80% of the children without DS demonstrated no laxity at all. Previous studies have also compared joint range of motions between children with DS and those with other forms of ID. Findings indicated that children with DS showed greater range of motion for hip flexion and abduction (Angelopoulou, Tsimaris, Chirstoulas, & Mandroukas, 1999). Difficulties with lower extremity hypermobility are not limited to the hip. Research has shown that individuals with DS are susceptible to patellar instability and disorders of the ankle, such as planus (Mik, Gholve, Scher, Widmann, & Green, 2008).

The presence of musculo-skeletal abnormalities may contribute to observed strength deficiencies in children with DS. For example, Mercer and Lewis (2001) found that children with DS demonstrated lower isometric hip abductor and knee extensor strength. Lower extremity strength deficiencies in DS have not been limited to isometric measures, though. Cioni found that isokinetic knee extensor strength was lower in children and adolescents with DS when compared to values in both children with TD and with non-specific ID (Cioni et al., 1994). Cioni's study also indicated that by the age of 14 years, individuals with DS failed to demonstrate gains in muscle strength that typically accompany adolescence. However, to my knowledge there have not been any studies of isokinetic leg strength in children with DS since Cioni's 1994 study.

Additional knowledge of leg strength in children with DS could be important for understanding motor development in children with DS (Ulrich & Ulrich, 1995). There is conclusive evidence that infants and toddlers with DS experience locomotor delays and gait abnormalities (Henderson, 1986; Ulrich & Ulrich, 1995; Ulrich, Ulrich, Angulo-Kinzler, & Yun, 2001; Ulrich, Lloyd, Tiernan, Looper, and Angulo-Barroso, 2008; Lydic & Steel, 1979; Parker, Bronks, & Snyder, 1986; Looper, Wu, Angulo-Barroso, Ulrich, & Ulrich, 2006). Less conclusive results have been documented for older children with DS. For example, Connolly & Michael (1986) found that children with DS score lower on functional motor tasks, including running speed, balance, strength, and visuo-motor control. However, results from Jobling (1998) indicated that children with DS were highly variable in their motor skill performances depending which skills were tested. For example, skills with a high balance component and items involving upper-limb

coordination were highly problematic for children with DS. On the other hand, running speed and agility were reported as being age-appropriate in children with DS, contrasting results from Connolly & Michael (1986). The majority of studies examining gross motor skills in children with DS have used the BOTMP or the Movement ABC assessment. There is limited research assessing gross motor skill ability in children with DS using the Test of Gross Motor Development-2nd Edition (TGMD-2) (Ulrich, 2000), which is widely used in school settings and in research with a variety of other pediatric populations.

Similar to the physical activity literature, studies regarding motor abilities in children with DS are inconsistent despite strong evidence for deficiencies in infancy. For instance, McKay and Angulo-Barroso (2006) reported that infants with DS spend more time in lower intensity activities relative to their peers. When older children with DS have been examined, however, the results have been mixed and largely dependent on the measure used. Sharav and Bowman (1992) compared the physical activity of children with DS to that of their siblings using a questionnaire and found that the children with DS were less active and showed a preference for indoor activities. In a sibling study using accelerometers, Whitt-Glover and colleagues (2006) demonstrated that children with DS (ages 3-10 years) accumulated less vigorous activity than their siblings and performed these activities for shorter durations. Children with and without DS in the Whitt-Glover study were found to spend similar times in moderate and lower intensity activities, however. Due to such discrepancies, researchers have recently

suggested that more than one method be employed when assessing physical activity in children with disabilities (Cervantes & Porretta, 2010).

The preceding discussion illustrates that despite widespread belief that children with DS experience significant challenges in the physical and motor domains the literature remains relatively inconclusive. Therefore, the primary objective of the current study was to utilize contemporary methodological considerations to comprehensively describe and compare the physical characteristics, motor abilities, and physical activity levels of school-aged children with and without DS. The following hypotheses were tested. First, children with DS will demonstrate decreased knee strength and increased ranges of motion compared to their unaffected peers. However, no group differences in body composition will be observed. Second, children with DS will show decreased locomotor and object control proficiency in relation to children without DS. Finally, children in the DS group will demonstrate less intense physical activity compared to the TD group. A secondary objective of the study was to explore the feasibility of using a combined heart rate monitor and accelerometer to assess physical activity in children with DS. Such devices have been validated and successfully used in children with typical development (Corder, Brage, Warehand, & Ekelund, 2005).

Methods

Participants

The sample consisted of twenty children with DS and twenty children with typical development (TD) who were matched on age and gender. Participants with DS were recruited from parent support groups in Michigan and northern Ohio. The TD sample was largely recruited from families and friends of the children with DS. A local summer camp and a University of Michigan research website were also utilized as secondary recruitment tools. Exclusion criteria for both groups included major health conditions that would be counter to mild exercise (e.g., cardiac problems, uncontrolled seizures). Neither race nor gender prohibited children from participating in the study as long as they met study criteria. However, willing volunteers with TD who did not match one of the children with DS on age or gender were placed on a waiting list until an appropriate match was found. If a DS match was not identified those volunteers were unable to participate.

Procedures

All children came to the motor development laboratory at the University of Michigan for testing. Prior to the visit, parents were called and given instructions with regard to proper attire and dietary restrictions. Upon arrival at the laboratory, informed consent and child assent (when appropriate) was obtained and documented. Parents were also asked to fill out a supplemental information sheet prior to child testing to gather socio-demographic information. Participating families received $20. All procedures were approved by the University of Michigan Medical Internal Review Board. The following assessments were conducted in the order presented below.

A) Body composition

Height and weight were assessed using a combined stadiometer/scale. Participants wore minimal clothing (t-shirts and shorts, socks) and no shoes for the weight and height measurements. For each measurement, children were instructed to stand still in the center of the scale platform with their hands at their sides. Participants were also instructed to stand up "tall" and to "look straight ahead." Some of the children had difficulty looking straight ahead and required additional assistance, such as being asked to look at a sticker or book that was being held in front of their face. Measurements were taken twice to ensure accuracy.

Body composition was assessed using bio-electrical impedance (BIA). BIA has been employed previously in children with DS (Luke, Sutton, Schoeller, & Roizen, 1996). The equipment implemented for this assessment was the Bio-Impedance Spectrum Analyzer (Hydra ECF/ICF, Model 4200). Prior to coming to the laboratory, parents were informed of conventional dietary and exercise restrictions to maximize the validity of BIA testing. Specifically, parents were instructed that their children should not exercise or eat for at least four hours prior to the assessment. However, children were allowed to drink a small amount of water if desired. Immediately following BIA testing, children were permitted to eat a small snack.

In preparation for the BIA testing, children were asked to lie in a tetrapolar configuration (supine with hands and legs slightly abducted) and to minimize their movements for ten minutes. During this time, the examiner measured the child's resting heart rate (RHR). Also during this period, rubbing alcohol was used to clean skin

areas on the hands and feet. Two electrodes were then placed on the cleaned and dried skin surfaces. Once the Hydra device was turned on, the child's height, weight, and gender was entered. The BIA procedure involved application of an electrical current (500 micro amps at a frequency of 50 kHz) across electrodes on the hands and feet of participant. Intracellular fluid (ICF), extracellular fluid (ECF), and total body water (TBW) were calculated by the analyzer in approximately one to two minutes, depending on the child's ability to remain still. To decrease the child's movements during both the BIA pre-testing and testing periods, books were available for the parents to read to their children.

B) Range of motion (ROM)

Passive ROM at the ankle, knee, and hip were assessed on the right side of the body using a standard goniometer. Measurements of dorsi-flexion, plantar flexion, knee hyperextension, and hip abduction were performed in accordance with joint measurement procedures in Norkin and White (1995). Ranges of motion at the ankle and knee were measured with the participant seated at the edge of an elevated table. Hip abduction was assessed with the child lying in a supine position. For hip abduction, parents were occasionally asked to assist with stabilizing the non-measured leg if the child had difficulty remaining still. Two measurements were made for each joint movement with the average value used in analysis.

C) Gross motor skills

The Test of Gross Motor Development (2nd ed.) (TGMD-2) (Ulrich, 2000) was used to assess gross motor skill ability of the participants. The TGMD-2 measures

locomotor and object control capabilities in children ages 3 to 10 years. The items of the locomotor subtest include running, galloping, hopping, leaping, horizontal jumping, and sliding. The object control subtest consists of striking a stationary ball, stationary dribbling, catching, kicking, overhand throwing, and underhand rolling. High test-retest reliability for the TGMD-2 has been established, with coefficients ranging from 0.86-0.94 for the age groups included in the current study (Ulrich, 2000).

Due to scheduling conflicts, the TGMD-2 was conducted in the motor development laboratory rather than a gymnasium, as recommended in the manual. As a result, some of the tasks were modified to accommodate the space constraints. For example, children were instructed to throw and kick the ball towards specific targets on the wall. The procedures complied with instructions provided by the TGMD-2 manual as much as possible given space limitations. Accurate demonstrations and verbal descriptions of the skills were provided to the participants. Each child was allowed a practice session in attempt to ensure that he or she understood the task. Two test trials were administered and scored based on the performance criterion set forth in the manual. All trials were videotaped. In addition to the standard TGMD-2 skills, participants were also measured on a timed run of 40 feet to obtain a measure of peak performance. Children were instructed to run as fast as they could. Two trials of the run were conducted.

D) Leg strength

Bilateral isokinetic knee flexor and extensor strength was measured using a Cybex 340 dynamometer (Cybex, New York, NY) with HUMAC (2004). This model

dynamometer has been used to assess isokinetic strength in individuals with DS previously (Pitetti, Climstein, Mays, & Barrett, 1992; Croce, Pitotti, Horvat, & Miller, 1996). Prior to testing, a researcher demonstrated the leg movements on the dynamometer. Pillows were placed behind the children when necessary to provide proper lumbar support. Subjects performed three practice sessions per movement (knee flexion, knee extension) per leg at a speed of 30 degrees per second. Following several minutes of rest, they performed three maximal efforts per movement per leg at a speed of 30 degrees per second. The children were instructed to push against the lever arm as powerfully and rapidly as possible following a verbal prompt. Throughout each trial, verbal encouragement was provided by the researcher and family members. One minute of rest was given between trials. The order of limb testing was randomized for each child to help minimize learning effects.

E) *Physical activity*

Physical activity was measured using two methods, a questionnaire and a combined heart rate monitor and accelerometer. The two measures provide different information about the quality and quantity of the participants' activity. While the questionnaire focuses on different dimensions of the children's participation in recreational and leisure activities, the accelerometer and heart rate monitor provides detailed information about the movement counts and heart rate of children throughout the day.

Children's Assessment of Participation and Enjoyment (CAPE)

The Children's Assessment of Participation and Enjoyment (CAPE) is a 55-item questionnaire that has been validated for individuals with and without disabilities between the ages of 6 and 21 years (King et al., 2007). It assesses 5 dimensions of participation: diversity, intensity, with whom, where, and enjoyment. The CAPE manual recommends that children fill out the measure themselves. However, anecdotal evidence from our previous projects suggests that children with DS significantly over-report their participation and enjoyment of activities. Therefore, parents of participants in the current study were asked to fill out the CAPE with their children. The completed CAPE was mailed back to the researchers in a self-addressed, postage-paid envelope.

Actiheart

The Actiheart (CamNtech, United Kingdom) was used in an exploratory manner to determine the feasibility of its use for children with DS. The device is a combined heart rate monitor and accelerometer that has been validated in children and used to measure physical activity intensity (PAI) (Corder, Brage, Warehand, & Ekelund, 2005; Brage, Brage, Franks, Ekelund, & Wareham, 2005; Parish, Rudisill, & St. Onge, 2007). The Actiheart samples at 32 Hz and measures activity counts (AC) and heart rate (HR). Both AC and HR are utilized to estimate PAI, allowing for more accurate estimations than the devices which only utilize activity counts or heart rate data (see analysis section for detailed explanation of the prediction equations).

The Actiheart monitor was attached to the chest of each child with two biocompatible electrocardiograph (ECG) electrodes. Electrodes were placed on both sides of the heart parallel to the space between the fourth and fifth ribs. Each

attachment site was cleaned with alcohol and dried prior to placement of the device. Reviews of pediatric activity literature suggest that accelerometers should be worn for a minimum of 4 days, with at least one day being a weekend day (Trost, McIver, and Pate, 2005). As a result, children were instructed to wear the device for 4 days. Further, all participants were scheduled in a manner to ensure they would wear the Actiheart on at least one weekend day. Families were sent home with an activity monitor log, which was used to document any times when the monitor was removed either intentionally (e.g. swimming, baths) or accidentally. The Actiheart device and activity log were returned via mail with the CAPE following the four-day assessment period.

Data Analysis

A. Data reduction

The socio-demographic variables of age, gender, parental education, number of siblings, and family income were included in analysis. With respect to physical variables, height and weight were used to determine body mass index (BMI) using the equation BMI= weight(kg)/height^2(m). In addition to BMI, measures of body composition were also calculated from the BIA output. Specifically, fat-free mass (FFM)(kg) was determined by dividing TBW (L) by specific hydrations constants based on the subject's height, age, and gender (Goran et al., 1993). The resulting equation was:

$$\text{FFM (kg)} = \frac{\text{TBW}}{0.769-(0.0025 \times \text{age}) - (0.019 \times \text{gender})}$$

Where age was entered in years and gender = "0" for females and "1" for males. Fat mass (FM) was then calculated by subtracting FFM from body weight. As a result, we

were able to calculate percent body fat by dividing FM by total body mass and multiplying by 100.

Measures of joint range of motion and strength were also used to characterize the physical characteristics of the sample. Mean ROM values for plantar flexion, dorsiflexion, knee hyperextension, and hip abduction were calculated across two trials. Isokinetic strength values were adjusted for body mass, using ratio standards (Davies & Dalsk, 1997; Sundegardh et al., 1988). Of the three trials, bilateral peak knee flexor and extensor strength (N·m/kg) were included in analysis.

Dependent variables of the motor domain included the raw scores of the locomotor and object control skills sections of the TGMD-2 along with scores on the individual items. Peak run time over the two trials was also included. Physical activity was represented in the dataset by variables from both the CAPE and Actiheart. For the CAPE, scores for 5 dimensions of participation (diversity, intensity, who, where, and enjoyment) were analyzed.

Preparing dependent variables from the Actiheart was a multi-step process. Once the monitors were returned to the laboratory, the data was downloaded using the Actiheart software. The software provided minute by minute output for activity counts (AC) and heart rate (HR) over the 4 days. The output was cleaned manually to remove any data points when the monitor was not worn and to omit outliers. Research has shown that maximal heart rate in children is stable up to 200 beats per minute (Rowland, 1993; Epstein, Paluch, Kalakanis, Goldfield, Cerny, & Roemich, 2001). Therefore, data points containing HR values above 200 beats per minute were removed

manually. The amount of valid data over the 4 days and the average number of missing data per day were calculated and recorded.

The cleaned Actiheart data was entered into a customized program developed by CamNtech to estimate physical activity energy expenditure (PAEE) (see Figure 2.1). The program is based on a branched equation model by Corder and colleagues (2005) who used a step-test to determine thresholds and parameters for children. In the model, child equations for HR and AC are extrapolated to go through sleeping heart rate and 0 counts per minute, respectively. Twenty-five counts per minute serves as the initial threshold in the model. For counts above 25, decisions are then made based on the transition heart rate, which was determined to be the highest walking and slowest jogging heart rate values above sleep during the step test. If counts are below 25 per minute, the second decision in the flow chart is based upon the flex heart rate. Flex heart rate is defined as the average above sleep of the highest heart rate during rest and the lowest heart rate during incremental exercise for children during treadmill testing. Depending on the outcomes in step 2, data is ultimately entered into one of four equations. These equations weight activity and heart rate data in varying proportions to predict PAEE in kJ/kg/min. In the current study, PAEE was converted to kcal/kg/min by dividing the value by 4.18. The conversion was performed in order to apply and interpret intensity cutpoints (see below). Due to the fact that some of the data points were removed, PAEE was totaled for each day and divided by the number of valid minutes per day. These values were then averaged by the number of days

included in analysis. Higher values for PAEE correspond to increased energy expenditure and, thus, more intense physical activity.

PAI was also operationalized by applying child-based, energy expenditure cutpoints to the PAEE values. The cutpoints, developed by Puyau, were as follows: sedentary (SEDPA) (< 0.015 kcal/kg/min), light (LPA) (≥ 0.015 and < 0.05 kcal/kg/min), and moderate to vigorous (MVPA) (≥ 0.05 kcal/kg/min) (Puyau, Adolph, Vohra, & Butte, 2002). Pediatric physical activity literature recommends using a combined MVPA category as opposed to separate moderate and vigorous categories, particularly when longer epoch lengths are utilized (Reilly et al., 2004). Each 60-second data point was assigned to an intensity category and summed each day. Percentages of time spent in SEDPA, LPA, and MVPA were calculated by dividing the daily totals per category by the number of valid 60-s data points per day. Daily averages were then calculated.

As a result of aforementioned procedures, three indicators of physical activity intensity were present in the current study: 1) intensity of participation from the CAPE; 2) PAEE using branched equation modeling; and 3) percentage of time spent in various intensities. Of note, methods 2) and 3) utilize equations and cutpoints that have not been validated in children with DS. Therefore, data using these methods should be viewed as exploratory at this time.

B. Statistics

Descriptive analysis by group (DS, TD) was conducted on the socio-demographic variables to characterize the sample. T-tests and chi-square statistics on socio-demographic data were also performed to determine if the groups differed on any of

these variables. Descriptive statistics and group comparisons using t-tests were also conducted on the physical, motor skill, and activity-related variables to address the hypotheses. In addition, effect sizes for the dependent variables were calculated and interpreted according to Cohen (1988) (0.2=small, 0.5=medium, 0.8=large). Alpha was set at $p<0.05$ for all analyses.

Results

Sample characteristics

Forty children (DS=20, TD=20) between the ages of 6 and 10 years were included in the study. The participants were successfully matched on age and gender, with both groups consisting of 4 girls and 16 boys and reporting an average age of 7.9 years. There were no significant differences between the groups on any of the socio-demographic variables. Children in both groups had between two and three siblings, on average. Chi-square statistics indicated no significant differences on parental education or family income, although families of children with DS reported slightly lower education but higher income compared to families without DS. Table 2.1 demonstrates the sample's socio-demographic characteristics.

Physical Domain

Children with DS demonstrated less desirable physical characteristics compared to children without DS (Table 2). Although the groups did not differ significantly on weight, the children with DS were shorter ($t(38)=-5.77$, $p<.001$) and demonstrated greater BMI scores ($t(38)=2.16$, $p=.037$), on average. Bioelectrical impedance analysis

also revealed group differences in body composition. Children with DS had less total body water ($t(32)=-2.79$, $p=.009$) and FFM ($t(32)=-2.71$, $p=.011$). As a result, significant differences were observed for percent body fat ($t(32)=3.10$, $p=.004$) with the DS group having a mean value of 20.6% compared to the TD group who had 12.9%. Consistent with expectations, children with DS also demonstrated significantly higher passive ranges of motion for sagittal plane ankle motion (dorsiflexion, $p=.002$; plantar flexion, $p=.006$), knee hyperextension ($p<.001$), and hip abduction ($p=.011$). In terms of strength, children with TD outperformed their peers with DS by greater than a 2:1 margin on bilateral knee flexor and extensor strength.

Motor Domain

A) Motor skills

Results from the TGMD-2 revealed that the DS group experienced significant gross motor skill deficits in relation to the TD group (Table 3). Mean scores for the locomotor subscale were twice as large for the children with TD compared to children with DS ($t(37)=-8.67$, $p<.001$). Similarly, the TD group nearly doubled the mean score of the DS group on the object control subscale ($t(38)=-6.24$, $p<.001$). Consistent with the subscale scores, children with DS scored significantly lower than children with TD on all items of the TGMD-2. Significant differences were also observed on the 40-foot timed run ($t(37)=7.77$, $p<.001$), evidenced by the fact that children with DS completed the run two seconds slower, on average. Of note, the DS group demonstrated considerably more variability in their motor performance as well. For example, standard deviations

for the DS group exceeded those of the TD group by a 3:1 margin for both the locomotor and object control subscales.

B) Physical activity

Results comparing the activity of the two groups are shown in Table 4. Findings from the CAPE indicated that children with DS experienced significantly less diversity in their activities compared to children with TD ($t(38)=-2.21$, $p=.033$). The groups did not differ on any of the other dimensions, however, including intensity. There was a moderate effect (Cohen's $d = 0.55$) for the Who dimension in the direction favoring children with TD. The difference was not statistically significant, though.

In terms of the Actiheart data, both groups ended up with between 3 and 4 days of valid data after cleaning. The children with DS had approximately a half a day less of data, on average, than the TD group but this difference was not statistically meaningful. Surprisingly, the groups were comparable with respect to physical activity intensity, as indicated by both PAEE and percent of time spent in MVPA. In fact, a scatter plot of the PAEE values (Figure 2) demonstrates that five of the six highest scores came from children with DS. The lone group difference in Actiheart data was observed on percent of time spent in LPA ($t(37)=2.403$, $p=.021$), with DS group spending approximately 36 percent of their day in LPA compared to 28% by the TD group.

Discussion

The current study was an attempt to comprehensively describe and contrast the physical and motor characteristics of school-aged children with and without DS matched

on age and gender. The primary objective was to reexamine previous questions while exploring relatively novel measurement techniques for assessing physical and motor abilities of school-aged children with DS. Largely consistent with expectations, our results indicated that children with DS had less desirable physical profiles and poorer gross motor skill abilities compared to their unaffected peers. Several interesting findings emerged upon examining the physical activity profiles of the two groups. First, physical activity levels of children with and without DS were comparable but the DS group was less diverse in their activities. Second, some children with DS may exhibit hyperactive behavior as part of their phenotype. Finally, while more research is necessary to assess the validity of Actiheart data in children, particularly for children with DS, the device is tolerated relatively well in this population and appears to provide reasonable group comparisons for PAEE.

Perhaps the most compelling findings in the current study involved physical activity intensity in the children with DS. While children with DS showed less diversity in their recreational and leisure activities, CAPE data indicated that they were similar to their peers with respect to the intensity for which they performed these activities. Our CAPE scores on diversity are slightly different than results from a recent study in Taiwan on participation in adolescents with DS (Wuang and Chwen-Yng, 2012). These researchers reported mean diversity scores on the CAPE to be 27.83, compared to our findings of 31.30. This minor discrepancy is not surprising, though, given numerous reports of declines in physical activity among adolescents (e.g., Kemper, Post, Twisk, & Van Mechelen, 1999). Interestingly, our Actiheart data produced similar findings to our

CAPE data with respect to intensity between the two groups. Children with DS spent more time than the TD group in LPA, a finding previously observed in infants with DS (McKay & Angulo-Barroso, 2006). However, we observed no group differences in percent time spent in MVPA or in PAEE. These results are somewhat inconsistent with prior studies that used accelerometers to assess physical activity in children with DS. For example, Whitt-Glover and colleagues demonstrated that children with DS accumulated less vigorous activity per day than their unaffected siblings (Whitt-Glover et al., 2006). They did not observe differences in moderate activity. It is important to note that they used a different accelerometer than what we employed in the current study. In addition, Whitt-Glover applied intensity cutpoints that were based on movement counts alone. Research indicates that accelerometry by itself is unable to account for increases in intensity that may correspond to activities like carrying a load or cycling (Corder et al., 2005). Therefore, the physical activity literature suggests using both HR and movement counts to get more accurate estimates of activity intensity (Brage, Brage, Franks, Ekelund, & Wareham, 2005; Haskell, Yee, Evans, & Irby, 1993; Corder et al., 2005). Further, Reilly has shown that applying various accepted intensity cutpoints to the same data results in statistically different results for SEDPA and MVPA (Reilly, Penpraze, Hislop, Davies, Grant, & Paton, 2008). Therefore, the discrepant findings between the current study and Whitt-Glover's can be attributed primarily to methodological differences. This reinforces the need for standardized measurement protocols when assessing physical activity.

Given previous physical activity results comparing children with and without DS, we expected to find group differences favoring the TD group for %MVPA and PAEE. There are several potential reasons for our unexpected Actiheart results. The first involves measurement and validity issues with the Actiheart in children with DS. When the device was placed on the children with DS, many of them were initially fearful and reluctant to wear it. In some cases, it took the parents several hours after they got home to get the child comfortable with the device. Further, some children took off the device on their own at various points during the 4-day assessment period. As a result, we had to remove this data from analysis and ended up with less data than we expected. Future studies should account for missing data when determining appropriate measurement durations.

Another methodological issue to consider is the fact that we may not have accurately captured physical activity intensity for children with DS. Neither the intensity cutpoints we applied nor the equations in the branched model for PAEE, both of which rely heavily on heart rate data, have been validated for children with DS. Our results demonstrated no differences in RHR between the two groups. However, there is evidence to suggest that individuals with DS may show increased heart rate responses when exercising, with peak heart rates in youth 30-35 beats per minutes lower than in individuals without DS (Varela & Pitetti, 1995; Fernhall et al., 1997). Therefore, it is possible that children with DS were actually performing tasks with a workload equivalent to light activity but, due to their elevated heart rates, the activity was classified as moderate to vigorous. If true, the current cutpoints and equations may

have overestimated intensity for the DS group. Recently, there has been concern that the Actiheart may also overestimate activity intensity in children with typical development during free-living activities (Moore, Pfeiffer, Aubrey, Vielbig, Peyer, & Trost, 2011). Although future research on pediatric activity equations and cutpoints is necessary, we believe the Actiheart provided a reasonable group comparison for PAEE. We are more cautious in interpreting the data for time spent in various intensities, however. MVPA, for example, was determined by applying cutpoints that have not been validated in DS to our PAEE data. Further, the branched model to determine PAEE also has not been validated in DS. Therefore, the intensity category data has two potential sources of validation error. As a result, our Actiheart data should be viewed as exploratory at this time and interpreted with these validity issues in mind, particularly for time spent in various intensities.

While the measurement errors may partially contribute to our activity results, we believe that alternative explanations are equally plausible. First, we must consider the possibility that children with and without DS are truly similar with respect to physical activity intensity when considering at least moderate activity. This argument is partially supported by our CAPE findings where we observed no group differences on the intensity dimension. It is important to note, however, that the intensity dimension on the CAPE measures frequency of participation as opposed to intensity defined by increased workload or energy expenditure. Therefore, we must be cautious in comparing the intensity variables from the CAPE and Actiheart. Another potential explanation for our activity results involves the behavioral characteristics of children

with DS. Specifically, a significant subsample of children with DS may demonstrate hyperactivity. If true, high levels of activity in these individuals could partially offset lower activity levels in the remaining individuals. In fact, examination of the individual Actiheart data revealed that five out of the top six PAEE scores belonged to children with DS. These children did not demonstrate similarly high levels of intensity on the CAPE. Anecdotally, these five children were also observed to be "lower" functioning in terms of their cognition, motor, and physical abilities and displayed high levels of hyperactivity during their visit to our laboratory. Increased levels of hyperactivity have been reported previously in children with DS (Dykens, 1997) and observed in DS mouse models (Altafaj et al., 2001; Galante et al., 2009). Further, rates of co-morbidity for DS and hyperactivity disorders range from 6% to over 50% (Myers & Pueschel, 1991; Gath & Gumley, 1986; Capone, Goyal, Ares, & Lannigan, 2006). Taken together, one must consider the possibility that children with and without DS are similarly active, on average, in terms of participation in leisure activities and overall physical activity. The latter may result from increased levels of hyperactivity in some children with DS. Additional research is needed to support these claims, however.

While our activity results were not expected, findings on the physical characteristics of children with DS were relatively consistent with previous literature. The lone exception was body composition, where past results have been inconclusive. Findings from the current study suggest that school-aged children with DS have increased BMI and body fat values compared to children with TD. Our results on BMI were remarkably similar to those from Whitt-Glover and colleagues (2006) who also

observed BMI of children with DS to be approximately 18 kg/m^2 compared to 16 kg/m^2 for children with TD. Findings in the current study on body composition using BIA differed from Luke et al. (1996), however. While the FFM values for children with TD in Luke's study were comparable to the TD group in the current study (23.2 kg vs. 23.6 kg), our sample of children with DS had substantially lower FFM values (19.6 kg vs. 22.5 kg). The discrepancy between the DS FFM findings is likely the result of sampling differences. Both studies included similar age ranges but the current study had twice as many participants as Luke's study. Further, Luke specifically recruited children with a range of body sizes in both groups, which could explain why both groups demonstrated similar FFM values. It is plausible to suggest that Luke's result would look similar to ours had they randomly selected children with DS irrespective of body shape.

As expected, our findings indicated that children with DS demonstrated increased lower extremity range of motion compared to children with TD. They also exhibited significant deficiencies in terms of leg strength. Several potential mechanisms for strength deficiencies in DS have been proposed. One possibility is that a general "hypofunctioning" of the neuromuscular system might be responsible for decreased force output in children with DS (Cioni et al., 1994). Cioni (1994) suggested that individuals with DS may exhibit a basic defect of pyramidal system, which could impact descending signals to the muscles. The authors arrived at a neuromuscular explanation after determining that children with DS fail to show typical strength gains coinciding with puberty despite demonstrating relatively normal ages for puberty and the appropriate hormonal increases.

Research involving mouse models suggest that dysfunction at the level of the muscle itself may account of strength deficiencies in DS. For example, myocardial cells from mouse fetuses with trisomy 16, a mouse model of DS, displayed altered action potentials (Orozco-Beunrostro, Godinez-Rodriguez, Winking, Arguello, & Torres, 2001). The authors argued that calcium influx may be decreased in such models, thereby impairing the contractibility of the muscle. A study by Rothermel and colleagues (2000) examining proteins in skeletal muscle also suggests that muscular dysfunction contributes to strength deficits in DS (Rothermal, Vega, Yang, Bassel-Duby, & Williams, 2000). Specifically, DSCR1, a gene associated with DS, encodes the protein MCIP1 (myocyte-enriched calcineurin interacting protein). The interaction of MCIP1 and calcineurin appears to alter calcineurin-dependent pathways. These pathways are believed to be responsible for hypertrophy and gene expression in striated muscle (Rothermal, et al., 2000). Our results on hypermobility and strength, in conjunction with previous findings, lead us to conclude that joint laxity, neuromotor control, and problems with contractility and hypertrophy constrain strength in children with DS.

Given the issues with joint laxity and muscle strength in children with DS, we were not surprised to also observe motor skill deficiencies despite an inconclusive body of literature. There appears to be consensus around poor motor performance on upper-limb motor skills in children with DS but some researchers have suggested that other skills, such as running and tasks of agility, may be age-appropriate in DS (Jobling, 1998). Our results are in agreement with more popular beliefs of a widespread deficit in gross motor skills (e.g., Connolly & Michael, 1986). We found that children with DS scored

substantially lower than the TD group on all items of the TGMD-2, which includes both locomotor and object control tasks. In addition, children with DS were extremely variable in their motor performances.

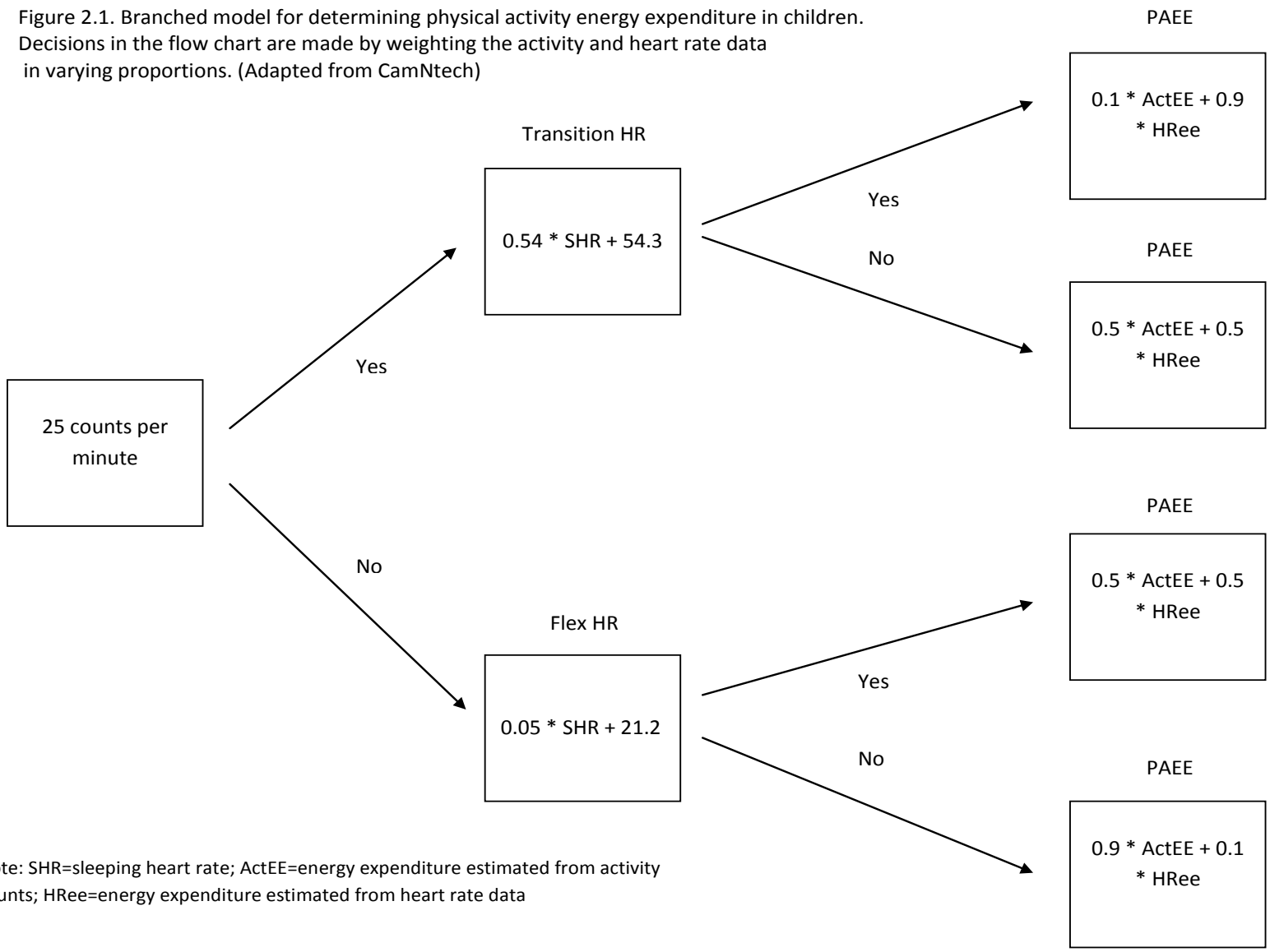
Results from our TGMD-2 assessments could have important implications for research and practice in adapted physical education. First, despite the popularity of the TGMD-2 in school settings, there is surprisingly limited research using this tool in children with DS. The current study may serve as a reference for teachers conducting TGMD-2 assessments in children with DS. Second, results suggest that significant effort should be devoted to improving fundamental motor skills in children with DS given the important role childhood motor skills may play in adolescent fitness (Barnett, Van Beurden, Morgan, Brooks, & Beard, 2008) and physical activity participation (Okely, Booth, & Chey, 2004). We were disappointed to observe such a wide gap in the gross motor skill abilities of children with and without DS considering that the Individuals with Disabilities Education Act (IDEA) has been in place since 1975. Our results suggest that elementary schools are not placing enough emphasis on gross motor skill instruction for children with DS. Therefore, parents should advocate for changes to adapted physical education programs to help foster these skills in their children. Similarly, our results indicated large degrees of variability in the gross motor skills of children with DS. This finding indicates that motor skill instruction should be highly individualized for children with DS. Given our findings on hyperactivity, educators should also be cognizant of the fact that some children with DS may display behaviors consistent with children who have ADHD or other hyperactivity disorders. This may have implications for instruction

as well since children with ADHD have been found to perform below average in the areas of gross motor skills and physical fitness (Harvey & Reid, 1997).

Collectively, results from the current study suggest that children with DS may experience wide-ranging deficits in their physical characteristics and motor skill abilities. Comparisons of physical activity and participation in recreational and leisure activities between children with and without DS remains less clear due to potential validity issues with the Actiheart, confounds of hyperactivity, and a limited sample. For example, we only observed significant group differences in participation on the diversity dimension. However, there was also a meaningful group difference on the who dimension as indicated by effect size, which could indicate that children with DS participate in activities with fewer individuals compared to children with TD. Perhaps this difference would have reached statistical significance in a larger sample. If so, there could be implications for increasing social participation in children with DS. In addition to sample size, several other limitations need to be acknowledged. Specifically, the study was advertised as a fitness and sports-related study. Therefore, many of the children with TD who volunteered may have been uniquely interested in sports and fitness and, therefore, had greater ability in these areas than randomly selected members of the population. This may have contributed to the large strength and motor skill differences between groups. Further, a cognitive assessment was not included. It is plausible that the observed motor skill, and possibly strength, differences may have been smaller had we controlled for cognition. With respect to physical activity, the Actiheart device needs to be validated for children with DS before we can confidently claim that children with

and without DS are similar with respect to their physical activity levels. Likewise, objective measures of participation in recreational and leisure activities are needed for children with DS. It is critical to determine if children with DS who have sufficient levels of motor skills are utilizing these skills during recreational activities. Therefore, researchers or teachers in the area of physical education should periodically assess the frequency of participation for students with DS once gross motor skill competency is achieved. Future research should also work towards identifying correlates to motor skill performance and physical activity in school-aged children with and without DS.

Figure 2.1. Branched model for determining physical activity energy expenditure in children. Decisions in the flow chart are made by weighting the activity and heart rate data in varying proportions. (Adapted from CamNtech)



Note: SHR=sleeping heart rate; ActEE=energy expenditure estimated from activity counts; HRee=energy expenditure estimated from heart rate data

Table 2.1. Participant characteristics

	DS	TD
Gender	4 Females, 16 Males	4 Females, 16 Males
Age, $\bar{x}$ (sd)	7.94 (1.25)	7.94 (1.57)
Number of siblings, $\bar{x}$ (sd)	2.38 (1.41)	2.95 (1.67)
Birth order, $\bar{x}$ (sd)	2.62 (1.63)	2.15 (1.27)
Mother's education (average)	Some college	Bachelor's degree
Father's education (average)	Bachelor's degree	Bachelor's degree
Household income (average)	$81,000-$100,000	$61,000-$80,000

Table 2.2. Physical characteristics

	$\bar{x}$ (sd)		P Value	Effect Size
	DS	TD		
Body composition				
Height (cm)	113.79 (6.41)	126.81 (7.79)	0.000	-1.82
Weight (kg)	23.66 (5.16)	26.94 (5.71)	0.065	-0.60
RHR (beats/min)	67.95 (8.39)	67.45 (4.94)	0.820	0.07
BMI (kg/m^2)	18.13 (2.80)	16.55 (1.70)	0.037	0.68
TBW (L)	14.07 (2.67)	17.30 (3.89)	0.009	-0.97
FFM (kg)	19.18 (3.77)	23.62 (5.50)	0.011	-0.94
Body fat (%)	20.62 (8.48)	12.87 (6.04)	0.004	1.05
Range of motion (degrees)				
Dorsiflexion	28.10 (13.47)	16.85 (6.44)	0.002	1.06
Plantar flexion	86.10 (7.69)	78.05 (9.75)	0.006	0.92
Knee hyperextension	7.30 (1.75)	2.08 (0.67)	0.000	3.94
Hip abduction	57.05 (19.48)	41.45 (17.36)	0.011	0.84
Leg strength (N·m/kg)				
Right knee extensor	0.50 (0.21)	1.38 (0.52)	0.000	-2.24
Left knee extensor	0.55 (0.27)	1.32 (0.34)	0.000	-2.48
Right knee flexor	0.40 (0.12)	0.93 (0.30)	0.000	-2.34
Left knee flexor	0.46 (0.11)	0.98 (0.24)	0.000	-2.78

Notes: Total body water, Percent body fat (DS, N=16; TD, N=18); Knee extensor strength (DS, N=19); Knee flexor strength (DS, N=18)

Table 2.3. Motor characteristics

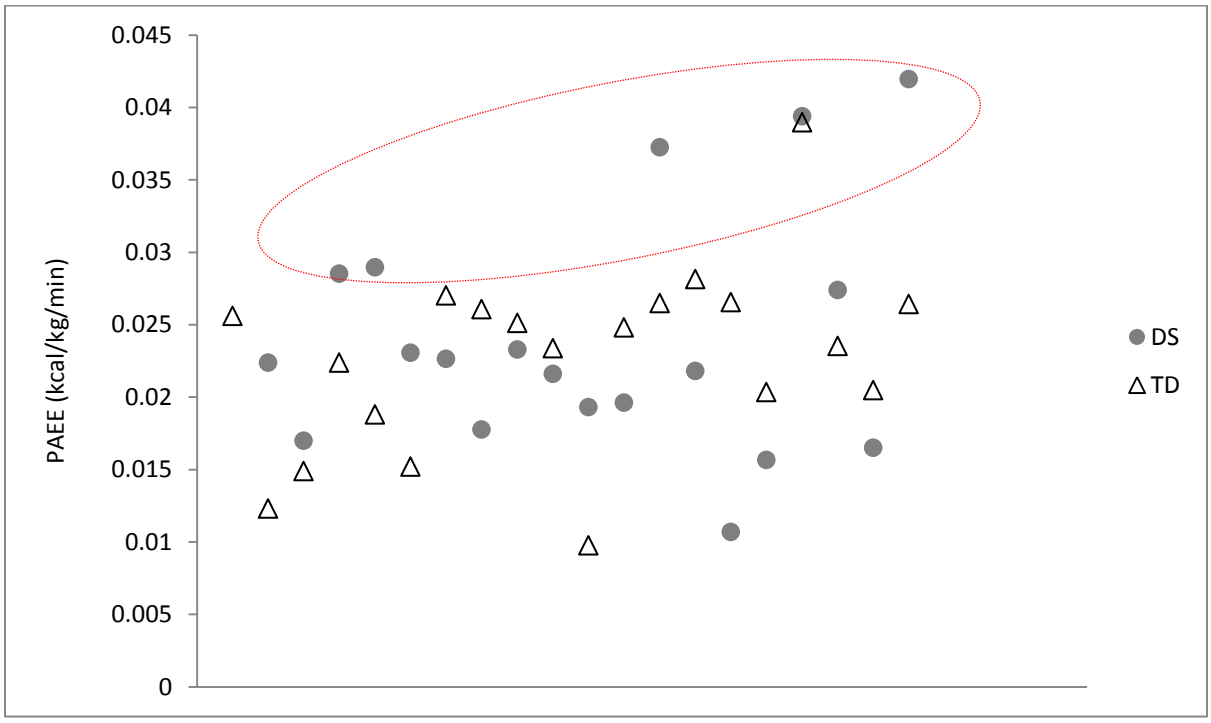
	$\bar{x}$ (sd)		P Value	Effect Size
	DS	TD		
Locomotor raw score	21.16 (11.88)	44.95 (3.02)	0.000	-2.74
Run	4.20 (2.19)	7.65 (0.74)	0.000	-2.11
Gallop	2.84 (2.71)	7.35 (0.88)	0.000	-2.24
Hop	2.47 (3.19)	9.45 (1.05)	0.000	-2.94
Leap	2.26 (2.05)	5.60 (0.75)	0.000	-2.16
Horizontal Jump	4.89 (2.73)	7.00 (1.45)	0.004	-0.96
Slide	4.37 (2.61)	7.90 (0.31)	0.000	-1.90
Object control raw score	24.80 (12.30)	42.85 (4.00)	0.000	-1.97
Strike	6.35 (2.52)	9.00 (1.21)	0.000	-1.34
Dribble	3.30 (2.75)	7.15 (1.27)	0.000	-1.80
Catch	2.90 (1.59)	5.70 (0.66)	0.000	-2.30
Kick	5.30 (2.36)	7.80 (0.62)	0.000	-1.45
Throw	3.45 (3.02)	6.05 (2.16)	0.003	-0.99
Roll	3.50 (2.89)	7.15 (1.39)	0.000	-1.61
40-foot run (sec)	5.13 (1.13)	3.10 (0.28)	0.000	2.45

Table 2.4. Physical activity

	$\bar{x}$ (sd)		P Value	Effect Size
	DS	TD		
Actiheart				
Days of data	3.26 (.99)	3.75 (.64)	0.075	-0.58
Missing data (minutes per day)	78.52 (114.87)	37.73 (39.43)	0.142	0.47
PAEE (kcal/kg/min)	0.024 (0.008)	0.023 (0.007)	0.640	0.15
SEDPA (%)	49.31 (17.10)	56.36 (10.29)	0.125	-0.49
LPA (%)	36.37 (12.94)	28.14 (8.00)	0.021	0.77
MVPA (%)	14.32 (6.80)	15.50 (6.16)	0.572	-0.18
CAPE (dimension scores)				
Diversity	31.30 (7.37)	36.05 (6.19)	0.033	-0.70
Intensity	2.76 (0.67)	3.05 (0.64)	0.163	-0.45
Who	2.36 (0.65)	2.69 (0.52)	0.089	-0.55
Where	2.72 (0.78)	2.75 (0.52)	0.887	-0.05
Enjoyment	4.03 (0.43)	3.86 (0.28)	0.144	0.47

Note: Actiheart data (DS, N=19)

Figure 2.2. Physical activity energy expenditure (DS vs. TD)



Note: N=19 for DS

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Chapter III

Exploring relationships between motor skills, physical activity, and physical characteristics in school-aged children with and without Down syndrome

Introduction

Dynamic systems theory (DST) proposes that a variety of subsystems (e.g., musculo-skeletal, cardiovascular, neural, etc.) self-organize to produce motor performance within a given task (Kamm, Thelen, & Jensen, 1990). Applications of DST have been used to understand critical factors, known as "control parameters," for locomotor development in infants with Down syndrome (DS) (Ulrich & Ulrich, 1995; Ulrich, Ulrich, Angulo-Kinzler, & Yun, 2001; Ulrich, Lloyd, Tiernan, Looper, & Angulo-Barroso, 2008). However, little is known about potential control parameters for gross motor skill ability in children with DS. It is important to understand such factors given the prevalence of gross motor skill deficiencies in school-aged children in this population (Connolly & Michael, 1986; Spano et al., 1999). Previous research suggests that children with DS may also demonstrate insufficient levels of physical activity, increased body mass index, and musculo-skeletal abnormalities, such as decreased leg strength and hypermobility (Whitt-Glover, O'Neill, & Stettler, 2006; Concolino, Pasquzzi, Capalbo, Sinopoli, & Strisciuglio, 2006; Cioni, Cocilovo, Di Pasquale, Araujo, Siqueira, & Bianco, 1994). Before one can determine whether or not physical characteristics and/or

physical activity serve as control parameters for gross motor skill ability in children with DS, relationships between these factors need to be explored. This is a necessary step since control parameters must be related to the behavior being modified.

There is a growing body of literature to support an association between musculo-skeletal difficulties and motor performance for children with typical development (TD). Following a review of strength training in adolescents, Webb (1990) concluded that increased strength enhances children's performance in athletic activities that require strength, power, or speed. Likewise, several studies have demonstrated improvements in long jump and vertical jump performances following muscle strengthening (Falk & Mor, 1996; Hetzler, DeRenne, Buxton, Ho, Chai, & Seichi, 1995; Weltman et al., 1986). Leg strength, in particular, has also been strongly implicated in the locomotor development of infants with DS (Ulrich et al., 2001, 2008). Whether or not strength is related to gross motor skill ability in school-aged children with DS remains relatively unexplored. Preliminary indications support such a relationship. Lewis and Fragala-Pinkham (2005) conducted a case study in a 10.5-year-old girl with DS to explore the effects of an exercise training program, which included strengthening. Following training, the child demonstrated improved gross motor skill performance.

In addition to strength, joint stability may play a role in gross motor skill abilities of children with DS. Research clearly establishes that children with DS experience joint laxity (e.g., Concolino, Pasquzzi, Capalbo, Sinopoli, & Strisciuglio, 2006). Intervention studies aimed at improving joint stability to facilitate motor development have

produced inconsistent findings, however. For example, Martin (2004) observed that children with DS significantly improved walking, running, and jumping performance with the use of a supramalleolar orthotic. More recently, though, Looper and Ulrich (2010) concluded that ankle orthotics may actually be detrimental to gross motor skill development in infants with DS. Besides these inconsistencies, previous research in this area has focused primarily on the ankle joint. Associations between motor skills and hypermobility at other joints are even more unclear. However, previous studies have shown that approximately one-third of children in the general population who demonstrate generalized hypermobility also experience gross motor skill delays; the degree of hypermobility and extent of motor delay do not appear to be related, though (Engelbert et al., 2005).

Similar to the musculo-skeletal and motor performance literature, there is a paucity of research examining relationships between motor skills and body composition in children with DS. In a large study of children with intellectual disabilities (ID), Frey and Chow (2006) found that body mass index (BMI) did not impact motor skills. More recently, though, Ulrich and colleagues (2011) suggested that learning to ride a bicycle may help to reduce BMI in children with DS (Ulrich, Burghardt, Lloyd, Tiernan, & Hornyak, 2011). Therefore, it is conceivable that the attainment of motor skills leads to increased physical activity, which subsequently influences body composition. Associations between body composition and physical activity in the pediatric literature have been observed but they are not as robust as one might expect. With respect to DS, specifically, Ordonez and Rosety-Rodriguez (2006) found that participation in a twelve-

week exercise program in adolescents with DS resulted in reduction of fat cells. However, little is known regarding the relationships between body composition and more generalized activity or participation in recreational activities in children with DS.

To date, many researchers contend that motor skill ability and physical activity levels are strongly linked in childhood. In fact, motor skill competence has been proposed as a primary underlying mechanism for physical activity engagement (Stodden, Goodway, Langendorfer, Robertson, & Rudsill, 2008, Du Toit & Pienaar, 2003). If this is true, motor skill competence may serve as an important control parameter for patterns of physical activity in children (Ulrich, Burghardt, Lloyd, Tiernan, & Hornyak, 2011). Several studies have observed significant relationships between motor skill ability and physical activity intensity, lending support for such claims (Wrotniak, Epstein, Dorn, Jones, & Kondilis, 2006; Fisher et al., 2005). Other studies illustrate the complexity of the motor-physical activity relationships. As evidence, Williams and colleagues (2008) found that locomotor skills, but not object control skills, were related to activity levels of preschool children (Williams et al., 2008). Further, some studies have failed to observe relationships between motor skills and physical activity altogether (Reed, Metzker, & Phillips, 2004; McKenzie et al., 2002). Inconsistent findings may be partially explained by methodological differences between studies, especially in how motor skills and physical activity were measured. Some of the aforementioned projects used accelerometers to assess activity while others utilized pedometers or survey measures. In addition, Raudsepp & Pall (2006) found that fundamental motor skills in elementary school children were related to skill-specific

physical activities but not to general levels of physical activity. Current recommendations for pediatric physical activity assessment include the use of multiple measures to account for these factors (Cervantes & Porretta, 2010).

The existence and extent of gross motor skill and physical activity associations in school-aged children with DS remains relatively unexplored. However, there is evidence that physical activity may be related to locomotor development in infants with DS. Specifically, results from Lloyd (2010) indicated that infants with DS who performed more intense leg movements walked earlier (Lloyd, Burghardt, Ulrich, & Angulo-Barroso, 2010). In conjunction with results from Ulrich's bicycle training study, it appears that physical activity may be related to selected motor skills in children with DS. Given the importance of fundamental motor skill development and the observed deficits in this area for children with DS, it is critical to understand potential associations between gross motor skills and other factors, such as physical activity.

The primary objective of the current study was to determine whether or not gross motor skill ability was related to physical activity and physical characteristics of school-aged children with and without DS. I hypothesized that motor ability would be significantly associated with activity, measured by both participation in activities and more generalized physical activity, in both populations. Second, I predicted that leg strength, and to a lesser degree body composition, would be related to motor skill ability for the TD and DS groups. In the DS group, I expected to find negative associations between hypermobility and motor performance. After exploring these

relationships, I also examined the relative importance of various physical characteristics, participation, and physical activity to motor performance for both groups using regression analyses. I expected to find that leg strength and intensity of participation would be the most influential factors in explaining motor performance for both groups. In addition, I hypothesized that hypermobility would be a significant contributor to motor skill models for the DS group. As a secondary question, I was interested in determining whether physical characteristics were related to physical activity and participation. I hypothesized that body fat would be negatively correlated with generalized activity and participation but other physical variables, such as range of motion or strength, would not be significant correlates to physical activity or participation in recreational or leisure activities.

Methods

Participants

The sample consisted of twenty children with DS and twenty children with typical development (TD) who were matched on age and gender. Participants with DS were recruited from parent support groups in Michigan and northern Ohio. The TD sample was largely recruited from families and friends of the children with DS. A local summer camp and a University of Michigan research website were also utilized as secondary recruitment tools. Exclusion criteria for both groups included major health conditions that would be counter to mild exercise (e.g., cardiac problems, uncontrolled seizures). Neither race nor gender prohibited children from participating in the study.

Procedures

All children came to the motor development laboratory at the University of Michigan for testing. Prior to the visit, parents were called and given instructions with regard to proper attire and dietary restrictions. Upon arrival at the laboratory, informed consent and child assent (when appropriate) was obtained and documented. Parents were also asked to fill out a supplemental information form prior to child testing to gather socio-demographic information. Participating families received $20. All procedures were approved by the University of Michigan Medical Internal Review Board. The following assessments were conducted in the order presented below.

E) Body composition

Height and weight were assessed using a combined stadiometer/scale. Participants wore minimal clothing (t-shirts and shorts, socks) and no shoes for the weight and height measurements. For each measurement, children were instructed to stand still in the center of the scale platform with their hands at their sides. Participants were also instructed to stand up "tall" and to "look straight ahead." Some of the children had difficulty looking straight ahead and required additional assistance, such as being asked to look at a sticker or book that was being held in front of their face. Measurements were taken twice to ensure accuracy.

Body composition was assessed using bio-electrical impedance (BIA). BIA has been employed previously in children with DS (Luke, Sutton, Schoeller, & Roizen, 1996). The equipment implemented for this assessment was the Bio-Impedance Spectrum

Analyzer (Hydra ECF/ICF, Model 4200). Prior to coming to the laboratory, parents were informed of conventional dietary and exercise restrictions to maximize the validity of BIA testing. Specifically, parents were instructed that their children should not exercise or eat for at least four hours prior to the assessment. However, children were allowed to drink a small amount of water if desired. Immediately following BIA testing, children were permitted to eat a small snack.

In preparation for the BIA testing, children were asked to lie in a tetrapolar configuration (supine with hands and legs slightly abducted) and to minimize their movements for ten minutes. During this time, the examiner measured the child's resting heart rate (RHR). Also during this period, rubbing alcohol was used to clean skin areas on the hands and feet. Two electrodes were then placed on the cleaned and dried skin surfaces. Once the Hydra device was turned on, the child's height, weight, and gender was entered. The BIA procedure involved application of an electrical current (500 micro amps at a frequency of 50 kHz) across electrodes on the hands and feet of participant. Intracellular fluid (ICF), extracellular fluid (ECF), and total body water (TBW) were calculated by the analyzer in approximately one to two minutes, depending on the child's ability to remain still. To decrease the child's movements during both the BIA pre-testing and testing periods, books were available for the parents to read to their children.

F) Range of motion (ROM)

Passive ROM at the ankle, knee, and hip were assessed on the right side of the body using a standard goniometer. Measurements of dorsi-flexion, plantar flexion, knee

hyperextension, and hip abduction were performed in accordance with joint measurement procedures in Norkin and White (1995). Ranges of motion at the ankle and knee were measured with the participant seated at the edge of an elevated table. Hip abduction was assessed with the child lying in a supine position. For hip abduction, parents were occasionally asked to assist with stabilizing the non-measured leg if the child had difficulty remaining still. Two measurements were made for each joint movement with the average value used in analysis.

G) Gross motor skills

The Test of Gross Motor Development (2nd ed.) (TGMD-2) (Ulrich, 2000) was used to assess gross motor skill ability of the participants. The TGMD-2 measures locomotor and object control skills in children ages 3 through 10 years. The items of the locomotor subtest include running, galloping, hopping, leaping, horizontal jumping, and sliding. The object control subtest consists of striking a stationary ball, stationary dribbling, catching, kicking, overhand throwing, and underhand rolling. High test-retest reliability for the TGMD-2 has been established, with coefficients ranging from 0.86-0.94 for the age groups included in the current study (Ulrich, 2000).

Due to scheduling conflicts, the TGMD-2 was conducted in the motor development laboratory rather than a gymnasium, as recommended in the manual. As a result, some of the tasks were modified to accommodate the space constraints. For example, children were instructed to throw and kick the ball towards specific targets on the wall. The procedures complied with instructions provided by the TGMD-2 manual as much as possible given space limitations. Accurate demonstrations and verbal

descriptions of the skills were provided to the participants. Each child was allowed a practice session in attempt to ensure that he or she understood the task. Two test trials were administered and scored based on the performance criterion set forth in the manual. Each trial was videotaped. In addition to the standard TGMD-2 skills, participants were also measured on a timed run of 40 feet to obtain a measure of peak performance. Children were instructed to run as fast as they could. Two trials of the run were conducted.

H) Leg strength

Bilateral isokinetic knee flexor and extensor strength was measured using a Cybex 340 dynamometer (Cybex, New York, NY) with HUMAC (2004). This model dynamometer has been used to assess isokinetic strength in individuals with DS previously (Pitetti, Climstein, Mays, & Barrett, 1992; Croce, Pitetti, Horvat, & Miller, 1996). Prior to testing, a researcher demonstrated the leg movements on the dynamometer. Pillows were placed behind the children when necessary to provide proper lumbar support. Subjects performed three practice sessions per movement (knee flexion, knee extension) per leg at a speed of 30 degrees per second. Following several minutes of rest, they performed three maximal efforts per movement per leg at a speed of 30 degrees per second. The children were instructed to push against the lever arm as powerfully and rapidly as possible following a verbal prompt. Throughout each trial, verbal encouragement was provided by the researcher and family members. One minute of rest was given between trials. The order of limb testing was randomized for each child to minimize learning effects.

E) *Physical activity*

Physical activity was measured using two methods, a questionnaire and a combined heart rate monitor and accelerometer. The two measures provide different information about the quality and quantity of the participants' activity. While the questionnaire focuses on different dimensions of the children's participation in recreational and leisure activities, the combined accelerometer and heart rate monitor provides detailed and objective information about the movement counts and heart rate of children throughout the day. We refer to the questionnaire data as an index of "participation" whereas data from the activity monitor represents "generalized physical activity."

Children's Assessment of Participation and Enjoyment (CAPE)

The Children's Assessment of Participation and Enjoyment (CAPE) is a 55-item questionnaire that has been validated for individuals with and without disabilities between the ages of 6 and 21 years (King et al., 2007). It assesses 5 dimensions of participation: diversity, intensity, with whom, where, and enjoyment. The CAPE manual recommends that children fill out the measure themselves. However, anecdotal evidence from our previous projects suggests that children with DS significantly over-report their participation and enjoyment of activities. Therefore, parents of participants in the current study were asked to fill out the CAPE with their children. This procedure worked well in our pilot work. The completed questionnaire was mailed back to the researchers in a self-addressed, postage-paid envelope.

Actiheart

The Actiheart (CamNtech, United Kingdom) was used to estimate intensity of generalized physical activity. The device is a combined heart rate monitor and accelerometer that has been validated in children (Corder, Brage, Warehand, & Ekelund, 2005; Brage, Brage, Franks, Ekelund, & Wareham, 2005; Parish, Rudisill, & St. Onge, 2007). The Actiheart samples at 32 Hz and measures activity counts (AC) and heart rate (HR). Both AC and HR are utilized to estimate physical activity energy expenditure (PAEE), an indicator of activity intensity. The Actiheart provides more accurate estimations of activity intensity than devices which only utilize activity counts or heart rate data (Brage, Brage, Franks, Ekelund, & Wareham, 2005; Haskell, Yee, Evans, & Irby, 1993; Corder et al., 2005) (see analysis section for detailed explanation of the prediction equations).

The Actiheart monitor was attached to the chest of each child with two biocompatible electrocardiograph (ECG) electrodes. Electrodes were placed on both sides of the heart parallel to the space between the fourth and fifth ribs. Each attachment site was cleaned with alcohol and dried prior to placement of the device. Reviews of pediatric activity literature suggest that accelerometers should be worn for a minimum of 4 days, with at least one day being a weekend day (Trost, McIver, and Pate, 2005). As a result, children were instructed to wear the device for 4 days. Further, all participants were scheduled in a manner to ensure they would wear the Actiheart on at least one weekend day. Families were sent home with an activity monitor log, which was used to document any times when the monitor was removed either intentionally

(e.g. swimming, baths) or accidentally. The Actiheart device and activity log were returned via mail with the CAPE following the four-day assessment period.

Data Analysis

A) Data reduction

Height and weight were used to determine body mass index (BMI) using the equation BMI= weight (kg)/height^2 (m). Measures of body composition were also calculated from BIA output. Specifically, fat-free mass (FFM)(kg) was determined by dividing TBW (L) by specific hydrations constants based on the subject's height, age, and gender (Goran et al., 1993). The resulting equation was:

$$\text{FFM (kg)} = \frac{\text{TBW}}{0.769-(0.0025 \times \text{age}) - (0.019 \times \text{gender})}$$

Where age was entered in years and gender = "0" for females and "1" for males. Fat mass (FM) was calculated by subtracting FFM from body weight. As a result, we were able to estimate percent body fat by dividing FM by total body mass and multiplying by 100.

Measures of joint range of motion and strength were also used to characterize the physical characteristics of the sample. Mean ROM values for plantar flexion, dorsiflexion, knee hyperextension, and hip abduction were calculated across two trials. Isokinetic strength values were adjusted for body mass, using ratio standards (Davies & Dalsk, 1997; Sundegardh, Bratteby, Nordesjo, & Nordgren, 1988). Of the three trials, peak bilateral knee flexor and extensor strength (N·m/kg) were included in the analyses.

Dependent variables of the motor domain included the raw scores of the locomotor and object control skills sections of the TGMD-2 along. Peak run time over the two trials was also included. Physical activity was represented in the dataset by variables from both the CAPE and Actiheart. For the CAPE, scores for 5 dimensions of participation (diversity, intensity, who, where, and enjoyment) were analyzed. These variables are referred to as DivPar, IntPar, WhoPar, WherePar, and EnjPar, respectively.

Preparing dependent variables from the Actiheart was a multi-step process. Once the monitors were returned to the laboratory, the data was downloaded using the Actiheart software. The software provided minute by minute output for activity counts (AC) and heart rate (HR) over the 4 days. The output was cleaned manually to remove any data points when the monitor was not worn and to omit outliers. Research has shown that maximal heart rate in children is stable up to 200 beats per minute (Rowland, 1993; Epstein et al., 2001). Therefore, data points containing HR values above 200 beats per minute were removed manually. The amount of valid data over the 4 days and the average number of missing data per day were calculated and recorded.

The cleaned Actiheart data was entered into a customized program developed by CamNtech to estimate PAEE (see Figure 3.1). The program is based on a branched equation model by Corder and colleagues (2005) who used a step-test to determine thresholds and parameters for children. In the model, child equations for HR and AC are extrapolated to go through sleeping heart rate and 0 counts per minute, respectively. Twenty-five counts per minute serves as the initial threshold in the model. For counts

above 25, decisions are then made based on the transition heart rate, which was determined to be the highest walking and slowest jogging heart rate values above sleep during the step test. If counts are below 25 per minute, the second decision in the flow chart is based upon the flex heart rate. Flex heart rate is defined as the average above sleep of the highest heart rate during rest and the lowest heart rate during incremental exercise for children during treadmill testing. Depending on the outcomes in step 2, data is ultimately entered into one of four equations. These equations weight activity and heart rate data in varying proportions to predict PAEE in kJ/kg/min. In the current study, PAEE was converted to kcal/kg/min by dividing the value by 4.18. Due to the fact that some of the data points were removed, absolute PAEE values were not used in analysis. Rather, PAEE scores (per minute) were totaled for each day and divided by the number of valid minutes per day. These values were then averaged by the number of days included in analysis. Higher values for PAEE correspond to increased energy expenditure and, thus, more intense physical activity. We use PAEE as an indicator of generalized physical activity and refer to this variable as "GenPA."

B) Statistics

Descriptive statistics by group (DS, TD) were previously conducted on the following variables to characterize the sample: age, gender, physical variables (body composition, ROM, leg strength), GenPA, participation (CAPE dimensions), and motor skills (locomotor/object control subscales, run time). In the current study, Pearson correlations were performed to determine bivariate associations between: 1) motor

skills (locomotor, object control, peak run) and physical activity (GenPA and participation dimensions; 2) motor skills and physical variables (BMI, body fat, ROM, leg strength); and 3) physical activity and physical variables. Stepwise linear regression analyses were also conducted for the dependent variables of locomotor score, object control score, and running performance. Independent models were run for the DS and TD groups. Age and gender were included as covariates in all regression models. Additional covariates were selected based on the results of the correlation analyses (see the Regression section of the Results). Alpha was set at $p < 0.05$ for all analyses.

Results

Descriptive statistics

Table 3.1 shows descriptive statistics for the DS and TD groups. Both groups consisted of four females and sixteen males. We were also successful in matching the groups by age, with mean ages of 7.94 years in both groups. As Table 3.1 illustrates, children with DS exhibited substantially lower motor skill and strength scores compared to the TD group. The DS group also demonstrated higher ROM and increased body fat. With respect to activity, the groups were similar in terms of GenPA. The TD group scored slightly higher on the participation domains of diversity, intensity, who, and where while the DS group recorded marginally higher scores on the enjoyment dimension. Previous analysis of this dataset indicated that group differences reached statistical significance ($p < 0.05$) on all of the motor and physical variables with the exception of weight. Besides the diversity dimension on the CAPE, which measures the

number of different activities they participated in, the groups were not significantly different on participation scores or on GenPA.

Motor skills and physical characteristics

Table 3.2 illustrates the correlations between motor performance and physical characteristics by group. For children with DS, leg strength was significantly correlated with motor performance. Right and left knee extensor strength, respectively, were positively correlated with locomotor ($r(17) = .525$, $p = .025$; $r(17) = .541$, $p = .020$) and object control ($r(17) = .589$, $p = .008$; $r(17) = .486$, $p = .035$) skills. Right knee extensor strength showed a negative association with running speed but the relationship failed to reach significance ($r(17) = -.428$, $p = .077$). In addition to leg strength, range of motion was significantly related to motor skill ability. Locomotor scores were negatively correlated with dorsiflexion ($r(18) = -.475$, $p = .040$) and knee hyperextension ($r(18) = -.549$, $p = .015$). Trends were observed for correlations between object control skills and both dorsiflexion ($r(18) = -.423$, $p = .063$) and hyperextension ($r(18) = -.433$, $p = .056$). There was also a trend for object control skills vs. weight ($r(18) = .692$, $p = .052$) in the DS group, such that higher object control scores were associated with increased weight. This result is likely due to the effect of age, however, since increased age corresponded to higher locomotor ($r(18) = .734$, $p < .001$) and object control ($r(18) = .692$; $p = .001$) scores and faster running times ($r(18) = -.487$, $p = .035$). Results of a partial correlation between object control scores and weight were non-significant when controlling for age.

Compared to the DS group, relationships between physical characteristics and running speed were much more robust in the TD group. Strong negative associations were observed between run time and bilateral knee flexor and extensor strength. Run time was also negatively correlated with both weight ($r(18) = -.452$, $p = .045$) and height ($r(18) = -.572$, $p = .008$). A trend was observed between percent body fat and running performance ($r(16) = .428$, $p = .076$). Physical characteristics were not highly related to locomotor or object control ability in the TD group. Exceptions included a significant negative association between body fat and locomotor skills ($r(16) = -.571$, $p = .013$) along with a significant positive correlation between object control skills and left knee extensor strength ($r(18) = .533$, $p = .016$). A trend was observed between object control skills and right knee flexor strength ($r(18) = .436$, $p = .054$). As was the case with the DS group, increased age was also associated with increased object control scores ($r(18) = .491$, $p = .028$) and improved run times ($r(18) = -.785$, $p < .001$). The relationship between age and locomotor scores just missed statistical significance for the TD group ($r(18) = .435$, $r = .055$).

Motor skills and physical activity

Pearson correlations revealed that physical activity and motor skills were strongly associated for children with DS. GenPA and participation in leisure and recreational activities were related to motor skills differently in our DS sample, however, with increased GenPA being associated with poorer motor performance and heightened levels of recreational and leisure activities corresponding to better motor scores. As evidence, GenPA was negatively correlated with locomotor scores ($r(17) = -.729$, $p =$

.001) and object control scores (r(17) = -.496, p = .031). Further, GenPA showed a strong positive association with time to run 40 feet (r(17) = .739), p < .001), such that children with higher GenPA took longer to run the distance. Conversely, run time was negatively correlated with the DivPar (r(18) = -.492, p = .032), IntPar (r(18) = -.546, p = .016), and EnjPar (r(18) = -.562, p = .012). These dimensions were also positively correlated with locomotor and object control scores, indicating that the increased diversity, intensity, and enjoyment of recreational activities in the DS group corresponded to better motor ability. Of note, nonlinear models were also explored to understand the nature of the relationships between GenPA and motor skills. Ultimately, linear models were most effective in describing the relationship between GenPA and our three motor variables.

Unlike the DS group, GenPA was positively associated with motor performance in the TD group. Only the correlation between GenPA and object control skills (r(18) = .479, p = .033) reached statistical significance, though. Surprisingly, locomotor skills were negatively associated with IntPar (r(18) = -.451, p = .046) and DivPar (r(18) = -.476, p = .034). Otherwise, motor performance was not correlated with participation in recreational or leisure activities for the TD group. Table 3.3 shows the correlations between motor skills and physical activity for both groups.

Physical activity and physical characteristics

In general, physical characteristics were not strongly related to either GenPA or participation in recreational or leisure activities in the DS group. DivPar and IntPar were positively associated with height and weight for children with DS. However, these

results are likely due to age effects since age was correlated more strongly with both DivPar ($r(18) = .657$, $p = .002$) and IntPar ($r(18) = .551$, $p = .012$). For the TD group, GenPA was positively associated with bilateral leg strength (right knee flexor, $r(18) = .455$, $p = .044$; left knee flexor, $r(18) = .512$, $p = .021$). InPar ($r(18) = .466$, $p = .038$) and DivPar ($r(18) = .495$, $p = .027$) were positively associated with dorsiflexion for the children with TD as well.

Regression analyses

Covariates for the regression models were selected on the basis of theoretical considerations and correlation analyses. All stepwise regression models included the covariates of age, gender, GenPA, and IntPar. Diversity of participation was also considered as a covariate but was excluded due to its strong correlation with the intensity dimension ($r(38) = 0.855$, $p < .001$). In addition to the aforementioned covariates, all DS models included the predictors of knee hyperextension and right knee extensor strength. Body fat, as opposed to knee hyperextension, was selected for inclusion in the TD models based on correlation results. The TD models also included left, rather than right, knee extensor strength. Of note, the TD models using left versus right knee strength were only marginally different. The left side was selected because it explained more of the variance in motor performance for the TD sample.

A. Locomotor skills

Results from stepwise linear regression analyses (Table 3.4) indicated that age, GenPA, and IntPar significantly predicted locomotor skills in our sample of children with DS ($F(3,13) = 24.49$, $R^2 = .840$) In step 1, age ($\beta = .793$, $p = .001$) explained 60.5% of the

variance in locomotor scores. The addition of GenPA (β = -.414, p = .013) increased R^2 by 12.6%. In the final step, IntPar (β = .428, p = .006) was added to account for additional 10.9% of the variance. The TD model of locomotor skills resulted in a single predictor, percent body fat (β = -.571, p = .013), which only explained 28.4% of the variance (F(1,16) = 7.74, p = .013).

B. Object control skills

Stepwise regression for the dependent variable of object control skills in DS resulted in an initial model (F(1, 16) = 20.88, p < .001, R^2 = .539) that included IntPar (β = .752, p < .001). The addition of right knee extensor strength (β = .395, p = .016) resulted in a final model (F(2,15) = 18.34, p < .001) explaining 67.1% of object control performance in children with DS. Object control ability in children with TD was significantly predicted by gender alone (β = .659, p = .003), which explained 39.9% of the variance. The DS and TD object control models are shown below in Table 3.5.

C. Running performance

Running performance in children with DS was solely predicted by GenPA (β = .683, p = .002), accounting for 43.1% of the variance. Stepwise regression for TD running performance resulted in a two-step model. In step 1, left knee extensor strength (β = -.753, p < .001) explained 54.0% of the variance. The inclusion of age (β = -.443, p = .043) in step 2 led to a final model (F(2,15) = 15.44, p < .001) accounting for 62.9% of the variance in running performance in children with TD. See table 3.6 for the DS and TD models of running performance.

Discussion

The primary goal of the current study was to examine correlates to gross motor skill ability in school-aged children with and without DS. To achieve this objective, we examined physical- and activity-related correlates to motor skill ability in forty children (20 with DS, 20 with TD) between the ages of 6 and 10 years of age. Results indicated several important findings. Though several factors were associated with motor performance in each group, regression analyses indicated that knee extensor strength, generalized physical activity, and intensity of participation in recreational and leisure activities were most relevant. Second, relationships between physical characteristics and the motor domain differed by group and were highly contingent on the motor variable in question (e.g., gross motor scores vs. peak running performance). Finally, participation and generalized activity related to motor skills in opposing fashion between and within groups.

Findings from the current study indicate that several factors in the physical and activity domains are closely related to gross motor skill development in children with and without DS. With respect to the physical domain, we observed significant positive correlations between knee extensor strength and both locomotor and object control scores in the DS group. A trend was also observed for correlations between knee extensor strength and running speed in this group. Our regression analyses confirmed the importance of knee extensor strength on gross motor skill scores, particularly for object control performance. These findings are consistent with those from Lewis and Fragala-Pinkham (2005), who observed increased gross motor skill performance in a

child with DS following strength training. They also lend support to motor interventions in DS that incorporate strengthening, such as treadmill training with ankle weights (Ulrich et al., 2008). Knee extensor strength has also been reported as a critical factor in learning to ride a two-wheel bicycle in youth with DS (Ulrich et al., 2011). Fortunately, leg strength is trainable and it should be a major component of individualized physical education programs for school-aged children with DS.

In addition to leg strength, ranges of motion at the knee and ankle were associated with motor skills in children with DS. Although ROM values were not statistically significant covariates in the DS regression models, they were modestly correlated with gross motor skill scores. Specifically, high degrees of dorsiflexion and knee hyperextension were related to poorer locomotor and object control scores. Our results are in partial agreement with those from Engelbert and colleagues (2005), who found that motor delay is present in a subsample of children with hypermobility. However, our findings differed from Engelbert in that the degree of motor deficits we observed appeared to correspond to the amount of hypermobility. We attribute the discrepancies to sample differences. Engelbert's study was not conducted on a sample of children with DS, but rather on children with a variety of hypermobility etiologies. Previous research suggests that preadolescent children with DS may have unique biomechanical constraints that become increasingly important with greater joint instability and task novelty (Ulrich, Haehl, Buzzi, Kubo, & Holt, 2004). Therefore, it is reasonable that children with DS who exhibit more joint laxity may demonstrate poorer motor performance, particularly if they are unfamiliar with tasks in question.

In contrast with the DS group, lower extremity range of motion was not associated with motor skills in the TD group. Similar to their peers with DS, though, leg flexor and extensor strength was positively correlated with gross motor skill scores. Further, strength was strongly associated with running speed in the TD group to the extent that it accounted for over half of the variance in run time. Our observation of a robust relationship between leg strength and peak motor performance is consistent with previous literature. For example, Webb (1990) concluded that strength was an important contributor to athletic skills requiring power and speed. In addition to strength, body composition was related to locomotor performance such that lower body fat values corresponded to higher locomotor scores on the TGMD-2 and faster running speeds. Body fat was the lone significant predictor in our TD regression model for locomotor scores. Percent body fat has been previously linked to locomotor performance in prepubescent boys (Ara, Calbet, Dorado, Jiminez-Ramirez, Serano-Sanchez, & Vicente-Rodriguez, 2004).

Though the relationships we observed between physical characteristics and motor skills in the two groups were expected, our findings on activity, participation, and motor skills were less anticipated. Previous literature has led to mixed results as to whether or not motor skills and physical activity are strongly related in children. For example, while Williams and colleagues (2008) argued for an association between physical activity and specific motor skills (e.g., locomotor only), some researchers failed to observe any significant relationship between activity and motor ability (Reed et al., 2004). Yet others have suggested that the relationship depends, not on the type of

motor skill, but rather on the type of activity measured (Raudsepp & Pall, 2006). In an attempt to mitigate these issues, we utilized multiple motor skill variables and included both generalized physical activity and participation in recreational and leisure activities. Despite these efforts, our results only further illustrate the complexity in delineating associations between activity and motor skills in children. The relationships become even less clear when examining children with disabilities, such as DS.

As anticipated, our findings indicated that diversity, intensity, and enjoyment of participation in recreational and leisure activities were positively correlated with motor performance in the DS group. Further, intensity of participation (i.e., how often they participate) significantly contributed to models of locomotor and object control scores in the DS group, even after controlling for age and gender. We also observed moderate, but non-significant, positive correlations for the who and where dimensions of participation versus object control scores in the DS group. It is possible that these correlations would have reached statistical significance with a larger sample, thereby lending support for more social participation in children with DS. Conversely, generalized physical activity was negatively correlated with all three of our motor variables. The observation that higher levels of GenPA corresponded to poorer motor performance in the DS group was inconsistent with our expectations. This finding warrants further discussion, though, since GenPA was a significant predictor in the locomotor and running speed regression models of DS. At first glance, the notion that increased GenPA could be detrimental to motor performance seems counterintuitive. However, increased levels of hyperactivity have been reported previously in children

with DS (Dykens, 1997) and in DS mouse models (Altafaj et al., 2001; Galante et al., 2009). Further, rates of co-morbidity for DS and hyperactivity disorders range from 6% to over 50% (Myers & Pueschel, 1991; Gath & Gumley, 1986; Capone, Goyal, Ares, & Lannigan, 2006). Therefore, it is plausible that high GenPA scores in our sample of children with DS are indicative of hyperactivity as opposed to high levels of functional, health promoting activity. We believe this to be true since some of the children with DS showed GenPA levels above and beyond those found in any of the TD children. Anecdotally, we observed several hyperactive children with DS during the testing sessions who also displayed high GenPA scores. These children tended to be lowest in terms of their overall level of functioning as well. As a result, we contend that high levels of "non-functional" activity could explain the negative association between GenPA and motor skills in the DS group. This claim is consistent with previous studies showing that hyperactive children display poorer motor performance (Harvey & Reid, 1997).

Results on associations between motor skills and activity in our TD group were in direct opposition with those from the DS sample. For instance, participation intensity and diversity were negatively correlated with gross motor scores. These correlations were marginal and only the relationships between participation and locomotor scores reached statistical significance. Nevertheless, the direction of the relationships is intriguing. We believe there are two contributing factors to our participation and motor skill correlations in the TD group. First, there is likely a ceiling effect on the locomotor subscale of the TGMD-2 for children with TD. Several of the locomotor skills were performed effortlessly by the majority of children in the TD group to the extent that

differences in locomotor scores were virtually undistinguishable. The mean locomotor score for the TD group was 44.9 ± 3.0 out of a possible 48. Second, and perhaps more importantly, only a subset of questions on the CAPE are representative of "movement-based" activities or "physical" activities. In fact, a large portion of the questions reflect social and self-improvement activities that are relatively sedentary in nature, such as talking on the phone, reading a book, or attending religious events. Therefore, it is conceivable that increased involvement in recreational or leisure activities could actually be detrimental to locomotor performance in some children, depending on the nature of their activities. This seems like a reasonable assertion given that we also observed that intensity of participation in these activities was negatively correlated with knee extensor strength in the TD group. However, further research examining the individual patterns of recreational activities would be necessary to support this claim.

When analyzing our activity-motor associations for the TD group, it is also important to consider that the CAPE excludes school activities. Many children may be the most physically active during periods of physical education or other school activities. If true, we might expect to find positive associations between motor skills and our GenPA data, which does not omit school activity. In fact, our results demonstrate that higher GenPA scores corresponded with better motor performance in children with TD. This suggests that children with TD engage in some form of physical activity, possibly during school rather than in recreational situations, that positively relates to their motor skills. We are cautious in interpreting our activity and motor skill relationships in the TD group, however. The correlations were marginal at best and neither GenPA nor

intensity of participation significantly contributed any of the three regression models for motor ability in the TD group.

There are several limitations to the current study that must be considered when interpreting our results. First, we utilized a small sample of convenience, which could explain why some of our correlations were marginal or failed to reach statistical significance altogether. In addition, some of the children in the TD group were responding to advertisements for a "sports-related" study. Therefore, it is plausible that they were exceptionally interested or competent with respect to motor skills. This argument is supported by the fact that children with TD in our sample (mean age = 7.9 years) reported slightly higher locomotor (44.9) and object control (42.8) scores than those reported for similarly aged children in the norm tables of the TGMD-2 (females, locomotor = 41, object control = 37; males, locomotor = 41, object control= 42). As a result, their high gross motor scores may not be representative of the general population. Future studies should address some of these issues by including a larger sample with randomly selected participations. Another limitation in the current study involves the activity data. Specifically, the equations we used for determining GenPA have not been validated in children with DS. Additional research is needed to establish whether the branched equation model used in the current study is suitable for assessing generalized physical activity in children with DS.

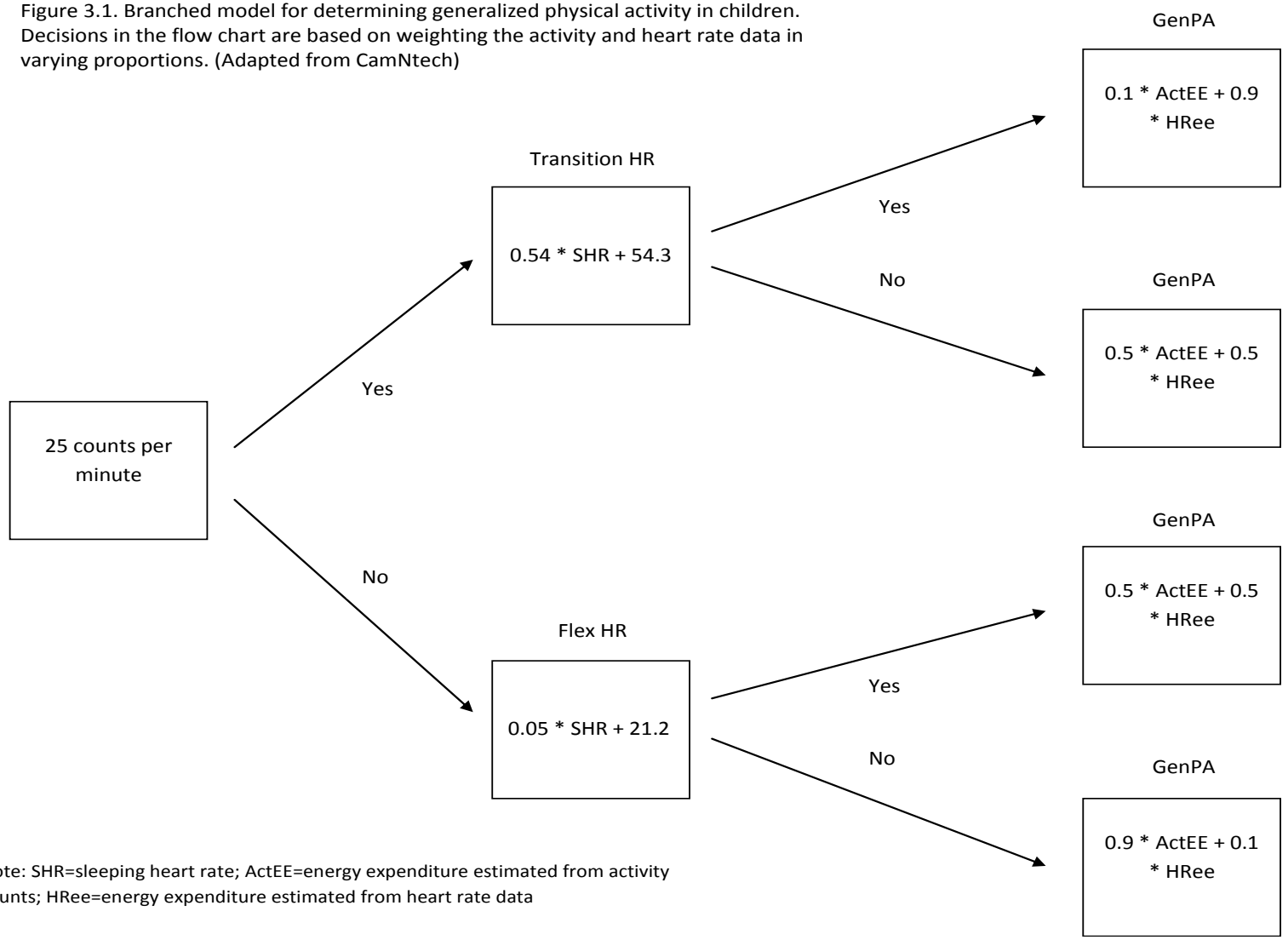
Despite these limitations, accumulated evidence from the current study suggests that physical characteristics, physical activity, and participation in recreational activities

are associated with motor skill ability to varying degrees in children with and without DS. Using concepts from dynamic systems, leg strength, generalized physical activity, and participation may be potential control parameters for motor skill development in school-aged children with DS. For children with TD, physical activity and participation appear to be less important to their motor capabilities. Physical characteristics, especially leg strength and body composition, are more likely candidates to serve as control parameters in this group. Other variables should also be considered as correlates to gross motor ability in both groups. For instance, recent studies in adults with DS suggest that support networks and motivation may serve as barriers to activity (Mahy, Shields, Taylor, & Dodd, 2010). Likewise, there is considerable evidence linking parental support with physical activity participation in children (e.g., Trost, Sallis, Pate, Freedson, Taylor, & Dowda, 2003). Cognition is also worth considering, although Connolly and Michael (1986) showed that children with DS have lower gross motor scores than children with other forms of intellectual disability. We contend that several of the variables highly related to gross motor skills (e.g., participation, leg strength) in the current study, in addition to factors such as parental support and motivation, all warrant further investigation with respect to their utility as control parameters for gross motor skill development in school-aged children with DS. Longitudinal examination of gross motor skills in children with DS is a necessary first step to identify windows for intervention (e.g., periods of heightened variability). Once these periods are established, the potential control parameters can be manipulated to determine their effects on gross motor skills.

The current study was primarily focused on exploring potential control parameters for gross motor skills. However, we cannot establish the direction of the relationships we tested so it is also possible that gross motor skills may serve as control parameters for physical characteristics (e.g., leg strength), generalized physical activity, and participation. An intriguing line of future research involves exploring gross motor skills as possible control parameters for increased physical activity and participation in recreational activities in children with DS. Recent empirical evidence and findings from the current study provide preliminary support for this idea. For instance, Ulrich and colleagues (2011) suggested that learning to ride a bicycle may decrease sedentary activity and increase moderate activity in youth with DS. Similarly, following a lengthy review of the literature, Bult and others (2011) concluded that gross motor skills were amongst the most important variables for increasing frequency of participation in recreational and leisure activities in children with disabilities, along with cognitive ability, communication skills, and manual ability (Bult, Verschuren, Jongsman, Lindeman, & Ketelaar, 2011). Given these findings and our observations of strong positive correlations between participation (intensity and diversity) and gross motor skills in children with DS, assertions of gross motor skills as control parameters for physical activity and participation seem reasonable. Of course, future research is needed to validate such statements. Studying the effects of gross motor skill interventions on physical activity and participation in children with DS would be beneficial, particularly during the school-aged years since there is substantial research showing a drop-off in physical activity once children reach adolescence (e.g., Kemper, Post, Twisk, Van

Mechelen, 1999). Future research should also investigate more close the issue of hyperactivity in children with DS. It is possible that "scaling down" hyperactivity through various treatments could positively impact motor skills in some children with DS. If so, level of hyperactivity could be an important control parameter for gross motor skill acquisition in children with DS. A study comparing the gross motor skills of children with DS without hyperactivity to those of children with DS with hyperactivity and to children with ADHD would be an important first step in that regard. Finally, just as we explored whether physical characteristics and physical activity relate to motor skills, future research should also examine whether motor skills and/or physical activity are associated with development in other domains (e.g., socio-emotional) for school-aged children with and without DS. Ultimately, researchers, educators, and clinicians should strive to improve physical, mental, and emotional health to positively impact overall well-being for children with DS. Enhancing motor skills and increasing physical activity may be two means to achieve this goal.

Figure 3.1. Branched model for determining generalized physical activity in children. Decisions in the flow chart are based on weighting the activity and heart rate data in varying proportions. (Adapted from CamNtech)



Note: SHR=sleeping heart rate; ActEE=energy expenditure estimated from activity counts; HRee=energy expenditure estimated from heart rate data

Table3.1. Sample description

	DS	TD
Gender	4 females, 16 males	4 females, 16 males
Age	7.94 (1.25)	7.94 (1.57)
Motor Skills		
Locomotor raw score***	21.16 (11.88)	44.95 (3.02)
Object control raw score***	24.80 (12.30)	42.85 (4.00)
40-ft timed run (sec)***	5.13 (1.13)	3.10 (0.28)
Physical Activity		
GenPA (kcal/kg/min)	0.024 (0.008)	0.023 (0.007)
Participation		
Diversity*	31.30 (7.37)	36.05 (6.19)
Intensity	2.76 (0.67)	3.05 (0.64)
Who	2.36 (0.65)	2.69 (0.52)
Where	2.72 (0.78)	2.75 (0.52)
Enjoyment	4.03 (0.43)	3.86 (0.28)
Body Composition		
Weight (kg)	23.66 (5.16)	26.94 (5.71)
Height (cm)***	113.79 (6.41)	126.81 (7.79)
BMI (kg/m^2)**	18.13 (2.80)	16.55 (1.70)
Body fat (%) **	20.62 (8.48)	12.87 (6.04)
Range of Motion (degrees)		
Dorsiflexion**	28.10 (13.47)	16.85 (6.44)
Plantar flexion**	86.10 (7.69)	78.05 (9.75)
Knee hyperextension***	7.30 (1.75)	2.08 (0.67)
Hip abduction*	57.05 (19.48)	19.48 (17.36)
Leg Strength (N·m/kg)		
Right knee extension***	0.50 (0.21)	1.38 (0.52)
Right knee flexion***	0.40 (0.12)	0.93 (0.30)
Left knee extension***	0.55 (0.27)	1.32 (0.34)
Left knee flexion***	0.46 (0.11)	0.98 (0.24)

Notes: Values represent mean (s.d.); Actiheart data (DS, N=19); TBW, Body fat (DS, N=16; TD, N=18); Knee extensor strength (DS, N=19); Knee flexor strength (DS, N=18); p-values (***p<.001; **p<.01; *p<.05)

Table 3.2. Correlations: motor skills vs. physical characteristics

		Locomotor skills		*Object control skills*		*Timed run*	
		DS	TD	DS	TD	DS	TD
Body composition							
Weight (kg)	*r*	.338	.238	.441	.355	-.079	-.452*
Height (cm)		.387	.255	.396	.333	-.261	-.572**
BMI (kg/m^2)		.201	.183	.330	.324	.073	-.225
Body fat (%)		-.193	-.571*	-.306	-.417	-.034	.428
Range of motion (degrees)							
Dorsiflexion		-.475*	-.262	-.423	-.036	.407	.051
Plantar flexion		.142	.216	-.085	-.118	-.311	-.271
Knee hyperextension		-.549*	-.218	-.433	-.317	.347	.190
Hip abduction		-.117	.202	-.084	-.017	-.068	-.194
Leg strength (N·m/kg)							
Right knee flexion		.253	.271	.533*	.436	-.258	-.717***
Right knee extension		.525*	.267	.589**	.366	-.428	-.809***
Left knee flexion		.347	.056	.460	.370	-.074	-.613**
Left knee extension		.541*	.375	.486*	.533*	.028	-.797***

Note: $p < 0.05$ (*), $p < 0.01$ (**), $p < 0.001$(***)

Table 3.3. Correlations: motor skills vs. physical activity and participation

		Locomotor skills		*Object control skills*		*Timed run*	
		DS	TD	DS	TD	DS	TD
Physical Activity							
Generalized activity	*r*	-.729***	.356	-.496*	.479*	.739***	-.393
Participation							
Diversity		.757***	-.476*	.805***	-.254	-.492*	.317
Intensity		.741***	-.451*	.752***	-.132	-.546*	.192
Who		.278	-.291	.437	.148	-.261	.152
Where		.154	-.113	.393	.029	-.227	.206
Enjoyment		.471*	-.258	.619**	-.339	-.562*	.424

Note: $p < 0.05$ (*), $p < 0.01$ (**), $p < 0.001$(***)

Table 3.4. Regression models for locomotor skills

DS				
Step	F	df	Beta	p-value
I	24.49	1, 15		
Age			.793	.000
R^2 =.605				
II	22.74	2, 14		
Age			.602	.001
Generalized physical activity			-.414	.013
$\Delta R^{2 =}$.126				
III	28.92	3, 13		
Age			.326	.038
Generalized physical activity			-.408	.003
Participation intensity			.428	.006
ΔR^2 = .109				
Final Model R^2=.840				
TD				
Step	F	df	Beta	p-value
I	7.74	1, 16		
Body fat (%)			-.571	.013
Final Model R^2=.284				

Table 3.5. Regression models for object control skills

DS				
Step	F	df	Beta	p-value
I	20.88	1, 16		
Participation intensity			.752	.000
R^2=.539				
II	18.34	2, 15		
Participation intensity			.643	.000
Knee extensor strength (R)			.395	.016
ΔR^2=.042				
Final Model R^2=.671				
TD				
Step	F	df	Beta	p-value
I	12.29	1, 16		
Gender			.659	.003
Final Model R^2=.399				

Table 3.6. Regression models for running performance

DS				
Step	F	df	Beta	p-value
I	13.14	1, 15		
Generalized physical activity			.683	.002
Final Model R^2=.431				
TD				
Step	F	df	Beta	p-value
I	20.93	1, 16		
Knee extensor strength (L)			-.753	.000
R^2=.540				
II	15.44	2, 15		
Knee extensor strength (L)			-.452	.040
Age			-.443	.043
ΔR^2=.089				
Final Model R^2=.629				

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Chapter IV

Investigating relationships between motor and socio-emotional characteristics in school-aged children with and without Down syndrome

Introduction

In most cases of Down syndrome (DS), every cell in the body is affected. Therefore, it is not surprising that children with this genetic condition experience challenges in several developmental domains. Although DS is most commonly associated with intellectual disability (ID), deficits also exist in socio-emotional functioning. For example, there is considerable evidence that infants and young children with DS show high rates of disorganized and insecure attachment (Ganiban, Barnett, & Cicchetti, 2000; Vaughn, Lefever, Seifer, & Barlow, 1989; van Ijaendoorn, Goldberg, Kroonenberg, & Frankel, 1992). Proper attachment formation is believed to be critical for an infant's willingness to explore her environment (Bowlby, 1969), which, in turn, affects cognitive and socio-emotional development (Campos, Anderson, Barbu-Roth, Hubbard, Hertenstein, & Witherington, 2000). In fact, several researchers have shown that young children with DS have difficulties with social referencing (Kasari, Freeman, Mundy, & Sigman, 1995; Knieps, Walden, & Baxter, 1994) and emotion recognition (Kasari, Freeman, & Hughes, 2001). The extent of socio-emotional difficulties and their correlates for school-aged children with DS are less understood. However, it is important to investigate these issues since socio-emotional problems and

psychopathology in children with DS are associated with severe behavioral disorders in adulthood (McCarthy, 2008).

Substantial research effort has been devoted to understanding the social development of children with DS. Significant research contributions by Kasari, among others, illustrate the complexity of this issue. Freeman and Kasari (2002) found that the majority of children with DS meet criteria for true friendships, including reciprocal interactions and stable relationships beyond six months. Generally speaking, children with DS also willingly engage in social interactions and demonstrate strong interest in their social environments (Fidler & Nadel, 2007; Kasari & Hodapp, 1996). Such findings, in conjunction with their own observations, have led teachers and parents to conclude that social characteristics may serve as a relative strength for children with DS (Gilmore, Campbell, & Cuskally, 2003; Hornby, 1995). However, other research indicates that "sociability" of children with DS may be employed inappropriately to compensate for inadequacies in other areas (e.g., cognition). Specifically, Kasari and Freeman (2001) found that 5- to 12-year-old children with DS looked to an adult and requested help more frequently, and often unnecessarily, compared to children with TD and other forms of ID during puzzle solving tasks. Social understanding may also be impacted in children with DS. In a series of studies, Wishart and colleagues observed lower levels of social cognition (e.g., emotion recognition) in children with DS compared to age- and developmentally-matched controls (Cebula & Wishart, 2008; Wishart, 2007; Wishart, Cebula, Willis, & Pitcairn, 2007). Collectively, these findings suggest that children with DS may overuse social behaviors and have difficulties with social competence.

Though the degree of social impairment in children with DS remains unclear, there is more of a consensus regarding the emotional and behavioral profiles of these individuals. While children with DS display less behavioral problems than children with other disabilities (e.g., Dykens & Kasari, 1997), they demonstrate increased rates compared to non-disabled children. As evidence, Coe and others (1999) used parental report to examine the behavioral characteristics of children ages 6 to 15 with and without DS. They found that one in three children with DS had significant behavioral problems (Coe et al., 1999). Of these, conduct disorders, social withdrawal, attention disorder, and psychotic disorders (e.g., repetitive speech and preoccupation of thought) were most prevalent. Coe also observed that behavioral problems for the DS group were nearly three times higher than the rates observed in children without DS. Findings from Coe are consistent with earlier studies, which identified significant emotional and conduct problems (e.g., aggression, oppositional behavior) in approximately one-fourth to one-third of school-aged children with DS (Cuskelly & Dadds, 1992; Menolascino, 1967; Myers & Pueschel, 1991). Issues of inattentiveness and hyperactivity have also been identified in children with DS. Dykens and Kasari (1997) found that many children with DS have difficulty concentrating. Further, rates of co-morbidity between DS and hyperactivity disorders have been placed in the range of 6% to over 50% (Myers & Pueschel, 1991; Gath & Gumley, 1986; Capone, Goyal, Ares, & Lannigan, 2006). These findings are consistent with mouse models of DS, which also suggest elevated rates of hyperactivity (Altafaj et al., 2001; Galante et al., 2009).

The findings on hyperactivity and behavioral problems in children with DS may indicate an association between motor activity and socio-emotional profiles. There is evidence suggesting that children with DS have deficits with gross motor skills (e.g., Connolly & Michael, 1986) and that they may spend less time performing vigorous physical activity than children with TD(Whitt-Glover, O'Neill, and Stettler, 2006). These are important findings given that numerous studies, primarily in older children and adolescents with TD, have identified social and emotional benefits with increased physical activity. For example, a study by O'dea (2003) found that children and adolescents with TD reported an array of social and psychological benefits (e.g., enhanced mood, self-esteem) from increased physical activity. Similarly, exercise treatments have been shown to increase positive mood and decrease negative mood in 9- and 10-year-old children with TD (Williamson, Dewey, & Steinberg, 2001). Past research also demonstrates that adolescents who engage in regular physical activity and participate in sports have lower anxiety-depression scores, display less social inhibition, and illustrate more positive emotional and behavioral attributes (Kirkcaldy, Shephard, & Siefen, 2002; Donaldson & Ronan, 2006).

Benefits of physical activity have not been limited to "typical" populations, however. Research suggests that children and adults with autism may improve their attention and focus (Rosenthal-Malek & Mitchell, 1997) and decrease problematic behaviors (e.g., stereotypies) with increased physical activity. With respect to socio-emotional and motor relationships in DS, Heller and colleagues (2004) examined psychosocial outcomes of a fitness and health education program for adults (Heller,

Hsieh, & Rimmer, 2004). They found increased exercise self-efficacy, improved life satisfaction, and marginally lower depression following the program. While research in this area is limited for children with DS, evidence from studies of children with other forms of disabilities suggests potential behavioral benefits to sports participation. For example, Gencoz (1997) observed decreases in maladaptive behavior following basketball training in 10- to 14-year-old children with ID. It is unclear if such associations exist in younger school-aged children with DS. Based on the literature, one might expect to find that increased physical activity or motor skills correspond to better social and behavioral profiles. However, it is also plausible that heightened levels of generalized activity could be detrimental to socio-emotional development, given the rates of behavioral problems and hyperactivity in children with DS. It is apparent that more research is needed to understand relationships between the socio-emotional and motor domains in school-aged children with DS.

The current study examined the socio-emotional characteristics of school-aged children with DS and investigated their relationships with gross motor skills, physical activity, and participation in recreational and leisure activities. A control group of children with TD matched on age and gender was included for comparison. Data on gross motor skills, physical activity, and participation was analyzed previously (see Chapters 2 and 3). I hypothesized that children with DS would demonstrate significantly more socio-emotional deficits than children with TD. In addition, I expected to find that socio-emotional problems (e.g., externalizing behaviors) would be negatively correlated with motor skill proficiency and physical activity participation in both groups. Finally, I

hypothesized that generalized physical activity would be associated curvilinearly (U-shaped) with social profiles and behavioral problems in children with DS such that too little or too much activity would correspond to poorer socio-emotional characteristics.

Methods

Participants

The sample consisted of twenty children with DS and twenty children with typical development (TD) who were matched on age and gender. Participants with DS were recruited from parent support groups in Michigan and northern Ohio. The TD sample was largely recruited from families and friends of the children with DS. A local summer camp and a University of Michigan research website were also utilized as secondary recruitment tools. Exclusion criteria for both groups included major health conditions that would be counter to mild exercise (e.g., cardiac problems, uncontrolled seizures). Neither race nor gender prohibited children from participating in the study as long as they met study criteria.

Procedures and measures

All children came to the motor development laboratory at the University of Michigan for gross motor skill assessment. Upon arrival at the laboratory, informed consent and child assent (when appropriate) was obtained and documented. Parents were also asked to fill out a supplemental information sheet prior to child testing to gather socio-demographic information. At the conclusion of this visit, children and their families were given instructions for the activity and socio-emotional assessments, which

were completed in their daily living environments (see below). Participating families received $20. All procedures were approved by the University of Michigan Medical Internal Review Board. The following assessments were performed.

I) Socio-emotional characteristics

The school-aged version (ages 6-18) of the Child Behavior Checklist (CBCL) (Achenbach & Rescorla, 2001) was used to assess socio-emotional characteristics of the participants. The CBCL is a widely accepted questionnaire that utilizes parental report. It has been used previously with pediatric populations of DS (Dykens & Kasari, 1997; Pueschel, Bernier, & Pezzullo, 1991) and reports high levels of inter-rater reliability (0.93-0.96). The instrument asks about the child's activities, social relations, and school performance, leading to competency scores in each of these areas as well as a total competency score. The CBCL also contains 113 items concerning the child's emotions and behaviors over the past six month. Each item is scored as 0 = not true at all, 1 = somewhat or sometimes true, or 2 = true or often true. As a result, scores are calculated for 8 cross-informant syndromes (aggressive behavior, anxious/depressed, attention problems, rule-breaking behavior, social problems, somatic complaints, thought problems, and withdrawn/depressed), externalizing problems (aggressive and rule-breaking behavior), internalizing problems (anxious/depressed, withdrawn/depressed, and somatic problems), and total problems. Profiles for several Diagnostic and Statistical Manual of Mental Disorders (DSM) can also be obtained through the CBCL but we chose not to pursue them in the current study. Parents were

provided instructions on filling out the CBCL prior to leaving the laboratory. They completed the form at home and sent it back in a self-addressed, postage-paid envelope that we provided.

B) Gross motor skills, physical activity, and participation

Procedures for the gross motor skill, physical activity, and participation assessments were the same as those described in Chapter 3. Briefly, gross motor skills were assessed using the Test of Gross Motor Development (2nd ed.) (TGMD-2) (Ulrich, 2000). Children wore a combined activity and heart monitor (Actiheart, CamNtech, U.K.) for four days to estimate generalized physical activity (GenPA). Participation in recreational and leisure activities data was measured with the Children's Assessment of Participation and Enjoyment (CAPE) (King et al., 2007). Please refer to the methods section of Chapter 3 for specific details of the procedures and data reduction techniques involved with these assessments.

Statistics

The socio-demographic variables of age, gender, parental education, number of siblings, and family income were included in analysis. Dependent variables of the socio-emotional domain included: competency scores (activity, social, school, total); total number of internalizing and externalizing problems; and scores for the 8 cross-informant syndromes (aggressive behavior, anxious/depressed, attention problems, rule-breaking behavior, social problems, somatic complaints, thought problems, and withdrawn/depressed). Raw scores of the locomotor and object control skills sections

of the TGMD-2, along with scores on the individual items, were selected as dependent variables representing the motor domain. For the CAPE, scores for 5 dimensions of participation (diversity, intensity, who, where, and enjoyment) were analyzed. These variables are referred to as DivPar, IntPar, WhoPar, WherePar, & EnjPar, respectively. Finally, the dependent variable of GenPA was selected as an indicator of generalized physical activity under everyday living conditions.

Descriptive statistics by group (DS, TD) were conducted on all of the aforementioned variables. T-tests and chi-square statistics on socio-demographic data were performed to determine if the groups differed on any of these variables. Group comparisons using t-tests were also conducted on the socio-emotional, motor, activity-related variables to address the hypotheses. In addition, effect sizes for these dependent variables were calculated and interpreted according to Cohen (1988) (0.2=small, 0.5=medium, 0.8=large). Please note that descriptive statistics for the motor- and activity-related variables were performed in Chapter 2. To examine linear relationships between the socio-emotional and motor (motor skills + activity) domains, bi-variate correlations (Pearson) were performed. Correlation analyses included the socio-emotional variables of social competence, social problems, attention problems, internalizing problems (anxious/depressed + withdrawn/depressed + somatic problems), and externalizing problems (aggressive + rule-breaking behavior). We investigated our hypothesis of a curvilinear (U-shaped) relationship between GenPA and the aforementioned CBCL variables by performing linear regression analyses of GenPA

and $GenPA^2$ on the CBCL variables and comparing it to models with GenPA as a lone predictor. Alpha was set at $p<0.05$ for all analyses.

Results

Sample characteristics

Forty children (DS=20, TD=20) between the ages of 6 and 10 years were included in the study. The participants were successfully matched on age and gender, with both groups consisting of 4 girls and 16 boys and reporting an average age of 7.9 years. There were no significant differences between the groups on any of the socio-demographic variables. Children in both groups had between two and three siblings, on average. Chi-square statistics indicated no significant differences on parental education or family income, although families of children with DS reported slightly lower education but higher income compared to families without DS. Table 4.1 demonstrates the sample's socio-demographic characteristics.

Socio-emotional characteristics

Results from the CBCL (Table 4.2) indicated that children with DS scored significantly lower than their unaffected peers on all competency scores (social, school, and total, $P<.001$; activities, $p<.01$). The DS group also demonstrated significantly more externalizing behaviors ($t(38)=4.03$, $p<.001$), with greater rule-breaking ($t(38)=3.69$, $p=.002$) and aggressive ($t(38)=3.69$, $p=.001$) behaviors. Likewise, group differences were observed for social ($t(38)=6.67$, $p<.001$), thought ($t(38)=5.65$, $p<.01$), and attention

(t(38)=5.65, p<.001) problems in the direction favoring the TD group. In contrast, there were no group differences on internalizing behaviors or any of its subscales (anxious/depressed, withdrawn, somatic problems), although moderate effect sizes were observed for anxious/depressed and somatic problems in favor of the TD group. Children with DS were most affected in the areas of externalizing behaviors, social problems, and attention problems. As a whole, children with TD demonstrated very few socio-emotional problems.

Gross motor skills, physical activity, and participation

Descriptive data for gross motor skills, physical activity, and participation for the two groups were conducted in Chapter 2 and presented in Chapters 2 (Tables 2.3, 2.4) and 3 (Table 3.1). Please refer to Chapter 3 for details regarding these results, as the variable names in that study match the terminology used in the current study. The descriptive data for motor skills, generalized physical activity, and participation are presented here again (see Table 4.3) because these variables were included in the correlation and regression analyses. Briefly, results indicated significant group differences favoring the children with TD on locomotor skills (t(37)=-8.67, p<.001), object control skills (t(38)=-6.24, p<.001), and diversity of participation ((t(38)=-2.21, p=.033) in recreational and leisure activities. The groups were not significantly different, however, on any other dimensions of participation or on generalized physical activity.

Socio-emotional characteristics vs. motor skills, participation, and generalized physical activity

Results from the bivariate correlations are presented in Table 4.4. In the DS group, social competence was related to gross motor skills and participation in recreational and leisure activities. Specifically, social competence was positively correlated with locomotor scores ($r(18) = .541$, $p<.05$), object control scores ($r(18) = .694$, $p<.01$), diversity of participation ($r(18) = .683$, $p<.01$) and intensity of participation ($r(18) = .552$, $p<.05$) for the children with DS. Figures 4.1 and 4.2 show the relationships between social competence and both object controls skills and participation diversity, respectively, in the DS group. For the children with typical development, social competence was also positively correlated with diversity of participation ($r(18) = .603$, $p<.01$) and intensity of participation ($r(18) = .631$, $p<.01$). Unlike the DS group, though, gross motor skills were not significantly associated with social competence. We failed to observe linear or curvilinear associations between social competence and GenPA in either group.

In contrast to the results on social competence, social problems were not significantly correlated with gross motor skills or participation for the DS group. We also did not observe linear relationships between social problems and GenPA in the children with DS. However, a regression model ($F(2,16)=3.315$, $\beta1=-1116.89$, $\beta2=19983.49$, $R^2=.305$) estimating a curvilinear, U-shaped association between GenPA and social problems showed a strong trend towards significance ($p=.054$) (see Figure 4.3). This suggests that too much or too little generalized physical activity may be associated with increased social problems for children with DS in this sample. For children with TD, social problems were not associated with generalized activity. We observed that social

problems were related to motor skills and social aspects of participation in the TD group, though. As evidence, social problems were negatively correlated with object control skills ($r(18) = -.577$, $p<.05$) and both the who ($r(18) = -.452$, $p<.05$) and where ($r(18) = -.484$, $p<.05$) dimensions of participation.

Surprisingly, we observed very few meaningful relationships between the remaining socio-emotional dimensions and our motor and physical activity variables in either group. The lone exception was a strong trend for a negative correlation between externalizing behaviors and object controls skills ($r(18) = -.440$, $p=.052$) in the TD group. The nature of the relationships was such that higher object control scores corresponded to less externalizing problems. For the DS group, motor skills and physical activity were not significantly correlated with externalizing problems, internalizing problems, or attention problems. We also explored curvilinear associations between these variables and found no significant relationships. Of note, examination of the raw data for externalizing behaviors and generalized physical activity suggests a U-shaped relationship between these variables for the DS group (see Figure 4.4). However, a curvilinear model to estimate such a relationship was non-significant ($F(2,16)=2.308$, $\beta1=-1926.34$, $\beta2=34623.73$, $R^2=.224$, $p=.132$).

Discussion

The current study investigated the socio-emotional profiles of school-aged children with and without DS and examined correlates to these profiles from the motor and activity domains. Results indicated that children with DS had significantly lower

social competence scores and more behavioral difficulties compared to children with TD, with externalizing behaviors, attention, and social problems being the most affected areas. In addition, our findings demonstrated the importance of participation in recreational and leisure activities for both groups, evidenced by the positive relationships we observed between participation and social competence. For children with DS, gross motor skills were also associated with social competence. Results suggest that generalized physical activity, as opposed to motor skills or participation, may be more closely related to social and behavioral problems in children with DS as evidenced by trends for curvilinear associations between these variables. This contrasts our findings from children with TD, who demonstrated significant negative correlations between gross motor skills and specific behavioral problems (e.g., externalizing behaviors).

Our results from the CBCL suggest that children with DS face challenges in terms of their social development when compared to their peers without DS. Specifically, findings revealed that children with DS had difficulties with social competence. On the CBCL, social competence is conceptualized by the number of organizations the children are involved in along with the quantity and quality of their relationships with other children (e.g., number of friends they have, how well they get along with other children, etc.) Similarly, children in the DS group demonstrated significantly higher scores for social problems, which included items such as being overly dependent on others, getting jealous easily, and preferring the company of younger children. Such findings are consistent with previous research by Kasari and Freeman (2001), which indicated that

children with DS overuse social behaviors and often rely unnecessarily on others. However, social development has also been considered to be a relative strength of children with DS. For example, Freeman and Kasari (2002) found that the majority of children with DS are able to develop friendships with other children. While it may be true that children with DS are able to establish and even maintain friendships, our results suggest that these relationships may be few in number or that the quality may not be optimal (e.g., more jealousy, preferring younger children, etc.). Therefore, it is critical for children with DS to be exposed to a wider range of activities that will promote positive social interactions with their peers.

In addition to social difficulties, children with DS in our sample also had elevated scores in the areas of maladaptive behaviors and attention problems. Results showed that externalizing behaviors, such as aggression and rule-breaking, were more problematic than internalizing behaviors (e.g., depression) in school-aged children with DS. These results are consistent with findings from Dykens and Kasari (1997) who used the CBCL to examine behavioral problems in children ages 4 to 19 years with DS. They reported internalizing and externalizing mean scores of 6.3 and 11.4, respectively. We observed similar differences between the two types of behaviors although our mean scores were lower (internalizing = 4.3, externalizing = 8.4). Discrepancies in magnitude likely result from age differences between our studies, considering that Dykens and Kasari found more behavioral problems with increased age in their sample of children and adolescents with DS. Dykens and Kasari also observed that children with DS had

difficulty concentrating, which is consistent with our observation of increased attention problems in the DS group.

There are several potential explanations for the socio-emotional difficulties in children with DS. One of the most compelling involves alterations in brain structure early in infancy. For example, Nadel (1999) reported that within the first months of life, infants with DS exhibit a variety of brain abnormalities compared to infants with TD. Most notably, perhaps, is decreased frontal lobe size. It is well documented that the frontal lobe is critical to emotional regulation. Interestingly, several studies show that there are no differences in the brains between individuals with and without DS at birth, however (e.g., Schmidt-Sidor, Wisniewski, Shepard, & Sersen, 1990). These findings suggest that something occurs within the first few weeks of life to alter, and possibly suppress, brain development in infants with DS. Several researchers have demonstrated reduced motor activity in infants with DS (e.g., Mazzone, Mugno, & Mazzone, 2004; Ulrich & Ulrich, 1995). Further, movement-based exploration of one's environment has been implicated in cognitive and socio-emotional functioning starting in infancy (Campos et al., 2000). Therefore, it is plausible that reduced motor activity in infants with DS contributes to early brain alterations, which in turn, may constrain socio-emotional development.

Our results suggest that motor and socio-emotional development may also be highly linked in school-aged children with DS. As evidence, we observed that social competence was positively correlated with locomotion and object control scores in the

DS group. These findings, in conjunction with our descriptive results on social and motor skill profiles, suggests that early intervention and physical education programs should prioritize gross motor skill instruction to promote motor and social development during the school-aged years for children with DS. It is also important for parents to recognize that gross motor skill competency could have a positive impact on their child's peer relationships and social development as the child ages. Therefore, they should advocate more strongly for gross motor skill instruction during the individualized education programs (IEP) process. In fact, gross motor skill training programs have previously been shown to improve socio-emotional profiles for youth with DS (Gencoz, 1997). Our data indicates that, in addition to gross motor skills, frequent participation in a variety of recreational and leisure activities is also associated with social competence for children with DS. Collectively, our findings suggest that children with DS should receive gross motor skill instruction and utilize these skills in various recreational settings to optimize social development.

Fostering gross motor skills and enhancing participation should be a goal for all children, regardless of whether or not they have a disability. Our results indicate that motor skill competency, in particular, may have an even broader impact for children with TD than it does on individuals with DS. In addition to being associated with social profiles, gross motor skills were significantly related to externalizing behaviors in the TD group, such that better object control scores corresponded to fewer problems. Contrary to expectations, we did not find evidence of a relationship between motor skills and internalizing behaviors for either group. A recent study by Rigoli and

colleagues may explain this observation. They found that self-perception mediated a relationship between motor coordination and internalizing behaviors in individuals with TD (Rigoli, Piek, & Kane, 2012). Therefore, given the limited sample in the current study, it would be difficult to uncover an indirect relationship of this nature. Rigoli's findings also indicate that it may be beneficial to examine whether gross motor skill competency or participation influence self-perceptions for children with and without DS. However, previous research indicates that perceived competence is not reliable in youth with intellectual disabilities (Ulrich & Collier, 1990). This is consistent with anecdotal evidence from our laboratory where we observed that children with DS tend to over-report their involvement and enjoyment in recreational and leisure activities. As a result, we would not expect associations between motor skill competency and self-perceptions for children with DS. Further, we are reluctant to use any self-report measures for participants with ID. The question of perceived competence and gross motor skills should be explored for school-aged children with TD and in children with other forms of disabilities where cognitive functioning may not be compromised (e.g., ADHD).

Besides the lack of an association between motor skills and internalizing behaviors, we were also surprised to find that attention problems were not related to gross motor skills or participation. Given previous findings of motor skill deficits in children with ADHD (Harvey & Reid, 1997) and numerous indications of hyperactivity in individuals with DS (Myers & Pueschel, 1991; Gath & Gumley, 1986; Capone, Goyal, Ares, & Lannigan, 2006), we anticipated an inverse relationship between attention

problems and gross motor skills. However, our failure to observe such an association is not entirely unexpected for two reasons. First, the CBCL captures attention problems rather than hyperactivity. Second, hyperactivity and inattentiveness do not always coincide, evidenced by the fact that one can be diagnosed with hyperactivity disorder without having an attention disorder. Results also indicated that participation in recreational and leisure activities was not related to attention. Therefore, it appears that we should consider potential correlates to attention problems for children with DS that were not included in the current study. Past findings indicate that a variety of socio-economic factors (e.g., living in an urban environment), genetics, and chronic health problems are associated with attention problems in children (Szatmari, Offord, & Boyle, 1989). Future studies should determine if these factors are also related to attention problems in children with DS. Research investigating associations between hyperactivity and motor skills for this population is also needed given the poor gross motor skill profiles and evidence of DS-hyperactive co-morbidity.

In addition to its potential association with motor skills, hyperactivity may be related to social problems and externalizing behaviors in school-aged children with DS. Some of the most interesting findings in the current study involved potential curvilinear associations between generalized physical activity and social and externalizing problems. Previously analyzed activity data in Chapters 2 & 3 suggests that some children with DS exhibited extremely high levels of generalized physical activity, possibly suggestive of hyperactivity. This claim is supported by various studies in the literature involving both humans with DS and mouse models (e.g., Capone et al., 2006; Galante et

al., 2009). As a result of our findings and the accumulated evidence, we hypothesized that GenPA would demonstrate U-shaped relationships with social and behavioral problems for the DS group whereby too little or too much generalized activity would be associated with more problematic behaviors. Our results indicated that, in fact, GenPA showed a curvilinear relationship with social problems. Though less convincing, it appears that GenPA may demonstrate a similar relationship with externalizing behaviors. If true, this suggests there is an optimal range of generalized physical activity for promoting socio-emotional health in school-aged children with DS. Therefore, managing generalized physical activity in youth with DS needs to be treated on an individual basis, with a universal goal of increasing functional, health promoting physical activities.

We must be cautious in our interpretations of the current data, however, due to several limitations. First, we utilized a small sample of convenience. It is certainly possible that some of the non-significant associations we observed, such as the curvilinear relationship between GenPA and externalizing problems in the DS group, may have been significant with a larger sample. Conversely, it is plausible that the curvilinear associations, in particular, could have resulted from a few outliers. We believe the latter is unlikely, though, given previous reports of hyperactivity in DS. In addition, our GenPA data needs to be viewed conservatively at this time since the equations we used have not been validated for youth with DS. Finally, children in the TD group reported very few behavioral problems. They also demonstrated motor skill scores above the age-expected norms reported in the TGMD-2 manual. Taken together,

it is highly probable that the children in our TD sample were a group of highly skilled, well-adjusted individuals who were not representative of the general population. For these reasons, one must exhibit additional restraint when interpreting our relationships in the TD sample.

Despite these limitations, we believe that the current study provides significant contributions to the literature on socio-emotional development in children with DS. In 2007, Dykens conducted a lengthy review on psychiatric and behavioral disorders in individuals with DS. She identified three critical directions for future research in this area. First, socio-emotional problems in DS should be studied from a lifespan perspective. Second, risk and protective factors for psychiatric and behavioral problems need to be identified. Third, interventions that both decrease symptoms and increase well-being should be investigated. We believe that the current study has implications for such concerns and lays a strong foundation for pursuing these three lines of research. By identifying socio-emotional challenges in young children with DS, we may be able to reduce some of the severe behavioral problems observed in adults with DS since deficits in childhood have been found to be predictive of problems in adulthood (McCarthy, 2008). Further, our data suggests that gross motor skills, participation, and even generalized activity, to some degree, may serve as protective or risk factors for socio-emotional health in children with DS. For example, it is possible that physical activity participation both constrains and is constrained by social and behavioral characteristics. Similarly, we propose that interventions to promote gross motor skills, enhance participation, and optimize physical activity are all highly plausible solutions for

decreasing problematic behaviors while, at the same time, promoting well-being in youth with DS. Collectively, results of the current study have wide ranging implications for future research and physical education practice by demonstrating the inter-relatedness between the motor, activity, and socio-emotional domains.

Table 4.1. Participant characteristics

	DS	TD
Gender	4 Females, 16 Males	4 Females, 16 Males
Age, $\bar{x}$ (sd)	7.94 (1.25)	7.94 (1.57)
Number of siblings, $\bar{x}$ (sd)	2.38 (1.41)	2.95 (1.67)
Birth order, $\bar{x}$ (sd)	2.62 (1.63)	2.15 (1.27)
Mother's education (average)	Some college	Bachelor's degree
Father's education (average)	Bachelor's degree	Bachelor's degree
Household income (average)	$81,000-$100,000	$61,000-$80,000

Table 4.2. Socio-emotional characteristics

	$\bar{x}$ (sd)		P Value	Effect Size
	DS	TD		
Competencies				
Activity Competence	9.40 (3.08)	11.68 (1.81)	0.007	-0.89
Social Competence	6.38 (2.51)	9.38 (1.68)	0.000	-1.40
School Competence	2.22 (0.70)	5.58 (0.46)	0.000	-5.65
Total Competence	18.00 (4.81)	26.62 (2.96)	0.000	-2.16
Externalizing Problems	8.35 (1.26)	2.80 (2.52)	0.000	1.27
Rule-Breaking Behavior	2.20 (1.70)	0.80 (0.89)	0.002	1.03
Aggressive Behavior	6.15 (4.56)	2.00 (2.20)	0.001	1.16
Internalizing Problems	4.30 (4.86)	4.15 (4.40)	0.919	0.03
Anxious/Depressed	1.40 (1.81)	2.75 (3.22)	0.111	-0.52
Withdrawn/Depressed	1.10 (1.65)	0.65 (1.27)	0.340	0.30
Somatic Complaints	1.80 (2.72)	0.75 (1.07)	0.117	0.50
Social Problems	5.00 (2.81)	0.55 (1.00)	0.000	2.11
Thought Problems	2.80 (2.69)	0.85 (1.27)	0.006	0.93
Attention Problems	7.55 (4.12)	1.80 (1.94)	0.000	1.78

Table 4.3. Motor skills, physical activity, & participation

	$\bar{x}$ (sd)		P Value	Effect Size
	DS	TD		
Motor skills				
Locomotor score	21.16 (11.88)	44.95 (3.02)	0.000	-2.74
Object control score	24.80 (12.30)	42.85 (4.00)	0.000	-1.97
Physical activity				
GenPA (kcal/kg/min)	0.024 (0.008)	0.023 (0.007)	0.640	0.15
Participation				
Diversity	31.30 (7.37)	36.05 (6.19)	0.033	-0.70
Intensity	2.76 (0.67)	3.05 (0.64)	0.163	-0.45
Who	2.36 (0.65)	2.69 (0.52)	0.089	-0.55
Where	2.72 (0.78)	2.75 (0.52)	0.887	-0.05
Enjoyment	4.03 (0.43)	3.86 (0.28)	0.144	0.47

Note: Physical activity data (DS, N=19)

Table 4.4. Correlations: Socio-emotional characteristics vs. motor skills, physical activity, and participation

		Social competence		*Social problems*		*Externalizing problems*		*Internalizing problems*		*Attention problems*	
		DS	TD	DS	TD	DS	TD	DS	TD	DS	TD
Gross motor skills											
Locomotor	*r*	.541*	-.239	.065	.184	.379	.102	-.074	.135	-.305	.034
Object control		.694**	.071	.204	-.577*	.246	-.440*	-.019	-.234	-.177	-.194
Physical activity											
GenPA		-.163	.010	-.089	-.171	-.064	-.262	.375	-.082	.284	-.112
Participation											
Diversity		.683**	.603**	.201	-.260	.234	-.252	.140	-.278	-.146	-.245
Intensity		.552*	.631**	.193	-.235	.236	-.335	.150	-.318	-.171	-.089
Who		.356	.361	.194	-.524*	.395	-.241	.311	-.214	.100	.180
Where		.270	.373	.065	-.484*	.267	-.358	.281	-.433	.128	.016
Enjoyment		.245	-.073	.216	.365	.264	.278	.102	.270	.023	.150

Note: $p < 0.05$ (*), $p < 0.01$ (**), $p < 0.001$(***)

Figure 4.1. Social competence vs. object control scores

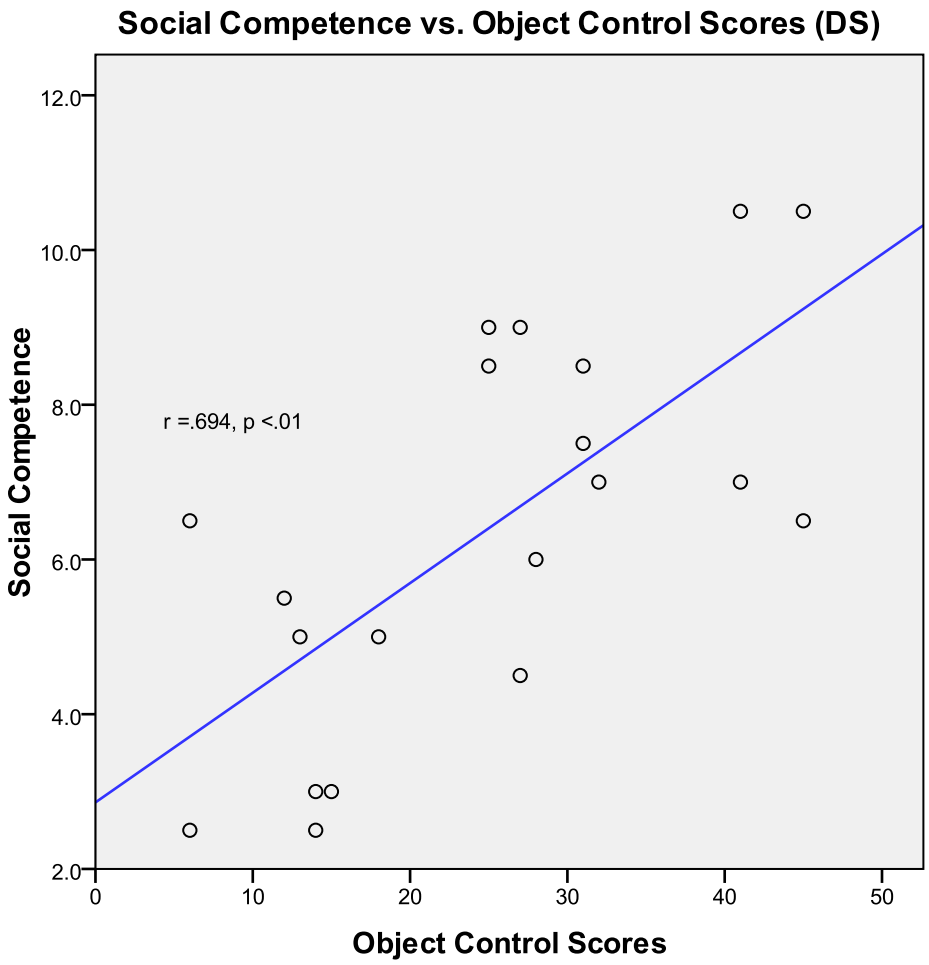


Figure 4.2. Social competence vs. participation

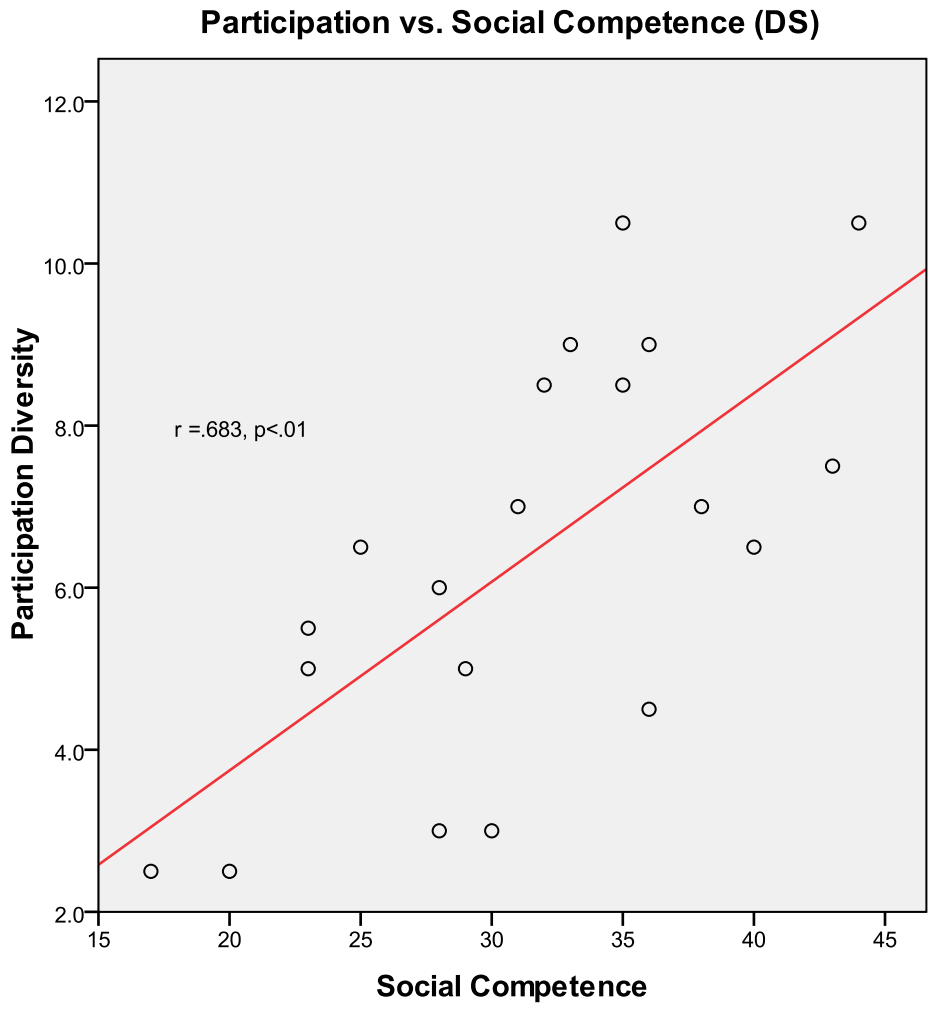


Figure 4.3. Social problems vs. generalized physical activity

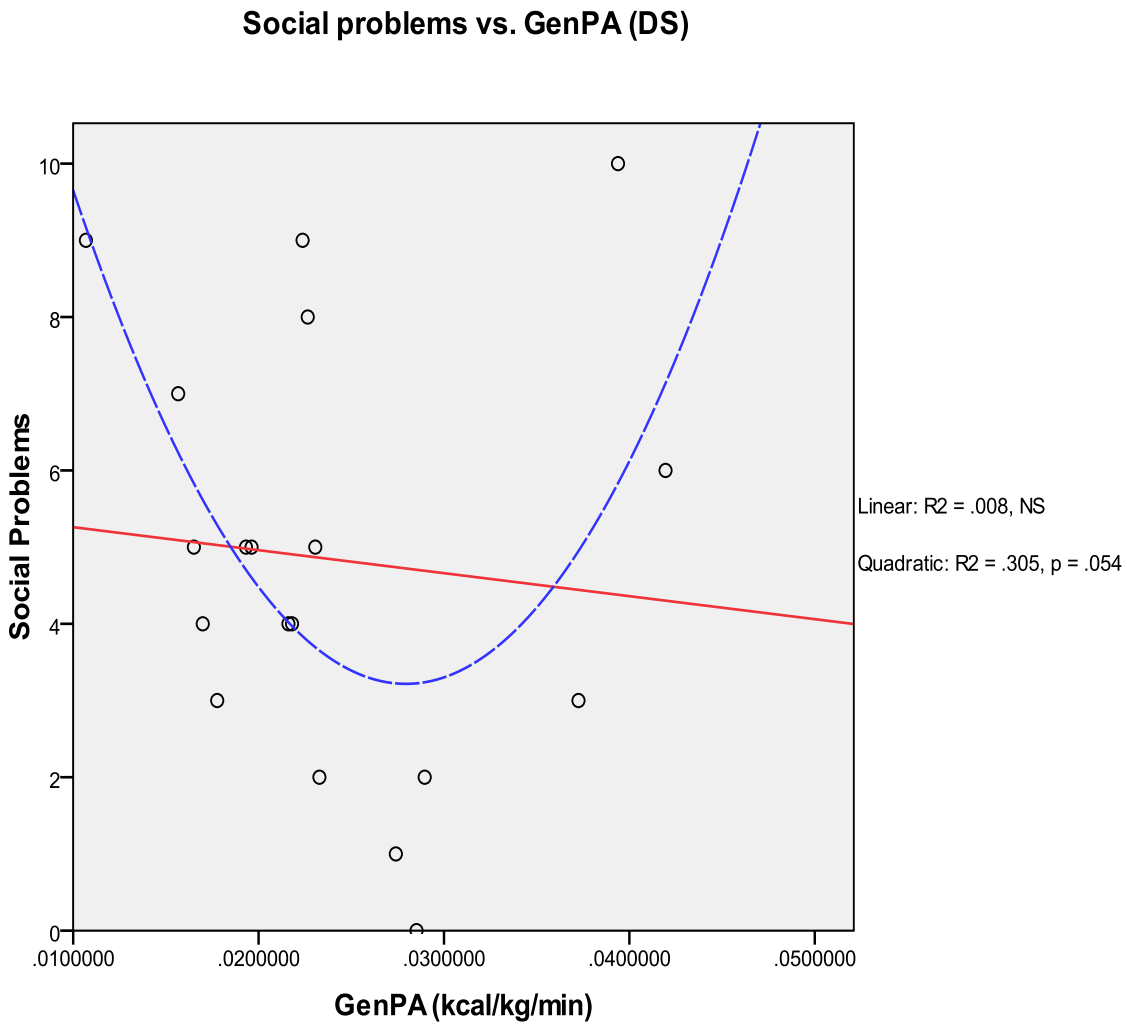
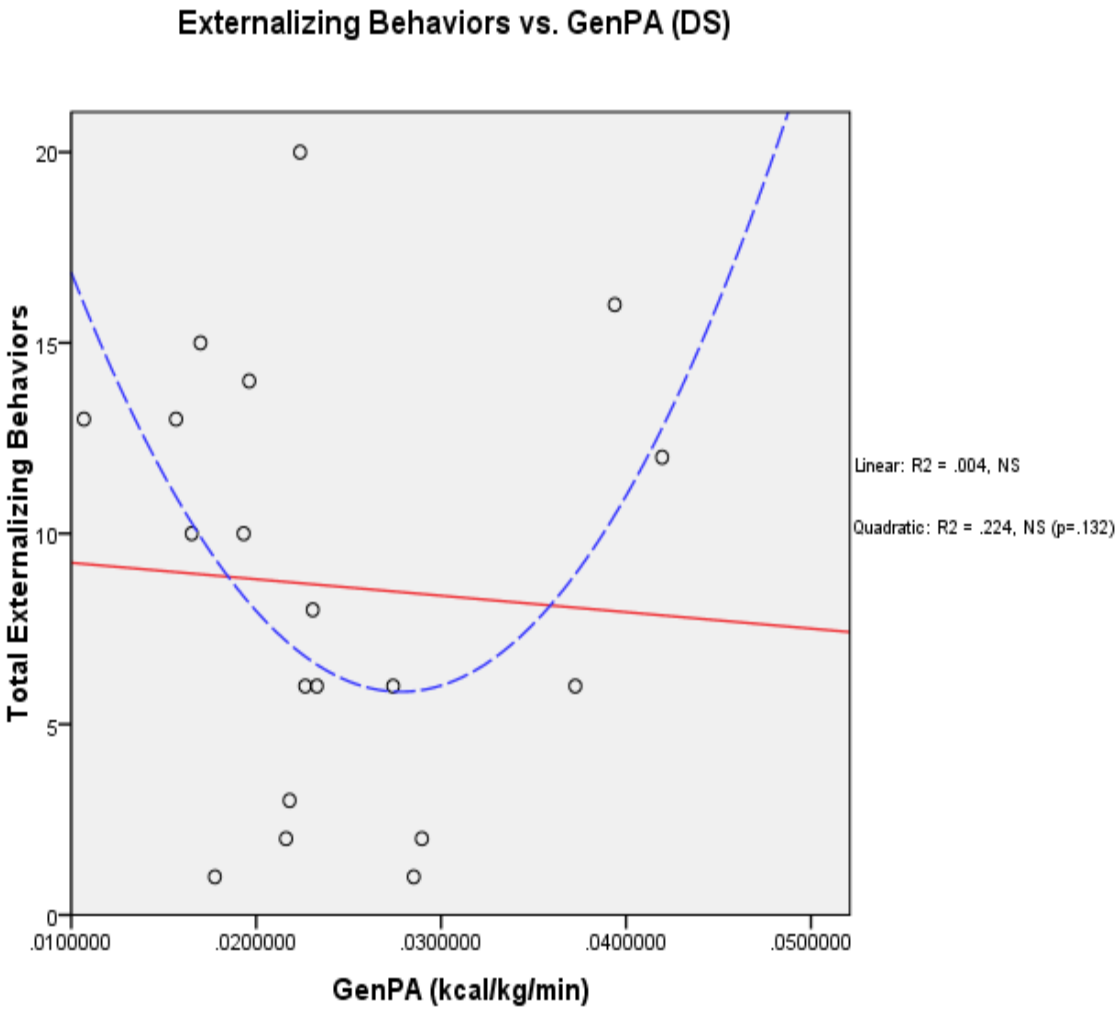


Figure 4.4. Externalizing behaviors vs. generalized physical activity



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