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**ASSESSMENT OF COORDINATION AND PROPRIOCEPTION IN YOUTH WITH
AUTISM SPECTRUM DISORDER**

by

Taylor Kristina Jones
B.S. May 2018, Old Dominion University

A Thesis Submitted to the Faculty of
Old Dominion University in Partial Fulfillment of the
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ABSTRACT

Autism spectrum disorder (ASD) is characterized by social and communicative delays. It is known that those with ASD exhibit lower activity levels and decreased proprioception to some extent. The biomechanics of movement in ASD has not been assessed thoroughly enough to provide information on ASD specific movement patterns, and no studies have been performed examining work and recovery. The purpose of this study is to examine whether 1) inter-limb and intra-limb coordination patterns during walking and running differ between youth with ASD and neurotypical sex, age, and BMI-matched controls. Youth with ASD (N=8) and their BMI, age, and sex matched controls (N=8) performed walking at their self-selected speed and also at a standardized speed of 1.3 m/s for at least five trials each. An eight-camera motion capture system was used to collect three-dimensional (3D) kinematics for each subject. After in-lab data collection, subjects were given an accelerometer to wear to measure physical activity levels over a span of at least four days. To analyze the data, angle-angle plots were constructed for the left upper-arm and right thigh, and right shank-foot. Vector coding was used to obtain coupling angle and coupling angle variability information. No significant differences existed in coordination patterns or physical activity levels between the two groups. Upper-arm dominance and anti-phase upper arm/thigh patterns were significantly related to minutes of vigorous physical activity (Rho: -0.63, $p < 0.01$ & Rho: 0.58, $p = 0.02$, respectively). According to these results, there are no differences in coordination between those with and without ASD.

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DEDICATION

First and foremost, this paper is dedicated to my parents and siblings who have encouraged me along my journey of schooling and especially now in grad school. This is also dedicated to all of my friends, loved ones, classmates, and professors who have helped me in any way these last few years at ODU.

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CHAPTER ONE

INTRODUCTION

Autism spectrum disorder (ASD) is a social developmental disability, characterized by deficits in communication and social interaction (American Psychiatric Association, 2013; Baio, Wiggins, Christensen, Maenner, Daniels, Warren, ... & Dowling, 2018). According to the APA (2013), those with an established diagnosis of autistic disorder, Asperger's disorder, or pervasive development disorder from previous years are now given the diagnosis of ASD. ASD is recognized by persistent deficits in social communication and interaction, and restrictive or repetitive patterns of behavior (APA 2013). The most widely known symptoms and effects of ASD include difficulty with language and interaction with others, restrictive and repetitive behaviors, and different self-stimulating or self-regulatory movements such as rocking and arm flapping (Kindregan, Gallagher, & Gormley, 2015). As of 2020, the CDC reported that about one in 54 children at eight years old is diagnosed with ASD (CDC 2020). Most research studies about ASD thus far pay most attention to the social deficits and developmental delays that occur with this condition without researching the possible physical aspects of the diagnosis.

Movement Deficiency

Until the past decade or so, ASD was mainly deemed as a social and communicative disorder (Kindregan et al, 2015). More recent research suggests that ASD not only affects communication and social interactions, but may affect movement (Dufek, Eggleston, Harry, & Hickman, 2017; Eggleston, Harry, Hickman, & Dufek, 2017). Some of the most widely known movement challenges that occur among those with ASD include, but are not limited to, altered muscle recruitment patterns (Damasio & Maurer, 1978; Kohen-Raz, Volkmar, & Cohen, 1992), impaired postural control (Kohen et al., 1992), dyspraxia (clumsiness) (Green, Baird, Barnett,

Henderson, L., Henderson, S., & Huber, 2002; Dziuk, Larson, Apostu, Mahone, Denckla, & Mostofsky, 2007; MacNeil & Mostofsky, 2012; Mostofsky, Dubey, Jerath, Jansiewicz, Goldberg, & Denckla, 2006; Provost, Heimerl, & Lopez, 2007), and deficits in locomotion and balance (Green et al., 2002). Studies have also noted that individuals with ASD exhibit less motor coordination, mainly at the visual level (Ghaziuddin & Butler, 1998; Fournier, Hass, Naik, Lodha, & Cauraugh, 2010); however, these previous examinations have only been performed via visual inspection. Symptoms of ASD may initially present as movement disorders rather than communicative disorders, considering that the common ages for diagnosis of this particular disorder is 2-4 years (Eggleston et al., 2017). A study performed by Weiss et al. (2013) comparing teenagers and younger adults with and without ASD found that variables such as step length, cadence, velocity, and gait cycle time were all lower in the ASD group compared to the control group. Though, considering that ASD varies from person to person, most studies are inconclusive in determining exactly what differences occur in those with ASD, and why.

Problem Statement

Those with ASD may experience motor impairments (Riquelme, Hatem, & Montoya, 2016; Weiss, Moran, Parker, & Foley, 2013), that may likely translate to impairments in walking gait. Proprioception plays a major role in walking, as limb awareness, position sense, and the ability to constantly shift weight are the main components needed for locomotion (Beets, Macé, Meesen, Cuypers, Levin, & Swinnen, 2012; Blanche et al., 2012). However, current literature has yet to determine if inter-limb coordination during locomotion differs between persons with ASD and neurotypical counterparts. In addition, it is unknown if these measures relate to a person's physical activity level.

The purpose of this study is to examine whether 1) inter-limb and intra-limb coordination patterns during walking differ between more and less physically active youth with ASD and neurotypical sex, age, and BMI-matched controls. Specifically, we compared 1) contralateral arm/leg swing, and 2) foot and shank coordination using modified vector coding and 3) between youth (defined by above and below the mean activity level) with ASD and matched controls. It is hypothesized that those with ASD will 1) have decreased inter-limb and intra-limb coordination, and 2) coordination were decreased all persons with lower physical activity levels.

Delimitations

The participation sample included boys and girls aged 13-18 years old that have a clinical ASD diagnosis. Inclusion criteria for participation required all participants have: no lower extremity injury within the last six months prior to the start of the study, no history of diagnosed joint disease or joint surgery, and should not be pregnant. In order to rule out cognitive disability in both populations, participants' IQ must be greater than 70. Lastly, all participants must be able to walk, run, and balance on one and two limbs.

Limitations

As with all laboratory research assessments, some study limitations must be acknowledged. One significant limitation is that individuals with ASD are not always willing to be involved in research that includes manual palpation, which was required in this study in order to record motion capture information. This could possibly decrease the pool of participants that were willing to participate. To alleviate any concerns participants may have had, visual references were used (via an iPad) with each participant prior to beginning the study protocol. As some persons with ASD favor visual stimuli, the visual reference should have assisted in participants' understanding and preparation for the study.

Another possible limitation is physical activity levels among both the ASD and neurotypical populations. It is known that most kids and adolescents are not engaging in the proper amount of moderate-to-vigorous physical activity as defined by the CDC, which could make it hard to differ between high and low levels of physical activity. When performing an analysis on the data, high and low physical activity groups may have to be defined in a different way.

Lastly, variations in movement patterns may exist amongst the function levels of ASD. This study did not control for specific function levels, so as not to limit the possible sample size. However, we obtained documentation on diagnosed level of function that can be used to assist in furthering our understanding of the findings.

CHAPTER TWO

LITERATURE REVIEW

The purpose of this thesis research is to assess the various kinematic elements of walking, proprioception, and physical activity in adolescents with Autism Spectrum Disorder. This review of literature will cover: 1) An overview of ASD, 2) Physical activity differences in those with ASD, 3) What is known about these individuals walking gait compared to that of normal developing controls, and 4) Proprioception in individuals with ASD.

Overview of Autism Spectrum Disorder

Autism spectrum disorder has been widely defined as a lifelong developmental disability that elicits deficits most notably in social communication and interaction (APA 2013). These deficits include, but are not limited to, deficits in social-emotional reciprocity, failure to initiate or respond to social interactions, deficits in nonverbal communicative behaviors, lack of facial expressions and non-verbal communication (American Psychological Association, 2013). Other possible patterns sometimes seen in individuals with ASD include stereotyped or repetitive movements, inflexible adherence to routines, abnormal and intense fixated interests, and hyper- or hyporeactivity to sensory input (American Psychological Association, 2013). Recent studies state that the incidence rate is 18.5 per 1000 children, or one in 54 (Maenner et al., 2020). Historically, children with ASD were overwhelmingly non-Hispanic white males that come from two-parent families living in large metropolitan statistical areas (Zablotsky, Black, Maenner, & Schieve, 2014), but over recent years there has not been a statistically significant difference between most race/ethnic groups, aside from Asian/Pacific Islanders (Maenner et al., 2020). In most cases, ASD is recognized by the age of three years old, if not earlier by either the parent or a health professional (Zablotsky et al., 2014). In some instances, although fairly rare, ASD can

go unnoticed and undiagnosed for months and even years, especially in the individuals who are eventually classified as “level one” and who likely present with mixed clinical presentation (Kamio, Moriwaki, Takei, Inada, Inokuchi, Takahashi & Nakahachi, 2013).

The functional limitations faced by those with ASD usually vary and can change over their lifespan (Wiggins et al., 2018). There are individuals who do not need much support that go on to lead regular lives, and there are individuals that require substantial amounts of support with more social, communicative, and even physical tasks than their counterparts that require less assistance. It is very possible that these deficits and behavioral patterns are not recognized as symptoms of ASD until later on in a child’s life, such as when they are unable to meet important life demands and developmental milestones (Wiggins et al., 2018).

Although ASD is most notably seen as a communicative and social disorder, more recent studies have found that it likely shows as movement differences initially before it can be recognized as a social and communicative disorder (Eggleston et al., 2017). Some of the most notable self-stimulating movements that occur include excessive rocking, finger flicking, and arm flapping (Kindregan et al., 2015). Various other motor problems including clumsiness, difficulties in fine and gross motor movements, and lack of fluency and coordination have also been found in those with ASD (Memari, Ghanouni, Gharibzadeh, Vahid, & Pouria, 2012). More recent studies have assessed walking and balance in children with ASD, though mixed results have been reported. In the next few sections, there were an overview of the recent findings about movement in those with ASD.

The Cerebellum and its Role

The cerebellum is a major part of the brain that’s role is to receive information from the spinal cord (sensory input), and it is mainly in charge of balance, coordination, speech, and

overall motor control. It acts as a processor that uses input to guide movement (Wang, Kloth, & Badura, 2014). The cerebellum is the most frequently disrupted part of the brain in persons with ASD, not only at the microscopic level but at the gross level as well (Wang et al., 2014).

Kindregan et al. (2015) explained that recent neuroimaging studies showed that children with ASD displayed reduced activation of the cerebellum during gross motor movements. Starting at early ages when the most noticeable ASD traits and deficits begin to appear, the cerebellum begins to show gross and cellular deficits, most notably in the vermis which has been found to have a decreased overall volume in those with ASD compared to their neurotypical counterparts (Scott, Schuman, Goodlin-Jones, & Amaral, 2009; Wang et al., 2014). It is suggested that the cerebellum provides an internal model, which is needed in order to refine the accuracy of movement. Alterations in the cerebellum and also the basal ganglia may cause motor impairments in different capacities in those with ASD (Nagy, Feher-Kiss, Barnai, Domjan-Preszner, & Angyan, 2007; Dufek, Eggleston, Harry & Hickman, 2017; Rinehart et al., 2006). If this is the case, it makes sense why persons with ASD exhibit problems in locomotion and balance, thus it is important to describe the known locomotion and balance alterations associated with ASD.

Walking Gait

Walking gait in neurotypical individuals is a topic that has been studied for many years now. Prior to the recent few decades, ASD was thought of as solely a social and communicative disorder and any possible physical differences were not addressed, so gait was not a significant focus within the research (Dufek et al., 2017; Eggleston et al., 2017). Within many of the more recent studies, there are inconclusive findings on the movement patterns in those with ASD (Eggleston et al., 2017). Those with ASD exhibit greater clumsiness, decreased motor

coordination, instability, hypotonia, and muscle rigidity compared to normally developing counterparts (Bauman & Kemper, 2005; Kohen-Raz, Volkman, & Cohen, 1992; Damasio & Maurer, 1978; Jones & Prior, 1985; Leary & Hill, 1996; Minshew, Sung, Jones, & Furman, 2004; Molly, Dietrich, & Bhattacharya, 2003). Some of the other issues include altered muscle recruitment patterns, impaired postural control, and various deficits in locomotion and balance (Dufek et al., 2017).

Spatiotemporal variables have been widely assessed in those with ASD compared to their neurotypical counterparts, but with mixed results (Nayate, Tonge, Bradshaw, McGinley, Iansek, & Rinehart, 2012; Nobile et al., 2011; Rinehart et al., 2006, Shetreat-Klein, Shinnar, & Rapin, 2014). Rinehart et al. (2006) assessed gait function in young children aged four to seven that were newly diagnosed with autism. The previous work found there were no significant differences found in velocity, cadence, stride length, double support, and heel-to-heel base of support between those with autism and the controls subjects (Rinehart et al. 2006). However, the previous study did find that those with ASD had a greater number of missteps, meaning that the foot did not fall on the line they were supposed to be walking on for the experiment, and exhibited a greater horizontal-axis range, meaning that they had greater difficulty walking in a straight line compared to their typically developing counterparts (Reinehart et al., 2006). Weiss et al. (2013) found that control participants walked with greater cadence, velocity, and gait cycle time, while those with ASD spent a greater percentage of time in the stance phase of the gait cycle and a shorter amount of time in the swing phase. In contrary, other studies have found higher cadences in those with ASD (Chester & Calhoun, 2012). Several studies have also found increased step width in those with ASD (Shetreat-Klein, Shinnar, & Rapin, 2014; Nayate et al., 2012; Nobile, Perego, Piccinini, Mani, Rossi, Bellina, & Molteni, 2011), while others found no

differences (Rinehart et al., 2006). Based upon the current literature, it appears that those with ASD walk with less balance and more sway, and exhibit greater difficulty walking along a straight path. Other variables may be subject-specific.

Walking gait in those with ASD has also been previously assessed using standard inverse dynamics-based biomechanics via three-dimensional motion capture and force platforms (Dufek et al. 2018; Eggleston et al., 2017; Rinehart et al., 2006; Weiss et al., 2013). Dufek et al. (2018) found that children with ASD exhibit unpredictable movement patterns and positioning during their gait cycle, such as varied vertical GRF and hip, knee, and ankle joint positions. The previous work found that differences occur between those with ASD and their neurotypical counterparts, but most notably are differences in the ankle at ground contact, as those with ASD tend to be more plantarflexed at this point in the gait cycle. Differences at each joint were assessed in a similar study, which found that there were significant differences in sagittal plane angles of the hip joint (39%), knee position (53%), and ankle position (45%), with the percentage differences representing a large effect size (Eggleston et al., 2017). These pattern differences were different in each child, so no general conclusions could be made about the time point in which the differences occur and why.

There are also many discrepancies regarding differences in kinematic variables between those with ASD and controls within the current literature. In the one study that was performed on adults, results showed “mild clumsiness” in those with ASD, but the only significant difference involved the range of ankle dorsiflexion (Hallet et al., 1993). The only consistency between various studies is that there is at least a slight reduction in range of motion (ROM) at the ankle joint in those with ASD at various points during the walking cycle (Ambrosini, Courchesne, & Kaufman, 1998; Nobile et al., 2011; Vilensky et al., 1981). Children with ASD have been found

to have reduced peak plantarflexor moments at the ankle, and decreased peak hip flexor moments (Chester et al., 2012). It also seems to be true that children with ASD exhibit normal ground reaction forces, except for a reduced vertical peak force during terminal stance, which would cause them to walk slower than neurotypical children (Ambrosini et al., 1998).

While research has examined several major areas of walking gait in those with ASD, one important area has yet to be investigated: gait energetics. From an energetics perspective, walking speeds of 1.1-1.4 m/s are the most efficient speeds for healthy adults (Cavagna & Kaneko, 1976; Mahaudens, Detrembleur, Mousny, & Banse, 2009; Willems, Cavagna, & Heglund, 1995). Efficiency is determined by the amount of energy that must be created/absorbed from step to step (i.e. work; (Cavagna et al., 1976; Mahaudens et al., 2009; Willems et al., 1995)), where less work per step would indicate a more efficient walking speed/pattern. When considering similarities in mass, age, and walking speed, an increase in the amount of positive work (i.e. increasing the energy of the system) to take a step would suggest that more muscular input/effort is required for that particular individual to walk. It could also suggest that muscular effort is wasted in some other capacity. For example, greater step widths in those with ASD (Shetreat-Klein, Shinnar, & Rapin, 2014; Nayate et al., 2012; Nobile, Perego, Piccinini, Mani, Rossi, Bellina, & Molteni, 2011) result in more mediolateral movement whereas the goal is to move forward, which may increase work during walking. In addition, the deficiencies in coordination or proprioception (Blanche et al., 2012; Weimer et al., 2001) in those with ASD may result in altered movement patterns of the limbs that are not efficiently counterbalanced. As counterbalanced limb movements are important for maintaining linear/angular momentum, issues with coordination could require muscular effort that does not produce forward motion. Thus, it becomes apparent that dissimilarities in the gait of those with ASD could ultimately

increase the work required to walk and could negatively impact walking as a mode of physical activity. However, previous work does not exist in the realm of gait energetics and/or limb coordination for those with ASD.

Variability

In human motion, including motion that is repetitively performed such as walking and running, humans display a certain level of coordination variability (Robertson, Caldwell, Hamill, Kamen, & Whittlesey, 2013). This can be defined as range of coordination patterns that a person exhibits while performing a movement (Robertson et al., 2013). Healthy, neurotypical individuals have a preferred coordination pattern, but they also possess the ability to access a variety of coordination patterns in order to respond to perturbations (Bernstein, 1966). For example, gait mechanics when walking up a hilly terrain tend to differ from walking on a smooth, level surface. Even when walking on the same surface and in the same conditions repeatedly, with the same goal in mind, kinematics and kinetics vary at least somewhat throughout those multiple repetitions (Miller et al., 2010). Variability in complex motor movements is a critical determinant of the quality of human movement and flexibility (Newell, 1985). Measuring variability has given insight to stability, fall risk (Newell, 1985), and injury status (Hamill, Palmer, & Van Emmerik, 2012). A traditional view of variability states that variability is “noise” coming from either error in performance, or the recording of the movement itself (Shannon, 1948). More modern views on the topic suggest that variability is not necessarily positive or negative, but it is more telling of the variety of coordination patterns used to complete whichever motor task is being observed (Haken, Kelso, & Bunz, 1985; Schoner and Kelso, 1988). With either view, it is still thought that variability will decrease with the level of skilled

performance and will increase with the level of injury or possible disease (Robertson, Caldwell, Hamill, Kamen, & Whittlesey, 2013).

Measuring variability can be used as a tool to identify movement patterns that differ from the norm (Hafer & Boyer, 2017). Children with ASD exhibit some abnormal motor patterns, such as clumsiness (Kanner & Lesser, 1958), unusual postures, toe-walking, and increased joint mobility (Filipek, Accardo, Baranek, Cook, Dawson, Gordon...&Minshew, 1999; Tsai, 1996). In a previous study (Shetreat-klein, Shinnar, & Rapin, 2014), researchers found that 68% of children with ASD exhibited some sort of gait abnormality during walking tasks, as compared to only 13% of controls. Their analysis was a simple visual observation, so it can be hypothesized that even more differences could be found with further analysis.

Over the past decade, vector coding has gained popularity as a way to measure and quantify movement variability during various motor tasks (Chang, Emmerik, & Hamill, 2008). Vector coding can be defined as the relative motion between angular time-series of two segments (Robertson et al., 2013). Vector coding uses spatial data only, which provides a metric that is more understandable to clinicians and could be a reason for its recent popularity (Hafer & Boyer, 2017). This technique involves creating angle-angle plots for motion between adjacent segments (Hafer & Boyer, 2017). These angle-angle diagrams usually depict the changes in angular rotations of the given segments or joints and can be examined for inter-limb or intra-limb coordination (Robertson et al., 2013). The use of vector coding requires examining a number of full stride cycles. Specifically, previous studies suggest anywhere from 5-15 cycles (Heiderscheit, Hamill, & van Emmerik, 2002; Silvernail, Boyer, Brüggemann, & Hamill, 2015; Needham, Naemi, & Chockalingam, 2014; Miller et al., 2010; Hafer, Freedman Silvernail, Hillstrom, & Boyer, 2016). It is important to use a correct number of strides so that the values

calculated are truly representative of the coordination variability within the group being studied (Hafer & Boyer, 2017).

Previous studies have not been performed assessing joint and limb variability in those with ASD. However, as noted earlier, children with ASD have been found to have reduced range of motion at the ankle joint along with inefficient ankle strategies (Kindregan et al., 2015; Blanche et al., 2012). This is a possible indicator that there is some sort of abnormality likely happening at the ankle joint, which could be displayed by plotting the movements of the shank and foot against each other. Previous studies have been done on adults assessing the shank and foot in the frontal and transverse while walking to give an idea of what is happening at the ankle complex. At heel strike, the shank is externally rotated which indicates that the rear foot is supinated compared to its relaxed position during standing (Cornwall & McPoil, 1995; Knutzen & Price, 1994; Lundberg, Svensson, Bylund, & Selvik, 1989; Mannon, Anderson, Cheetham, Cornwall, & McPoil, 1997). Most studies agree that during midstance, the angles in the frontal plane and transverse planes remain the same with ankle inversion and shank external rotation beginning around 50-55% of the gait cycle (Kepple, Stanhope, Lohmann, & Roman, 1990; Mannon, Anderson, Cheetham, Cornwall, & McPoil, 1997; Pierrynowski, Smith, & Mlynarczyk, 1996), while only one reports that external rotation of the shank starts earlier, around 16% of the gait cycle (Nester, Hutchins, & Bowker, 2000). There is not much data for what happens later on, at least in healthy adults, but it is reported that during early swing the rear foot begins to pronate but remains in a supinated position relative to its relaxed state (Nester et al., 2000). Being able to alternate between flexibility and rigidity in the foot joint is essential for adaptation during walking and running and assisting in forward progression (Stefanyshyn & Nigg, 1997;

Whittle, 1999). Therefore, if those with ASD have decreased adaptability, then it is likely that their variability at the ankle joint will differ from that of the neurotypical children.

Another characteristic typical of those with ASD are self-stimulating arm movements, mostly noted as arm flapping (Kindregan et al., 2015). The extent to which these arm movements may affect walking and running efficiency is still unknown. Although it is possible to walk without any arm swing, it is thought that arm swing may reduce the metabolic cost of walking or running by enhancing stability (Ortega, Fehلمان, & Farley, 2008; Umberger, 2008; Ford, Wageenar, & Newell, 2007). It is also known that arm swing helps to generate a horizontal torque at the upper trunk, which may do three things – counteract pelvis rotation, minimize angular momentum, and help reduce reaction moments at the foot and ankle joint (Umberger, 2008; Ortega et al., 2008; Li, Wang, Crompton, & Gunther, 2001; Park, 2008). A previous study assessed coordination between arm and leg movements during locomotion, and found that the arms and legs move at a 2:1 ratio (arm:leg) at lower velocities, but decreases towards 1:1 ratio with increased speeds (Donker, Beek, Wagenaar, & Mulder, 2001). This means that the arms oscillate either twice as fast as the legs, or at about the same speed. Considering the known arm movement differences, it is likely that the subjects with ASD will not consistently match this ratio. As mentioned earlier, improper coordination at the arms and legs can increase angular momentum, and increase the metabolic cost of locomotion, which could then in turn lead to decreased levels of physical activity especially in populations that are already compromised. Using vector coding, it would be possible to quantify the differences in coordination between those with ASD and neurotypical controls.

Summary

ASD is a lifelong developmental disability that elicits deficits in not only social communication and interaction, but also possibly in movement (American Psychiatric Association, 2013). It is suggested that alterations in the cerebellum play a large role in these deficits, creating issues with locomotion, proprioception, and balance (Wang et al., 2014). Those with ASD exhibit similar patterns such as greater clumsiness, decreased motor coordination, and instability compared to their typically developing counterparts (Damasio & Maurer, 1978; Jones & Prior, 1985; Bauman & Kemper, 2005; Kohen-Raz et al., 1992; Leary & Hill, 1996; Molly et al., 2003; Minshew et al., 2004). However, there is disagreement in the literature as to what specific variables are different in those with ASD (Eggleston et al., 2017). These movement deficiencies seem to have translated to those with ASD engaging in less physical activity (Healy et al., 2017; Tyler et al., 2014; Memari et al., 2012), which is causing obesity rates in these individuals to continuously rise (Egan et al., 2013; Tyler et al., 2014). Thus, it is important that future research determine what motor deficiencies are present in those with ASD, including coordination and proprioception differences, and to determine if the variables are related to the lower level of physical activity within this population.

CHAPTER THREE

METHODOLOGY

Participants

We obtained Old Dominion University Institutional Review Board approval for all aspects of this study prior to recruitment of participants. This study included eight youth, age 13-18, clinically diagnosed with ASD and eight age, sex, and mass matched neurotypical controls. To be involved in this study, each participant (both groups) must have an IQ of at least 70 or above, no recent (within 6 months) musculoskeletal injury, and no history of joint surgery or replacement. Additional criteria for all participants include being able to walk, run, jump, and land. Participants were recruited via word of mouth. Upon arrival, participants (and their parents) were informed of the study procedures and asked to sign consent forms. Next, the parents filled out a brief medical and physical activity questionnaire. Participant demographics are provided in Table 1.

Protocol

Walking and Running Gait Testing:

Prior to beginning any data collection, participants were shown pre-recorded videos of all preparation and experimental procedures. The goal for the video was to increase participant familiarity, so that participants were comfortable and less apprehensive to perform this unfamiliar activity. After viewing the videos, participants were asked to don standardized lab shoes and spandex shorts.

To determine movement patterns during walking and running tasks, biomechanical data were collected using a 10-camera motion capture system (200 Hz, Vicon Motion Analysis Inc., UK) and three force platforms (2000 Hz, FP4060, Bertec Inc., USA). Retroreflective markers

were placed on the upper and lower extremities and trunk (total of twelve body segments) of each participant. Anatomical markers were placed bilaterally on the ulnar and radial styloid processes, lateral and medial humeral epicondyles, acromion processes, iliac crests, anterior superior iliac spines (ASISs), posterior superior iliac spines (PSISs), greater trochanters, lateral and medial femoral epicondyles, lateral and medial malleoli, 1st and 5th metatarsal heads, and 2nd toes. Clusters of four tracking markers were placed on the forearm, upper arm, posterior pelvis, and bilateral thighs, shanks, and shoe heels.

Participants with ASD were asked to perform walking under two conditions: at self-selected speeds and at a standardized speed of 1.3 m/s. Each participant's self-selected speeds were recorded using two sets of electric timing gates during warm-up trials, then averaged, and then used to set boundaries at $\pm 5\%$ average self-selected speeds. A trial was deemed successful when performed at the required speed and with full foot contact within the force platform boundaries. Control participants were asked to walk at the previously defined standardized speeds and matched speeds of their ASD counterpart in that order. The speed boundaries for each trial were $\pm 5\%$ of each given speed, and a trial was deemed successful when performed at that required speed and with full foot contact within force platform boundaries. Five successful trials were recorded for each condition.

Accelerometer Data

After completing the on-site data collection, participants wore ActiGraph GT3X+ accelerometers (ActiGraph, Pensacola, FL) on their waistband for seven consecutive days. Participants were instructed to wear the accelerometers throughout the day and only remove it when going to sleep or engaging in activities that expose it to water. Data were collected with a

60 Hz sample frequency and analyzed using 15 second epochs. To be included in the final analysis, participants needed to accumulate a minimum of 10 hours/day of wear time for at least four days, including one weekend day. Previously validated count thresholds for children by Evenson, Catellier, Gill, Ondrak, & McMurray (2008) were used to establish sedentary activity (0 – 100 counts per minute), light physical activity (101-2295 counts per minute), moderate physical activity (2296-4011 counts per minute), and vigorous physical activity (4012+ counts per minute). Accelerometers were returned in-person to the researchers.

Data Processing

Motion capture and force data were imported into the biomechanical software suite Visual3d (Version 6, C-Motion, USA). Marker and force data were filtered using zero-lag 4th order recursive Butterworth filters at 6 Hz for walking (Chiu & Wang, 2008). An inverse dynamics-based model was created for each participant. Shoulder joints were defined using the equation $Marker_{Radius} + 0.17 * Distance(LAC, RAC)$ as defined by the Visual 3d software guidelines. The elbow and wrist joints were defined as the midpoint of the medial and lateral humeral epicondyles and styloid processes, respectively. The Davis method was used to determine hip joint centers (Davis, 1953). Knee and ankle joints were defined as the midpoint of the femoral epicondyles and malleoli, respectively. An X-Y-Z (flexion-adduction-internal rotation) Cardan rotational sequence and the right-hand rule were used for 3D angular kinematics computations and polarizations. A full gait cycle consisted of right heel strike (vertical GRF > 10 N) to subsequent right heel strike.

Angle-Angle Plots & Vector Coding

Angle-angle plots were constructed for contralateral arm/thigh and ipsilateral foot-shank segment groupings in the sagittal plane (Figure 1). The proximal and distal segments comprise

the x and y-axes, respectively. Coordination and variability were determined using previously defined methodology (Chang, Emmerik, & Hamill, 2008), and is briefly described below.

Sagittal plane upper arm, thigh, shank, and foot data were imported into Matlab (MATLAB. (2018). 9.7.0.1190202 (R2019b). Natick, Massachusetts: The MathWorks Inc.) The relative motion between the two segments was quantified using the coupling angle, represented as (γ). The coupling angle was found by using the following equation (Robertson et al., 2013), which determines the quantification of a vector adjoining two successive time points:

$$\gamma_{j,i} = \tan^{-1}\left(\frac{y_{j,i+1} - y_{j,i}}{x_{j,i+1} - x_{j,i}}\right)$$

The coupling angle was modulated to exist between 0° and 360° . Within the equation, i represents consecutive data points in a cycle, and j is indicative of the multiple gait cycles. Mean coupling angles ($\bar{\gamma}$) were computed with circular statistics (Batschelet 1981; Fisher 1993). Within each subject, $\bar{\gamma}$ is calculated from mean horizontal (\bar{x}_i) and vertical components (\bar{y}_i) across the gait cycles (j) for each percentage (i) of the gait cycle:

$$\bar{x}_i = \frac{1}{n} \sum_{j=1}^n (\cos \gamma_{j,i})$$

$$\bar{y}_i = \frac{1}{n} \sum_{j=1}^n (\sin \gamma_{j,i})$$

Next, the mean vector's length is defined as

$$r_i = \sqrt{\bar{x}_i^2 + \bar{y}_i^2}$$

for each percentage of the gait cycle. The mean vector will have a defined angle versus the positive horizontal axis, which is known as the *coupling angle* γ . Mean coupling angles across multiple gait cycles will again be defined for each percentage (i) of the cycle:

$$\bar{\gamma}_i = \begin{cases} \arctan\left(\frac{\bar{y}_i}{\bar{x}_i}\right), & \text{if } \bar{x}_i \geq 0 \\ 180 + \arctan\left(\frac{\bar{y}_i}{\bar{x}_i}\right), & \text{if } \bar{x}_i < 0 \end{cases}$$

The coupling angles were then be organized into four separate bins, separated by 45 degrees (Chang et al., 2008), across stance and swing phases of the gait cycle. The four bins represent segmental dominated (n=2), anti-phase, and in-phase coordination patterns that can be expressed for each coupling: thigh-upper arm and foot-shank. The frequency of each coordination pattern (out of 100%; e.g. where 100% denotes that coupling pattern existed for the entirety of stance) were determined for each participant and condition. Frequencies were determined for early (0-50%) and late (51-100%) portions of the stance phase.

Statistical Analysis

As the participant pool was small for each group, non-parametric analyses were assumed to be necessary. Wilcoxon RankSum Tests ($p < 0.05$) were performed to analyze differences, if any, in coordination patterns (four patterns per coupling group) between youth with ASD and matched controls at each speed (matched self-selected and 1.3 m/s). Spearman's Rho correlation analyses were performed to determine if any of the four coordination patterns for each couple during the stance phase or sagittal plane thigh, upper arm, or ankle joint range of motions were significantly related to physical activity level. Due to the small sample size, these relationships were analyzed across the entire participant pool (n=16) and only for the standardized walking speed.

CHAPTER IV

FINDINGS

DESCRIPTION OF COORDINATION PATTERNS

Coupling angle and angle-angle plots are provided for the upper arm-thigh and shank-foot for each group at matched and standardized speeds (Figure 1). For upper arm-thigh coupling, dominance in the proximal segment would show as the plot having more movement towards the right side, while dominance in the distal segment would show as more movement to the left side. The opposite would be true for shank-foot coupling. These plots also show whether the limb pairings are in-phase or anti-phase. In-phase is defined as simultaneous movement of both limbs performing the same movement (e.g. flexion or extension), while anti-phase is defined as opposite movement of the limbs (e.g. thigh flexion and upper arm extension).

PATTERN DIFFERENCES BETWEEN GROUPS

For the shank-foot plots, both groups begin in the stance phase with flexion (positive angle for the foot, negative angle for the shank according to right-hand rule) occurring at both the shank and foot. Shortly after, peak flexion is reached and both limbs begin to extend (decreasing positive/increasing negative angles) until toe-off. The ASD group displays slightly reduced flexion at both segments, but the overall range of motion is consistent with their neurotypical counterparts.

Although the general coordination is similar in the figures, the greatest differences occur in the upper arm-thigh plots. Both groups follow a similar pattern beginning with flexion at both the upper arm and thigh, but the ASD group is slightly more extended at both limbs. At both the self-selected and standardized speeds (Figure 1C/1D), it appears that those with ASD present

with greater thigh and upper arm extension over the whole gait cycle, while the control group is flexed more in both limbs. The pattern in both groups shows that while one limb is flexing, the other limb is generally performing the same motion, which is expected.

The only instance where coupling angles are noticeably different occur between the upper-arm and thigh at the standardized speed (Figure 1D), where the ASD group showed a considerably lower peak angle around 50-60% of stride compared to the control group. None of the frequency values were significantly different between the two groups. Figures 2 and 3, and tables 2, 3, 4, and 5 display the frequency results for both groups under both conditions.

Correlation Analyses

Upper-arm dominance and anti-phase upper arm/thigh-dominance patterns were significantly related to decreased minutes of vigorous physical activity (Rho: -0.63, $p < 0.01$ & Rho: 0.58, $p = 0.02$, respectively; Figures 4 and 5). No other coordination patterns were significantly related to minutes of vigorous physical activity ($p > 0.05$). No coordination pattern for either coupling was significantly related to steps ($p > 0.05$). No significant relationships were found between upper arm, thigh, or ankle joint ranges of motion and minutes of vigorous physical activity or steps ($p > 0.05$).

Table 1. Participant Demographics

	Age (yrs.)	Mass (kg)	Height (m)	BMI
ASD (n=8)	15.6+1.5	63.87+20.29	1.67+0.13	22.0+4
CON (n=8)	15.0+1.0	64.44+15.90	1.72+0.09	22.0+4
T-test (p-value)	0.707	0.951	0.408	0.756

Note. There were no significant differences between the ASD and control groups in age, mass, height, or BMI. A Mann-Whitney test for two independent samples was used to obtain the p-value in each category.

Table 2. Frequency data for walking at self-selected speeds for the thigh (Th) and upper arm (UA).

GRP	Early Stance				Late Stance			Anti-Phase
	Th	UA	In-Phase	Anti-Phase	Th	UA	In-Phase	
ASD	34.3±30.3	15.9±11.0	47.0±24.1	8.9±7.9	41.3±32.6	8.9±10.0	45.2±30.1	6.3±7.5
CON	22.2±24.2	14.9±9.3	54.4±23.9	8.5±6.4	41.5±32.5	4.0±4.1	55.2±30.6	2.4±2.3
Wilcoxon Ranksum Test	W(76), p=0.425	W(69), p=0.945	W(54.5), p=0.168	W(67), p=0.963	W(66.5), p=0.897	W(81), p=0.174	W(55.5), p=0.200	W(84), p=0.096

Table 3. Frequency data for walking at self-selected speeds, for the shank (Sh) and foot (Ft).

GRP	Sh	Ft	In-Phase	Anti-Phase	Sh	Ft	In-Phase	Anti-Phase
ASD	56.5±13.2	6.5±6.9	31.0±16.1	5.0±8.7	26.4±18.5	3.2±3.5	72.2±18.0	1.4±3.9
CON	58.5±13.0	6.0±6.8	33.5±18.6	2.0±3.4	22.2±17.5	2.4±2.9	77.0±18.4	1.6±4.6
Wilcoxon Ranksum Test	W(67), p=0.944	W(71), p=0.824	W(63), p=0.628	W(75.5), p=0.439	W(78.5), p=0.289	W(75), p=0.497	W(56.5), p=0.248	W(67.5), p=1.000

Table 4. Frequency data for walking at standardized speeds (1.3 m/s), for the thigh (Th) and upper arm (UA).

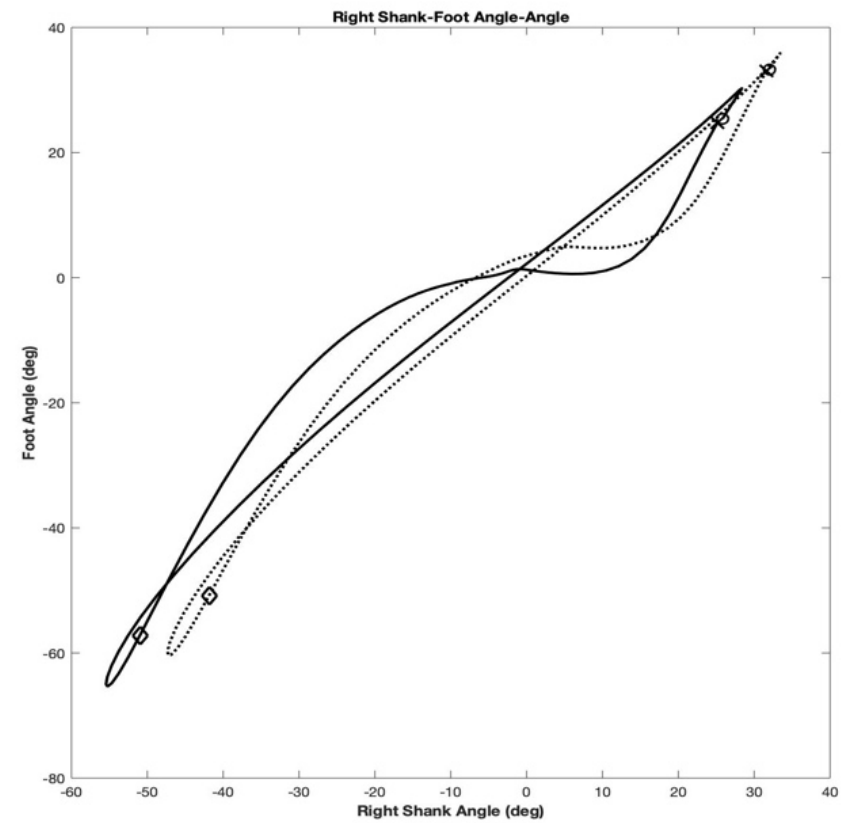
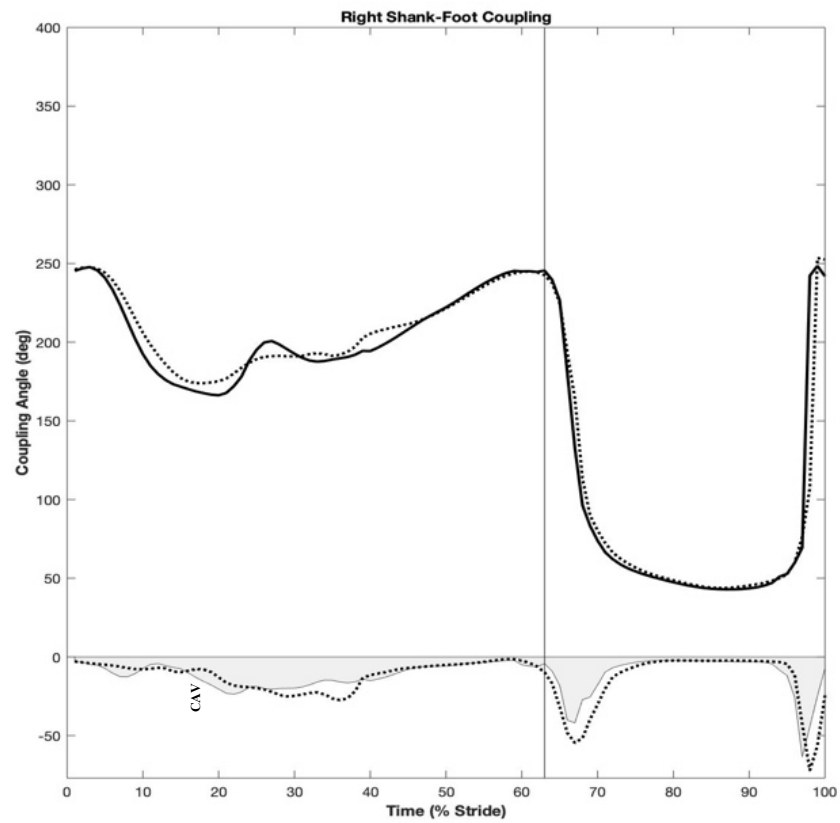
GRP	Early Stance				Late Stance			
	Th	UA	In-Phase	Anti-Phase	Th	UA	In-Phase	Anti-Phase
ASD	41.1±34.9	15.3±10.1	45.8±27.6	9.3±8.2	42.3±35.7	4.0±4.6	53.4±34.8	3.4±3.4
CON	18.1±21.7	16.9±8.4	56.5±24.3	8.5±7.9	33.5±26.8	3.6±2.1	61.7±27.4	4.4±3.8
Wilcoxon Ranksum Test	W(79.5), p=0.246	W(62), p=0.552	W(54), p=0.152	W(72), p=0.725	W(71.5), p=0.738	W(63), p=0.682	W(62), p=0.556	W(58), p=0.312

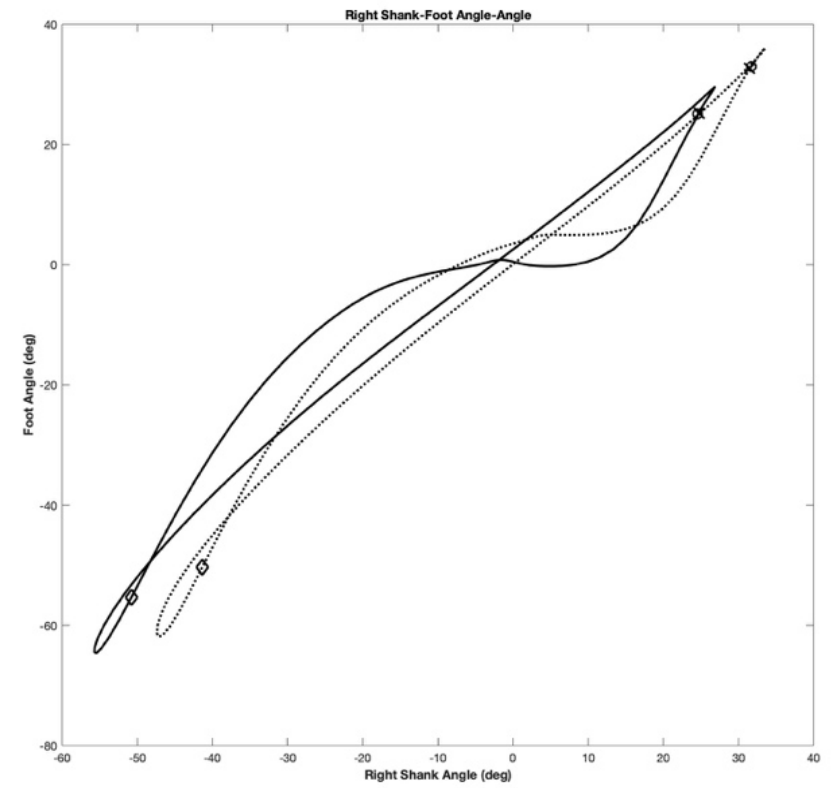
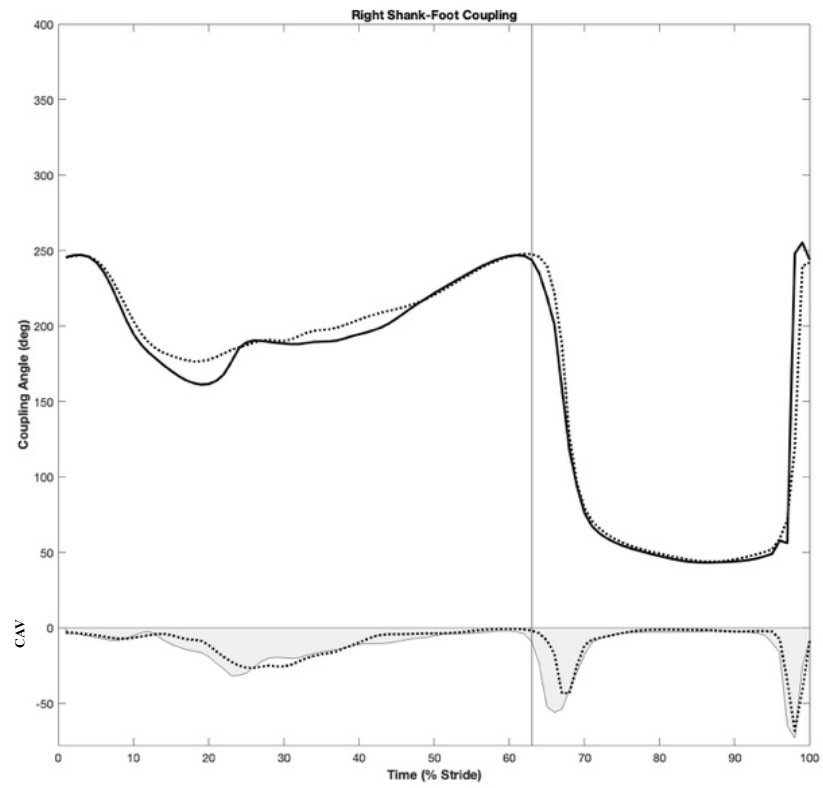
Table 5. Frequency data for walking at self-selected speeds, for the thigh (Th) and upper arm (UA).

GRP	Early Stance				Late Stance			
	Sh	Ft	In-Phase	Anti-Phase	Sh	Ft	In-Phase	Anti-Phase
ASD	54.8±11.9	6.7±5.9	30.4±16.5	6.7±10.5	26.2±19.2	3.4±4.0	73.2±21.2	0.4±1.6
CON	57.7±15.8	6.0±5.6	33.1±18.9	3.2±9.1	20.2±15.5	3.2±3.0	79.0±18.7	0.8±2.3
Wilcoxon Ranksum Test	W(66.5), p=0.888	W(72), p=0.723	W(61.5), p=0.519	W(78.5), p=0.282	W(83), p=0.118	W(67.5), p=1.000	W(56), p=0.220	W(64), p=1.000

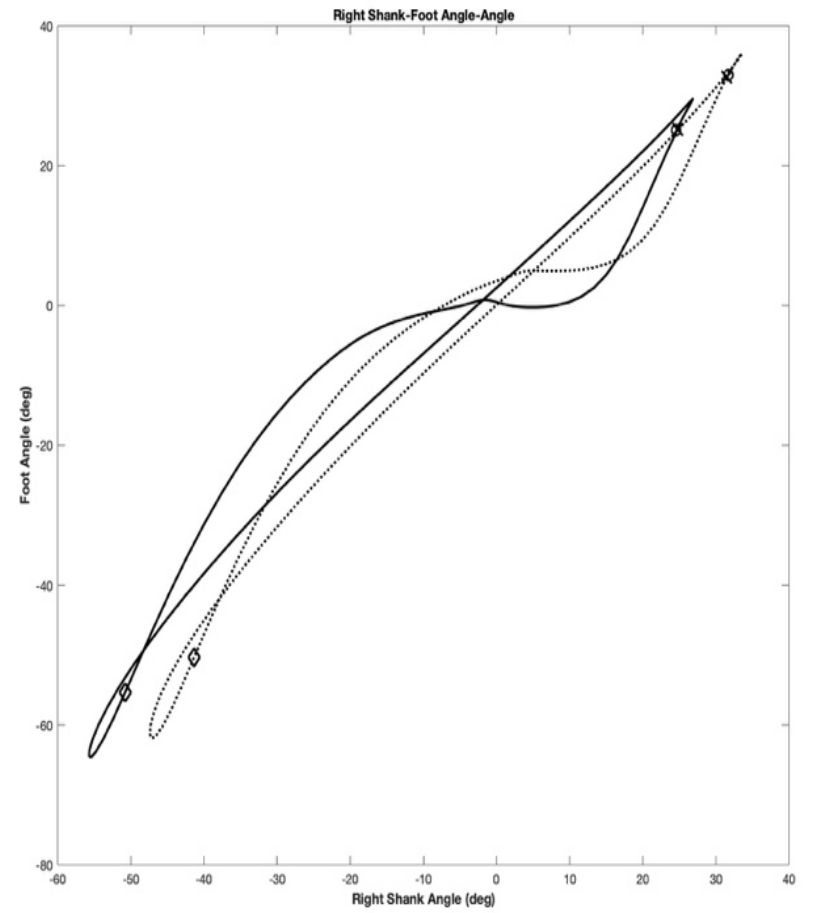
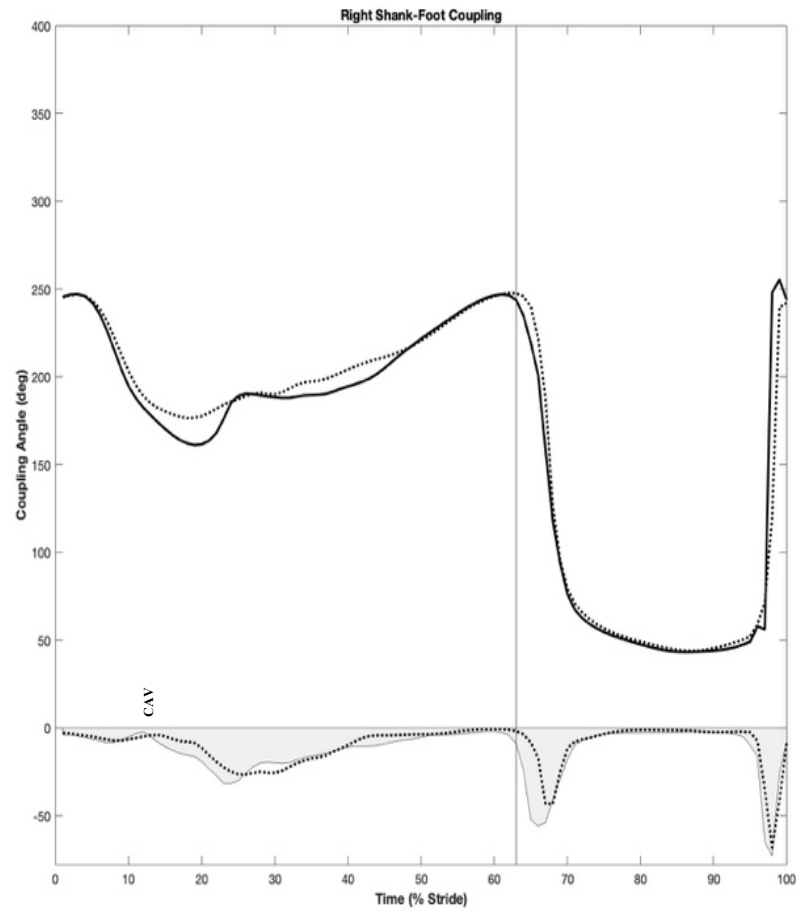
Figure 1. Angle-angle, coupling angle, and coupling angle variability plots for each group and condition.

A.



B.

C.



D.

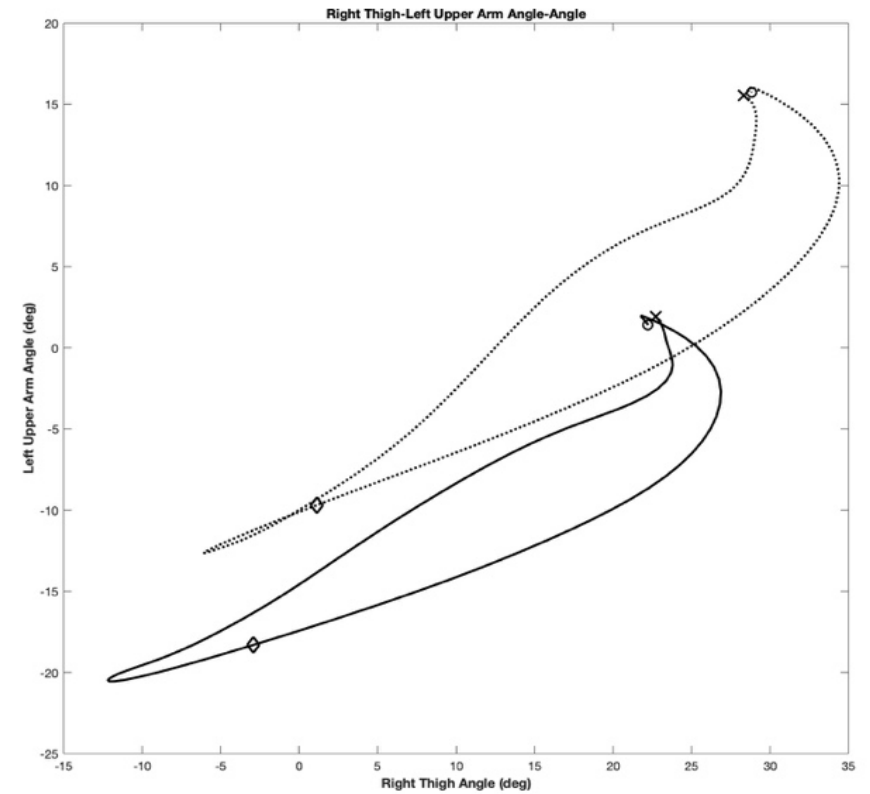
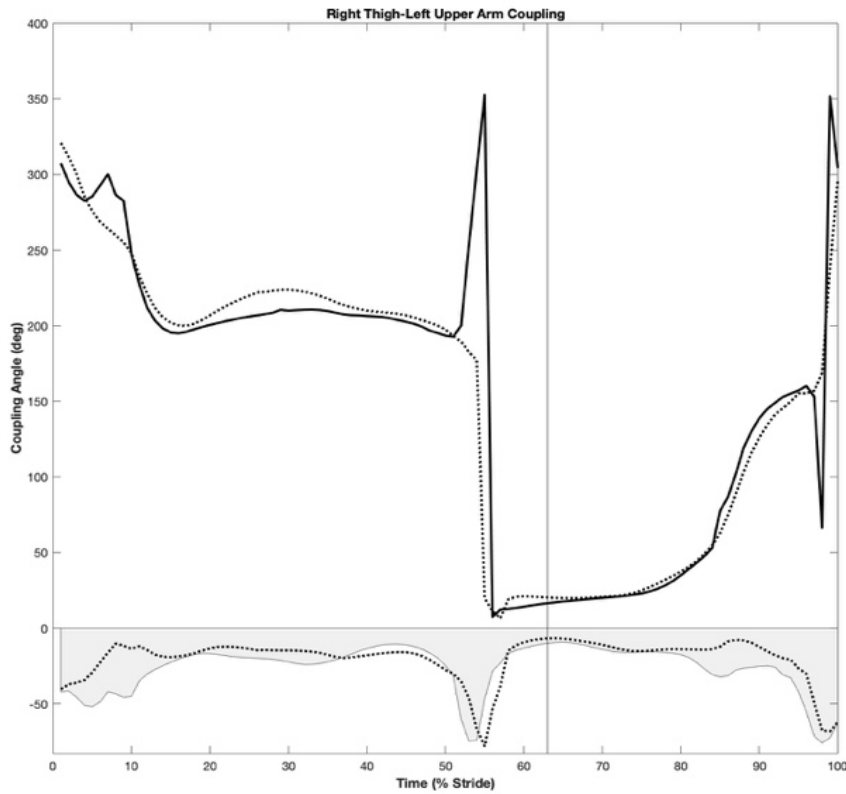
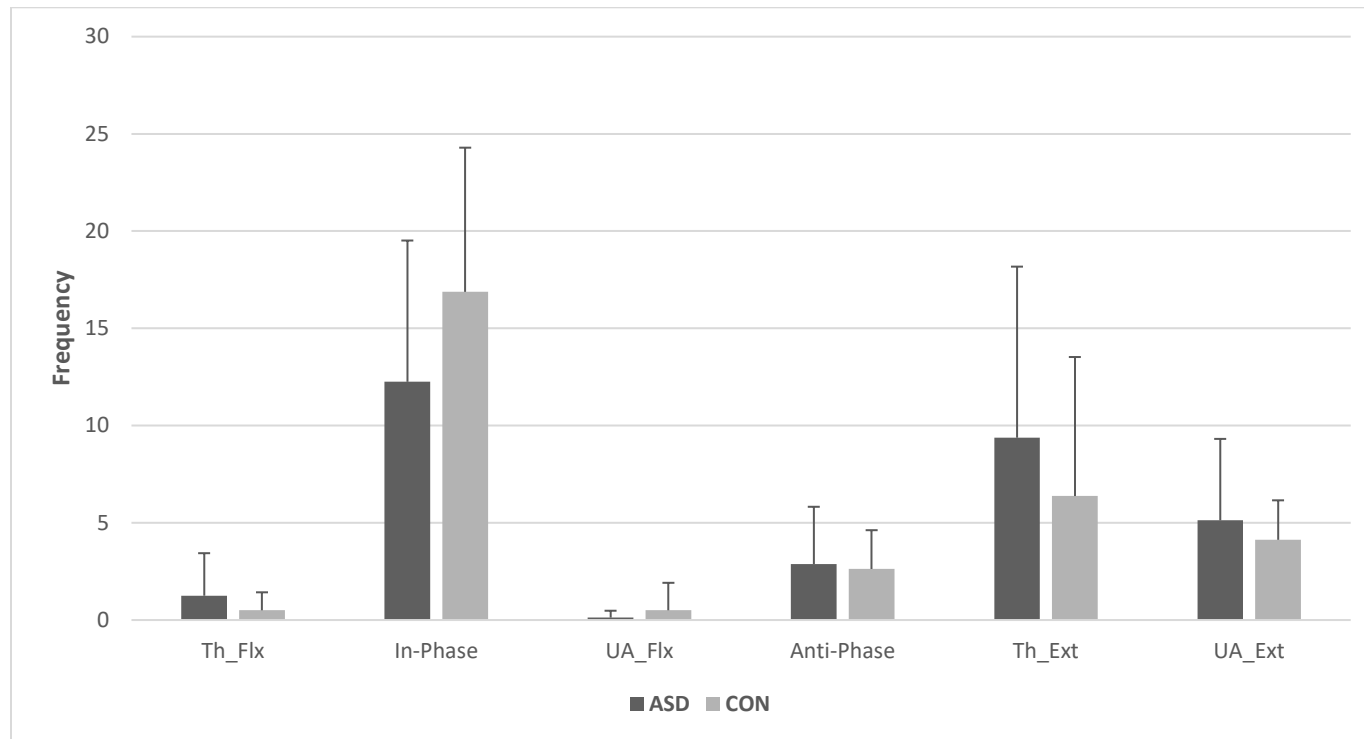
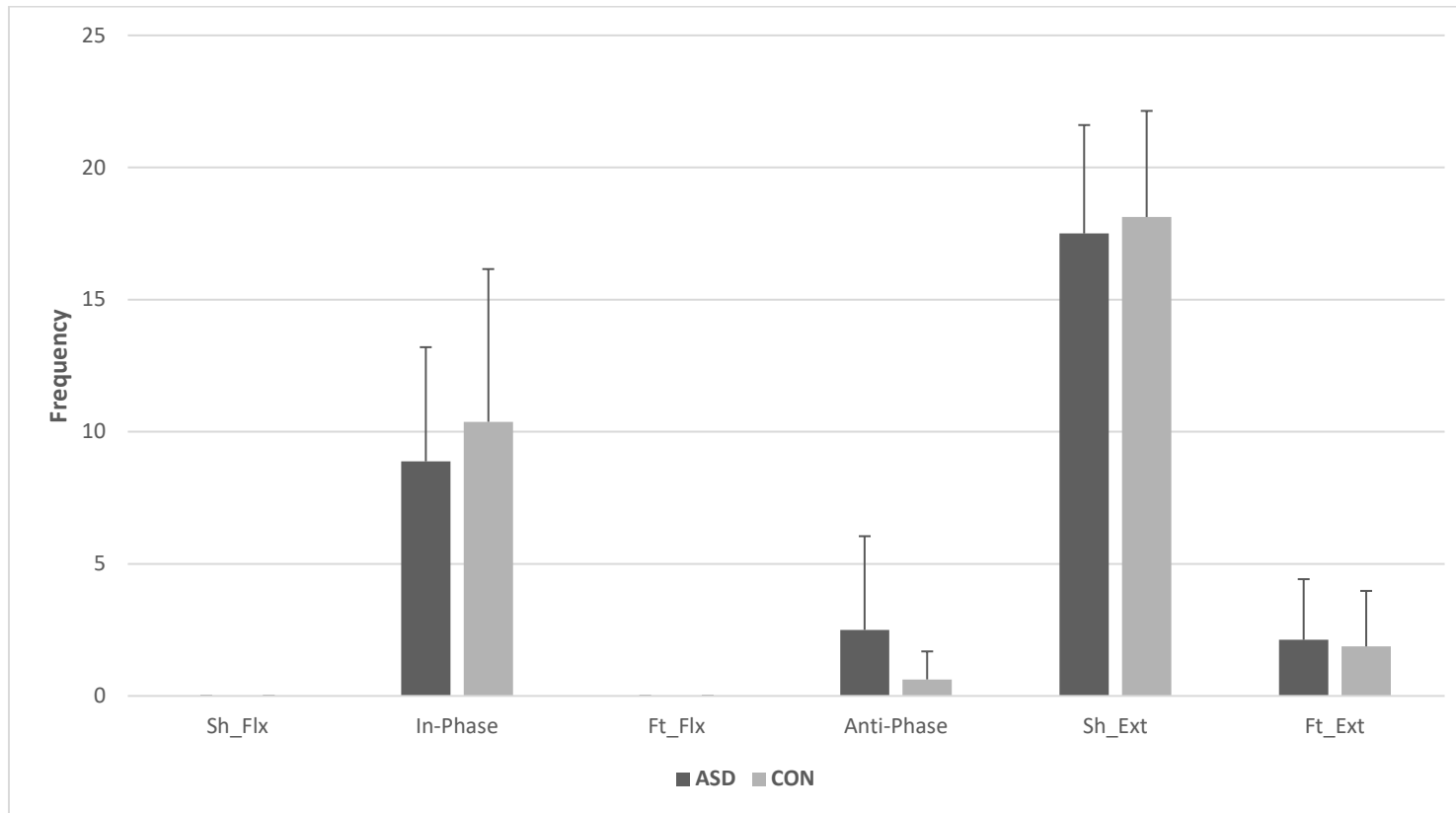
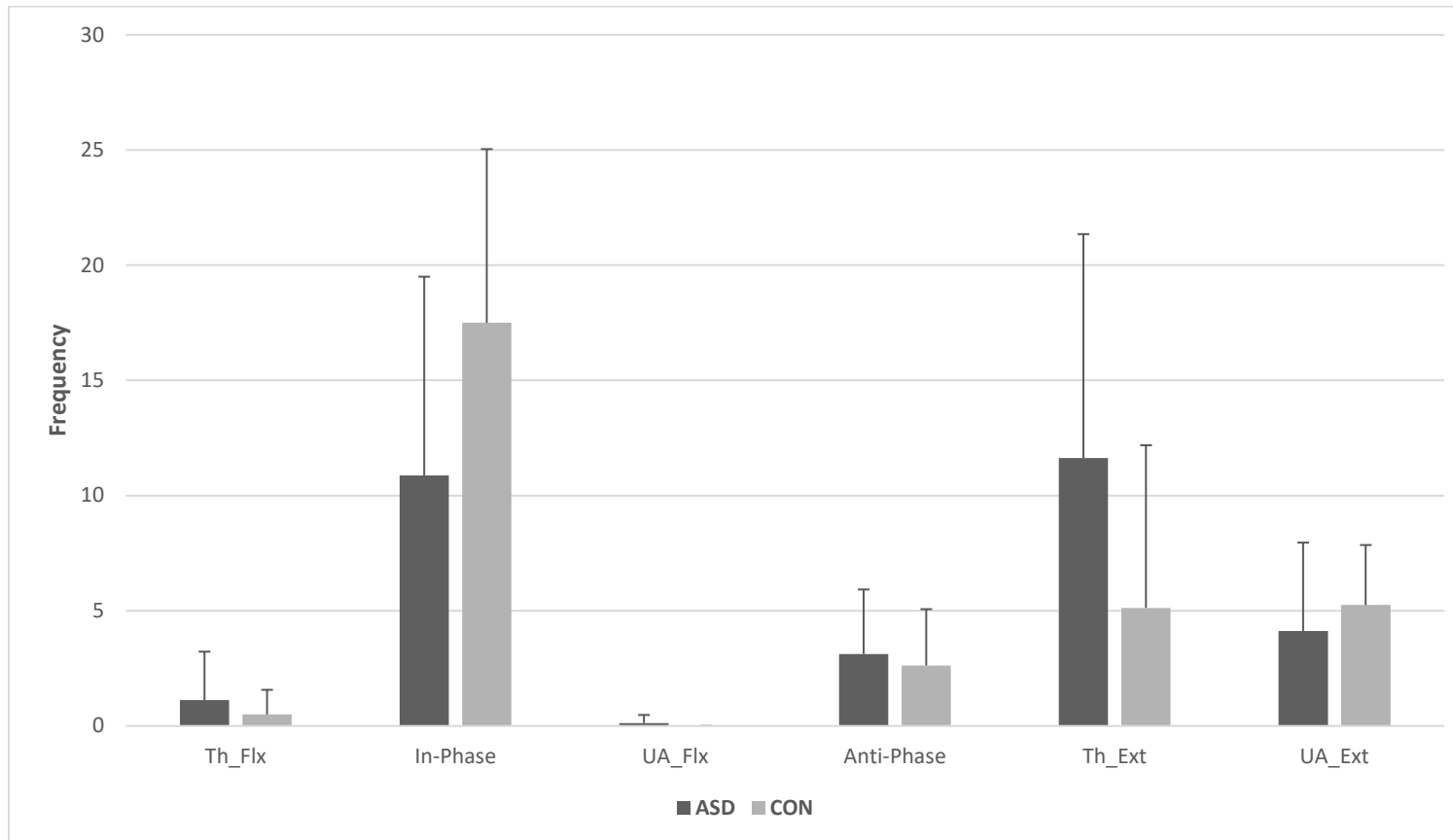


Figure 1. Ensemble angle-angle and coupling angle plots for right shank-ankle at self-selected speed (1A), right shank-ankle at standardized speed of 1.3 m/s (1B), right thigh-left upper arm at self-selected speed (1C), and right thigh-left upper arm at standardized speed of 1.3 m/s (1D) are presented for the persons who with ASD (solid lines) and controls (dashed lines) at matched speeds during walking. The “x” and diamond notations on angle-angle plots represent heel strike and toe-off events, respectively. The solid vertical line on coupling angle plots represents toe-off. Coupling angle variability (CAV; deg) are also provided for each angle-angle grouping (persons with ASD: solid line and shading, controls: dashed line).

Figure 2. Early stance frequency data.**A.**

B.

C.



D.

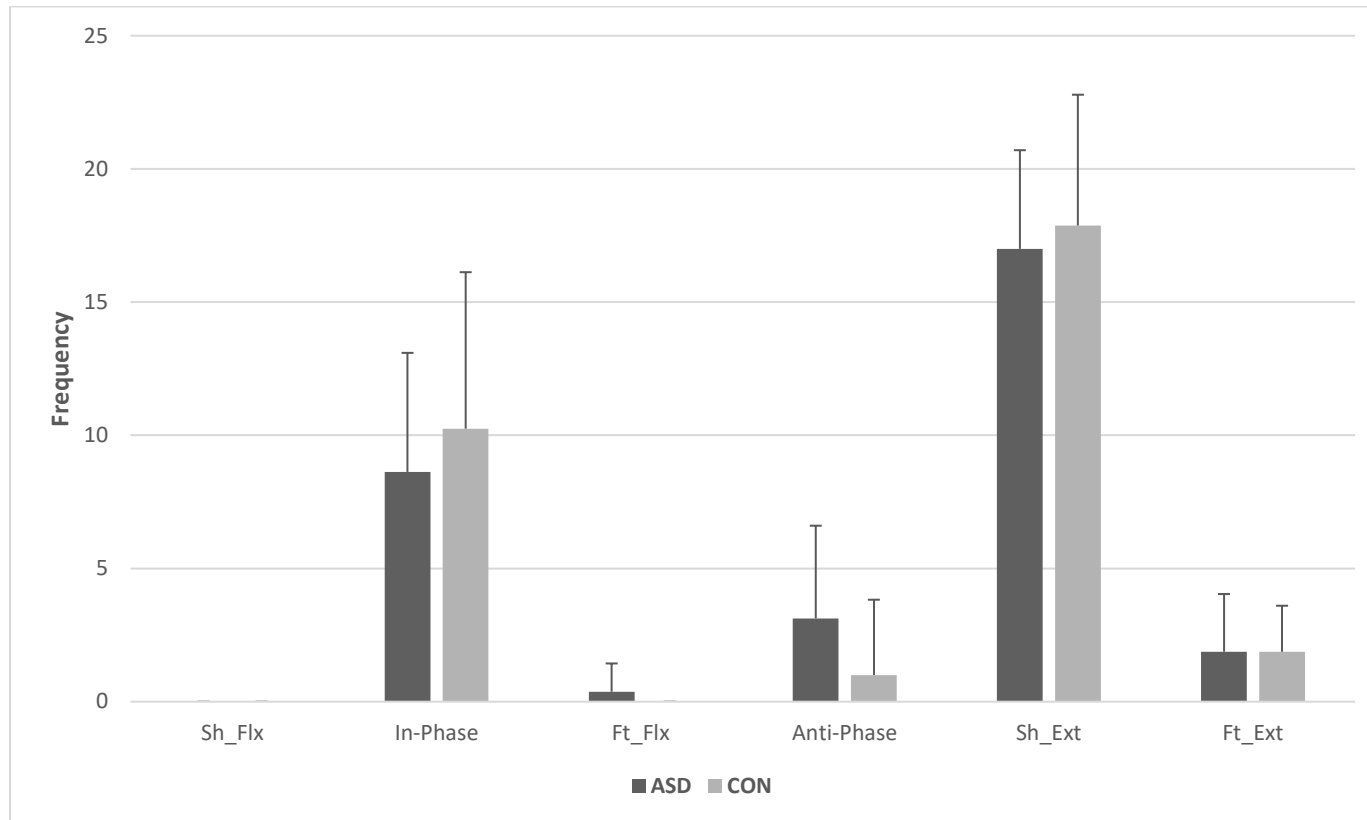
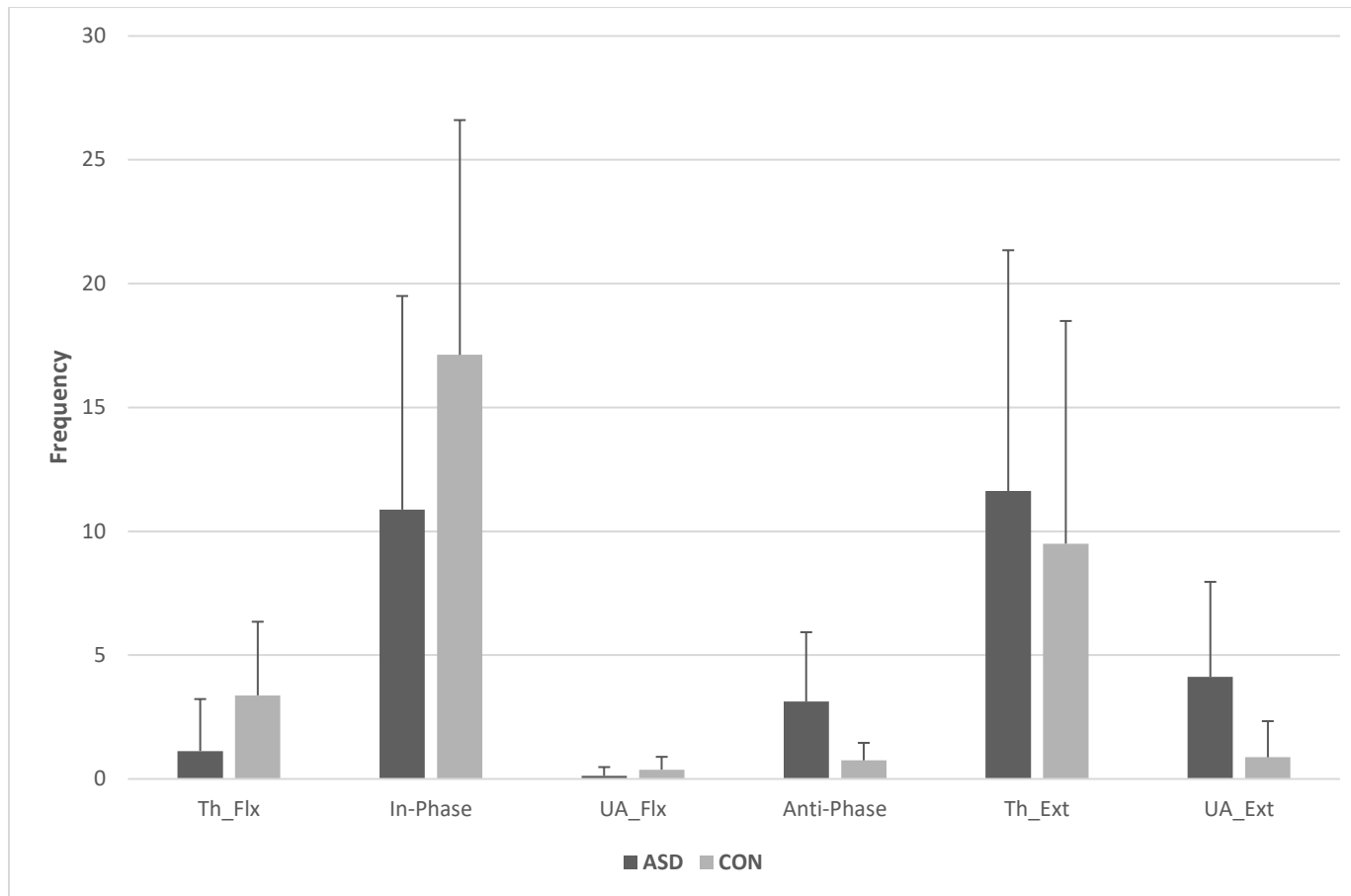
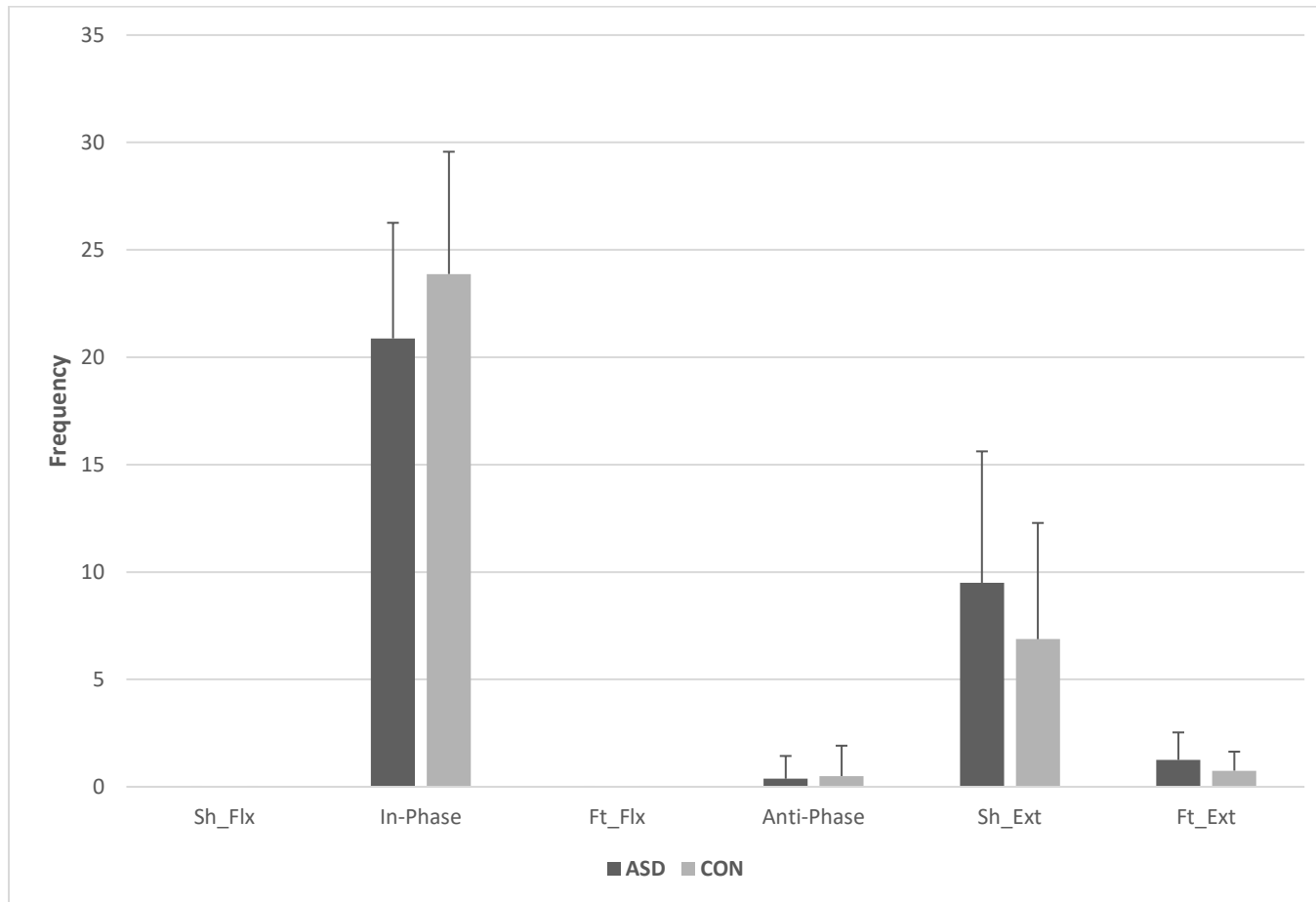


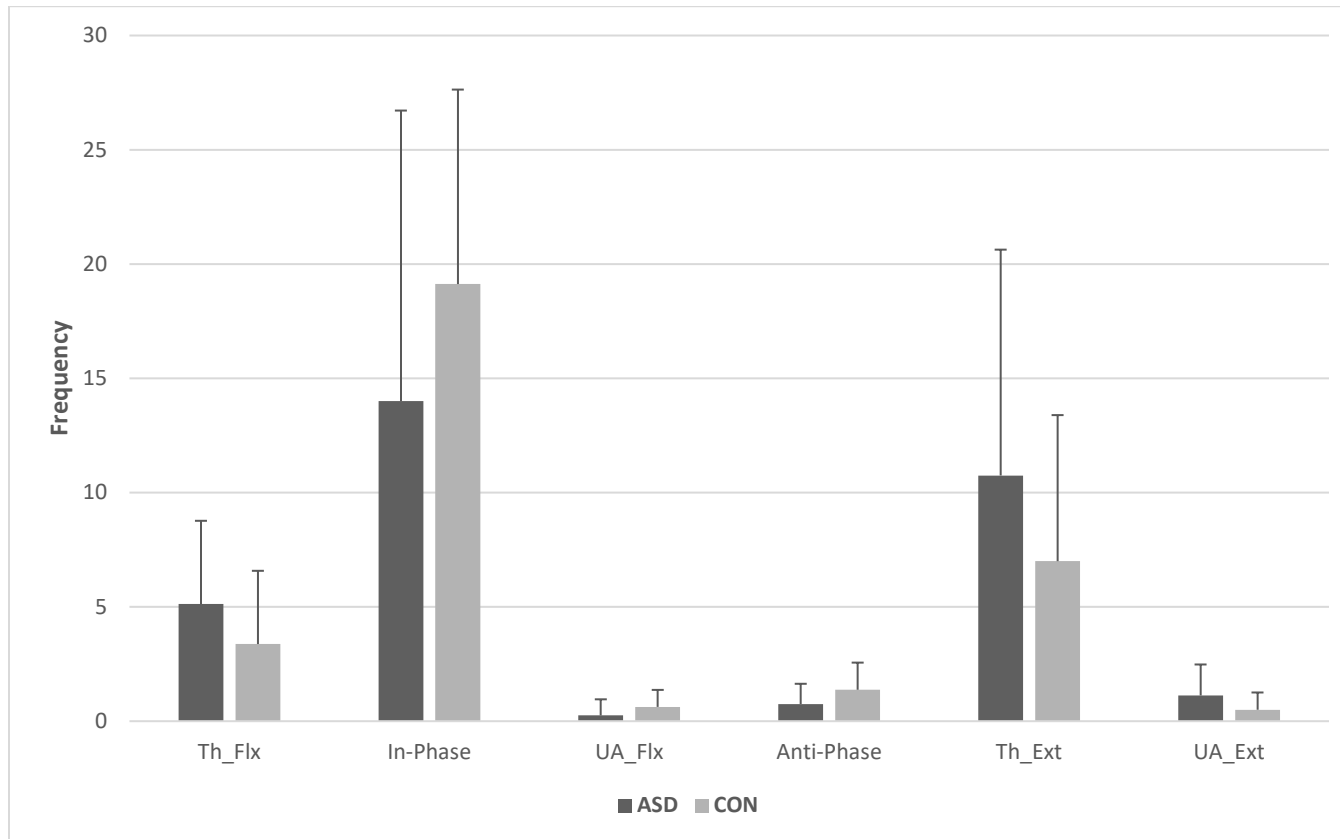
Figure 2. Frequency plots for both control and ASD groups during early stance (1-50% of stance). Plots are shown for upper arm-thigh at self-selected speed (2A), shank-foot at self-selected speed (2B), upper arm-thigh at standardized speed (2C), and shank-foot at standardized speed (2D). The plots show at how many points out of a total of 31 that the specified limbs were flexing or extending, and whether they were in-phase or anti-phase.

Figure 3. Late stance frequency data.
A.



B.

C.



D.

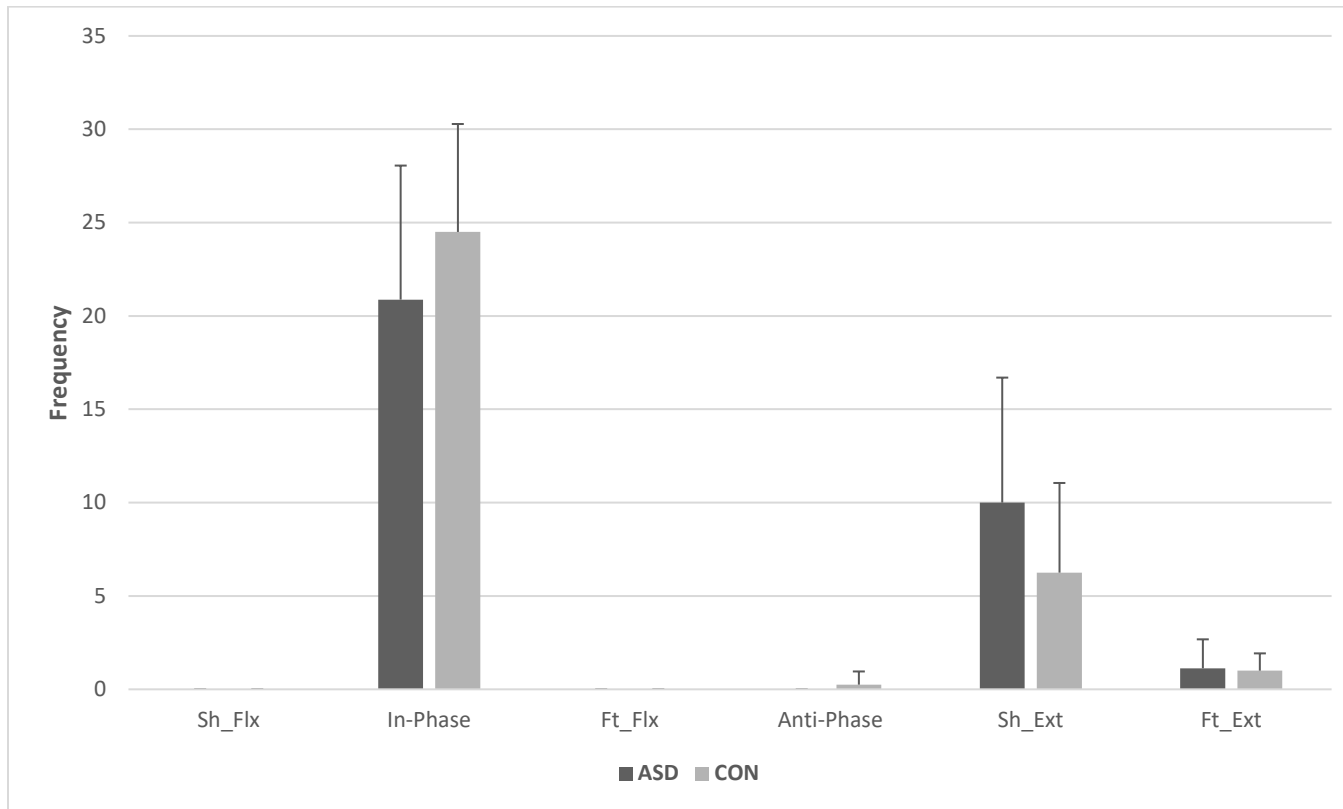


Figure 3. Frequency plots for both control and ASD groups during early stance (51-100% of stance). Plots are shown for upper arm-thigh at self-selected speed (3A), shank-foot at self-selected speed (3B), upper arm-thigh at standardized speed (3C), and shank-foot at standardized speed (3D). The plots show at how many points out of a total of 31 that the specified limbs were flexing or extending, and whether they were in-phase or anti-phase.

Figure 4. Scatter plot of Upper Arm Dominance and Minutes of Vigorous Physical Activity (MVPA).

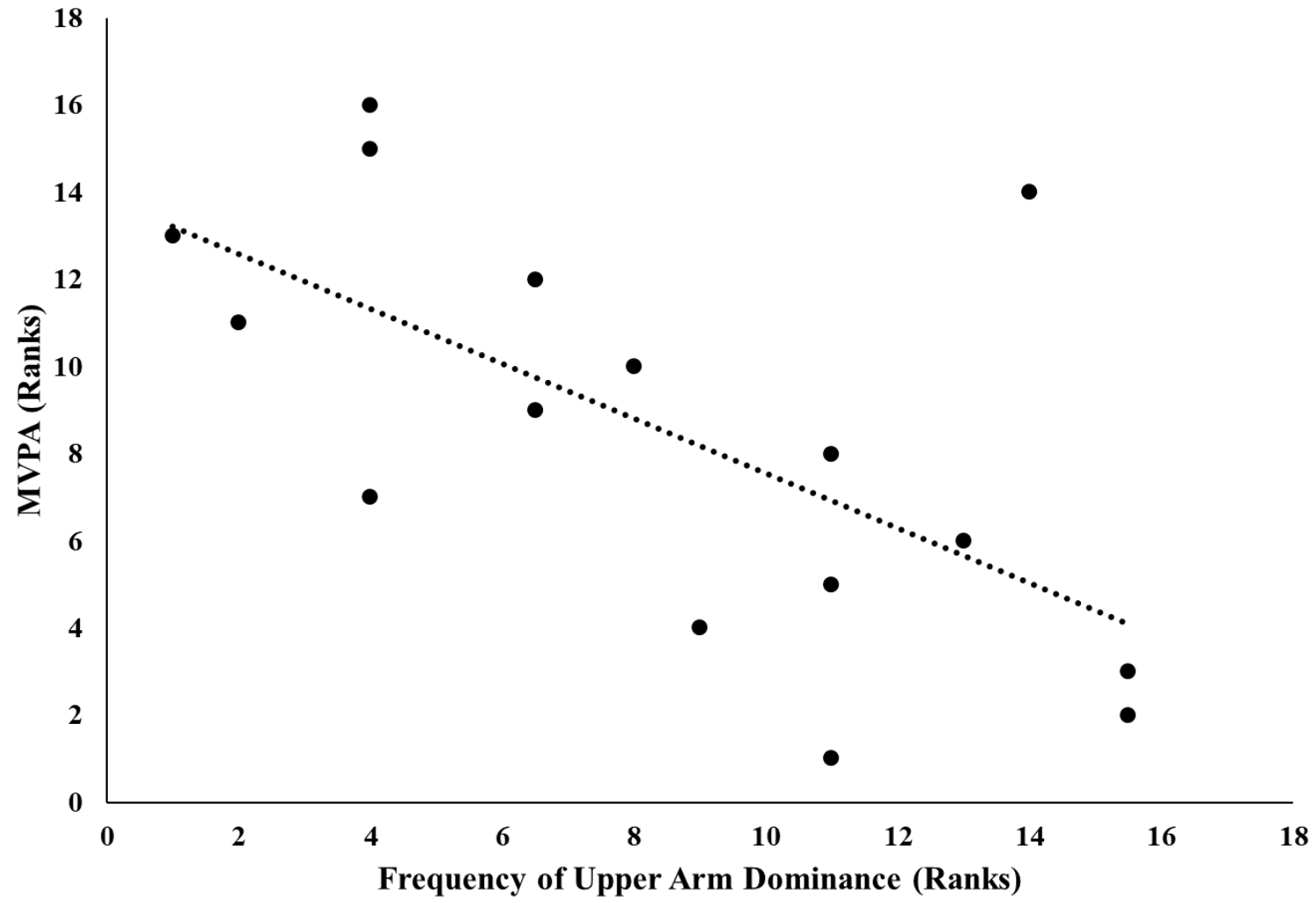
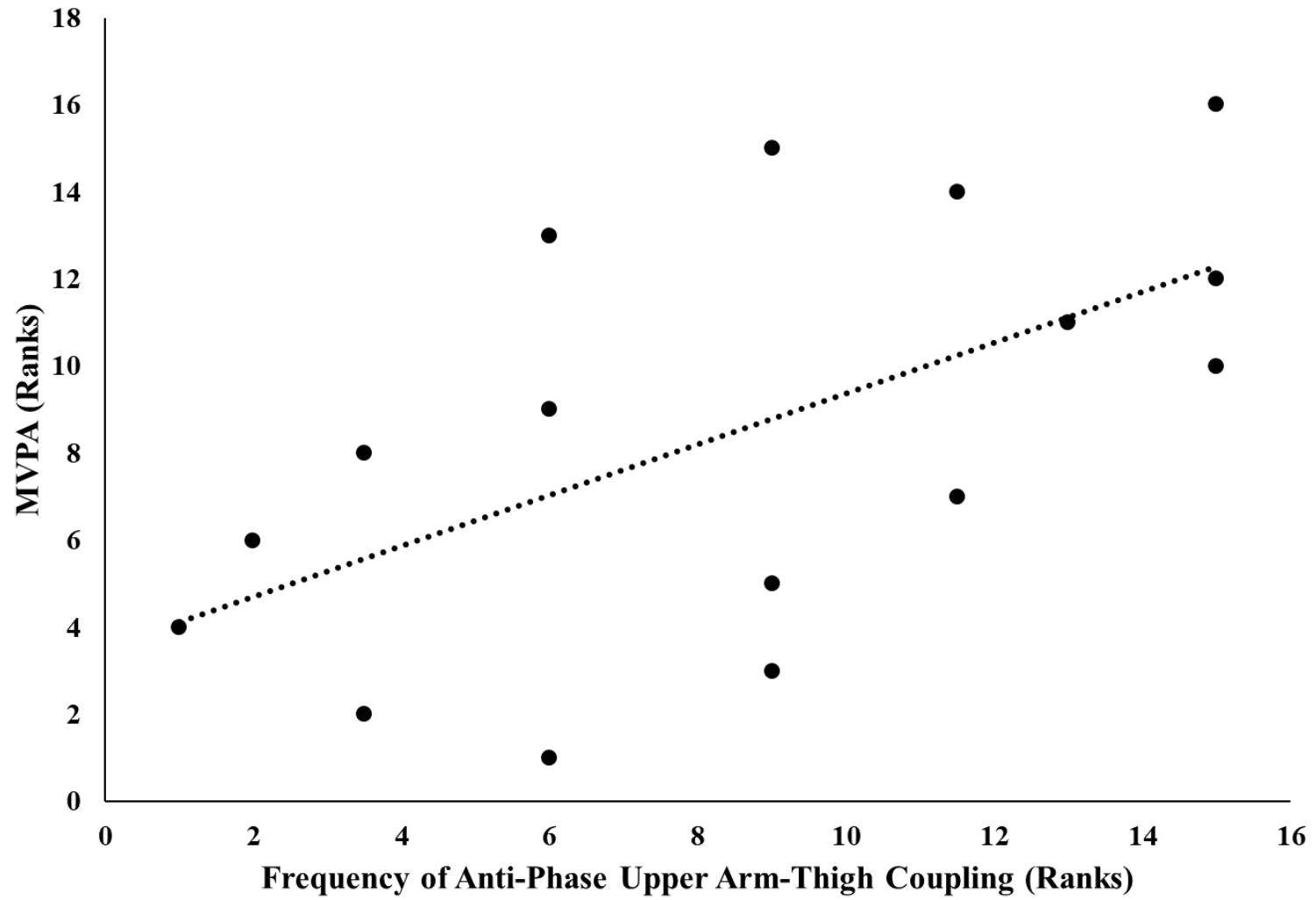


Figure 5. Scatter plot of Anti-phase Upper Arm and Thigh Coupling and Minutes of Vigorous Physical Activity (MVPA)

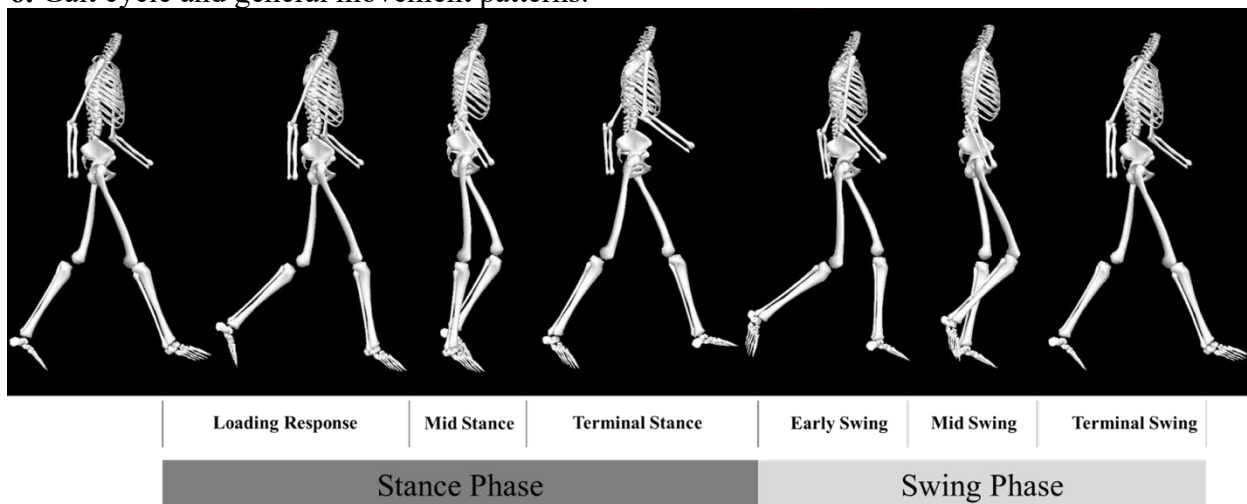


CHAPTER V

DISCUSSION

The purpose of this study was to assess differences in inter-limb and intra-limb coordination in those with ASD and age, sex, and BMI matched controls. We hypothesized that youth with ASD would have decreased inter-limb and intra-limb coordination. This hypothesis was rejected, as there were no significant coordination differences in either category between the two groups.

Figure 6. Gait cycle and general movement patterns.



In typical walking gait, during heel contact and early stance phase is when the right foot contacts the ground, the opposite upper arm (left), right thigh, shank, and foot are all generally in a flexed position. As the rest of the body transitions from behind to in front of the stance limb (i.e. loading response through terminal stance), the right lower limb segments extend. The extended limbs are then quickly flexed to produce and prepare for forward swing. The upper arm and thigh generally remain in line with each other, moving at a 1:1 ratio while walking as can be seen in Figures 1A and 1B. The shank and foot move either in-phase or anti-phase with each other as shown in Figure 1C and 1D, to produce ankle dorsiflexion or plantarflexion as needed.

According to the frequency plots, during early stance phase at both the self-selected and standardized speeds most of the movement occurred at the ankle consisted of shank extension and little foot extension, in both the ASD and control group. No flexion occurred in either limb. Also, during early stance for both conditions, the upper arm and thigh were mainly extending in both groups, with most of the movement as in-phase.

Walking speed did not appear to affect frequency of movement at the ankle. Though, there is a slightly higher frequency of shank and foot extension, and in-phase frequency. For the upper arm and thigh, the ASD group at their self-selected speed exhibited similar extension and in-phase frequency values during late stance as they did during early stance. The control group at their self-selected speed showed a little bit more thigh flexion points than the ASD group, and a slightly greater percentage of the time was spent in-phase, but these values were similar to what they did during early stance. Similar values were seen at the standardized speed, with slightly more thigh flexion and greater percentage of the thigh and upper arm being in-phase.

Shank-foot angle-angle plots (Figure 1A and 1B) show that both the ASD and control group exhibit similar angles and movement patterns from heel-strike to toe-off, at both the self-selected and standardized speeds. The average angles at each limb only differ by a few degrees. The coupling angles were also extremely similar and coupling angle variability (CAV) was not significantly different between the two groups.

Surprisingly, none of the variables were significantly different in either group at either condition. This was unexpected especially at the ankle, where it has been noted in previous literature that those with ASD exhibit some kinematic and kinetic differences at the ankle (Blanche et al., 2012; Chester et al., 2012; Dufek et al., 2018; Eggleston et al., 2017).

Moderate-to-vigorous physical activity levels (MVPA) were not different between the ASD and control groups. However, there was a significant relationship between MVPA and upper arm activity (Figure 4). Those with increased upper arm movement generally had reduced MVPA. This could be related to the self-stimulating extra arm movements that are sometimes seen in those with ASD, such as arm flapping (Kindregan et al., 2015). Excessive aberrant arm movement could possibly lead to increased energy expenditure in these individuals, causing them to expend more energy during normal, everyday movements, which in turn leads to a decrease in more vigorous physical activity.

Though variability has not been widely assessed in youth with ASD, especially to this degree, it has been noted that this population is known to exhibit behaviors such as increased clumsiness (Kanner & Lesser, 1958) and toe-walking (Filipek et al., 1999; Tsai, 1996). Most studies performed on subjects with ASD have also noted that the majority of children with ASD exhibit some sort of differences in gait compared to their neurotypical counterparts (Dufek et al., 2018; Eggleston et al., 2017; Shetreat-klein et al., 2014), thus it was surprising that increased variability was not found at the shank-foot or between the upper arm and thigh. A vector coding technique has not been performed amongst this population, so any observations made about gait differences in previous studies were strictly visual or from kinematic variables in the form of percent differences. Although there were some slight differences in arm-thigh angles in those with ASD, none of the differences were significant despite the known differing arm movements that some individuals with ASD exhibit (Kindregan et al., 2015).

One reoccurring conclusion in previous studies was that although it is known that those with ASD exhibit movement differences, high inter-subject variability made it difficult to pinpoint consistent differences. Based on the frequency data in this study, it does not appear that the

individual subjects in the ASD group exhibited any more limb variability than their neurotypical counterpart, though group variability was not assessed so any similarities or differences are not concrete. The lack of differences could be because the sample size is relatively low. It is also possible that the ASD subjects in this study had a lower severity level (though this data was not directly collected) compared to those reported in the previous literature.

Studies assessing these same variables have not been performed before in this population. In the future, it would be helpful for more studies to assess these variables in a wider range of subjects, also testing different joints. It is possible that previously noted ankle differences could be due to abnormalities at the knee or hip, so this is something that can be determined in further research. Aside from walking, it would also be helpful for movement to be assessed in a wide range of activities. Since physical activity levels are a concern among adolescents in general, especially those with ASD, observing and collecting data of various other movements involved in various activities (such as cutting or pivoting, in the context of an activity that would be performed at recess or after school, such as soccer) could possibly be helpful in determining why adolescents with ASD have slighter higher rates of obesity compared to their neurotypical counterparts. Also, in future research studies it should be noted if subjects require any assistance in day to day activities, and how much assistance they require, if any. With enough subjects, the ASD group can be divided up and comparisons can be made between different subjects with different assistance levels.

CHAPTER VI CONCLUSION

Within the given sample of youth with ASD and their matched neurotypical controls, there are no significant differences in coordination patterns, variability, or physical activity levels. This research suggests that ASD itself may not be a physical disorder, but the other factors involved with the condition may contribute to physical differences in some youth, given that it is an extremely heterogenous condition. Future research addressing coordination patterns should consider looking at other limbs and joints, such as the hip and knee, and possibly tracking physical activity over a longer period of time.

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VITA

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