MOVEMENT PATTERNS AND PHYSICAL ACTIVITY IN CHILDREN WITH NEURODEVELOPMENTAL DISORDERS

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DECLARATION

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SUMMARY

Autism Spectrum Disorder (ASD) is a group of complex neurodevelopmental disorders characterised by difficulty with social communication and restricted and repetitive behaviours. Developmental Coordination Disorder (DCD) is a neurodevelopmental disorder characterised by motor impairments which interfere with activities of daily living. Children with ASD and/or DCD exhibit signs of poor motor function, with potential impacts on physical activity, and are at higher risk of overweight/obesity than typically developing (TD) children.

Two literature reviews were carried out: one examining gait deviations in ASD and one examining physical activity (PA) in children with ASD or DCD. The results of both reviews were inconclusive as many studies had contradictory results. However, there was evidence or deviations in gait patterns and PA levels in the literature.

Chapter 4 measured PA levels in children with ASD and/or DCD in comparison to TD peers through survey means and Chapter 5 objectively measured PA and fitness in children with ASD and/or DCD compared to TD children. Children with ASD and/or DCD were found to be less active, more sedentary and less fit than TD peers.

Chapter 6 determined gait patterns and motor function in children with ASD and/or DCD compared to TD children. Several differences in gait parameters between children with ASD and comorbid DCD and TD controls were noted including increased step width, reduced step length, and increased knee flexion and foot progression angle. Significant differences in motor function scores were evident with the TD controls scoring consistently higher on all categories of the coordination test.

These results indicated barriers to PA in children with neurodevelopmental disorders. When this was examined further in Chapter 7, children with neurodevelopmental disorders were found to face similar barriers to TD children (as described in published research) and additional barriers. Those specific to children with neurodevelopmental disorders included tactile sensitivity and unsupportive environments.

This thesis highlighted deficits in physical activity and fitness in children with neurodevelopmental disorders that indicate risk of poor health outcomes. Some deficits in gait were detected that appear to be more pronounced in the dual diagnosis group. This study added to the literature by examining the impact of comorbidity. Significant additional barriers to exercise exist for individuals with neurodevelopmental disorders that are clearly impacting their ability to meet recommended levels of physical activity.

This research has investigated a relatively understudied topic which has the potential to increase risks for significant negative health outcomes. Further studies would help to clarify how best to encourage engagement with exercise in order to promote physical health and wellbeing, and to investigate the relationship between motor impairment and physical activity.

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Kindregan, D, Gallagher, L, Gormley, J. Gait Deviations in Children with Autism Spectrum Disorders: A Review. *Autism Research and Treatment.* Volume 2015. Article I.D: 741480

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Kindregan D., Gallagher L., Gormley J. Physical Activity in Children with Autism Spectrum Disorders and Developmental Coordination Disorder. Proceedings of the International Meeting for Autism Research, Baltimore, 2016.

LIST OF ABBREVIATIONS

ACSM – American College of Sports Medicine

ADHD – Attention Deficit Hyperactivity Disorder

ASD – Autism Spectrum Disorder

ASIS – Anterior Superior Iliac Spin

BMI – Body Mass Index

CDC – The United States Centre for Disease Control and Prevention

CODA – Cartesian Optoelectronic Dynamic Anthrompometer

CPM – Counts Per Minute

DCD – Developmental Coordination Disorder

DSM – Diagnostic and Statistical Manual of Mental Disorders

DSM5 – Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition

FAI – Football Association of Ireland

HSE – Health Service Executive

ID – Intellectual Disability

IQR – Interquartile Range

GAA – Gaelic Athletic Association

LED – Light Emitting Diode

LPA – Light Physical Activity

MABC-2 – Movement Assessment Battery for Children, 2nd Edition

MPA – Moderate Physical Activity

MVPA – Moderate to Vigorous Physical Activity

ODD – Oppositional Defiant Disorder

PA – Physical Activity

PSIS – Posterior Superior Iliac Spine

RRB – Restricted and Repetitive Behaviours

SD – Standard Deviation

SPD – Sensory Processing Disorder

TD – Typically Developing

VPA – Vigorous Physical Activity

WHO – World Health Organisation

CHAPTER 1: INTRODUCTION

1.1 NEURODEVELOPMENTAL DISORDERS

The term neurodevelopmental disorders refers to a group of conditions defined by a disruption to the development of the central nervous system (Goldstein and Reynolds, 1999). As the name suggests, these conditions manifest in the developmental period (American Psychiatric Association (APA), 2013). Neurodevelopmental disorders normally present as delays in achieving developmental milestones, for example commencement of speech or walking and may also include 'symptoms of excess', such as the restricted and repetitive behaviours characteristic of ASD (APA, 2013). Examples of neurodevelopmental disorders include autism spectrum disorder (ASD), intellectual disability (ID), developmental coordination disorder (DCD), global developmental delay and attention deficit hyperactivity disorder (ADHD). Many neurodevelopmental disorders have overlapping phenotypes and comorbidity is common (Kadesjö, 2000, Landgren et al., 1996). Motor impairment has been well documented in children with ASD and is implicit in those diagnosed with DCD as it is a disorder characterised by poorly developed motor skills (Bhat and Landa, 2011, Brasic and Gianutsos, 2000, Dewey et al., 2007). For this reason, ASD and DCD are two neurodevelopmental disorders of particular interest in the author's field of study, physiotherapy.

1.2 AUTISM SPECTRUM DISORDER

ASD is a lifelong developmental disorder which presents before three years of age and is found across all ethnic cultures and economic groups (Rogers, 2000, King and Bearman, 2011, Becerra et al., 2014). ASD is characterised by deficits in social interaction and communication and by the presence of restricted and repetitive behaviours (RRBs) (APA, 2013). The disorder was first described in the 1940s by Leo

Kanner in the USA and Hans Asperger in Germany (Kanner, 1943, Asperger, 1944). Kanner and Asperger, both physicians, documented social idiosyncrasies and communication deficits in child patients independently. With the revision of the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) in 2013, all children with autism, Asperger's syndrome or other pervasive developmental disorders receive one umbrella diagnosis of Autism Spectrum Disorder (APA, 2013). There has been a steady increase in the prevalence of ASD over the last twenty years (Fombonne, 2009, Lenoir et al., 2009). It has been suggested that this may however be due to increased access to diagnostic services, widening of diagnostic criteria or increasing awareness of the condition, which is a source of continuing debate in the autism research community (Weintraub, 2011, Wing, 1993, Matson and Kozlowski, 2011). Historically, autism was thought of as a rare disorder in Ireland and the UK, but a 2006 report by the Department of Education and Skills (UK) stated that this was no longer the case (Wilkinson, 2010). A study published in 2016 on prevalence rates of ASD in Ireland, the first of its kind, reported that ASD affects approximately 1% of the Irish population which is a similar figure to that reported in the US and the UK (Boilson et al., 2016, Fombonne, 2003, Baron-Cohen et al., 2009). Worldwide, ASD affects more males than females with a wide range of male: female ratios reported from 1.33:1 to 15.7:1 (Fombonne, 2009). The most commonly referenced ratio is approximately 4:1 (Ehlers and Gillberg, 1993, Fombonne, 2003, Yeargin-Allsopp et al., 2003). The causes of ASD have not been fully explained although it is widely thought to be influenced by the interaction between genetic and environmental risk factors (Szatmari, 2003, Samaco et al., 2005, Marshall et al., 2008, Herbert, 2010).

1.2.1 Diagnostic Criteria for ASD

The diagnostic criteria for ASD stated in the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5; APA, 2013) are displayed in Figure 1.1.

Figure 1.1: Diagnostic Criteria for ASD

A. Persistent deficits in social communication and social interaction across multiple contexts, currently or by history.

B. Restricted, repetitive patterns of behaviour, interests, or activities, currently or by history.

C. Symptoms must be present in the early developmental period (but may not become fully manifest until social demands exceed limited capacities, or may be masked by learned strategies in later life).

D. Symptoms cause clinically significant impairment in social, occupational or other important areas of current functioning.

E. These disturbance are not better explained by intellectual disability (intellectual developmental disorder) or global developmental delay. Intellectual disability and autism spectrum disorder frequently co-occur; to make comorbid diagnoses of autism spectrum disorder and intellectual disability, social communication should be below that expected for general developmental level.

1.3 DEVELOPMENTAL COORDINATION DISORDER

DCD is a neurodevelopmental disorder characterised by impaired fine and gross motor function which disrupt the affected individual's ability to carry out tasks of daily life. It is classed as a motor disorder and is also known as childhood dyspraxia, developmental dyspraxia, clumsy child syndrome or specific developmental disorder of motor function (APA, 2013). DCD is reported to affect approximately 6% of school-aged children (Fox and Lent, 1996). Similarly to ASD, more males than females are affected by DCD with published male: female ratios ranging between 2:1 and 7:1 (APA, 2013). The causes of DCD are not well understood, perhaps due to the heterogeneity of the condition (Barnhart et al., 2003). It has been suggested that DCD is "part of the continuum of cerebral palsy" as there is evidence of a prenatal delay in brain development (Barnhart et al., 2003, Hadders-Algra, 2003). DCD is more prevalent following pre-natal exposure to alcohol and in preterm children (APA, 2013).

1.3.1 Diagnostic Criteria for DCD

The diagnostic criteria for DCD stated in the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5; APA, 2013) are displayed in Figure 1.2.

Figure 1.2: Diagnostic Criteria for DCD

A. The acquisition and execution of coordinated motor skills is substantially below that expected given the individual's chronological age and opportunity for skill learning and use. Difficulties are manifested as clumsiness (e.g., dropping or bumping into objects) as well as slowness and inaccuracy of performance of motor skills (e.g., catching an object, using scissors or cutlery, handwriting, riding a bike or participating in sports).

B. The motor skills deficit in Criterion A significantly and persistently interferes with activities of daily living appropriate to chronological age (e.g., self-care and self-maintenance) and impacts academic/school productivity, prevocational and vocational activities, leisure and play.

C. Onset of symptoms is in the early developmental period.

D. The motor skill deficits are not better explained by intellectual disability (intellectual developmental disorder) or visual impairment and are not attributable to a neurological condition affecting movement (e.g., cerebral palsy, muscular dystrophy, degenerative disorder).

1.3.2 Autism Spectrum Disorder and Developmental Coordination Disorder

ASD and DCD commonly co-occur (APA, 2013). As neurodevelopmental disorders, ASD and DCD have some similarities in terms of phenotype, but are characterised by separate specific diagnostic criteria (see sections 1.2.1 and 1.3.1). In previous editions of the Diagnostic and Statistical Manual for Mental Disorders (DSM), a dual-diagnosis of ASD and DCD was not permitted. With the most recent edition, the DSM5, both diagnoses can be given if criteria for both conditions are met.

1.4 PHYSICAL ACTIVITY

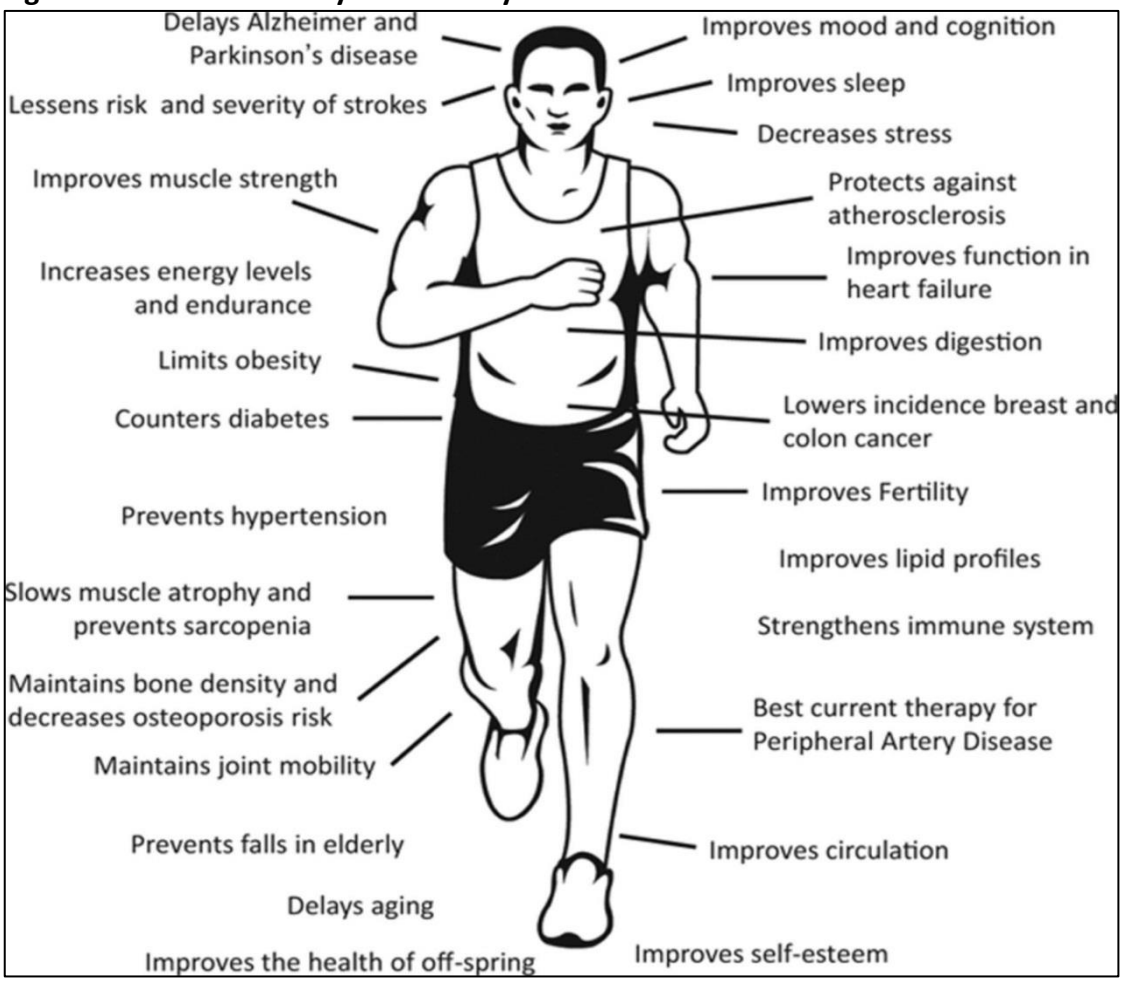
Physical activity (PA), exercise and fitness are terms which are often used together but describe three separate concepts. The World Health Organization (WHO) defines PA as "any bodily movement produced by skeletal muscles that requires energy expenditure." PA and exercise do share some common components: both are bodily movements produced by skeletal muscles which require energy expenditure. The energy expenditure required varies with the intensity of movement and both PA and exercise are positively correlated with physical fitness (Petajan et al., 1996, McGuire and Ross, 2011, Caspersen et al., 1985). The difference is that exercise is a planned and structured endeavour with an objective to improve fitness or skill (Caspersen et al., 1985). In other words, all exercise would be considered PA but not all PA could be considered exercise. Fitness has been described as "the ability to carry out daily tasks with vigour and alertness, without undue fatigue and with ample energy to enjoy leisure-time pursuits and to meet unforeseen emergencies" (Caspersen et al., 1985).

1.4.1 Physical Activity and Health

The health benefits of PA are widely known and well documented, including reducing rates of all-cause mortality (Brown et al., 2012, Johnsen et al., 2013, Loprinzi et al., 2016, Paffenbarger Jr et al., 1986). Physical benefits include a reduced risk of pathologies such as cardiovascular disease or type II diabetes, reduced hypertension, reduced adiposity and increased strength and fitness (Tremblay et al., 1994, Paffenbarger et al., 1993, Pate et al., 1995, Carpenter et al., 1999, Slentz et al., 2007, Bouchard et al., 1993). During a recent keynote address at the International Olympic Committee World Conference on Prevention of Injury and Illness in sport, Prof. Willem van Mechelen stated "there is not a single organ system not responsive to physical

activity" and made reference to a diagram which depicts many of the health benefits of PA (see Figure 1.3). PA also has many psychological and emotional benefits including reduced stress and anxiety, improvement of mood and self-confidence (De Matos et al., 2009). Conversely, a sedentary lifestyle is reported to have negative effects on health, including increased adiposity, increased risk of obesity, type II diabetes, cardiovascular disease and some cancers (Slentz et al., 2007, Bernstein et al., 1994, Carpenter et al., 1999).

Figure 1.3: Benefits of Physical Activity and Exercise



(Rowe et al., 2014)

1.4.2 Measurement of Physical Activity

Measurement of PA is becoming commonplace across many platforms including scientific research and personal fitness tracking. The latter may be due to the increasing commercial availability of simple tools for monitoring activity including pedometers and smart phone apps. Many research studies utilise triaxial accelerometers to objectively measure PA as it is a non-invasive, cost-effective and reliable method of measuring activity (Vanhelst et al., 2012).

Metabolic equivalents (METs) are commonly used to describe exercise intensity. METs are defined as "the ratio of the work metabolic rate to the resting metabolic rate" (Ainsworth et al., 1993). One MET is defined as the oxygen costs of sitting at rest and is commonly reported to be equal to 3.5 millilitres of oxygen per kilogram of body weight per minute (Jette et al., 1990, Strath et al., 2013).

A detailed compendium of physical activities first developed in 1993 and periodically updated has been used extensively in research to classify the energy costs of PA (Ainsworth et al., 1993, Ainsworth et al., 2000, Ainsworth et al., 2011). The compendium includes a comprehensive list of activities and gives descriptions of the type and category of PA for each activity in addition to their corresponding MET values. It is a useful tool for classifying activities into the following intensity categories: sedentary behaviour (≤1.5 METs), light activity (1.6-2.9 METs), moderate activity (3-5.9 METs) and vigorous activity (≥6 METs). The adult compendium was used as a partial basis for the development for a compendium of the energy costs of daily activities in children (Ridley and Olds, 2008). Examples of sedentary behaviours and activities of light, moderate and vigorous intensity are presented with their associated METs value in Table 1.1.

Table 1.1: Energy Costs of Activities for Children

ACTIVITY	METs
Sedentary Behaviours	**≤ 1.5 METs**
Watching television (sitting)	1.2
Colouring with crayons	1.4
Light Intensity Activities	**1.6 – 2.9 METs**
Catching a ball	2.6
Dressing and undressing	2.7
Moderate Intensity Activities	**3 – 5.9 METs**
Sweeping	3.6
Playground games	4.9
Vigorous Intensity Activities	**≥ 6 METs**
Rollerblading	6.5
Soccer	8.8

(Ridley and Olds, 2008)

1.4.3 Sedentary Behaviour and Physical Inactivity

Sedentary behaviour may be defined as any waking activity in a seated or reclined position with an energy cost of ≤1.5 METs (Owen et al., 2010). In current research, there is a clear division between sedentary behaviour and physical inactivity (Pate et al., 2008). Physical inactivity is used to describe failure to meet recommended exercise guidelines whereas sedentary behaviour is used to describe activities which incur a very low energy cost and are characterised by sitting or lying (Bauman et al., 2011, Hardy et al., 2013). Common sedentary behaviours in childhood include watching television and playing video games. Independent of PA levels, sedentary behaviours have been identified as a risk factor for pathologies such as cardiovascular disease and type II diabetes (Healy et al., 2016, Starkoff et al., 2014, Ekelund et al., 2006, Song et al., 2015). This suggests that regardless of the time spent in PA, prolonged periods of time spent in sedentary behaviours could be detrimental to children's current and future health (Starkoff et al., 2014).

1.4.4 Physical Activity in Childhood

Although there are numerous guidelines for PA in children, there is currently no gold standard. There is, however, a general consensus that children should be physically active for at least 60 minutes most days of the week. The American College of Sports Medicine (ACSM) recommends that children and adolescents engage in at least 60 minutes of moderate to vigorous PA (MVPA) per day, with vigorous-intensity PA, bone-loading activities and resistance training on at least three days per week (ACSM, 2013).

Children and adolescents tend to be more physically active than adults. However, US statistics have shown that it is mainly only younger children who are meeting recommended guidelines for PA, while the majority of children over the age of ten years are not as physically active as recommended (ACSM, 2013). In fact, only 8% of those aged 12-19 years were reported to meet PA guidelines (ACSM, 2013). A similar figure of 7% was reported in Canadian children (Colley et al., 2011).

Several longitudinal studies have found that PA levels in childhood and adolescence are related to PA levels in adulthood (Telama et al., 2005, Makinen et al., 2010). Additionally, PA, specifically weight-bearing exercise, in childhood and adolescence is vital for bone development and density (Hind and Burrows, 2007, MacKelvie et al., 2002). The results of one study suggested that although those who were more active in childhood have been shown to be more active in adulthood, those who were forced to participate in sports or have had negative experiences with exercise as a child or adolescent may be less active as adults (Taylor et al., 1999).

A review of correlates of PA in children revealed that age and gender were associated consistently with PA during and after school (Stanley et al., 2012). Boys tended to be

more active than girls, and age was negatively correlated with PA such that as children got older, PA decreased (Stanley et al., 2012). Many studies have reported that children with disabilities, including neurodevelopmental disorders, are less likely to participate in sustained bouts of vigorous PA compared to peers without disability (Srinivasan et al., 2014, Rimmer and Rowland, 2008, Rimmer et al., 2007).

1.4.5 Physical Activity in Children with ASD

Extensive research has been carried out examining PA in children with ASD with methods varying from questionnaires (both parent-report and self-report) to the use of triaxial accelerometers. Several studies have reported that children with ASD were less physically active than typically developing peers and further studies have suggested a low proportion of children with ASD are as physically active as guidelines recommend (Borremans et al., 2010, Srinivasan et al., 2014, Pan et al., 2016). Published research which has objectively measured PA in ASD compared to typically developing controls is reviewed in more detail in Chapter 2.

Studies examining the effects of age and gender on PA in children with ASD report a similar pattern to typically developing children. Girls with ASD were less active than boys with ASD and adolescents with ASD were less active and spent more time in sedentary behaviours than their younger counterparts (Memari et al., 2013, Memari et al., 2015, Macdonald et al., 2011, Pan and Frey, 2006). One study has examined PA in Irish adolescents with ASD (Healy et al., 2016). They found that those with ASD spent significantly less time in light and moderate to vigorous PA compared to typically developing controls (Healy et al., 2016). Overall participation in sports, both team and individual, was reduced in adolescents with ASD compared to typically developing

controls, as was the variety of sports participated in by individuals with ASD (Healy et al., 2016).

1.4.6 Physical Activity in Children with DCD

The number of studies examining PA in children with DCD has been steadily growing over the last number of years. Children with DCD have been found to be significantly less active in comparison to typically developing peers (Silman et al., 2011, Visser et al., 1998, Bouffard et al., 1996, Cermak et al., 2015). Several qualitative studies have shown that participation in sports and exercise activities are associated with the severity of motor impairment in children with DCD (Fisher et al., 2005, Castelli and Valley, 2007, Hay and Missiuna, 1998). Another similarity to children with ASD was demonstrated in a research study which found that participation in team sports was reduced in children with DCD compared to typically developing children (Christiansen, 2000).

A longitudinal study of PA in children with DCD reported lower levels of MVPA compared to typically developing children (Kwan et al., 2016). The study also found that boys with DCD were more physically active than girls with DCD, as is reported in both typically developing children and children with ASD (Kwan et al., 2016). PA of children with DCD declined at a similar rate to typically developing children, suggesting that the relative inactivity of children with DCD compared to typically developing children begins earlier in childhood in DCD (Kwan et al., 2016). Further published research which has objectively measured and compared PA in DCD and typically developing controls is reviewed in Chapter 2.

1.5 FITNESS

The Oxford dictionary defines the word 'fitness' as "the condition of being physically fit and healthy" (Stevenson, 2010). In some circumstances it is a relative term – for example an elite swimmer will generally have excellent cardiorespiratory fitness, pulmonary function and tolerance for aerobic and anaerobic exercise but may be a poor runner due to the additional weight-bearing component. Oxygen consumption (VO_2) is a measure of the energy costs of exercise. Aerobic fitness and maximal oxygen consumption (VO_2^{max}) are directly related. Increased fitness implies an increased oxygen uptake. There are many commonly used methods of measuring fitness ranging from maximal aerobic tests such as the Bruce Protocol to maximal strength and power tests such as the one repetition max test. In children, fitness testing is commonly carried out through field tests such as the 20 Metre Multistage Fitness test (Hamlin et al., 2014).

1.5.1 Components of Fitness

The components of fitness may be divided into two categories: health-related fitness and skill-related fitness (Caspersen et al., 1985). Health-related components of fitness include cardiorespiratory endurance, muscular strength and endurance, body composition and flexibility (Caspersen et al., 1985). Skill-related components of fitness include agility, balance, coordination, power, reaction time and speed (Caspersen et al., 1985).

1.5.2 Fitness in Childhood

There is some evidence to suggest that measures of health- and skill-related fitness show moderate positive correlations with age from childhood into adolescence (Malina, 1996). A review of the effects of physical fitness on health outcomes in

childhood and adolescence reported a negative association between cardiorespiratory fitness and abdominal adiposity (Ortega et al., 2008). Associations for both cardiorespiratory fitness and muscular fitness (strength and endurance) with cardiovascular disease risk factors have also been established (Ortega et al., 2008, Twisk et al., 2002).

1.5.3 Fitness in Children with ASD

There is growing concern over declining fitness rates and increasing prevalence of overweight and obesity in children with ASD (Dickinson and Place, 2014, Corvey et al., 2016). Several studies have assessed cardiorespiratory fitness in children with ASD and the majority have found lower levels compared to typically developing children (Pan, 2014a, Pan et al., 2016). However, one study did not find differences in cardiorespiratory fitness between ASD and typically developing children but did find that children with ASD scored significantly lower on strength tests (Tyler et al., 2014).

ASD children have been found to score significantly lower than typically developing children across a battery of aerobic and strength tests, including a cardiorespiratory fitness test, isometric push-ups, curl-ups and sit-and-reach tasks (Pan et al., 2016). The authors of this study concluded that these results suggested lower levels of cardiovascular endurance, abdominal and upper body muscular strength and endurance and lower reduced flexibility than typically developing peers (Pan et al., 2016).

1.5.4 Fitness in Children with DCD

Similar to ASD, cardiorespiratory fitness has been shown to be lower in children with DCD in comparison to typically developing counterparts (Haga, 2008, Silman et al., 2011, Chia et al., 2010, Rivilis et al., 2011, Wu et al., 2010, Cermak et al., 2015). One

large longitudinal study examining fitness in children with DCD found that children had a lower peak VO_2 values at baseline compared to typically developing controls (Cairney et al., 2011). Furthermore, fitness was shown to have declined at a steeper rate over the two-year period studied (from 9-11 years of age) in children with DCD than in typically developing children at follow-up (Cairney et al., 2011). Measures of power and muscular fitness have also been found to be significantly lower in children with DCD compared to typically developing children (Raynor, 2001, Schott et al., 2007, Kanioglou, 2006).

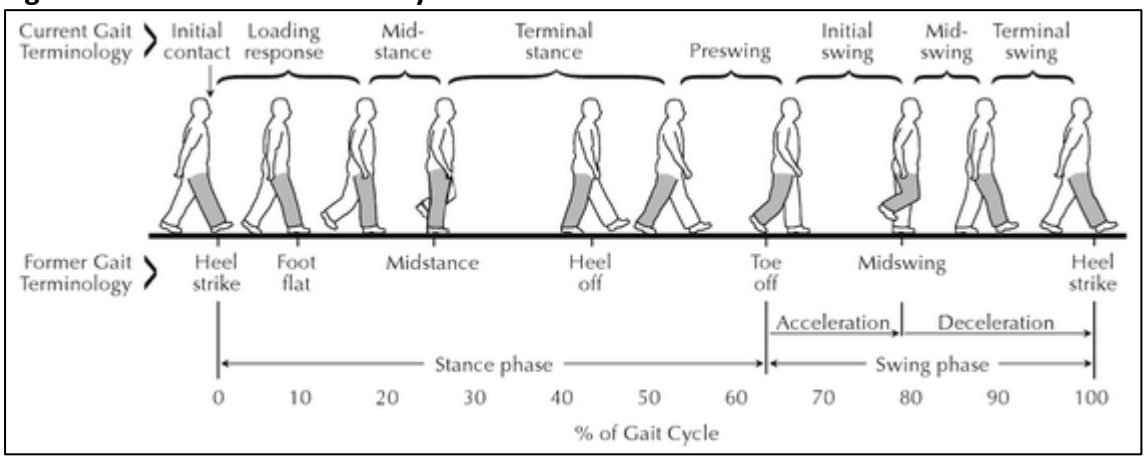
1.6 GAIT

1.6.1 Introduction to Gait

The term 'gait' describes the 'manner or style of walking' (Levine et al., 2012). Gait is made up of a series of repetitive events and the gait cycle is defined as the time interval between one of these events and the next occurrence of the same event. The most commonly referenced event by which to describe the start and end of a gait cycle is initial contact. Initial contact, as the name suggests, occurs when the foot first touches the ground. In normal gait, the heel is the part of the foot which makes first contact with the ground. The gait cycle is normally described in two main phases: the stance phase and the swing phase. The stance phase occurs when the foot is placed on the ground. The swing phase occurs when the foot is off the ground and it is this phase which produces forward progression of the body. Using the right initial contact as a start point, one right gait cycle will continue until the next right initial contact. The distance covered by this right cycle is known as the right stride length, whereas step length refers to the distance between the initial contact on the right foot and the left foot (and vice-versa). Step width is the horizontal distance between the right and left

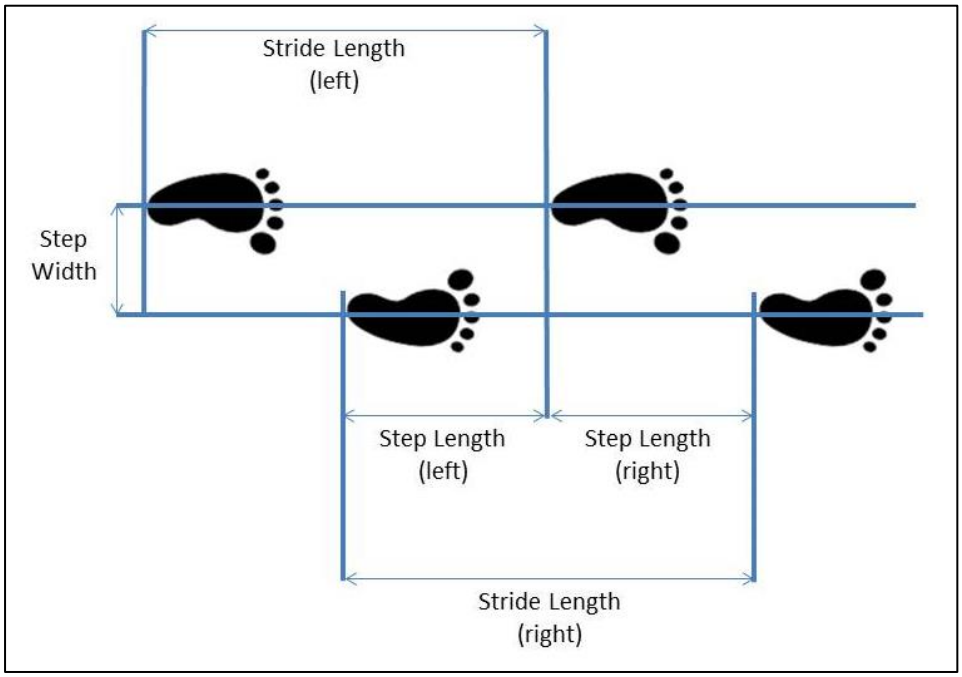
foot placement. Figures 1.4 and 1.5 show the phases of the gait cycle and a description of step length, step width and stride length, respectively.

Figure 1.4: Phases of the Gait Cycle



(Miller et al., 2008)

Figure 1.5: Step Width, Step Length and Stride Length



1.6.3 Gait Analysis

The movement of humans and animals has been studied as far back as 350BC when Greek Philosopher Aristotle wrote *De Motu Animalium*, translated as 'the movement of animals' or 'the gait of animals'. The earliest scientific gait analysis on record was

carried out in the 17th century by an Italian scientist named Giovanni Alfonso Borelli. Borelli's ideas, based on Newtonian principles of mechanics, have been attributed as the first descriptions of gait's distinct phases and the displacement of the body's centre of gravity as imperative for progression and balance (Al-Zahrani and Bakheit, 2008, Baker, 2007).

Gait analysis is commonplace in clinical and research practice. The methods in each of these settings, however, tend to differ dramatically due to cost, time consumption and availability of scientific gait analysis systems. Although the clinical use of scientific gait analysis systems has been steadily increasing since 1980, it is still an expensive method of analysing patients' gait (Whittle, 2014). The most common method of clinical gait analysis is the use of observational or visual analysis as digital video cameras have become affordable and widely available tools (Sutherland, 2001, Sutherland, 2002, Sutherland, 2005).

1.6.2 Requirements for Normal Gait

As with any complex movement of the human body, there are a number of systems working cohesively to produce normal gait. Skeletal muscles contract to generate movement at the joints, powered by the cardiovascular system and coordinated by the central nervous system. Motor control, proprioception and balance, thus, are vital components in producing fluid movements and precise, coordinated foot placement. Therefore, deviations from normal gait patterns may be expected in disorders in which information processing and sensory-motor deficits have been documented, such as ASD or DCD (Wilson and McKenzie, 1998, Piek and Dyck, 2004).

1.6.4 Gait Development in Childhood

Gait in infancy and childhood is different to adult gait in several ways. Typically in young children initial contact is made by the flat foot rather than the heel, there is little knee flexion during the stance phase and during the swing phase the hip is externally rotated (Levine et al., 2012, Sutherland et al., 1988). These differences normally mature to match the adult pattern by the age of two years (Sutherland et al., 1980). Young children also tend to have a wider base of support and lack reciprocal arm swing, they adapt the adult pattern for these parameters by the age of four years (Sutherland, 1997, Levine et al., 2012). Several parameters continue to adapt with growth until plateauing around the age of 15 years including cadence (steps per minute), stride length, walking velocity and the length of the gait cycle. Cadence decreases with increasing age while stride length, velocity and cycle time increase (Levine et al., 2012, Sutherland et al., 1988).

1.6.5 Gait in Children with ASD

Gait abnormalities are well documented in children with ASD. There is a higher prevalence of idiopathic toe-walking in children with ASD compared to typically developing children (Esposito et al., 2011, Barrow et al., 2011). Gait symmetry (i.e. comparison of parameters of the right and left lower limbs) has been shown to be similar in children with and without ASD (Chester and Calhoun, 2012). Major findings of gait analysis studies in children with ASD suggest a tendency to augment walking stability with a wider base of support and shorter steps compared to typically developing children (Shetreat-Klein et al., 2012, Nobile et al., 2012, Nayate et al., 2005). The gait patterns of children with ASD are reviewed in greater detail in Chapter 2 (see Section 2.1, page 22).

1.6.6 Gait in Children with DCD

Few studies have comprehensively analysed gait in children with DCD. As of yet, no characteristic pattern has been described. This may be in part due to a high variability of gait parameters between subjects with DCD (Rosengren et al., 2009, Wilmut et al., 2016, Du et al., 2015). Asymmetry of gait parameters has also been documented (Wilmut et al., 2017). Wider step width was documented in one study, prompting the author to conclude that children with DCD have difficulty in controlling their centre of mass (Wilmut et al., 2016). These factors combined suggest instability and a lack of coordination and balance in the gait patterns of children with DCD.

1.7 MOTOR SKILLS AND COORDINATION

1.7.1 Development of Motor Function and Coordination in Childhood

Motor development refers to the development of a child's musculoskeletal and nervous system leading to the ability to move and manipulate their environment (Marotz and Allen, 2015). The American Academy of Pediatrics recommends that all children are screened for general development at ages 9, 18 and 24 or 30 months of age (CDC, 2015). In Ireland, infants are screened at birth and at ages 3, 7 and 9-12 months and 2 and 3 years of age. These 'Developmental Examinations' are carried out by public health nurses (HSE, 2016).

Motor function is usually described in terms of gross motor and fine motor function. Gross motor function refers to the body's ability to carry out large movements of the limbs and/or trunk in a coordinated and fluid manner. Examples of gross motor function include kicking a ball, throwing and catching objects, jumping, walking and running. Fine motor function refers to the body's ability to carry out small (fine)

movements of the hands in a smooth coordinated fashion. Examples of fine motor function include handwriting, colouring, tying shoelaces and threading a needle.

1.7.2 Motor Function and Coordination in Children with ASD

Extensive research has been carried out across a variety of scientific fields examining motor impairments in ASD. Motor deficits present early in childhood and may persist into adulthood (Matson et al., 2011). Motor impairments have been found to be more common in children with ASD than typically developing children, with motor apraxia presenting in approximately one-third of children with ASD (Ming et al., 2007). A comprehensive review on the subject concluded that motor coordination deficits were a "cardinal feature of ASD" (Fournier et al., 2010).

1.7.3 Motor Function and Coordination in Children with DCD

Motor impairment, i.e. the loss or deficit of motor function, is inherent in the diagnosis of DCD and is undisputed in the literature. Thus, the literature is more concerned with examining interventions to improve motor function or identifying associations between motor function and other variables such as physical fitness and PA.

1.8 RATIONALE FOR THESIS

Many of the areas of interest in this thesis have been studied in children with ASD and also separately in children with DCD. There is, however, a subset of children with ASD who have more severe motor deficits leading to a secondary diagnosis of DCD. Severe motor impairments combined with deficits in social communication and interaction could make it even more difficult to engage in PA and therefore leads to an increased risk of inactivity and increased likelihood of engaging in sedentary behaviours. As shown earlier in the introduction, inactivity and sedentary behaviours increase the risk of obesity and related health consequences. PA patterns in childhood and adolescence

are known to track into adulthood patterns, further compounding the health risks in later life. There are inconsistencies in the literature relating to DCD and ASD that need to be further clarified. There is a deficit in the literature regarding ASD comorbidity that needs to be further explored. The studies in this thesis include cohorts with sub-groups of children with a diagnosis of ASD, children with a diagnosis of ASD and comorbid DCD, children with a diagnosis of DCD and typically developing children for comparison. This allows the author to tease out any potential differences between differing diagnoses and also with typically developing children.

1.9 AIMS OF PHD

The overarching aims of this thesis are to further investigate the impact of neurodevelopmental disorders, namely ASD and DCD and comorbidity of these disorders on physical activity, fitness, gait, motor coordination and barriers to exercise and physical activity. Specifically:

1. To determine frequency, intensity and duration of physical activity in children with ASD and/or DCD compared with typically developing controls
2. To compare fitness levels in children with ASD and/or DCD with typically developing controls
3. To compare the efficiency of gait in children with ASD and/or DCD with typically developing controls
4. To compare temporal-spatial gait parameters in children with ASD and/or DCD with typically developing controls
5. To compare kinematic gait parameters in children with ASD and/or DCD with typically developing controls

6. To compare motor coordination in children with ASD and/or DCD with typically developing controls
7. To investigate the relationship between physical activity, fitness, gait and motor coordination in children with ASD and/or DCD
8. To investigate the barriers to and benefits/motivators for PA and exercise in children with neurodevelopmental disorder

CHAPTER 2: LITERATURE REVIEWS

This chapter describes two literature reviews which were carried out by the author. Both reviews were narrative in nature but a systematic approach was applied for clarity.

2.1 LITERATURE REVIEW 1: GAIT DEVIATIONS IN CHILDREN WITH AUTISM SPECTRUM DISORDERS

2.1.1 Introduction

Children with ASD have difficulty with social interaction, communication and language skills with many children demonstrating restrictive and repetitive behaviours (APA, 2013). These behaviours may include rocking, finger flicking or arm flapping (Lord and Jones, 2012). Motor stereotypies are defined as 'involuntary, coordinated, patterned, repetitive, rhythmic, and purposeless but seemingly purposeful movements (Marsden and Fahn, 2013). Children with ASD have been found to demonstrate numerous gait stereotypies such as pacing, jumping, hopping, skipping and spinning and it has been suggested that these may also be considered restrictive and repetitive behaviours (Goldman et al., 2009).

The 'manner or style of walking' or gait, is described as a method of locomotion using reciprocal placement of the lower limbs to provide both propulsion and support by Levine et al. (2012). Alterations in movement patterns in children with ASD were noted as far back as 1943 by Kanner et al., (1943) who found that those with ASD often demonstrated "clumsy" gait and gross motor patterns. In more recent years, Ghaziuddin and Butler (1998) found that children with ASD demonstrated poorer coordination than those with Asperger's disorder. Many studies have subsequently

examined motor coordination in children with ASD and a recent review, by (Fournier et al., 2010), provided further evidence that children diagnosed with ASD may be "less coordinated and show fewer motor capabilities." This may therefore suggest that gait disturbances may be common among children with ASD. For example, it has been observed that children with ASD are more prone to idiopathic toe walking than age-matched healthy controls (Barrow et al., 2011, Marcus et al., 2010). This, however, is only evident in children under six years of age (Barrow et al., 2011).

The motor deficits reported in association with ASD, i.e. impaired vestibular control and fine and gross motor abnormalities have been likened to patients with known cerebellar deficits (Freitag et al., 2007). Deficits in smooth pursuit and saccadic eye movements are reported in ASD and are suggestive of vermal dysfunction in the cerebellum (Takarae et al., 2004a, Takarae et al., 2004b). Neuroimaging studies in children with ASD show reduced ipsilateral activation of the cerebellum during gross motor movement and more diffuse activation in lobules VI-VII (Mostofsky et al., 2009, Allen and Courchesne, 2003). Cerebellar deficits are widely reported in ASD, reduced Purkinje and granule cells, and vermal hypo and hyperplasia have been reported and both pre and post-natal processes have been implicated (Arin et al., 1991, Bailey et al., 1998, Bauman and Kemper, 2005, Courchesne et al., 2005, Whitney et al., 2008). The cerebellum has been implicated in the motor deficits in ASD via connections with the parietal lobe and wide ranging connections with cortical and subcortical brain regions serve to modulate multiple brain functions that are impaired in ASD (Schmahmann et al., 2004). Deficits in postural control and gait in ASD have been linked to dysfunction in sensory integration to the cerebellum or to the basal ganglia due to similarities with

gait abnormalities observed in Parkinson's Disease (Fournier et al., 2010, Minshew et al., 2004, Rinehart et al., 2006a). Abnormalities in basal ganglia shape have also been associated with motor deficits in ASD (Qiu et al., 2010).

In adults, the first study on kinematic and kinetic gait patterns in ASD was carried out by Hallett et al. (1993). Adults with ASD were found to demonstrate "mild clumsiness" during gait but the only significant deviation in pattern from typically developed controls was a reduced range of motion at the ankle joint (Hallett et al., 1993).

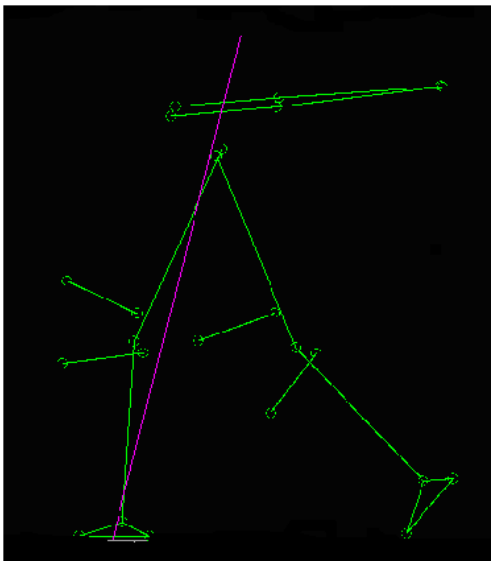
Following on from this, in the last twenty years there have been several papers published which examined gait patterns in children with Autism Spectrum Disorders. Therefore, the objective of this review is to survey and summarise the findings of these studies with regard to ASD.

2.1.2 Methods

Terminology

The terms kinematics and kinetics are used to describe gait. Kinematic analysis of gait describes the linear and angular displacement, velocities and accelerations of motion. Inherent in kinematic analysis is the description of motion from a temporal-spatial (i.e. timing and distance) perspective which describes for example step or stride length as well as cadence (steps per minute) and gait velocity. Kinetics is the study of the forces that cause motion. In movement analysis, kinetic parameters define the forces causing the movement. The most common force acting on the body during gait is the ground reaction force (GRF – see Figure 2.1), which is the force exerted by the ground on the foot (Winter, 1984). Another kinetic parameter commonly described is the joint moment which is the turning effect of a force generated by a muscle across a joint.

Figure 2.1: Ground Reaction Force Vector (generated by Codamotion software)



The downwards force exerted by the foot on the ground is equalled by the force exerted on the foot by the ground.

The purple line shows the upwards direction of this ground reaction force.

Search strategy

Articles included in this review were retrieved from five online databases (PubMed, EmBase, Medline, CINAHL and Web of Science) by a single investigator. The following key terms were used: gait, autism. Reference lists of the articles retrieved in this search were then manually searched.

Selection criteria

Studies which assessed the relationship between temporal-spatial, kinematic or kinetic gait parameters and ASD in children aged between four and 18 years were selected for inclusion in this review. Studies written in English and published between January 1970 and October 2016 were included. Of 127 studies retrieved in the search, 12 studies met the inclusion criteria and were therefore selected and included in this review. Table 2.1 shows the studies included for review and their major findings.

Table 2.1: Studies Included in Literature Review 1

Author	**Sample Size** Total (ASD)	**Average Age in Sample (years)**	**Gait Analysis Method**	**Major Findings**
Lim et al. (2016)	30 (15)	11.2	GAITRite	Increased cycle time, stance time and double support time, reduced cadence and velocity and a reduced step width in ASD.
Shetreat-Klein et al. (2012)	76 (38)	4.58	Video Analysis	Gait with wide-base of support common in ASD
Weiss et al. (2013)	19 (9)	19	GAITRite	Reduced stride length and increased stance time in ASD
Chester and Calhoun (2012)	36 (14)	6.06	8-Cam Vicon	No significant differences in mean temporal-spatial gait parameters
Nayate et al. (2005)	33 (11)	12.75	GAITRite	Increased step width in ASD; Visual cues increased stride length variability in ASD
Calhoun et al. (2011)	34 (12)	6.06	8-Cam Vicon	Increased cadence, reduced peak ankle plantarflexion and hip flexion moments in ASD
Nobile et al. (2011)	32 (16)	10.28	ELITE	Increased step width, reduced ankle plantar flexion and knee flexion-extension at toe-off, and a reduced hip range of motion in ASD
Rinehart et al. (2006a)	30 (10)	10.69	Clinical Stride Analyzer	Increased variability in stride length in ASD
Rinehart et al. (2006b)	22 (11)	5.79	GAITRite	Increased variability in stride length and stride time in ASD
Vernazza-Martin et al. (2005)	15 (9)	5	ELITE	Reduced step length in ASD
Ambrosini et al. (1998)	8 (8)	10.8	5 Cam Vicon	Reduced stride length, increased step width and reduced ground reaction forces during terminal stance in ASD
Vilensky et al. (1981)	41 (21)	7.73	Video Analysis	Reduced stride length and increased stance time in ASD. Reduced ankle dorsiflexion and knee extension at initial contact in ASD

2.1.3 Results

Temporal-spatial parameters

Temporal and spatial parameters refer to gait parameters which are related to timing and displacement or distance. The temporal-spatial parameters that have been examined in this review are stride length, step length, step width, cadence (steps per minute), velocity, stance time and double support. Of the ten studies that examined stride/step length, five found that stride length/step length was significantly reduced in children with ASD compared to healthy controls (Weiss et al., 2013; Nobile et al., 2011; Vernazza-Martina et al., 2005; Ambrosini et al., 1998; Vilensky et al., 1981), which is consistent with the study by Hallett et al. (1993) who found a reduced stride length in adults with ASD. The other six studies, however, found no significant differences in stride length between children with ASD and controls (Lim et al., 2016; Rinehart et al., 2006a; Nayate et al., 2005; Calhoun et al., 2011; Rinehart et al., 2006b; Chester & Calhoun, 2012). Step width was assessed in five studies and has been found to be significantly increased in children with ASD in four (Lim et al., 2016; Nobile et al., 2011; Shetreat-Klein et al., 2014; Nayate et al., 2005), but no significant differences were found by another (Rinehart et al., 2006b).

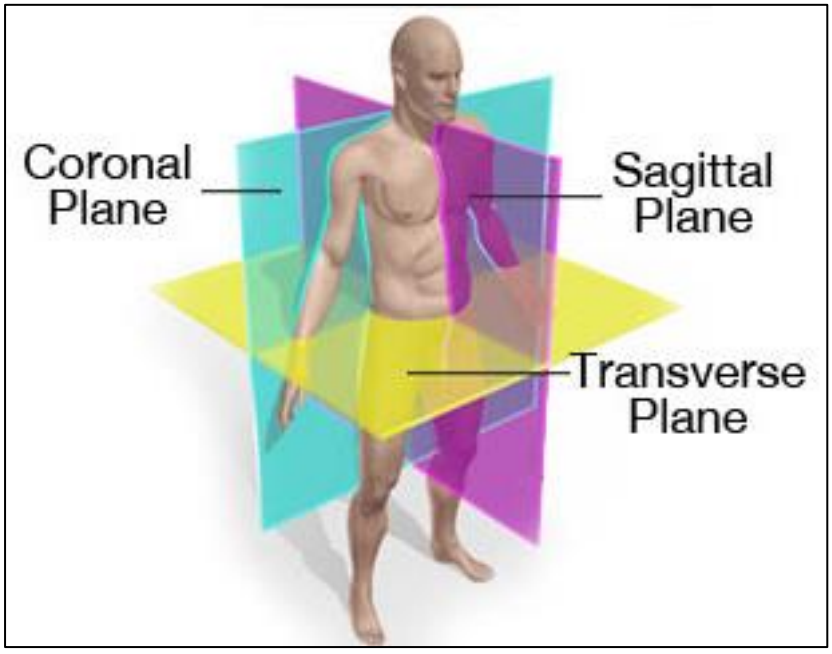
Of the 12 studies, nine assessed cadence. Calhoun et al., (2011) found that children with ASD had, on average, a higher cadence than the controls, contrasting with Weiss et al., (2013) and Lim et al., (2016) who found it to be reduced in the ASD group and the six studies which found no significant difference between groups (Rinehart et al., 2006a; Nobile et al., 2011; Vernazza-Martin et al., 2005; Vilensky et al., 1981; Nayate et al., 2005; Rinehart et al., 2006b; Chester & Calhoun., 2012).Velocity was assessed by ten studies, but no significant differences between children with ASD and controls

were found by six of them (Rinehart et al., 2006a; Vernazza-Martin et al., 2005; Nayate et al., 2005; Calhoun et al., 2011; Rinehart et al., 2006b; Chest & Calhoun, 2012). Velocity was, however, found to be significantly reduced in the ASD in three studies (Lim et al., 2016; Weiss et al., 2013; Ambrosini et al., 1998) and slightly reduced in a fourth (Nobile et al., 2011). In three studies, stance time was found to be significantly increased (Lim et al., 2016; Weiss et al., 2013; Ambrosini et al., 1998), but no significant difference was found in four other studies (Nobile et al., 2011; Vernazza-Martin et al., 2005; Calhoun et al., 2011; Chester & Calhoun, 2012). Double support was examined by seven studies, with two finding significant differences. Weiss et al., (2013) and Lim et al., (2016) found that double support time was significantly increased in the ASD group when compared to controls.

Kinematic parameters

Kinematics is the study of motion without regard to the forces that cause it. In gait, kinematic parameters refer to joint motions and angles at specific points in the gait cycle. These motions occur within three planes – the sagittal plane, the coronal plane and the transverse plane (Figure 2.2). The sagittal plane is the plane through which most of the motion of gait occurs. As such it is the plane that splits the human body into right and left and the plane through which forward/backward motion occurs – for example flexion/extension of the hip. The results will be discussed by joint in order of ankle, knee and hip.

Figure 2.2: Anatomical Planes of the Human Body



Children with ASD were found by Vilensky et al., (1981) to have reduced dorsiflexion of the ankle joint at ground contact but found other ankle joint angles to be within normal limits. At toe off, significantly reduced plantarflexion was detected by Nobile et al., (2011) in children with ASD compared to controls. Children with ASD had overall reduced range of motion at the ankle joint but not significantly so (Ambrosini et al., 1998). Ambrosini et al., (1998) also found that children with ASD had slightly, but not significantly, increased dorsiflexion during mid-stance and toe-off. This may be interpreted to mean they had reduced plantarflexion at toe-off, as found by Nobile et al., (2011).

At the knee, children with ASD were found to have significantly reduced ROM with a decreased flexion-extension angle at toe-off when compared to healthy controls (Nobile et al., 2011). Significantly reduced knee extension in children with ASD was also found by Vilensky et al., (1981) only this time at initial contact. At the hip joint there is no consensus with Nobile et al., (2011) finding a significantly reduced range of

motion at the hip but Vilensky et al., (1981) found that the children with ASD had increased hip flexion at toe-off.

Kinetic parameters

Kinetic gait parameters are those which are concerned with the forces involved in the production of movement. Calhoun et al., (2011) and Ambrosini et al., (1998) are the only studies to investigate kinetic gait parameters in children with ASD. In the study by Calhoun et al., (2011) children with ASD were found to have reduced peak plantarflexion moments at the ankle but all other ankle kinetics were within normal ranges. No significant differences were found in knee joint kinetics, but one significant difference was found in hip joint kinetics between groups. The children with ASD had decreased peak hip flexor moments compared to the control group.

Ground reaction forces are the forces exerted by the ground on an object or body in contact with it. Children with ASD were found to have relatively normal ground reaction forces, with the exception being the second vertical peak which was reduced in children with ASD when compared to normative data (Ambrosini et al., 1998). The second vertical peak refers to the ground reaction force during the period of terminal stance, which ends with toe-off.

Variability in gait

Three studies investigated the effect of external factors on the variability of gait in children with ASD. One study by Nayate et al., (2005) examined the effect of self-determined speed on the temporal spatial gait patterns of children with ASD. They asked the children to walk at their normal pace, and then asked them to walk at faster

and slower rates, and found that the children with ASD widened their base of support while walking at increased speed. They also studied the effect of cueing and concurrent tasks on gait in children with ASD. Their results showed that visual cues significantly increased stride length variability in children with ASD, but effects of dual tasks – tapping while walking (motor task) and counting while walking (cognitive task) – were not statistically significant. Some studies found that children with ASD had significantly increased variability in their stride lengths (Rinehart et al., 2006a; Rinehart et al., 2006b), walking velocities and stride times (Rinehart et al., 2006b). One study found that children with ASD demonstrated an unusual cadence-stride length relationship in their gait pattern, with an increased stride length at a given cadence compared to controls (Nayate et al., 2005).

In summary, between-group gait parameter varied across the different studies. Stride/step length was found to be increased in ASD in half of the studies that examined it. Step width was assessed in two studies and was found to be increased in both. Cadence was found to be increased in children with ASD in one study, but decreased in another, and no significant differences were found by six other studies. Stance time has been found to be increased in ASD in two studies, but other studies found no differences. Joint ranges of motion were found to be significantly different in children with ASD compared to controls in all studies that presented kinematic data. However the joints affected and the periods of the gait cycle in question varied between studies. Two studies assessed kinetic gait parameters. The ASD groups demonstrated altered joint moments at the ankle and the hip in one study and ground reaction forces were found to be reduced in another.

2.1.4 Discussion

Although there are few studies completed in the area of gait analysis in children with ASD, some emerging commonalities were identified. Some studies have found differences in temporal and spatial gait parameters between children diagnosed with ASD and healthy controls. The most common deviations found are an increased step width, and debatably, a decreased step length and stride length. Nayate et al., (2012) revealed an increase in stride length at a given cadence when compared to controls, which may account for the disagreement between studies, as this may not have been taken into account. Cadence was found to be increased in children with ASD in one study, and since the children were found to take smaller steps in many studies, this is not surprising. An increased step width gives the children a wider base of support, and reduced step and stride lengths allows them to keep their centre of gravity firmly within this base of support. This, combined with reduced velocity and increased time in the stance phase of gait, suggests a tendency to augment their stability during walking. This may be partially explained by issues with balance, proprioception or behavioural anxiety (Whyatt and Craig, 2012, Torres et al., 2013, Rinehart et al., 2002).

Of the studies in this review, three which studied kinematic parameters have found reduced range of motion during gait in children with ASD, especially ankle dorsiflexion. Nobile et al., (2011) and Ambrosini et al., (1998) found that there was increased ankle dorsiflexion (i.e. decreased plantarflexion) at toe-off. Vilensky et al., (1981) found that children with ASD had reduced dorsiflexion at heel strike, but did not find any significant differences at toe-off. It is not possible therefore to draw a conclusive picture of changes at the ankle joint during gait in children with ASD. At the knee, children with ASD were observed to have reduced ROM in two studies (Nobile et al.,

2011; Vilensky et al., 1981). However, there was no agreement at the hip joint with Nobile et al., (2011) stating that children with ASD had reduced hip ROM but Vilenksy et al., (1981) suggesting an increase in hip flexion in children with ASD. An increase in hip flexion would fit in with the above findings of reduced plantarflexion at toe-off as increased hip flexion would compensate for the reduced propulsive force and aid in foot clearance as the ipsilateral limb enters the swing phase.

Overall, the kinematic findings of these studies are sparse and often contradict each other. There is a lack in comparability in results as analysis differs. For example, Vilensky et al., (1981) concentrates on the angles at initial contact, whereas Nobile et al., (2011) focuses on those at toe-off. These possibly suggest that there is a general reduction in range of motion at the joints of the lower limbs in children with ASD during gait. However, little research has been done solely on joint mobility in ASD so it is unclear whether children with ASD have reduced range of motion or simply have a more rigid gait pattern than healthy controls. This would fit in with the aforementioned idea that children with ASD seek to stabilise their gait. Further research in the area should aim to clarify this.

Only two studies investigated kinetic gait parameters and yielded few results of significance. The only significant differences in joint moments between children with ASD and healthy controls were in peak ankle plantarflexion moments and peak hip flexor moments. The children with ASD had reduced peak plantarflexion moments, meaning that the forces acting around the ankle joint during flexion were not as high. Since children with ASD have been shown by Vilensky et al., (1981) to have reduced plantarflexion at toe-off, it makes sense that the forces generated would also be

reduced. This finding may imply a weakness of the plantarflexor muscles or may be due to the group's reduced peak plantarflexion angles as less force is required to generate a smaller movement in the joint. The author suggested that it may also be caused by hypotonia, which was confirmed in one-third of the children diagnosed with ASD. Children with ASD were also found to have reduced peak hip flexor moments but had increased hip flexion angles. This may imply weakness in the hip flexor muscles as they are unable to generate the same amount of force as those that healthy controls can, and the increased angles may imply weakness or a lack of control of the hip flexor muscles.

The findings of many studies in children with ASD concluded that gait abnormalities observed are indicative of widespread dysfunction in cerebellar and frontostriatal basal ganglia circuitry (Rinehart et al., 2006a; Weiss et al., 2013; Nobile et al., 2011; Vernazza-Martin et al., 2005; Ambrosini et al., 1998; Vilensky et al., 1981; Shetreat-Klein et al., 2014; Nayate et al., 2005; Rinehart et al., 2006b). This is in agreement with Hallett et al. (1993), who found that adults with ASD had a gait pattern similar to patients with Parkinson's and suggested cerebellar involvement. A study comparing brain images of children with ASD to healthy controls showed abnormal cerebellar maturation in the ASD group (Rinehart et al., 2002). The kinematic gait pattern exhibited by children with ASD shares common characteristics with 'crouch gait', a pattern commonly elicited in Parkinson's disorder which involves changes to the cerebellum. As referenced previously a wide range of studies point towards cerebellar deficits in ASD based on post-mortem histopathological studies, structural and functional imaging. It has been argued that cerebellar dysfunction may explain the

heterogeneous deficits observed in ASD, both sensory motor and cognitive (Schmahmann, 1997).

The increased variability of gait parameters exhibited however by children with ASD, as examined by three studies, may suggest an association with extensive neurobiological dysfunction which is unlike adult-onset disorders such as Parkinson's (Rinehart et al., 2006a; Nayate et al., 2005; Rinehart et al., 2006b). Furthermore, the lack of improvement with visual cues and increase in variability of gait parameters with dual-task observed in children with ASD lends strength to the argument for ASD to be viewed as a 'disorder of complex information processing' as initially proposed by (Minshew and Goldstein, 1998).

Several study design considerations are noteworthy from this review. ASD is a spectrum rather than one specific condition with differing sub-types and varying levels of severity. These high levels of variability in this group, combined with small sample sizes, may lead to masking of between-group differences. Several studies cited this as a limitation to their research and recommended that future studies include a larger cohort (Ambrosini et al., 1998; Nayate et al., 2005; Calhoun et al., 2011; Chester & Calhoun et al., 2012).

Furthermore ASDs frequently are accompanied by a range of comorbid conditions such as attention deficit hyperactivity disorder (ADHD), developmental coordination disorder (DCD) and anxiety disorders. The Fifth Edition of the Diagnostic and Statistical Manual of Mental Disorders by the American Psychiatric Association now permits the diagnosis of comorbid ADHD and DCD with ASD. It remains unclear the extent to which gait abnormalities in ASD are unique or associated with other comorbid conditions and

existing studies have not addressed this issue. This is a question therefore for future study.

Overall, the studies reviewed had significant differences in terms of methodology, thus reducing comparability of their results. This may explain some of the inconsistencies found, for example Vilenksy et al. (1981) found that the ASD group had increased time in stance phase whereas Rinehart et al. (2006a) found no significant difference in the same parameter; the split opinion on stride/step length. These may be accounted for by the differing inclusion criteria such as age – the mean age of the children with ASD in the study by Vilensky et al., (1981) was 6.1 years, with the youngest being just three, compared to the group studied by Rinehart et al., (2006a) which had a mean age of 10.7 years and the youngest was six years old. Gait patterns develop as a child grows so patterns will be different with differing age ranges, making it very difficult to compare the gait patterns observed. There were also differences in IQ between the groups of both studies, with Rinehart et al., (2006a) only including "normally intelligent" children but Vilensky et al., (1981) had seven children who were classed as "severely retarded" which may influence movement patterns, and the two studies used different gait analysis systems so differences in detection of patterns may arise.

Many children with ASD are prescribed anti-psychotic medications, which may have effects on certain parts of the brain responsible for the control of movement such as the cerebellum. Only Nobile et al. (2011) specified that all the children included in the study were drug naïve. Nayate et al. (2012) cited this as a limitation to their research as three children in the study were on a mood-altering drug called sodium valproate which, at high doses, may have clinical effects on the cerebellum, which in turn may

affect gross motor control and, therefore, movement patterns (McCandless et al., 1979).

The temporal-spatial patterns exhibited by children with ASD were similar to the gait of children with obesity, with a wide base of support and shorter strides (Sheehan and Gormley, 2012). None of the studies reviewed examined the effects of BMI or body weight as a confounding factor. To date, no study has been published which compares gait patterns in children with ASD to healthy controls and also to children with obesity. It may also be of clinical importance to compare gait patterns in children with ASD to children with obesity as it has been shown that children with ASD have, by temporal-spatial parameters, a similar gait to children with obesity and have, on average, a significantly higher body fat percentage and lower lean tissue masses than healthy controls (Roke et al., 2012).

There are several factors that may affect research in this area such as intellectual ability, behavioural problems and severity of the condition. Most studies in the area examined gait patterns in children with high-functioning ASD. This means that the current research cannot provide an overall picture of gait deviations in children across the Autism Spectrum Disorders. The question of whether changes in gait patterns become more obvious with increasing severity of the disorder was raised by Weiss et al. (2013). They suggested that future research should be carried out including more low-functioning individuals.

The studies reviewed are helping to provide clinical practitioners, across a variety of disciplines, with a description of physical characteristics of ASD. The knowledge and understanding of this aspect of ASD may increase routine referral to services, such as

physiotherapy, and allow for better intervention and treatment planning. Since children with ASD were found by three studies to have reduced range of motion during gait (Nobile et al., 2011; Ambrosini et al., 1998; Vilensky et al., 1981), there may be underlying weakness within muscles of the lower limb and the wider base of support found by five (Lim et al., 2016; Nobile et al., 2011; Ambrosini et al., 1998; Shetreat-Klein et al., 2014; Nayate et al., 2005) may imply issues with balance and/or proprioception.

The overall findings of the studies conducted in the area are inconclusive, due to a number of confounding factors as discussed, however some results suggest an emerging pattern. The current perspective on gait patterns in children with ASD is that there are a number of deviations present in terms of temporal-spatial, kinematic and kinetic parameters and that gait, along with other movement pattern changes, may be used to allow for earlier diagnosis of ASD. There is, however, some consensus regarding the involvement of the cerebellum and basal ganglia in children with ASD and the relationship with observed motor deficits. Any altered biomechanical patterns, such as deviations from normal gait, may have impacts on an individual's ability to be physically active in daily life. The increased flexion observed may be an indication of tonal issues or an inclination to lower the centre of gravity to compensate for poor balance. Further studies should aim for larger sample sizes and examine other factors which may influence gait, such as motor function.

2.2 LITERATURE REVIEW 2: PHYSICAL ACTIVITY IN CHILDREN WITH ASD AND/OR DCD

2.2.1 Introduction

As discussed in Chapter 1, PA is an integral factor in health. For children with neurodevelopmental disorders, physical activity may have additional benefits including self-regulation, improving problem behaviours and stress relief (Lang et al., 2010, Pan and Frey, 2006, Allison et al., 1991).

Physical activity is typically described in terms of intensity and categorised as light-, moderate- and vigorous intensity activity. For health-related PA guidelines, moderate and vigorous PA is grouped together as moderate to vigorous physical activity (MVPA) as activity at the intensity of moderate and higher has been shown to have a positive impact on overall health (Hallal et al., 2006, Paffenbarger et al., 1993, Lee and Paffenbarger, 2000). Guidelines for PA and exercise in childhood and adolescence recommend at least 60 minutes of MVPA per day (ACSM, 2013) (see Section 1.4.4, page 10).

The objective of this review is to survey and summarise the literature regarding physical activity levels in ASD and DCD in comparison to TD children as measured by accelerometry. The review is narrative in nature but a systematic approach has been taken.

2.2.2 Methods

Accelerometers are compact, lightweight, non-invasive, precise and reasonably cost-effective methods for objectively measuring PA (Toschke et al., 2007, Vanhelst et al., 2012). As the name suggests, accelerometers are designed to measure the body's

acceleration (i.e. the change of speed with relation to time). Uniaxial accelerometers measure the body's vertical acceleration, whereas triaxial accelerometers measure the body's acceleration on three axes – vertical, anteroposterior and mediolateral (Chen and Bassett, 2005). Most accelerometers consist of piezoelectric sensors which creates a charge in response to acceleration. The charge builds up and a voltage signal proportional to the applied acceleration is generated (Chen and Bassett, 2005). This voltage signal is converted into digital values and summarised over a user-defined period of time (i.e. an epoch) (Chen and Bassett, 2005, Vanhelst et al., 2012). Cut-points divide levels of physical activity counts into the following categories: light, moderate and vigorous. The term 'wear time' is used to describe the time interval in which the accelerometer was worn.

Search strategy

Articles included in this review were retrieved from five online databases (PubMed, EmBase, Medline, CINAHL and Web of Science) by a single investigator. The following key terms were used: physical activity, autism, developmental coordination disorder. Reference lists of the articles retrieved in this search were then manually searched.

Selection criteria

Studies which compared objectively measured PA using accelerometry in children with a clinical diagnosis of ASD and/or DCD aged between four and 18 years with typically developing peers were selected for inclusion in this review. Studies written in English and published up to and including October 2016 were included. Studies which assessed PA through subjective methods of data collection, such as self-report or

parent report questionnaires, and those which did not include a comparison group of typically developing controls were excluded. A total of 426 studies were retrieved in the search. Fifteen studies met the inclusion criteria. Three of these studies did not include a control group and were excluded (Pan and Frey, 2006, Macdonald et al., 2011, Memari et al., 2013). Two further studies included a cohort of children with probable DCD but not clinically diagnosed with the disorder and were also excluded. A total of ten studies were therefore included in this review (Silman et al., 2011, Kwan et al., 2016). A flow diagram of the search and selection process adopted from the Prisma Statement (Moher et al., 2009) is displayed in Figure 2.3. Details of the studies included in this review and their main findings are presented in Table 2.2.

Many of the studies examining PA in this cohort focused on a particular time of day/week. The differences in PA levels across all days and times of the day or week were of interest to the author. Therefore, the results will be presented by group in the time points studied:

- Weekly (seven-day)
- Weekdays
- Weekends
- School break-times
- School Physical Education Class

Quality assessment of the papers included in this review was carried out using the checklist proposed by Downs and Black (1998). As the studies included were observational cross-sectional non-intervention studies, several items of the checklist did not apply and were not included in quality assessment.

Figure 2.3: PRISMA Flow Diagram of Search and Selection Process

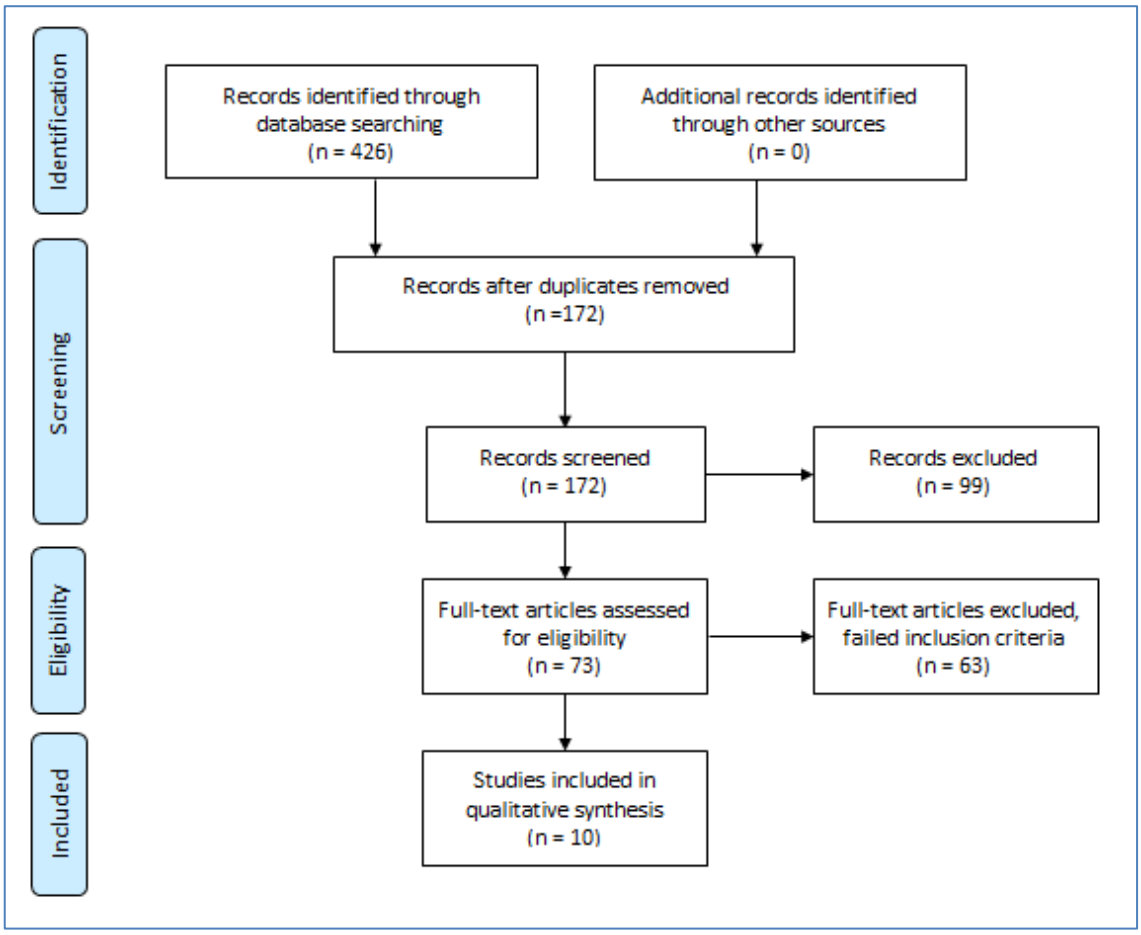


Table 2.2: Main Findings of Articles Included in Literature Review 2

Study	Cohort Diagnosis	Sample	Age Range of Sample	Device	Main Findings
Sandt & Frey (2005)	ASD	15 ASD (10 males) 13 TD (8 males)	5-12 years	MTI 7164 (uniaxial)	No differences in PA between children with ASD and TD children. 66.6% of ASD group met PA guidelines
Pan, (2008a)	ASD	24 ASD (23 males) 24 TD (23 males)	7-12 years	Actigraph GT1M (uniaxial)	Children with ASD spent less time in MVPA in school breaks than TD children
Pan, (2008b)	ASD	24 ASD (23 males) 24 TD (23 males)	7-12 years	Actigraph GT1M (uniaxial)	Children with ASD and TD children spent a similar amount of time in MVPA in PE class
Baerg et al., (2011)	DCD	32 DCD (20 males) 48 TD (30)	12-13 years	Actical (uniaxial)	No significant differences in step count between children with DCD and TD children
Pan et al., (2011)	ASD	19 ASD (19 males) 76 TD (not specified)	14 years	Actigraph GT1M (uniaxial)	Steps per minute in PE class lower in ASD group than TD group
Bandini et al., (2013)	ASD	53 ASD (45 males) 58 TD (44 males)	3-11 years	Actical	No differences in PA levels between ASD and TD groups. 23% of ASD group met PA guidelines.
Beutum et al., (2013)	DCD	9 DCD (6 males) 9 TD (6 males)	7-11 years	Actigraph GT3X (triaxial)	Significantly less MVPA in DCD group compared to TD controls
Tyler et al., (2014)	ASD	17 ASD (9 males) 12 TD (6 males)	9-18 years	Actigraph GT3X+ (triaxial)	Time spent in light PA, moderate PA and MVPA reduced in ASD group compared to TD group
Cermak et al., (2015)	DCD	53 DCD (35 males) 65 TD (not specified)	6-11 years	Actical	Total activity counts and MVPA reduced in DCD group compared to TD group
Pan et al., (2016)	ASD	35 ASD (35 males) 35 TD (35 males)	12-17 years	Actigraph GT1M (uniaxial)	TD group overall more active than ASD group. 37% of ASD group meeting PA guidelines

2.2.3 Results

Weekly Physical Activity (across weekdays and weekends)

ASD

Three studies compared weekly PA levels in children with ASD to TD children (Pan et al., 2016; Tyler et al., 2014; Bandini et al., 2013). One study found no difference in weekly averages and percentages of time spent in light PA, moderate PA, vigorous PA and MVPA between children with ASD and TD children (Bandini et al., 2013). No other studies published results on vigorous PA. Pan et al., (2016) found significant differences in overall PA between children with ASD and typically developing children. Activity counts per minute and the percentage of time spent in MVPA were significantly reduced in children with ASD compared to TD children.

Tyler et al., (2014) found that children with ASD were significantly less active than their typically developing peers. They found that the daily average time spent in light PA, moderate PA and MVPA were significantly lower in the ASD group than the TD group, and that the daily average time spent in sedentary behaviour was increased in the ASD group.

DCD

Two studies investigated weekly PA in children with DCD (Baerg et al., 2011; Beutum et al., 2013). Baerg et al., (2011) analysed step count and activity energy expenditure (AEE) and found no between group differences in either variable. The study did not report on activity counts or differentiate between light, moderate and vigorous PA. Beutum et al., (2013) found that MVPA was reduced in children with DCD compared to those with TD. They, however, found that children with DCD spent a similar amount of

time in structured and unstructured PA and engaged in a similar amount of sedentary behaviour as controls.

Physical Activity During Weekdays

ASD

Two studies examined weekday PA (Pan et al., 2016; Bandini et al., 2013) and both studies found that children with ASD were less active than TD children during weekdays. One study found total moderate counts and time spent in moderate PA were significantly reduced in the ASD group compared to the TD group. The percentage of time spent in moderate PA and in MVPA were also found to be reduced with p-values approaching significance ($p<0.07$ and $p<0.08$ respectively). However, another study found significant differences between children with ASD and TD children when the percentage of time spent in MVPA was analysed (Pan et al., 2016). This study also found fewer weekday activity counts per minute in the ASD group compared to the TD group.

DCD

One study examined weekday PA in children with DCD and found that overall counts per minute and the time spent in MVPA per weekday were significantly reduced in children with DCD compared to their typically developing peers (Cermak et al., 2015).

Physical Activity At Weekends

ASD

Two studies examined weekend PA levels in children with ASD compared to TD children (Pan et al., 2016; Bandini et al., 2013). Both studies found that children with ASD were similarly active to TD children at weekends. Pan et al. (2016) found that children with ASD had significantly less activity counts per minute at weekend in comparison to weekday counts per minute, but that there was no significant difference in MVPA between weekends and weekdays for children with ASD. TD children had reduced counts per minute and less MVPA at weekends in comparison to weekday PA.

DCD

Studies which examined PA in children with DCD did not report on weekend PA.

Proportions of Children Meeting Recommended Guidelines for PA

ASD

Bandini et al., (2013) examined the proportions of children with ASD and TD children who were meeting PA guidelines of at least 60 minutes of MVPA per day. Their results showed that 43% of TD children met guidelines compared to 23% of children with ASD. The difference between the groups was approaching significance. Pan et al., (2016) found a significant difference in the proportions of children in each group meeting guidelines – 37% of children with ASD were meeting recommended guidelines compared to 60% of TD children. Conversely, Sandt and Frey (2005) reported that the

majority of children with ASD (66.6%) and TD children (92%) were meeting PA guidelines.

DCD

No studies included in this review studied the proportions of children with DCD meeting recommended PA guidelines.

Physical Activity Levels During Break-times in School

ASD

Three studies examined PA during break-time in children with ASD compared to control children (Pan, 2008a; Pan, 2008b; Sandt & Frey., 2005). Pan, (2008a) and Pan, (2008b) shared the same cohort and found that MVPA was significantly lower in the ASD group compared to the control group. Sandt & Frey (2005) did not find any differences between the groups in break-time MVPA.

DCD

No studies included in this review examined PA during break-time in children with DCD.

Physical Activity Levels During Physical Education (PE) Class

ASD

Three studies examined PA during PE class in children with ASD compared to TD children (Pan et al., 2011; Pan, 2008b; Sandt & Frey., 2005). All three studies found that children with ASD were similarly active to TD children during PE class. Pan, (2008)

found that both children with ASD and TD children were moderately active for less than 50% of the time spent in P.E class and that P.E class contributed to approximately 30% of the recommend 60 minutes of MVPA per day for all children. Pan et al., (2011) found that children with ASD were similarly active in terms of MVPA counts but had a significantly reduced step count compared to TD children during PE class. One study compared results with data previously published by Pan, (2008b) and found that children with ASD aged 12-17 years spent a smaller percentage of time in MVPA during PE class than children with ASD aged 7-12 years (37% and 46%, respectively).

Pan et al., (2011) also examined the percentage of time spent in MPA and VPA during PE class and also the PA during bouts lasting 5, 10 and 20 minutes, respectively. No significant between-group differences were found in these variables.

DCD

No studies included in this review examined PA during PE class in children with DCD.

<u>The Effect of Age and Gender</u>

ASD

The effect of gender on PA in children with ASD was analysed in one study (Sandt and Frey, 2005). This study found no differences in PA levels between males and females with ASD, or between TD males and females (Sandt and Frey, 2005). Group-gender interactions were not reported by any of the studies included in this review. Pan, (2008a) reported an age-effect on school-time PA in children with ASD. Early primary children with ASD were more active than late primary children in the first morning

break-time, but late primary children with ASD were then more active than early primary children in the an afternoon break-time (Pan, 2008a).

DCD

The effect of gender on PA in children with DCD was analysed in one study (Baerg et al., 2011). Their results showed no significant difference in PA between males and females with DCD, but found significant differences in the TD group where males were more active than females (Baerg et al., 2011). No studies in this review examined the effect of age on PA levels in children with DCD.

2.2.4 Discussion

This review examined the reported activity levels of children with DCD and ASD in comparison with typically developing controls. All studies reviewed looked at each condition separately. None of the articles examined activity levels in children with ASD and comorbid DCD. The majority of the studies in this review showed that children with ASD or DCD were less active than their typically developing peers (Pan, 2008a, Pan et al., 2011b, Bandini et al., 2013, Beutum et al., 2013a, Tyler et al., 2014, Cermak et al., 2015, Pan et al., 2016). Studies which examined school-time PA showed that children with ASD were similarly active during PE class but were less active during break-time and therefore were less active in total during school (Pan et al., 2008). This may imply that children with ASD are less likely to choose to be physically active in the absence of supervision.

There were inconsistent results reported in relation to weekly PA activity. Several studies reported that both children with ASD and children with DCD were less active than children with TD (Pan et al., 2016; Beutum et al., 2013; Tyler et al., 2014); other

studies found no difference between groups (Bandini et al., 2013; Baerg et al., 2011). Differences in the samples included in the studies reviewed here may contribute to diverging findings. Notably different age ranges were reported in samples. Some studies focused on younger children or adolescents only and other included children and adolescents. This may limit the comparability of several of these studies as there is evidence to suggest that PA decreases with age from childhood into adolescence (Trost et al., 2002, Telama et al., 1997). Several studies included only male subjects while others had both male and female subjects.

Few studies reported proportions of children meeting exercise guidelines, but those who did showed fewer children with ASD or DCD were meeting guidelines than typically developing children (Pan et al., 2016, Bandini et al., 2013). Pan et al., 2016 reported that the majority of children with ASD did not meet the guidelines and that these children had significantly poorer cardiovascular endurance and upper body strength than the typically developing children who also did not meet guidelines. Children with ASD who met PA guidelines scored significantly lower on cardiovascular endurance and lower body flexibility than typically developing children who met the guideline which may imply that more PA activity is needed for children with ASD to gain the same health and fitness benefits as typically developing children with the standard guidelines. None of the studies examining PA in DCD reported on PA guidelines.

Several studies have shown that the cut-points employed for activity count analysis and epoch length may impact results (Guinhouya et al., 2006, Pulsford et al., 2011, Romanzini et al., 2014, Banda et al., 2016). For example, one study found that using

different cut-points to analyse the same data produced significantly different results when MVPA was analysed (Guinhouya & Hubert, 2006). Consistency is important for allowing comparison across different studies but also when it comes to determining when individuals are meeting recommended guidelines for PA. Lower cut-points will give false-positive results in which children who are inactive may be shown to be sufficiently active. This decreases the ability to compare results between studies and may account for differences in reported findings.

At first glance the apparatus used may seem to make results incomparable between studies with different brands and models of accelerometers. However validation studies have shown that data collected by several models including Actigraph GT3X, the Actigraph GT1M and the Actical have significant and high correlations (Esliger et al., 2007). Another study found high correlations between data collected with uniaxial accelerometers and that collected with triaxial accelerometers (Vanhelst, 2012).

There are currently no available guidelines or a gold standard for collection and analysis of PA data. However, one recommended approach for determining habitual PA levels in children is to collect data for seven days and to record at least four valid days, including three weekdays and one weekend day (Trost et al., 2005). A valid day should include at least 10 hours (600 minutes) of wear time and data should be collected in 10 second epochs to ensure accurate recording of the short but intense bursts of activity exhibited in childhood (Trost et al., 2005). Most of the studies in this review which examined weekly PA levels adhered to these criteria, although some did not report precise inclusion and exclusion criteria for analysis of PA data.

Gender presents a confounding factor for PA analysis in children with many studies showing that girls are less active than boys, with the difference becoming greater with increasing age through adolescence and this gender difference is present across all ethnic groups (Owen et al., 2009). The results of the study by Baerg et al., (2011) show an interesting trend whereby the gender difference is present as expected in the typically developing group, but not in the DCD group. The reason for this is unclear, but may be due to the apparent reduction in PA in both girls and boys with DCD to the point that they are similarly inactive.

Many children with neurodevelopmental disorders exhibit behavioural problems including aggression, anxiety, opposition and sleep disturbance (Brand et al., 2015, Farmer and Aman, 2011, van Steensel et al., 2011, Kadesjö, 2000). Moderate PA has been shown to alleviate symptoms of anxiety, reduce aggressive behaviours and improve sleep patterns (De Matos et al., 2009; Nouri & Beer, 1989; Kanchana et al., 1993; Loprinzi & Cardinal, 2011; Brand et al., 2015). Children with either ASD or DCD are more likely to be overweight than their typically developing peers (Curtin et al., 2015; Cairney et al., 2005) with related health concerns implied so it is vital to accurately ascertain whether children with these conditions are meeting exercise guidelines, and if so whether the guidelines for the general population are suitable for these children.

The overall findings are inconclusive but there is evidence to suggest reduced PA in children with ASD or DCD. To the best of the author's knowledge, no cut points have been described specifically for children with neurodevelopmental disorders and this may be an area of interest for future studies. Studies which objectively measure PA in

these children should adhere to recommendations for collection of PA data proposed by (Trost et al., 2006b) as discussed earlier in the discussion.

CHAPTER 3: METHODS

This chapter outlines the recruitment process, clinical diagnoses, and inclusion and exclusion criteria for subjects and details the equipment used in data collection. Reliability and validity of measures used and introduction to data analysis is also provided. Details of design, sampling, measures and data analysis relevant to the individual studies are provided in the study chapters which follow.

3.1: ETHICAL APPROVAL

Ethical approval for Chapter 4, Chapter 5 and Chapter 6 was obtained from the Irish Health Service Executive Linn Dara Child and Adolescent Research Ethics Committee, the St. James's Hospital/The Adelaide Meath National Children's Hospital Ethics Committee, the National Children's Research Centre Ethics Committee and the School of Medicine Research Ethics Committee, Trinity College, Dublin (TCD). Data collection for Chapter 4Chapter 4, Chapter 5 and ChapterChapter 66 were covered in one ethical application. Ethical approval for Chapter 7Chapter 7 was obtained from the TCD School of Medicine Research Ethics Committee. Approval letters are enclosed in Appendix I.

3.2: RECRUITMENT OF PARTICIPANTS

For Chapter 5Chapter 6Chapter 5 and Chapter 6, male and female children aged between 6 and 18 years with ASD and/or DCD were recruited through schools, advocacy groups, HSE assessment and intervention teams, social media and child and adolescent mental health services. Both case and control participants were recruited through schools, social media sites, volunteer websites, local businesses and educational programmes. Recruitment through schools, clinical settings, local business and educational programmes involved posters and information leaflets with contact

details of the lead investigator. Parents were invited to contact the lead investigator to discuss the research further. Appointments were made at least seven days after the parents first received the information leaflets. Recruitment through social media was carried out using the "TCD Research: Autism and Related Neurodevelopmental Disorders" Facebook page. Digital posters were uploaded to the page and sponsored to appear as an ad. The posts were targeted to reach the appropriate audience by using keywords such as 'autism', 'developmental coordination disorder' etc. These posts would then appear as Facebook advertisements for people who actively engage with related pages and groups or relevant advocacy groups, for example the "Irish Autism Action" Facebook page.

All subjects were required to have borderline or average intellectual functioning defined as Intelligence Quotient (IQ) above 70. Inclusion criteria for ASD subjects included a confirmed research diagnosis using two formal diagnostic tests (see Section 3.3.1 and 3.3.2, page 56-57). Subjects included in the dual diagnosis group (ASD/DCD) also met these diagnostic criteria for ASD. Exclusion criteria included known medical or genetic disorders associated with ASD such as Fragile X syndrome, comorbid neurological or psychological disorders or current use of psychoactive medication. Sensory processing disorder (SPD) is a disorder which causes affected individuals to experience sensory input differently (Miller and Schaaf, 2008, Tomchek and Dunn, 2007). Sensory processing disorder or dysfunction has been reported to be prevalent among children with ASD (Tomchek and Dunn, 2007, Watling et al., 2001, Lord, 1995). This was a consideration for exclusion as the close-fit of gait analysis wands and the snug fit of the gas analysis face mask combined with tactile sensitivity may cause

distress to the child. This was discussed with parents/guardians prior to appointments. For the typically developing control cohort, additional exclusion criteria included a history of developmental delay, a score > 12 on the social communication questionnaire (SCQ), a score > 50 in the social responsiveness scale (SRS) or first-degree relatives with a diagnosis of an autism spectrum disorder as it has been shown that there is a 30% chance of broader phenotype ASD in those with a first-degree relative with ASD. For all groups, serious injury to the lower limbs in the past 6 months, any additional condition which may alter gait (e.g. cerebral palsy) or affect the child's ability to exercise safely, cardiorespiratory disease or poorly controlled asthma were also exclusion criteria. Both parental written consent as well as written assent from participants was obtained from all participants. Participants were given information and the opportunity to ask questions prior to commencing. Information leaflets and consent/assent forms are presented in Appendix II and Appendix III.

For Chapter 4Chapter 4 and Chapter 7Chapter 7, parents of children aged 6+ with neurodevelopmental disorders were recruited through social media and advocacy groups as mentioned above, and from a database of parents who had consented to being contacted regarding future research studies. Professionals who work with children were recruited for Chapter 7Chapter 7 via clinical managers of HSE assessment and intervention teams and special needs clinics.

3.3: DIAGNOSTIC ASSESSMENTS

Diagnostic assessments were carried out by a clinical psychologist and psychology graduates who were trained in the administration and interpretation of the tests

outlined in sections 2.3.1-2.3.3 below. The numbers of each test carried out by these researchers are presented in Table 3.1.

Table 3.1: Diagnostic Assessors

Name	ADOS-2	ADI-R	WASI
Codruta Sudrijan (Clinical Psychologist)	20	29	57
Sarah-Marie Feighan (Psychology Graduate)	29	20	65
Ana McLaughlin (Psychology Graduate)	10	10	21

3.3.1: Autism Diagnostic Observational Schedule, 2nd Edition (ADOS-2)

The Autism Diagnostic Observation Schedule, 2nd Edition (ADOS-2) (Lord et al., 1989, Gotham et al., 2008) and the Autism Diagnostic Interview-Revised (ADI-R) (Lord et al., 1994) were used together to confirm a research standard clinical diagnosis of autism spectrum disorder (ASD) for all ASD participants prior to recruitment. The Autism Diagnostic Observational Schedule, 2nd Edition (ADOS-2) is a semi-structured diagnostic observational assessment developed for the research setting, which enables the examiner to observe the social interaction, social communication and repetitive behaviours of the participants. The ADOS-2 contains four modules, one of which is selected for the assessment based on prior knowledge of the participant's age and expressive language. Module four is most suitable for verbal older children, adolescents and adults with fluent speech. The diagnostic algorithm has a three-step cut-off. Individuals must score at least 2 on the communication sub-domain, a minimum of 4 on the reciprocal social interaction sub-domain and at least a score of 7 for the combined scores of the sub-domains to receive a diagnosis of ASD. A score of 3 on communication, 6 on reciprocal social interaction and a combined score of 10 on

both domains will yield a diagnosis of autism. Scores of stereotyped behaviours and restricted interest are also recorded but are not required for clinical diagnosis. The ADOS-2 has been found to be a reliable and valid measure of autism severity (Lord et al., 2000; Gotham et al., 2008).

3.3.2: Autism Diagnostic Interview, Revised Edition (ADI-R)

The ADI-R is a structured interview conducted with parents or caregivers and focuses on both previous and current behaviour. The ADI-R consists of 93 questions exploring developmental history, language functioning, communication, social development, interests and clinically relevant behaviours. The diagnostic algorithm consists of four subdomains; qualitative abnormalities in reciprocal social interaction, qualitative abnormalities in communication, restricted, repetitive and stereotyped patterns of behaviour and abnormality of development evident before 36 months. Participants must score above the cut-off in each behavioural sub-domain to receive a clinical diagnosis of ASD, and this was the criterion used in the current study.

3.3.3: Wechsler Abbreviated Scale of Intelligence, 1st Edition (WASI)

The Wechsler Abbreviated Scale of Intelligence (WASI; (Wechsler, 2004) was used to evaluate intelligence quotient (IQ) for both ASD and controls participants. The WASI consists of four subtests; vocabulary, similarities, block design and matrix reasoning which collectively provide a measure of full scale IQ. Verbal IQ can be ascertained from the vocabulary and similarities subtests. The vocabulary subtest measures word knowledge and verbal concept formation while the similarities subtest measures verbal reasoning and concept formation. Performance IQ can be established from the block design and matrix reasoning subtests. The block design subtest assesses nonverbal concept formation, visual perception and visual-motor coordination while

the matrix reasoning subtest assesses abstract reasoning in addition to visual information processing. The WASI has been shown to be a valid brief measure of IQ (Canivez et al., 2009).

3.3.4: Social Responsiveness Scale (SRS)

The SRS is a 65-item rating scale designed to measure the severity of ASD symptoms as they occur in a child's everyday life and social setting (Constantino, 2013). It can be completed by a parent or teacher and describes a child's social impairment, including their ability in reciprocal social communication and their social awareness. It is sensitive and reliable across a variety of symptom severity and is suitable for use with children aged 4-18 years. A Raw score and T-Score are calculated for five subscales: Social Awareness, Social Cognition, Social Communication, Social Motivation and Autistic Mannerisms. Total score results are converted to T-Scores by gender. T-Scores above 70 indicate a likely ASD diagnosis. Higher T-Scores indicate a higher level of severity. This was completed by all parents of participants in Chapter 5 and Chapter 6 as a screening tool to exclude the presence of ASD in the control and DCD-only groups. The SRS has been found to be a valid quantitative measure of autistic traits which is feasible for clinical use or for research in ASD (Constantino et al., 2003, Bolte et al., 2008b). The SRS was used to provide a quantitative measure of ASD traits across all four groups.

3.3.5: Social Communication Questionnaire (SCQ)

The SCQ is a brief parent-reported instrument intended to evaluate social and communication skills in children who may have autism or ASD (Rutter et al., 2003). It is comprised of 40 Yes/No questions on the child's social communicative behaviour and is derived from the ADI-R. The SCQ is used clinically to ascertain whether an

individual should be referred for further assessment and is suitable for use with children over four years of age. It is commonly used in a research setting to screen individuals without a diagnosis of autism or ASD, for example when determining an individual's eligibility for participation as a typically developing control in a study. Scores above 15 would indicate a likely diagnosis of ASD. This was completed by all parents of participants in Chapter 5 and Chapter 6 as a screening tool to exclude the presence of ASD in the control and DCD-only groups, and as a tool to investigate behavioural correlations with physical activity and motor function findings in all cohorts. The SCQ has been found to be a valid and reliable screening tool for ASD (Bolte et al., 2008a). The SCQ was used to evaluate ASD symptoms, particularly in control subjects as an exclusion criterion, as referenced above.

3.3.6: Child Behaviour Checklist (CBCL)

The CBCL is a parent-reported questionnaire comprised of 112 items (Achenbach and Edelbrock, 1991). It is not an autism-specific scale but is used widely in child psychology to determine problem areas for the individual. The items were factor-analysed to identify types of psychopathology that occur in childhood (Achenback and Edelbrock, 1991). Raw scores can be converted to t-scores and may be put into problem scales or used as individual behaviour syndrome scores. The CBCL has been found to be a valid tool for screening problem behaviours in children across all cultures (Crijnen et al., 1999, Ivanova et al., 2007).This checklist was completed by all parents of participants in Chapter 5 and Chapter 6 as a behavioural screening tool.

The above described SRS (Section 3.3.4), SCQ (Section 3.3.5) and CBCL (Section 3.3.6) were completed by parents of all participants and were used for assessment of eligibility for inclusion in the studies outlined in Chapters 5 and 6. They were used to

confirm ASD diagnoses in children with a diagnosis of ASD for inclusion in the ASD/ASD&DCD groups and to confirm the absence of ASD or ASD traits in the DCD and TD groups. Scores for the completed questionnaires were collated by the author and children were included as appropriate. Scores from these measures were not analysed.

3.4: PHYSICAL ACTIVITY AND FITNESS

3.4.1: Gas Analysis

The Cosmed K4b^2 portable metabolic measurement system was used to assess oxygen consumption and, therefore, energy expenditure. The K4b^2 provides cardiopulmonary gas exchange analysis on a breath-by-breath basis, and allows for monitoring of heart rate with integrated sensors which receives a signal from a heart monitor on a chest strap.

Calibration:

The K4b^2 must be turned on approximately 45 minutes prior to allow for warm-up and calibration. There are four calibrations that must be carried out prior to each testing session: Air Calibration, Reference Gas Calibration, Turbine Calibration and O_2/CO_2 Delay Calibration. A user guide with instructions for the set up and calibration process was written and developed by the author (see Appendix IV).

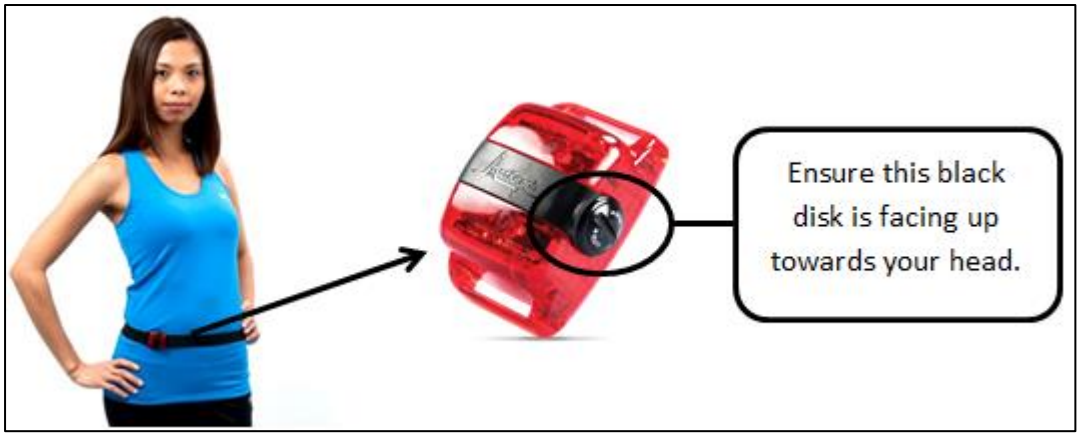
The Cosmed K4b^2 has been found to be a valid measure of oxygen uptake across a range of exercise activities in adults (McLaughlin et al., 2001; Eisenmann et al., 2003). The Cosmed K4b^2 has been used in numerous studies examining children and adolescents (Innerd and Azevedo, 2016, Trost et al., 2006b, Harrell et al., 2005, Treuth et al., 2004, Schmitz et al., 2005). Harrell et al. (2005) compared VO_2 data obtained

from 14 children with the K4b^2 with the Parvo Medicas Truemas 2400 and found that the K4b^2 exhibited a small positive bias but that the differences in VO_2 were less than 6% ($P<0.05$).

3.4.2: Objective Measure of Physical Activity – Actigraph GT3X+

Physical activity was measured objectively using the Actigraph GT3X+. The GT3X+ is a triaxial accelerometer (i.e., activity is assessed in the horizontal, vertical and perpendicular axes) designed to monitor human physical activity and record energy expenditure values. It can be worn either on the waist, wrist or ankle. GT3X+'s are initialised using the Actilife software with the participants name or I.D number, weight, height, date of birth and hand dominance (only applicable when worn on the wrist) entered, and can be set to record data immediately or on a delayed start date and time. Data is collected at a sampling rate between 30Hz and 100Hz. The Actigraph GT3X has been used extensively in studies examining children and adolescents (Diouf et al., 2016, Gaba et al., 2016, Johansson et al., 2016, Hjorth et al., 2012, Robusto and Trost, 2012, Trost et al., 2013) and has been shown to be a valid and reliable measure of physical activity in children (O'Neil et al., 2014, Johansson et al., 2016). Studies have shown that accelerometers worn on the hip have better accuracy than wrist-worn accelerometers (Rosenberger et al., 2016, Swartz et al., 2014). For the purposes of this study, participants were asked to wear the monitor on the waist, in front of the right hip and upright (i.e. with the USB cap facing up – see Figure 3.1) for seven days.

Figure 3.1: Actigraph Fitting/Placement



3.4.3: Subjective Measure of Physical Activity

A previously validated questionnaire, the Modifiable Activity Questionnaire for Adolescents (MAQ-A), formed the basis of a subjective measure of physical activity (Aaron et al., 1995). Some changes were made to facilitate use as a survey on physical activity in children with ASD and other neurodevelopmental disorders. This included changing it from a self-report questionnaire to parent reported, and the addition of several questions necessary to allow evaluation of responders' eligibility for inclusion. For example, questions were added which ascertained present clinical diagnosis, the child's age and gender. Questions were also modified to capture modern sedentary behaviours and sports that would be commonplace in Ireland. Modifications applied to this questionnaire are discussed in Chapter 4 (see Section 4.2.1, page 78). The MAQ-A has been found to be a reliable and valid measure of physical activity in children and adolescents (Delshad et al., 2015, Aaron et al., 1995).

3.4.4: Body Composition

Body Mass Index

Body Mass Index (BMI) is defined as the weight in kilograms divided by the square of the height in metres. It is a simple index which is commonly used to classify

underweight, overweight and obesity. In children, it is not a stable measure and varies throughout development. BMI percentiles are calculated using gender and age to the nearest month and have been recommended for use in assessing body composition in children and adolescents aged 2-22 years, and provides a reasonably reliable screening tool which is convenient and cost-effective to use on large populations (Cole, 2010, Flegal et al., 2009, Kuczmarski et al., 2002, Kuczmarski et al., 2000, Ogden et al., 2010).

Bioelectrical Impedance

Body composition was assessed using the Tanita Body Composition Analyzer MC-180MA (Tanita, Tokyo, Japan). MC-180MA provides accurate and reliable information by multi-frequency bioelectrical impedance (BIA). An electric current is passed through the body from electrodes in contact with the palms of the hands and soles of the feet. The resistance to this current in various tissues is known as the bioelectrical impedance. Tissues with higher water content, such as muscle and bone, provide less resistance than those with lower water content, such as fat tissues, and therefore impedance to the current. By measuring the impedance to the current as it travels around the body, the amount of different tissues in the body can be determined. BIA has been found to be a reliable measure of body fat percentage in children, with high correlations to DEXA scan measures (Kettaneh et al., 2005).

3.4.5: 20 Metre Multistage Fitness Test

The 20 Metre Multistage Fitness Test first developed in 1982 and further refined in 1988, was used to assess fitness (Leger and Lambert, 1982, Leger et al., 1988). It is a maximal aerobic running fitness test. A 20 metre straight course is set out and an audio recording is played. Participants must run back and forth and touch the line at

either end following the pace of the recording. The initial pace is 8.5km/h and is increased by 0.5km/h every minute. When the participant can no longer keep to the pace, the last stage number achieved is recorded as their score. Léger et al., (1988) described predicted maximal oxygen consumption (VO_2max) based on age and gender in a table (please see Appendix V). The 20 Metre Multistage Fitness Test has been shown to be a valid predictor of $VO2_{max}$ in children and adolescents (Boreham et al., 1990, van Mechelen et al., 1986, Naughton et al., 1996, Liu et al., 1992) and has been used in many studies assessing aerobic fitness in school-going children (Tomkinson et al., 2016, Boreham et al., 1993, Tomkinson et al., 2003, Guerra et al., 2002, Hussey et al., 2007). The 20 Metre Multistage Fitness Test was also found to be a reliable measure of cardiorespiratory fitness in 12 year old children and had consistent test-retest reliability (Mahoney, 1992). In this study, participants wore the Cosmed $K4b^2$ during the test so the rise in oxygen demand may be captured objectively.

3.5: Motor Function

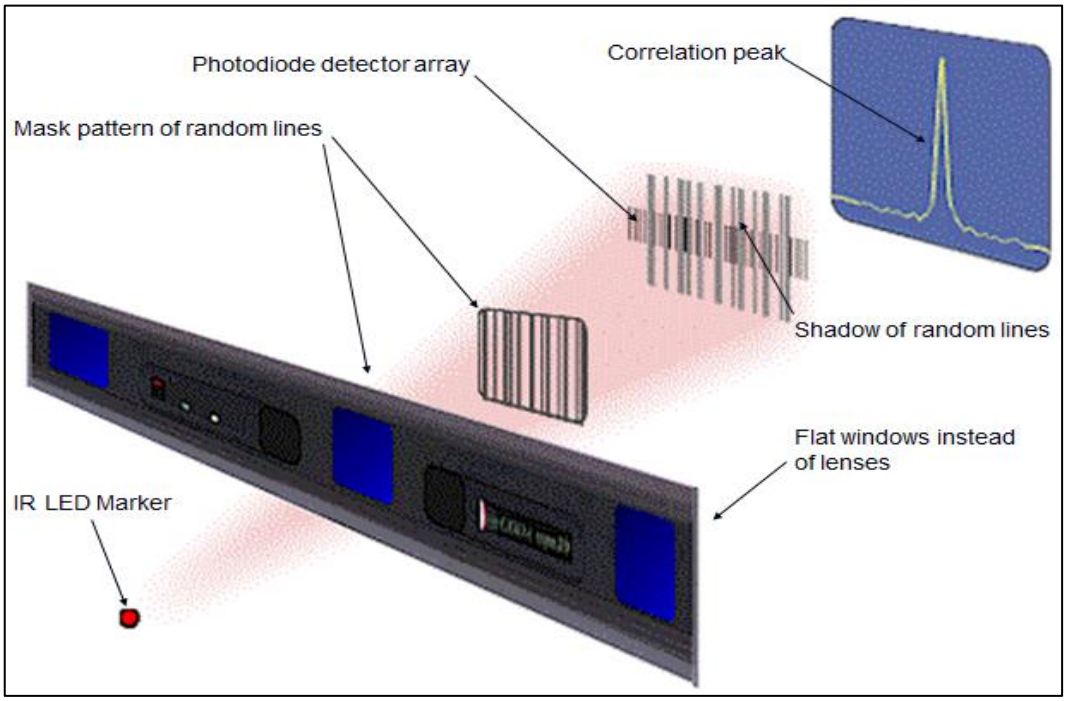
3.5.1: Gait Analysis

Motion Capture System

Gait analysis was carried out using the Codamotion Dual CX1 and Gait Wand System (Charnwood Dynamics Ltd., Leicestershire, UK). This system enables 3D motion capture and analysis. The system is comprised of two CX1 sensor units on tripods, an ActiveHub computer, gait wands and light emitting diode (LED) active marker powered by rechargeable driveboxes. Each sensor unit has three masked linear arrays (MLA's) aligned vertically and covered by sensor windows. The top and bottom MLA's capture horizontal movement, and the middle MLA captures vertical movement. Each MLA contains a series of black lines, or a mask, in front of a photodiode sensor array (see

Figure 3.2). Twenty-two LED markers are fitted to predefined anatomical locations on the lower limbs, either on the wands or placed directly onto the skin. Pulsed by the CX1, markers flash infrared light individually in a specific order and the photodiode detects these flashes and converts them into current. MLA's calculate the location of individual active markers by measuring the pattern produced when the marker flashes and casts a shadow on the sensor array through the mask. As an individual moves with the markers in place, this process continues and data is stored on the ActiveHub for analysis.

Figure 3.2: LED marker flash across photodiode sensor array in CX1



Physical Assessment

Each participant underwent a physical clinical assessment prior to gait analysis. Height and weight were measured and recorded. The following anatomical points were marked bilaterally using a hypoallergenic skin marker: anterior superior iliac spine

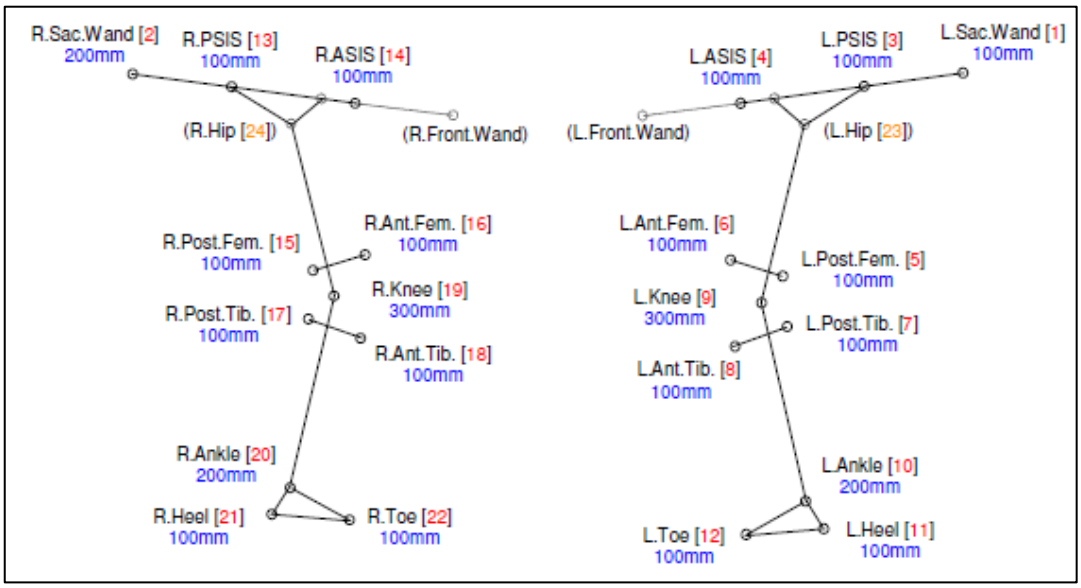
(ASIS), posterior superior iliac spine (PSIS), the superior and inferior aspects of the lateral knee joint line, the fibular head, the medial and lateral malleoli, the lateral aspect of the calcaneal tuberosity and the head of the fifth metatarsal. Pelvic width was defined as the distance between the right and left ASIS and was measured using large clinical width callipers. Pelvic depth was measured unilaterally using Martin Breadth Callipers. One arm was placed at the right ASIS and the other arm was placed at the right PSIS. Thigh length was defined as the distance between the most prominent point of the greater trochanter to the superior aspect of the lateral knee joint line. Shank length was defined as the distance between the inferior aspect of the lateral knee joint line and the lateral malleolus of the ankle. Foot length was defined as the distance between the most posterior point of the calcaneal tuberosity at the distal aspect of the 2nd toe. Thigh, shank and foot length were measured bilaterally using a non-stretch clinical measuring tape. Knee width was defined as the distance between the lateral and medial femoral condyles. Ankle width was defined as the distance from the lateral malleolus to the medial malleolus. Knee and ankle width were measured bilaterally using a large clinical width callipers. See Appendix VI for clinical assessment form.

Marker and Wand Placement

The gait wand system was comprised of a pelvic frame with a sacral wand, two femoral wands, two tibial wands, Velcro straps, driveboxes, 14 wand markers and eight surface markers. Markers and driveboxes were attached to the wands and skin with double sided tape. The pelvic frame was made up of two pelvic wands which were inserted into a holder, to which the sacral wand was attached in midline

posteriorly. A drivebox was attached to each wand, with a marker attached at each wand's superior anterior and superior posterior corners. Markers were attached either side of the posterior end of the sacral wand. The femoral and tibial wands had a drivebox and two markers attached at the superior anterior and superior posterior corners. The tibial wands also had a drivebox attached at mid-tibial level which powered markers at the knee and ankle joint centres. The knee joint centre was found by locating the fibular head, measuring a point 1.5 centimetres anterior using a non-stretch clinical measuring tape and drawing a line to meet the superior aspect of the lateral knee joint line. The intersection of the two lines was considered knee joint centre and the knee marker was placed there (Protocol from the Central Remedial Clinic, Dublin; Walsh et al, 2000). Ankle joint centre was defined as the line connecting the lateral and medial malleoli. The ankle marker was placed at the most prominent bony point of the lateral malleolus, as previously marked. The foot was defined by two markers with one placed at the calcaneal mark and one placed at the head of the fifth metatarsal. An outline of marker placement locations is shown in Figure 3.3.

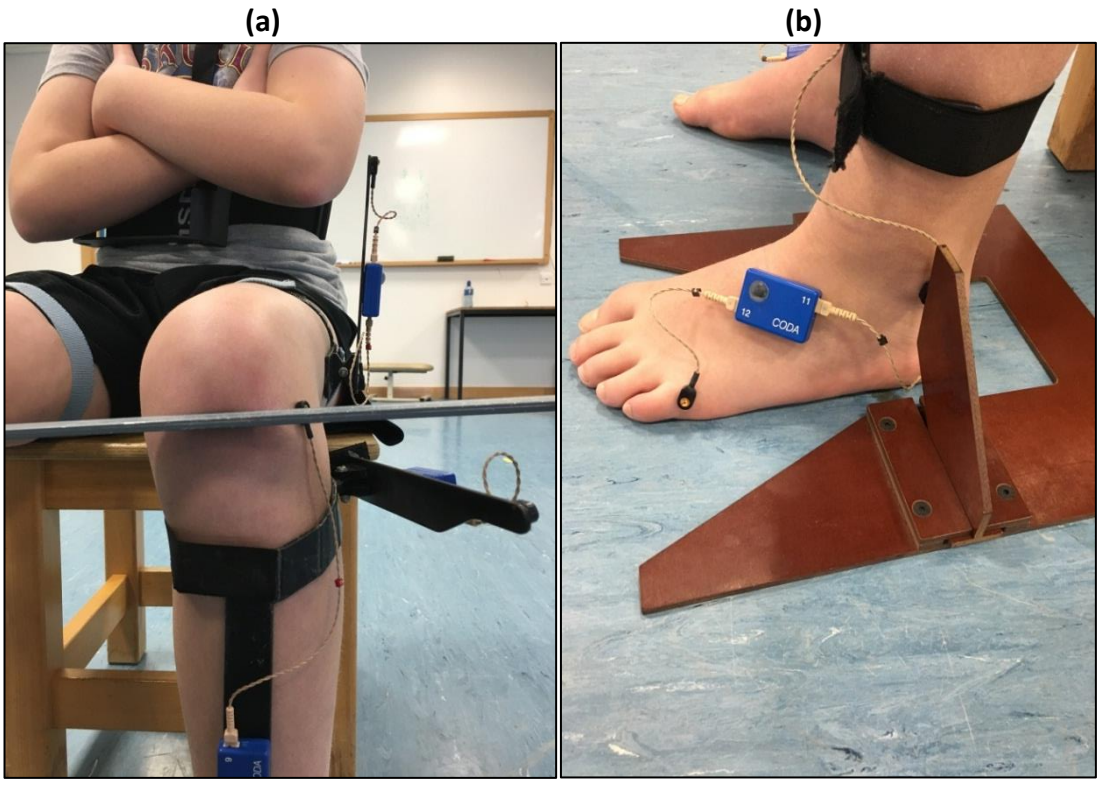
Figure 3.3: LED Marker Placements



Wand Alignment Procedure

The pelvic frame was aligned horizontally parallel to bilateral PSIS and angled inferiorly towards the ASIS. The femoral wand was aligned perpendicular to the transcondylar axis (i.e. the line joining the medial and lateral femoral condyles as marked). With the participant sitting with their knees pointed anteriorly and flexed at 90 degrees, a ruler was held in line with the transcondylar axis and the femoral wands were set perpendicular to the ruler. The tibial wands were aligned using the ankle alignment jig. With the participant still sitting with their knees flexed at 90 degrees, the ankle alignment jig was positioned around the foot with the arms of the jig at the medial and lateral malleoli. The tibial wand was aligned parallel with the lateral edge of the jig. Figure 3.4 (a) and (b) show the femoral wand alignment procedure and the tibial wand alignment procedure, respectively.

Figure 3.4: Femoral (a) and Tibial (b) Wand Alignment



Inter-rater and intra-rater reliability

Optical-based motion capture systems such as the Codamotion CX1 may be affected by system, inter-rater and intra-rater reliability (Monaghan et al., 2007, Maynard et al., 2003). To improve system reliability, maintenance checks were carried out bi-annually as recommended by Charnwood Dynamics Ltd., Leicestershire, UK. An alignment protocol was carried out on each test day to ensure the laboratory co-ordinate frame was consistent for all tests. Laboratory set up, alignment and clinical assessment were carried out by one investigator for all participants to eliminate inter-rater variability. To minimise intra-rater variability, the protocol for the clinical assessment, joint centre identification and marker placement, as described in the previous paragraph, was strictly adhered to.

Gait Terminology

This section defines the gait parameters analysed in Chapter 6 and describes the gait cycle and significant events at which kinematic parameters are described. Definitions of general gait terms such as cycle, stride and step can be found in Chapter 1 (see Section 1.6.1, page 15).

The gait cycle can be divided into two main phases: the stance phase when the foot is in contact with the ground and the swing phase where the foot is off the ground and moving forwards. The cycle is normally described from the initial contact (i.e. when the foot first touches the ground) on one side until the following initial contact on that same side. The stance phase begins at initial contact and ends at toe-off (i.e. when the

foot leaves the ground). The swing phase then begins at toe-off and ends at initial contact. The stance phase normally accounts for 60% of the total cycle time.

Temporal-spatial parameters

Temporal-spatial parameters are those which are concerned with timing and space or distance. The temporal-spatial parameters examined in Chapter 6 are described in Table 3.2.

Table 3.2: Temporal-spatial Parameters Examined in Chapter 6

Parameter	Definition
Step Width	The horizontal distance between the left and right foot placement. This is also known as base of support. And is usually measured in millimetres (mm) or centimetres (cm).
Step Length	The distance from the left heel strike (initial contact) to the right heel strike (and vice versa). Usually measured in cm or metres (m).
Stride Length	The distance from the left heel strike to the following left heel strike (or right). Usually measured in cm or m.
Stride Time	The time it takes for one stride to be completed. Usually measured in seconds.
Stance Percent Time	The percentage of the gait cycle spent in the stance phase.
Cadence	The numbers of steps taken in a period of time. Reported as steps per minute.
Velocity	The distance covered per unit time. Usually reported in metres per second (m/s).

Kinematic parameters

Kinematic parameters are those which are concerned with the range of motion of joints during gait but not the forces which cause motion. The sagittal plane is the plane through which the majority of motion of the gait cycle occurs. This plane splits the body into right and left and as such, forwards/backwards motions such as flexion/extension of the hip, knee and ankle occur in this plane. The terms ipsilateral

and contralateral are used to describe the angles of the joints on the same side as the gait event and the angles of the joints on the opposite side to the gait event, respectively.

From the evidence presented in the literature review, motion of these three joints in the sagittal plane was of interest to the author, particularly at initial contact and toe-off. The conclusions of the literature review suggest a pattern similar to a mild crouch gait, which is characterised by excessive flexion and the hip and knee and dorsiflexion at the ankle during the stance phase (Khamis et al., 2015). As such, mid-stance (i.e. the halfway point between initial contact and toe-off) was also deemed to be a gait event of interest in this cohort. To get a clear picture of the gait cycle, the joint angles on the ipsilateral side were measured at initial contact, mid-stance and toe-off and also on the contralateral side opposite these events.

The kinematic parameters analysed in Chapter 6 describe the angles of the hip, knee and ankle at the following events of the gait cycle: initial contact, opposite initial contact, mid-stance and toe-off. The overall range of motion observed in the joint throughout the gait cycle (i.e. knee range of motion refers to the difference between the angle of the knee at its most flexed position in gait and the angle at its most extended position in the gait cycle). Table 3.3 explains these terms further.

Table 3.3: Points of the Gait Cycle Examined in Chapter 6

Gait Cycle Event	Definition
Initial Contact	Joint angles at the point at which the foot first makes contact with the ground
Opposite Initial Contact	Joint angles when the opposite limb is at initial contact
Mid-Stance	joint angles at the mid-point between initial contact and toe-off
Opposite Mid-Stance	Joint angles when the opposite limb is at mid-stance
Toe-Off	Joint angles at the point the foot leaves the ground and enters the swing phase
Opposite Toe-Off	Joint angles when the opposite limb is at toe-off
Range of Motion	The full arc of movement the joint moves through during the gait cycle.

These parameters and time points were chosen as areas of interest for the author based on the results of the literature review described in Chapter 2 (Section 2.1) and on advisement from the Codamotion lab technicians. The author was interested in examining specific details (e.g. the degree of flexion exhibited at each of the main joints of the lower limbs at mid-stance may provide additional evidence for a gait pattern similar to crouch gait as discussed in Chapter 2, Section 2.1.4) rather than an overall summary measure of gait quality such as the Gait Deviation Index or the Gait Profile Score. The author acknowledges that such a summary measure may provide a score which is more easily understood by individuals without knowledge of gait analysis.

3.5.2: Motor Coordination

The Movement Assessment Battery for Children, 2nd Edition (MABC-2), was used to assess motor coordination and function. The MABC-2 is an eight-item battery which identifies and describes motor impairment in children. There are three categories in the test: Manual Dexterity, Aiming & Catching, and Balance. There are three separate

age bands for testing. Age Band 1, 2 and 3 assess children aged three to six years, seven to ten years and eleven to sixteen years, respectively. The items for each age band are summarised In Table 3.4. Item raw scores are converted to standard scores using a separate table for the age of the child by year. The sum of the standard scores for each category give the component score for the category, which can be converted to a standard score and percentile using one table for all ages. Standard scores for all nine tasks are summed to give the total test score. The total test score is converted to a standard score and percentile rank. The standard scores for the categories and total test results allow comparison across age ranges. A total test percentile score below the 5^{th} percentile is classified as definite motor impairment and is consistent with DCD. Scoring from the 5^{th}-16^{th} percentile is classified as at risk of motor impairment. The MABC-2 has been found to be a valid and reliable measure of motor ability in children (Croce et al., 2001, Van Waelvelde et al., 2007, Tan et al., 2001) and has been used in many studies examining children with neurodevelopmental disorders, including ASD and DCD (Siaperas et al., 2012, Piek et al., 2006, Piek and Edwards, 1997, Green et al., 2002, Green et al., 2009, Wilson et al., 2000).

Table 3.4: Items in Movement Assessment Battery for Children, 2nd Edition (by age band)

Movement Assessment Battery for Children, 2nd Edition				
Category	Item No.	Age Band 1	Age Band 2	Age Band 3
Manual Dexterity	1	Posting Coins*	Placing Pegs*	Turning Pegs*
	2	Threading Beads	Threading Lace	Triangle with Nuts and Bolts
	3	Drawing Trail 1	Drawing Trail 2	Drawing Trail 3
Aiming & Catching	4	Catching Beanbag	Catching with Two Hands	Catching with One Hand*
	5	Throwing Beanbag onto mat	Throwing Beanbag onto mat	Throwing at Wall Target
Balance	6	One-Leg Balance*	One-Board Balance*	Two-Board Balance
	7	Walking Heels Raised	Walking Heel-to-Toe Forwards	Walking Heel-to-Toe Backwards
	8	Jumping on Mats	Hopping on Mats*	Zig-Zag Hopping*

* = tested bilaterally

3.6 Testing Protocol for Chapter 5 and Chapter 6

Data for Chapter 5 and Chapter 6 were collected simultaneously and separated for analysis and write-up. All children with a diagnosis of ASD and their parent/guardian attended for two sessions. The first session consisted of the ADI-R, ADOS-2 and an IQ test (WASI). All physical tests and measurements took place in the second visit. Children with DCD and typically developing children attended for one session. IQ was assessed once introductions were completed and written informed consent was obtained. The order of events for data collection is presented in Figure 3.5 below.

Participant set-up for gait analysis is shown in Figure 3.6.

Figure 3.5: Flow chart of order of testing for data collection of Chapter 5 and Chapter 6 (chapters 5 and 6)

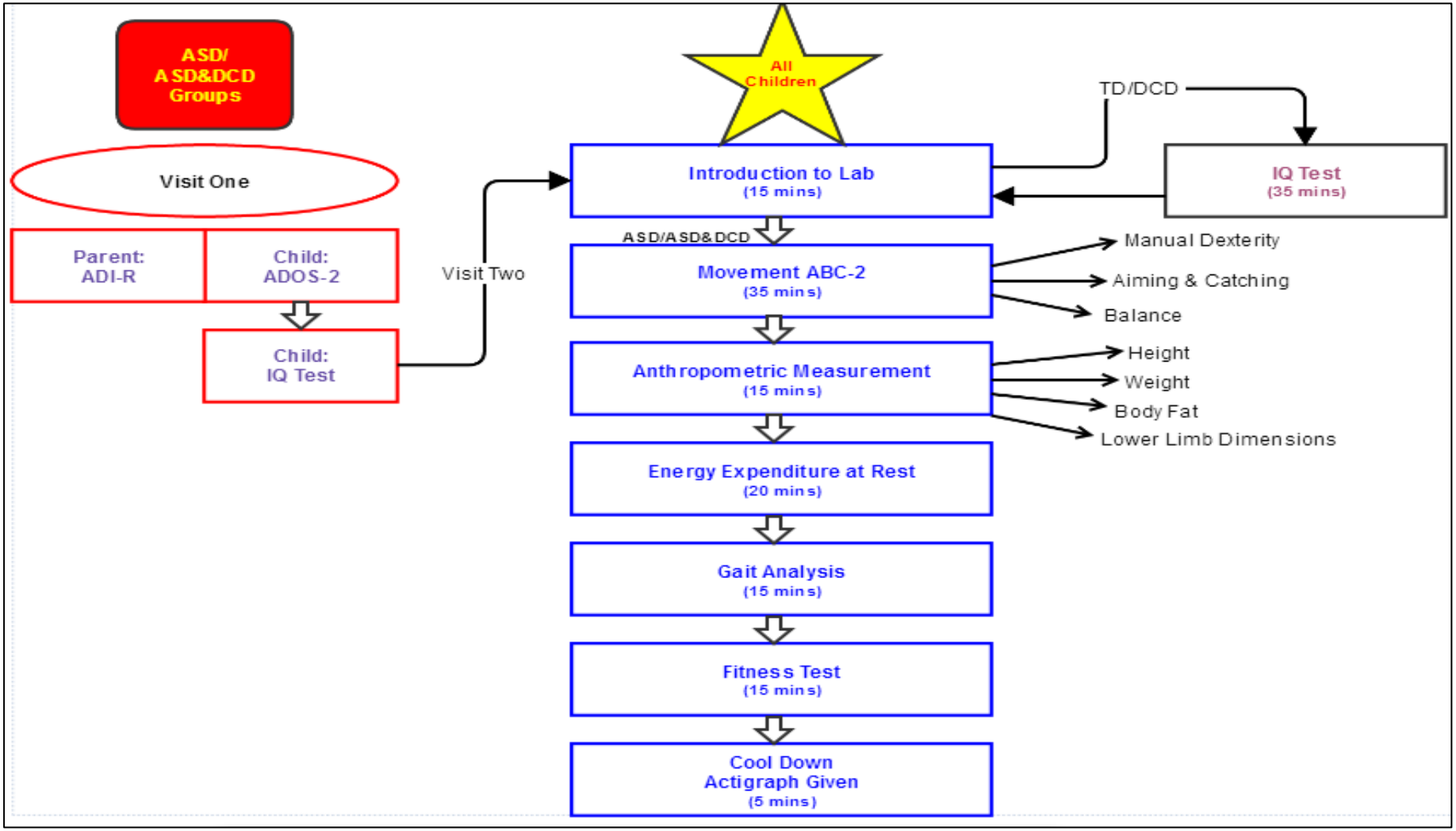
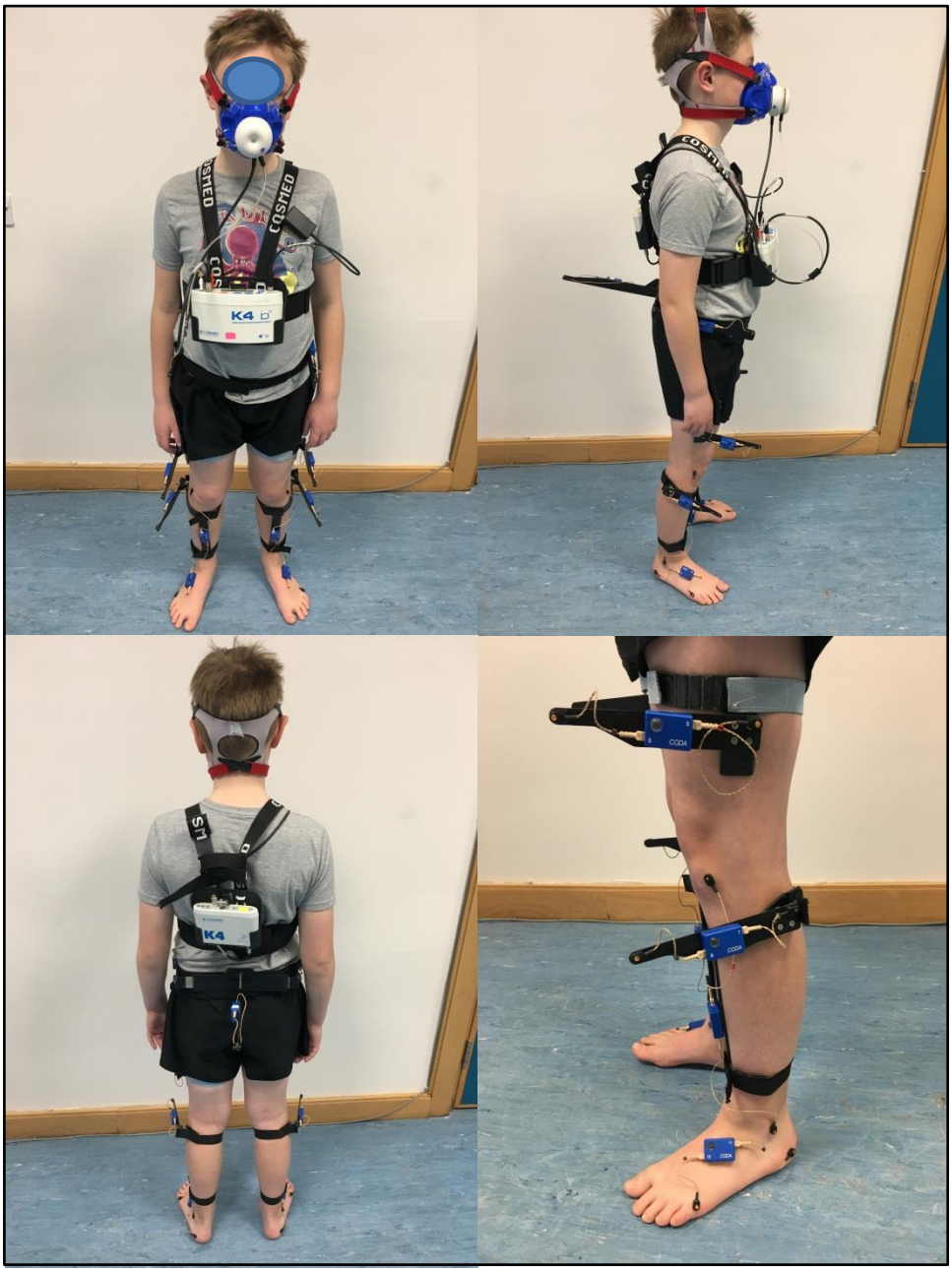


Figure 3.6: Participant Set-up for Gait Analysis



CHAPTER 4: PHYSICAL ACTIVITY IN CHILDREN WITH AUTISM SPECTRUM DISORDER AND/OR DEVELOPMENTAL COORDINATION DISORDER.

4.1 INTRODUCTION

Children with ASD typically have difficulty with social communication and language, and may exhibit restricted and repetitive behaviours (RRBs) (Leekam et al., 2011). Some RRBs may be smaller movements of the upper limb such as finger flicking or arm flapping, or may be larger full-body movements such as rocking back and forth (Marsden & Fahn, 1994). These may also be described as motor stereotypies, defined as 'involuntary, coordinated, patterned, repetitive, rhythmic, and purposeless but seemingly purposeful movements' (Goldman et al., 2009). Other examples of such stereotypies are pacing, jumping and hopping. These atypical movements may have an impact on motor coordination and, in turn, physical activity and participation in sport (Radonovich et al., 2013).

Physical activity is an important factor in a healthy lifestyle as it is known to have overall health benefits and promotes psychological well-being. Physical activity may also improve mood while reducing symptoms of perceived anxiety, which has been reported to affect approximately 40% of children and adolescents with ASD (De Matos et al., 2009, van Steensel et al., 2011). Studies which have examined physical activity in children with ASD or DCD have found that they spend a reduced amount of time in moderate-to-vigorous physical activity (MVPA) than typically developing peers (Pan, 2008, Sandt & Frey, 2005, Tyler et al., 2014, Cermak et al., 2015). Possible reasons for this include motor impairment and difficulty with social integration. These factors may also impact the child's ability to integrate into a team environment.

As discussed in Chapter 1 (see Section 1.4.4, page 10), the American College of Sports medicine recommends that children are moderately to vigorously active for at least 60 minutes per day. Several studies have reported that a low proportion of children with ASD are sufficiently active to meet these recommended guidelines (Bandini et al., 2013, Pan et al., 2016). Furthermore, children with ASD have been reported as less likely to participate in sports compared with children with other neurodevelopmental disorders (Blanchard et al., 2006).

The objectives of this study, therefore, were:

1. to determine the frequency and duration of physical activity (PA) in children with ASD, DCD and comorbid ASD and DCD
2. to determine the frequency and duration of sedentary behaviour in children with ASD, DCD and comorbid ASD and DCD
3. to determine the types of physical activities and sedentary behaviours engaged in by children with ASD, DCD and comorbid ASD and DCD
4. to compare variables from objectives 1, 2 and 3 with typically developing controls

4.2 METHODS

4.2.1 Modification of a Validated Questionnaire for Use as an Online Survey

The Modifiable Activity Questionnaire for Adolescents (MAQ-A) is a questionnaire designed to assess PA in youth (Aaron et al., 1993, Aaron et al., 1995). It has been shown to be a valid and reliable tool for collecting habitual PA data in a cost-effective manner (Aaron et al., 1993, Aaron et al., 1995, Delshad et al., 2015). The questions used in data collection for this study were based on questions from the MAQ-A. The questionnaire had been modified by a previous PhD student to capture PA data in Irish

children and included questions regarding the child's use of video games and this version of the MAQ-A formed the basis for this study (see Appendix VII). This version of the MAQ-A was a 17-item self-report questionnaire.

In this study I modified the questions to be appropriate for parental response and additional items were added while some questions were combined and others were excluded (see Appendix VIII for online survey). Modifications included the addition of questions 1-3 to determine basic demographic information and eligibility for inclusion – the child's gender, age and clinical diagnosis were reported in these questions. Four questions regarding mode of transport to school (questions 1-4 in Appendix VII) were combined into one question which assessed mode of transport and one question which assessed duration (questions 4 and 5 in Appendix VIII). The list of physical activities (question 5 in Appendix VII) was updated to include current popular sports in Ireland in question 7 of the survey (Appendix VIII). Hockey, martial arts and rugby were added to this list. Ballet was combined with dance classes. In question 8 of the survey, parents were asked to give an overall estimate of time spent in these activities in an average week as the tabular form (question 6 in Appendix VII) was incompatible with the online survey layout. Determining the amount of time in each type of sedentary behaviour was not an objective of this study. Therefore, questions 8-17 of the questionnaire (Appendix VII) were combined into two succinct questions – one assessed the sedentary behaviours engaged in by the child in the past week and one probed the total amount of time spent in these behaviours (questions 9 and 10, Appendix VIII).

Survey Monkey® is an online platform for the design, dissemination and collection of responses for surveys. It is user-friendly and a convenient method of collecting a large number of responses to surveys. This study utilised Survey Monkey® as a means of obtaining a large sample to provide a more representative overview of the physical activity habits of children with ASD and/or DCD.

4.2.2 Recruitment methods

Recruitment was conducted through social media. The TCD Autism and Rare Neurodevelopmental Disorders Research Group maintain a social media account (https://www.facebook.com/AutismTCD/) for dissemination and communication regarding ongoing research projects and to support recruitment.

The link to the survey was disseminated through the Facebook page using a paid targeting strategy. Paid advertising on Facebook ("Boost Post") allows specification of the target audience. For this post we targeted the audience using the following terms: "Autism", "Asperger Syndrome", and "Dyspraxia".

Sufficiently completed responses from parents of school-aged children with ASD, DCD, comorbid ASD and DCD or typical development were included. Responses were automatically excluded if details of the child's age, gender and diagnosis were not given (i.e. if questions 1-3 were not answered). Responses were also excluded where conflicting responses were given in the diagnosis question (e.g. if the child was reported to have both ASD and typical development). Where there were missing data in questions regarding PA and sedentary behaviour, the results are presented as proportions of those who completed the question rather than of the whole sample.

4.2.3 Data Analysis

Responses were exported from Survey Monkey® in excel files. Data was cleaned using Microsoft Excel 2010. This process involved allocating responses into four study groups: ASD, DCD, ASD&DCD and TD by reported diagnosis. Data was then imported into IBM's Statistical Package for the Social Sciences (SPSS) for analysis. Chi-squared tests of homogeneity were performed to evaluate significant between-group differences. Where necessary to meet sample size requirements for these tests, data were pooled appropriately and Fisher's exact test was carried out where applicable. Post hoc analysis involved pairwise comparisons using the z-test of two proportions with a Bonferroni correction for multiple comparisons. Ordinal regressions were carried out to determine gender and age interactions with ordinal dependent variables (weekly PA, weekly sedentary behaviour and weekly P.E.). Binomial regression was carried out to determine gender and age interaction with nominal dependent variables (method of school transport). Continuous variables were analysed using Kruskal-Wallis H tests and Wilcoxon Signed Rank Tests.

4.3 RESULTS

Of 886 total parental responses, 285 were insufficiently completed and were excluded from analysis. A further 51 responses contained conflicting reports of the child's diagnosis and were also excluded. The remaining responses (n=550) met the inclusion criteria and were assigned to one of four groups based on the subject's diagnosis. Table 4.1 shows the distribution into the four groups and group characteristics. There was no statistically significant difference in age across the groups (p=0.110), and gender proportions were similar across the ASD, DCD and ASD&DCD groups. However, the TD group had a higher proportion of females in comparison to the other three

groups ($p<0.0005$). The effects of gender and age are discussed later in the results section.

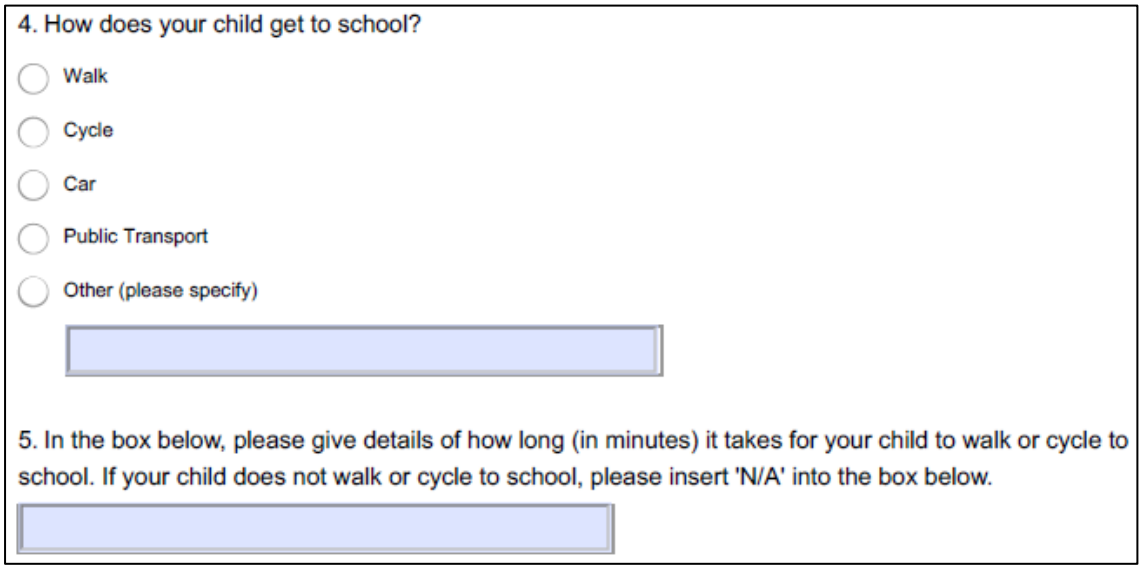
Table 4.1: Group Characteristics

Group	ASD	DCD	ASD&DCD	TD	Total	p-value
N	294	94	81	81	550	N/A
Female: Male (% Males)	58:236 (80%)	21:73 (78%)	10:71 (88%)	35:46 (57%)	124:426 (77%)	<0.0001
Age in years (mean ± SD)	9.43±3.56	9.44±3.21	9.44±3.38	8.43±2.93	9.29±3.4	0.112

4.3.1 School Transport

Questions 4 and 5 assessed children's modes of transport to school and are displayed in Figure 4.1.

Figure 4.1: Questions Related to School Transport



As proportions of children reported to walk or cycle to school were low and violated requirements for chi-squared analysis, data were pooled into two categories: active methods (walking and cycling) and passive methods (car and public transport). There

were no significant between-group differences in methods of school transport (p=0.230) and no gender by group effect was observed. A low proportion of all groups travelled to school by active methods (ASD 20.4%; ASD&DCD 27.8%; DCD 22.6%; TD 30%). These results are presented in Table 4.2.

Table 4.2: Methods of Transport to School

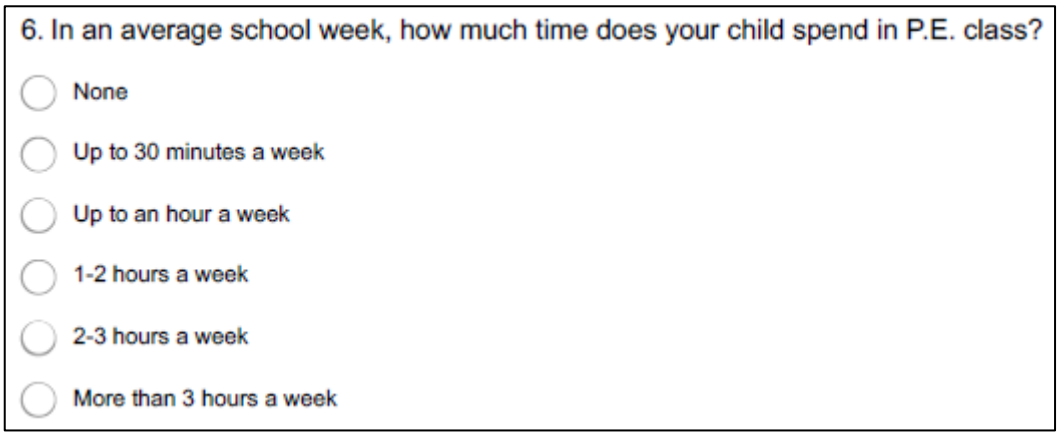
School Transport		Group			
		ASD	ASD&DCD	TD	DCD
Active	Count	57_a	22_a	24_a	21_a
	% within Group	20.40%	27.80%	30.00%	22.60%
Sedentary	Count	223_a	57_a	56_a	72_a
	% within Group	79.60%	72.20%	70.00%	77.40%

Each subscript letter denotes a subset of Group categories whose column proportions do not differ significantly from each other at the .05 level.

4.3.2 Physical Education Class (PE)

Question 6 assessed the time spent in P.E. class per week and is displayed in Figure 4.2.

Figure 4.2: Question Assessing Time Spent in PE Class Per Week



There were no significant between-group differences in the amount of time spent in P.E. class per week (see Table 4.3). No gender or age effects or interactions were observed. The majority of the sample (57.4%) had less than one hour of P.E. class per week.

Table 4.3: Time Spent in Physical Education Per Week

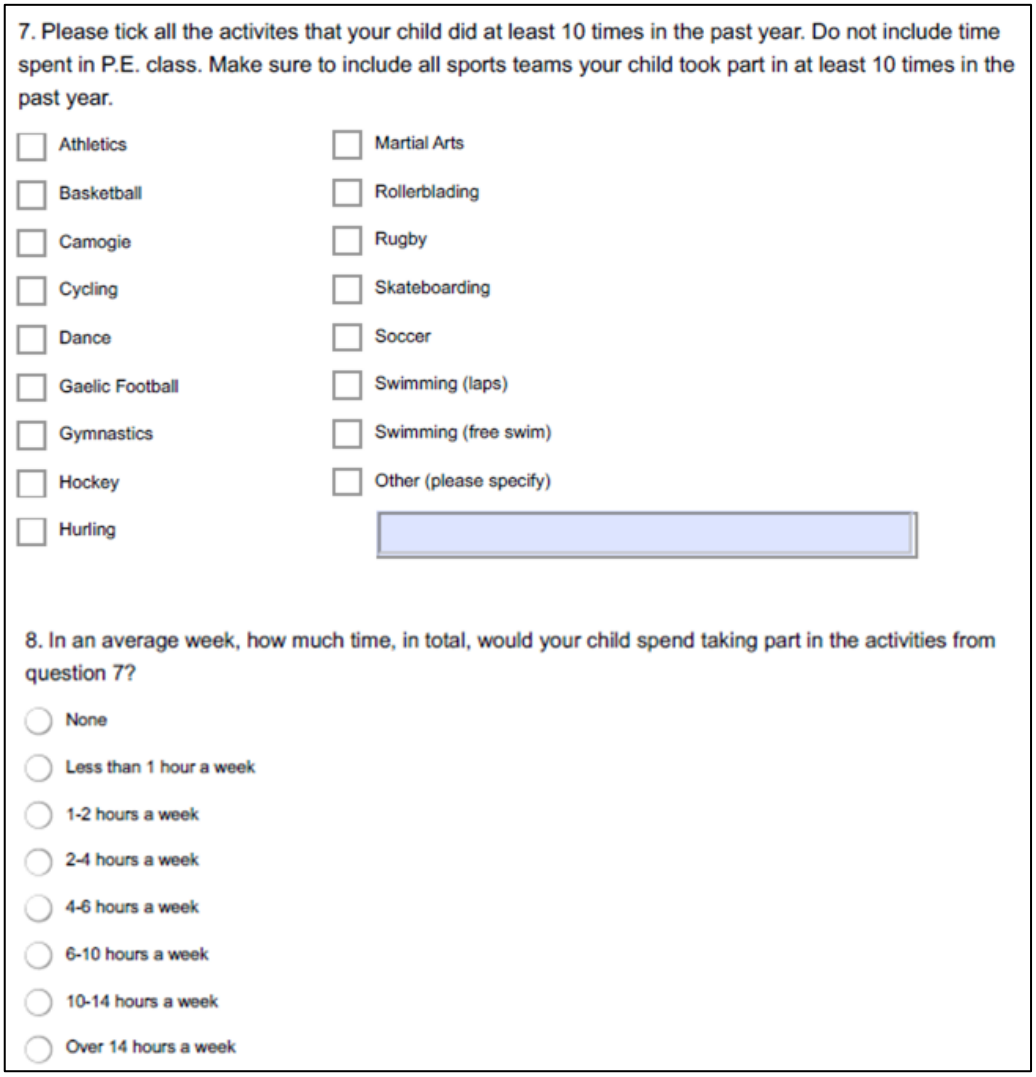
P.E. Class Per Week		Group			
		ASD	ASD&DCD	TD	DCD
<30 minutes	Count	47_a	16_a	15_a	25_a
	% within Group	18.70%	21.60%	19.70%	28.40%
< 1 hour	Count	112_a	34_a	36_a	31_a
	% within Group	44.40%	45.90%	47.40%	35.20%
1-2 hours	Count	65_a	20_a	18_a	25_a
	% within Group	25.80%	27.00%	23.70%	28.40%
> 2 hours	Count	28_a	4_a	7_a	7_a
	% within Group	11.10%	5.40%	9.20%	8.00%

Each subscript letter denotes a subset of Group categories whose column proportions do not differ significantly from each other at the .05 level.

4.3.4 Leisure-time Physical Activity

Leisure-time PA was assessed in questions 7 and 8 as displayed in Figure 4.3. Due to violations of requirements for chi-squared analysis, data were pooled into three categories for analysis: 1) < 1 hour, 2) 1-6 hours and 3) >6 hours. These categories were chosen for the following reasons: category 1 allows analysis of proportions of children with very low levels of PA; category 2 and 3 allows the division between those meeting recommended guidelines of at least 60 minutes of MVPA per day.

Figure 4.3: Questions Assessing Leisure-time PA



Significant between-group differences were found ($p<0.0005$). Post-hoc z tests revealed a significantly greater proportion of children in the ASD&DCD group (41.9%) spent less than one hour taking part in structured PA than their TD peers (14.3%). No between-group differences were found in the one to six hours category. A significantly larger proportion of TD children (15.6%) participated in more than six hours of structured PA per week than children in the ASD&DCD group (1.4%) and the DCD group (1.2%). The ASD group (9.3%) was not significantly different to the other three

groups in this category. Results are presented in Table 4.4. No gender or age effects/interactions were observed.

Table 4.4: Leisure- time Physical Activity (PA) Per Week

Weekly PA		Group			
		ASD	ASD&DCD	TD	DCD
< 1 hour	Count	$71_{a, b}$	31_{b}	11_{a}	$19_{a, b}$
	% within Group	27.40%	41.90%	14.30%	22.90%
1-6 hours	Count	164_{a}	42_{a}	54_{a}	63_{a}
	% within Group	63.30%	56.80%	70.10%	75.90%
> 6 hours	Count	$24_{a, b, c}$	1_{c}	12_{b}	$1_{a, c}$
	% within Group	9.30%	1.40%	15.60%	1.20%

Each subscript letter denotes a subset of Group categories whose column proportions do not differ significantly from each other at the .05 level.

There were significant differences between the groups in the proportions of children meeting recommended guidelines for PA. A significantly larger proportion of TD children (15.6%) met guidelines than those with ASD&DCD (1.4%) and those with DCD (1.2%). No other significant differences were found with any other group combination (see Table 4.5).

Table 4.5: Proportion of Children Not Meeting Recommended Physical Activity Guidelines in Each Group

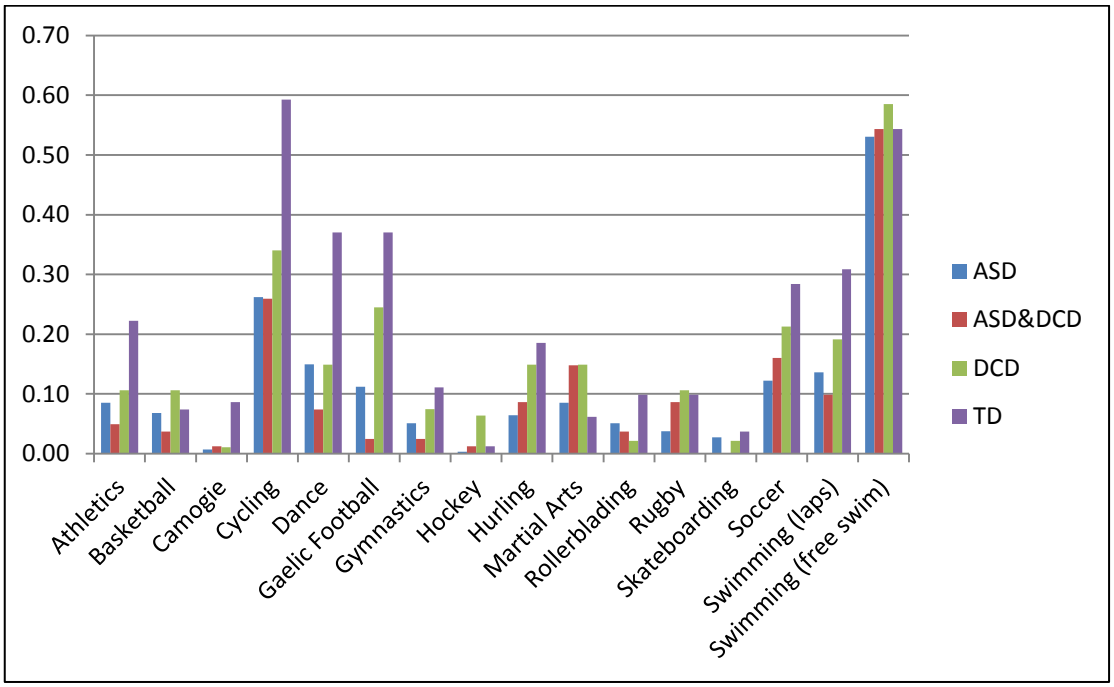
PA Per Week		Group			
		ASD	ASD&DCD	TD	DCD
Not Meeting PA Guidelines	Count	$235_{a, b, c}$	73_{c}	65_{b}	$82_{a, c}$
	% within Group	90.70%	98.60%	84.40%	98.80%

Each subscript letter denotes a subset of Group categories whose column proportions do not differ significantly from each other at the .05 level.

4.3.5 Number of Sports

A simple count of the number of sports children participated in was analysed using a Kruskal-Wallis H Test. Significant between-group differences in the number of sports participants took part in ($p<0.0005$). Post-hoc tests revealed that the TD group participated in a greater number of PA activities than the ASD group ($p<0.0005$), the ASD&DCD group ($p<0.0005$) and the DCD group ($p=0.02$). The DCD group participated in significantly more activities than the ASD group ($p=0.001$) and the ASD&DCD group ($p=0.012$). There were no significant differences detected between the ASD group and the ASD&DCD group. The different sports participated in by group are illustrated in Figure 4.4.

Figure 4.4: Participation in Sports by Group



4.3.6 Individual and Team Activities

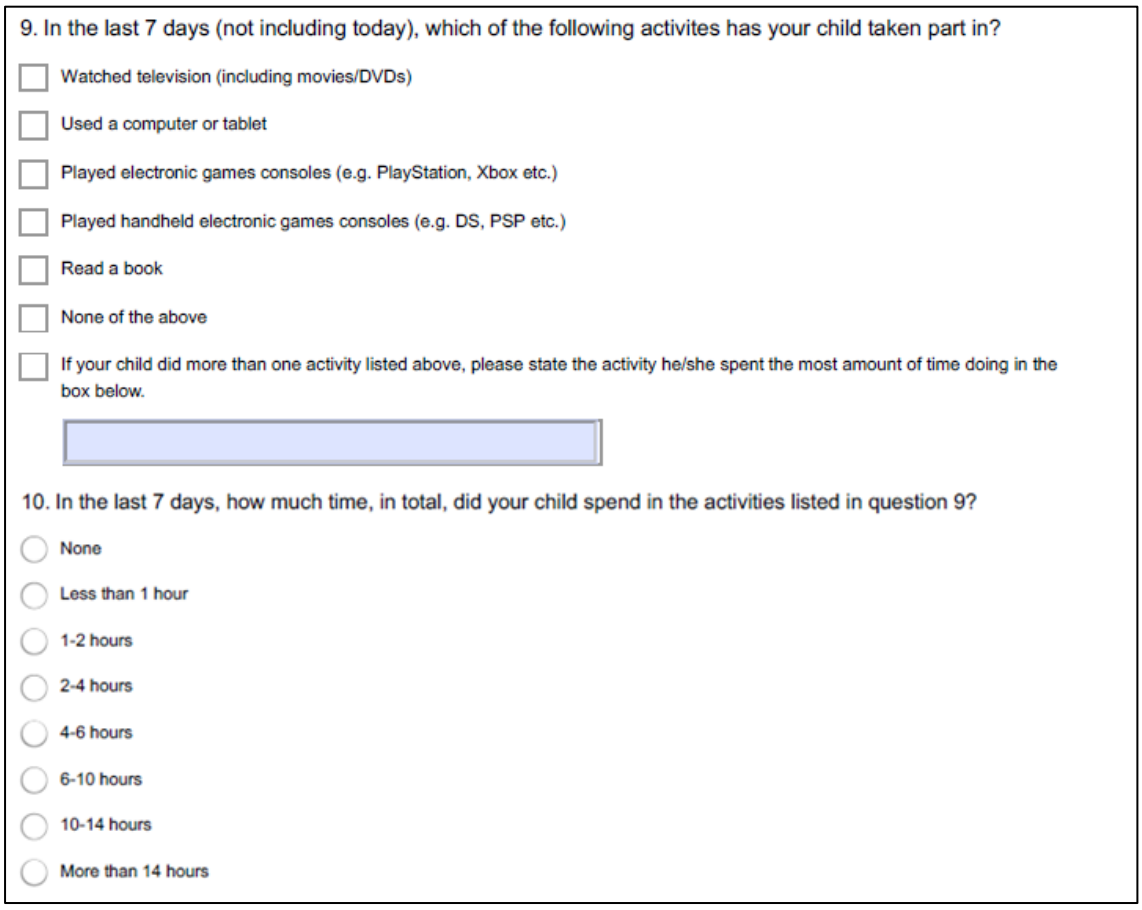
Sports were categorised into team and individual activities. As there were more individual sports listed in question 7, the number of sports each child took part in from each category was divided by the total number of sports in each category and analysed

using Kruskal-Wallis H Tests. Significant difference between the groups in the number of individual sports activities were revealed, with the TD group participating in more individual sports than the ASD group ($p<0.0005$), the ASD&DCD group ($p<0.0005$) and the DCD group ($p=0.008$). No significant differences were found between the ASD, ASD&DCD and DCD groups. Similarly, significant differences were found in the number of team sports activities the groups partook in. The TD group participated in more team sports than the ASD group ($p<0.0005$) and the ASD&DCD group ($p<0.0005$. The DCD group took part in more team sports than the ASD group ($p=0.001$). There were no significant differences in any other pairwise comparisons.

4.3.7 Leisure-time Sedentary Behaviour

Questions 9 and 10 assessed sedentary behaviours and are displayed in Figure 4.5.

Figure 4.5: Questions Assessing Sedentary Behaviour Per Week



Data were pooled into two categories for analysis: less than six hours per week and greater than six hours per week. Significant between-group differences were found (p=0.028). Post-hoc tests revealed no significant differences between the ASD group and the ASD&DCD group. However, a significantly higher proportion of the ASD group (57.8%) and the ASD&DCD group (61.5%) spent more than six hours per week engaging in sedentary behaviours such as screen time or reading than the TD group (40.3%). Chi-squared analysis of the type of sedentary behaviour most often engaged in showed no significant differences between the groups (p=0.142). These results are presented in Table 4.6. No gender effects or interactions were observed. An age by group interaction was observed suggesting that for each increase of one year in age, time spent in sedentary behaviour increased by 50% (p=0.009). However, this trend was only evident in the TD group (p=0.041) and no age effect was observed in the ASD, ASD&DCD or DCD groups.

Table 4.6: Leisure-time Sedentary Behaviour Per Week

Weekly SB		Group			
		ASD	ASD&DCD	TD	DCD
< 6 hours	Count	113_a	30_a	46_b	$40_{a, b}$
	% within Group	42.20%	38.50%	59.70%	47.10%
> 6 hours	Count	155_a	48_a	31_b	$45_{a, b}$
	% within Group	57.80%	61.50%	40.30%	52.90%

Each subscript letter denotes a subset of Group categories whose column proportions do not differ significantly from each other at the .05 level.

4.4 DISCUSSION

This survey aimed to assess the physical activity and sedentary behavioural habits based on parental report of children with ASD and/or DCD in an average school week

when compared with typically developing children. Physical activity is a vital factor in overall health. Research indicates that children are becoming less active and spend more time in sedentary behaviour, especially screen time (Sisson et al., 2009)(Sisson et al., 2009; Riddoch et al., 2004; Houghton et al., 2015). Children with ASD and/or DCD face extra obstacles to engaging in physical activity and sports in comparison to typically developing children, including social communication deficits in ASD and motor coordination deficits, which have been documented in both conditions (Van Waelvelde et al., 2010; Van Damme et al., 2015; Dewey et al, 2007).

The findings indicated that overall the majority of children surveyed were not meeting guidelines in relation to participation in moderate to physical activity. A large proportion of all four groups participated in structured physical activity for less than six hours per week. Children with ASD were well below recommended exercise guidelines, with 27.4% being active for less than one hour per week and only 9% carrying out at least one hour of physical activity per day. Children in the ASD&DCD group were the least active with 41.9% participating in less than one hour of PA per week and approximately 1% of children were meeting recommended exercise guidelines. Just under a quarter of children with DCD were active for less than one hour per week and, similar to the ASD&DCD group, approximately 1% were meeting guidelines. The TD group was more active but still a small proportion (14%) of the group were meeting guidelines, with a further 14% of this group participating in less than one hour of physical activity per week. Several studies have shown that the levels of PA in childhood and adolescence is related to PA levels in adulthood, such that the benefits of being physically active in youth carry into adult health and activity,

including contributing to the formation of healthy lifestyles (Makinen et al., 2010; Hallal et al., 2006; Biddle et al., 2004). The data in this study illustrate a serious deficit in activity levels in these children which may leave them at risk of inactivity in their adult lives.

Reduced engagement in physical activity is likely to lead to greater morbidity and mortality in adult life. In relation to ASD, a recent study highlighted that premature mortality rates were higher in people with ASD than the general population (Hirvikoski et al., 2016). At time of death, those with ASD were, on average, 16 years younger than peers in the general population. Cardiovascular disease, cancer and suicide were reported as the three leading causes of death in those with ASD (Hirvikoski et al., 2016). There is strong evidence to suggest that increased physical activity leads to a decrease in risk of cardiovascular events and disease (Pate et al., 1995; Paffenbarger et al., 1993; Myers et al., 2002) and that physical activity can reduce mortality in a variety of cancers (Bernstein et al., 1994; Carpenter et al., 1999; Thune & Fuberg, 2001). Data on PA was not available for the data in the study by Hirvikoski et al., (2016). However, given the results presented in this chapter, a possible risk factor has been identified. Future studies might investigate the link between reduced physical activity and ASD premature mortality rates. To the best of the author's knowledge, no published studies have examined mortality rates in DCD.

In recent research, a gender difference has been found in physical activity levels, with girls tending to be less physically active and spend more time in sedentary activity than boys (Jago et al., 2005; Vilhjalmsson & Thorlindsson, 1998; Slater & Tiggemann 2011; Vilhjalmsson & Kristjansdottir, 2003). This has also been shown in an Irish population

where boys were found to be more active than girls (Layte and McCrory, 2011). Possible explanations for this trend have been reported as reduced support at home and at school in terms of encouraging girls to participate in PA compared to their male counterparts (Telford et al., 2016). No significant gender differences were observed in these data. However, girls were underrepresented in all four groups and so the female sample may not have been sufficiently powered.

Many studies have examined the association between PA and mental health. A 2015 paper by Sibold et al., suggested that physical activity was inversely related to sadness and suicide ideation in adolescents and this may carry on to adulthood. This prompted an editorial by Stein & Dubowitz (2015) which cites a growing evidence base examining the relationship between mental health and exercise. They note that in recent years there has been a focus on integrating mental and physical health but that the action is somewhat unilateral, with primary caregivers being encouraged to recognise and intervene in mental health issues but less emphasis is put on mental health practitioners addressing physical health issues in their patients. In other words, the emphasis has been on the ability of practitioners such as physiotherapists and occupations therapists to recognise signs and symptoms of impaired mental health rather than on the ability of mental health practitioners to identify individuals who are physically inactive and means of encouraging physical activity for overall health promotion. The awareness of recommended PA/exercise guidelines of practitioners across a variety of disciplines has not yet been studied.

The results show an increased amount of time spent in sedentary behaviour and a decreased amount of time spent in physical activity among children with

neurodevelopmental disorders, suggesting a higher risk of overweight and obesity and related health problems, especially as the children grow into adolescence, which is in agreement with research by Curtin et al., (2015), Hinckson et al., (2013) and Silman et al., (2011). Curtin et al., (2015) found that children with ASD were 40% more likely to be overweight or obese than typically developing peers. Encouragement of physical activity in children with ASD may play a pivotal role in reducing this premature mortality phenomenon as it has been linked to a reduced risk of the top three causes of death highlighted by Hirvikoski et al. (2016), and may also reduce risk of other causes of premature death such as respiratory disease, bowel disease or endocrine conditions such as diabetes mellitus (Gleeson et al., 2011; Handschin & Spiegelman, 2008; Slentz et al., 2007; Knowler et al., 2002; Tuomilehto et al., 2001). One study found that children with ASD who were meeting exercise guidelines scored lower on measures of lower body flexibility and cardiovascular endurance than their TD peers, perhaps suggesting that meeting the guidelines does not provide the same amount of fitness-related benefits for children with ASD as it does for TD children (Pan et al., 2016). It may be the case that children with ASD may require more exercise to acquire the same benefits, or may need individually-prescribed recommendations for exercise. Increasing awareness of the benefits of physical activity and the risks of sedentary behaviour may also help reduce the relatively high prevalence of overweight and obesity in children with ASD and/or DCD.

The results of this study suggest that children with ASD participated in generally the same sports as typically developing peers, which may be due to the relatively narrow range of popular sports and accessibility to facilities in Ireland. However, there was a

reduction in the variety of sports and exercise activities that children with ASD participated in and in the number of team sports children with ASD participated in. This may be due to difficulty with social integration in a team environment, or social anxiety. It may also be due to the suggestion that individual sports tend to be more repetitive (e.g. swimming, running or cycling laps) and require less behavioural flexibility (i.e. to react to a situation within a game), which has been shown to be reduced in children with ASD who tend to be more rigid in their approach to situations (D.Cruz et al., 2013; APA, 2013). Furthermore, children with neurodevelopmental disorders are more likely to exhibit aggressive behaviours than typically developing peers (Shroeder et al., 2014; Farmer et al., 2015). A study by Kanne & Mazurek (2010) found that 68% of parents of children with ASD reported that their child had acted aggressively towards their caregiver and 49% to non-caregivers. Mental health issues such as sleep disturbance, anxiety and self-harm have also been documented in children with ASD (Shroeder et al., 2014;). physical activity has been shown to be inversely related to aggression, anxiety, and self-harm and to improve quality of sleep (Nouri & Beer, 1989; Kanchana et al., 1993; Loprinzi & Cardinal, 2011; King et al., 1997; Driver & Taylor, 2000; Foti et al., 2011). Screen time and sedentary behaviour have conversely been shown to be positively correlated with aggression, anxiety, self-harm and quality of sleep (Kelishadi et al., 2015; Liu et al., 2016; Parent et al., 2016; Foti et al., 2011). Therefore, increasing PA and reducing sedentary behaviour may help to alleviate the effects of these challenging behaviours.

A strength of this study was the large sample size. This is in part due to the ease of participation from the online survey design. However, a limitation of this study is that

the survey did not quantify time spent sitting in class, but this would also be considered sedentary activity. This, however, would be expected to be similar across the groups since school attendance is unlikely to differ. There was an age by group interaction in sedentary behaviour, such that with increasing age, sedentary behaviour increased. However, this trend was observed in the TD sample but not in the other groups. This implies that children with ASD and/or DCD do not follow the same pattern with increasing age as TD children and suggests that children with ASD and/or DCD tend to be more sedentary at a younger age and remain sedentary. It is important to note that children with these conditions were also more sedentary across all age groups which may indicate a floor-effect in the ability of this survey to quantify differences between younger children and adolescents in terms of the amount of time they spend in sedentary behaviour.

Similarly, this survey is limited to assessing structured PA as unstructured PA is difficult to assess through survey means. This may have impacted the results and may account for the extremely low proportions of children with ASD and DCD who were meeting guidelines. This was a limitation in this study as it was unable to quantify exercise through play which may contribute to meeting PA guidelines for children (ACSM, 2014). The subjective methods employed in this study also limit any analysis of the effect of variables such as BMI on PA and sedentary behaviour. There is presently a worrying epidemic in Ireland with childhood sedentary behaviour, overweight and obesity increasing. The recent 'Growing Up in Ireland' national longitudinal study of children examined overweight and obesity among nine-year-olds (Layte & McCrory, 2011). They found that 19% of children in Ireland were overweight and a further 7%

were obese. Their results suggest that the prevalence of childhood overweight and obesity in Ireland is increasing. Since children with neurodevelopmental disorders are at a higher risk of overweight and obesity, this has great implications for these children in the future. Interestingly, children from all groups, including typically developing controls exhibited poor adherence to recommended PA guidelines. This has not been previously reported in the Irish paediatric control population.

Overall, this study found that children with ASD and/or DCD were less active and more sedentary than typically developing peers. It also showed that children with ASD and/or DCD participate in a narrower variety of physical activities which has also been reported in other studies (Memari et al., 2015, Askari et al., 2015). However, there is evidence to show that subjective measures of PA, such as the questionnaire used in this study, may overestimate PA in typically developing children but may underestimate PA in children with ASD as they are more likely to accumulate time in MVPA in unstructured activity which is difficult to quantify through survey means (Bacardi-Gascón et al., 2012, Must and Tybor, 2005, Bandini et al., 2013). Therefore, there is a need for further study employing objective measurement of PA levels, such as triaxial accelerometers, to quantify PA in children with ASD and comorbid DCD.

CHAPTER 5: PHYSICAL ACTIVITY, FITNESS AND ENERGY EXPENDITURE

5.1 INTRODUCTION

As described in Chapters 1, 2 and 4, there is evidence to suggest that children with ASD or DCD have altered physical activity patterns in comparison to their typically developing peers. Chapter 4 adds some strength to this argument, and suggests that children with ASD and a dual diagnosis of DCD may be even less active and more sedentary. To date, no study has been published which has objectively measured PA levels in children with this dual diagnosis.

Fitness and energy expenditure are known to be related to PA levels and sedentary behaviour in typically developing individuals (Kohl et al., 1988, Cordain et al., 1998, Ravussin and Bogardus, 1989, Ekelund et al., 2005) and there are few studies examining the relationships between physical activity, energy expenditure and fitness in children with neurodevelopmental disorders.

The objectives of this study, therefore, were:

1. to determine the frequency and duration of physical activity in children with neurodevelopmental disorders
2. to determine the frequency and duration of sedentary behaviour in children with neurodevelopmental disorders
3. to determine fitness in children with neurodevelopmental disorders
4. to determine energy expenditure in children with neurodevelopmental disorders at rest, during gait and during maximal exercise

5. to compare the above parameters in children with neurodevelopmental disorders to typically developing controls

5.2 METHODS

Details of recruitment are given in Chapter 3 (see Section 3.2, page 53).

5.2.1 Data Collection

Physical activity was measured using Actigraph GT3X+ triaxial accelerometers (see Section 3.4.2, page 60) Participants were asked to wear the accelerometer on the waist, in front of the right hip and upright (see Figure 3.1, Section 3.4.2, page 61) for seven days. Parents were given a log diary to record the date, time and duration the accelerometer was worn (see Appendix IX). A minimum of 600 minutes wear time per day was required and individuals were required to record at least four valid days (including three weekdays and one weekend day) for their PA data to be included for analysis. These criteria for inclusion were based on methods described by Trost et al. (2006a). Wear time was analysed using the protocol developed by Choi et al. (2011) which defines non-wear time as zero activity counts for 90 minutes or more. This wear time validation was chosen as children with ASD or DCD are reported to engage in more sedentary behaviour than typically developing children and therefore may be inactive for up to 90 minutes at a time (Must et al., 2014, Cermak et al., 2015). Therefore, use of more conservative wear time validation protocols may have identified periods of sedentary behaviour as non-wear time. Activity was categorised into sedentary behaviour, light, moderate and vigorous PA based on cut-points defined by the number of activity counts recorded per minute. Table 5.1 shows the cut points used as proposed by Pulsford et al., (2011). These cut-points were chosen for use in this study as they were deemed to be the most appropriate for use with the

cohort examined. Pulsford et al., (2011) determined the cut-points on data obtained from children of a similar age to the cohort in this study and were deemed in pilot analysis to give an accurate picture of activity of children of this age where other derived cut-points were deemed too conservative or too strict (e.g. Freedson et al., (2005) and (Puyau et al., 2002)). It was not deemed within the scope of this project to independently derive cut-points for use with the specific cohort.

Table 5.1: Cut Point Used for Classification of Physical Activity by Intensity

Cut-Points:	**Counts Per Minute**
Sedentary	0-99
Light	100-2240
Moderate	2241-3840
Vigorous	3841+

(Pulsford et al., 2011)

Fitness was assessed using the 20 Metre Multistage Fitness Test (Leger et al., 1988) (see Section 3.4.5, page 63 and Appendix V).

Energy expenditure (EE) was assessed using the Cosmed K4b^2 portable indirect calorimeter (see Section 3.4.1, page 59). Participants were introduced to the face mask and given time to acclimatise to it prior to testing. EE was measured at rest, during gait and during the fitness test.

5.2.1 Data Analysis

Data was imputed into Microsoft Excel throughout data collection and imported into IBM's Statistical Package for the Social Sciences (SPSS) version 22 for statistical analysis. Between-group differences were analysed using one-way ANOVA or Kruskal-Wallis H Tests as appropriate for distributions. Between-group differences in total time

(mins), daily time (mins) and percentage of time spent in sedentary, light, moderate, vigorous and moderate-to-vigorous PA were the main PA variables analysed. Fitness test results and EE in kilocalories per minute (kcals/min) and oxygen consumption per kilogram of body weight (ml/min/kg) at rest, during gait and during the fitness test were also analysed by group. Post-hoc tests (Tukey's for ANOVA; Mann Whitney U-Tests for Kruskal-Wallis H Test) were carried out on significant findings and were adjusted for multiple pairwise comparisons using the Holm-Bonferroni method (Holm, 1979) whereby the p-value is divided by the number of groups included in pairwise comparisons. In the case of this research, the p-value produced by post-hoc tests was automatically divided by 4 in SPSS and this value is reported in pairwise comparisons throughout the results sections in this chapter and in Chapter 6. Wilcoxon Signed Ranks tests were used to determine differences in weekday and weekend PA. Fitness, EE and VO_2 values were plotted against PA data to examine potential trends. The significance level was set as $p<0.05$.

5.3 RESULTS

A total of 143 children who met inclusion criteria were recruited and divided into the four study groups by diagnosis: ASD (n=29; 23 males), ASD&DCD (n=30; 21 males), DCD (n=41; 24 males) and TD (n=43; 25 males). Group characteristics are shown in Table 5.2 with median and interquartile ranges presented where datasets are not normally distributed. There were no significant differences in age, gender, height, weight, body fat percentage or BMI. However, the groups differed significantly in BMI percentile (p=0.035). Post hoc pairwise comparisons revealed that the TD group had significantly lower BMI percentiles than the ASD&DCD group (p=0.025). No other significant differences were observed in pairwise comparisons.

ADOS-2 comparison scores were not significantly different between the ASD and the ASD&DCD group (p=0.233). However, significant between-group differences in IQ were observed (p<0.0001). Post hoc analysis revealed that the TD group scored significantly higher on the IQ test than the ASD group (p<0.0001), the ASD&DCD group (p<0.0001) and the DCD group (p<0.0001). No other pairwise comparisons yielded significant results.

Table 5.2: Participant Characteristics

Group	ASD (n=29) (mean ± SD)	ASD&DCD (n=30) (mean ± SD)	DCD (n=41) (mean ± SD)	TD (n=43) (mean ± SD)	Significance
Age (years)	9.42; 3.6†	9.29; 4.0†	9.42; 3.0†	9.42; 4.25†	*p*=0.407
Height (m)	1.33; 0.2†	1.35; 0.2†	1.37; 0.2†	1.35; 0.3†	*p*=0.326
Weight (kg)	30.65; 15.5†	33.85; 17.0†	34.15; 20.5†	29.70; 13.0†	*p*=0.265
BMI (kg/m^2)	17.22; 2.5†	17.53; 6.0†	16.76; 4.4†	16.27; 2.2†	*p*=0.112
BMI Percentile	64; 56†	77; 49†	50; 56†	39; 35†	*p*=0.035*
Body Fat Percentage	21.8; 5.4†	24.1; 11.2†	20.9; 6.4†	20.1; 6.9†	*p*=0.067
IQ	101 ± 15.5	99 ± 15.7	102 ± 13.1	116 ± 13.2	p<0.0001*
ADOS-2 Comparison Score	7.0 ± 2.3	7.5 ± 2.0	N/A	N/A	p=0.233

† = median; interquartile range (nonparametric data)
p = Independent Samples T-test p-value with significance at alpha ≤ 0.05.
p = Kruskal Wallis p-value with significance at alpha ≤ 0.05.
* = significant difference between groups.

5.3.1 Physical Activity (PA)

Results of wear time analysis revealed that 119 of participants had sufficient wear time for inclusion in data analysis. There were no significant differences between the

groups in the number of calendar days recorded, overall wear time, the number of epochs or total activity counts. These results are presented in Table 5.3.

Table 5.3: Results for Wear Time Analysis

Group	ASD (n=27) (median; IQR)	ASD&DCD (n=25) (median; IQR)	DCD (n=36) (median; IQR)	TD (n=31) (median; IQR)	Significance
Wear Time (mins)	4804; 1709	5121; 1093	4992; 877	5200; 1010	$p = 0.39$
Days	7; 2	7; 1	7; 1	7; 1	$p = 0.53$
Epochs	19216; 6836	20484; 4370	19968; 3508	20800; 4040	$p = 0.39$
Counts	7228012; 3774595	7763934; 5019958	8451937; 4033915	9850870; 4816478	$p = 0.06$

p = Kruskal Wallis p-value with significance at alpha ≤ 0.05

PA results are presented in Table 5.4. Seven-day analysis revealed no significant differences in PA variables and intensities. Weekday analysis also revealed no significant differences in these variables. Weekend analysis revealed significant between-group differences in total time spent in vigorous PA (p=0.028) and the percentage of time spent in vigorous PA (p=0.026). Post-hoc analysis revealed that the TD groups spent more time in total in vigorous activity than the ASD group (p=0.035) and also spent a greater percentage of time in vigorous activity than the ASD group (p=0.026). There was also a difference in weekend vigorous PA between the TD group and the ASD&DCD group and also between the TD group and the DCD group but these differences did not survive post-hoc p-value adjustment. No other pairwise comparisons revealed significant differences.

A gender effect was observed for MVPA ($p=0.01$). In all four groups, males spent more time in MVPA than females. No group by gender effect was observed ($p=0.98$). No gender effect or interaction was found for sedentary behaviour ($p>0.05$).

Table 5.4: Results for Physical Activity by Group

Group	Time Frame	ASD (n=27) (mean ± SD)	ASD&DCD (n=25) (mean ± SD)	DCD (n=36) (mean ± SD)	TD (n=31) (mean ± SD)	Sig.
Total Sedentary (mins)	Weekly	2844 ± 717	2887 ± 714	2911 ± 730	2888 ± 639	p = 0.99
	Weekday	2017 ± 562	2107 ± 552	2175 ± 612	2161 ± 527	p = 0.95
	Weekend	775; 609†	853; 575†	811; 486†	702; 364†	*p* = 0.90
Daily Sedentary (mins)	Weekly	459 ± 80	453 ± 88	450 ± 83	444 ± 75	p = 0.91
	Weekday	466 ± 80	456 ± 93	454 ± 83	461 ± 91	p = 0.95
	Weekend	440 ± 128	449 ± 89	430 ± 124	404 ± 89	p = 0.39
Percentage of Time in Sedentary (%)	Weekly	61 ± 7	59 ± 10	59 ± 9	59 ± 9	p = 0.71
	Weekday	61 ± 7	59 ± 11	59 ± 9	59 ± 9	p = 0.80
	Weekend	62; 14†	60; 10†	61; 16†	60; 18†	*p* = 0.84
Total Light (mins)	Weekly	1460 ± 373	1600 ± 427	1609 ± 318	1620 ± 426	p = 0.37
	Weekday	1080 ± 265	1165 ± 343	1216 ± 315	1204 ± 308	p = 0.34
	Weekend	384; 240†	487; 314†	407; 263†	421; 323	*p* = 0.63
Daily Light (mins)	Weekly	236 ± 47	251 ± 58	253 ± 47	248 ± 53	p = 0.63
	Weekday	242 ± 44	252 ± 62	259 ± 54	255 ± 51	p = 0.62
	Weekend	232 ± 68	250 ± 60	226 ± 73	230 ± 68	p = 0.58
Percentage of Time in Light (%)	Weekly	32 ± 6	33 ± 8	34 ± 7	33 ± 7	p = 0.73
	Weekday	32 ± 5	33 ± 8	34 ± 7	33 ± 7	p = 0.73
	Weekend	31; 11†	34; 7†	31; 11†	32; 13†	*p* = 0.71
Total Moderate (mins)	Weekly	187; 129†	223; 145†	209; 107†	269; 158†	*p* = 0.55
	Weekday	148; 82†	164; 112†	160; 107†	188; 115†	*p* = 0.51
	Weekend	56; 51†	54; 42†	43; 38†	57; 61†	*p* = 0.60
Daily Moderate (mins)	Weekly	34; 17†	37; 22†	33; 17†	39; 21†	*p* = 0.71
	Weekday	33; 18†	36; 24†	32; 20†	38; 20†	*p* = 0.72
	Weekend	29; 20†	35; 19†	33; 20†	30; 27†	*p* =0.77
Percentage of Time in Moderate (%)	Weekly	4; 2†	4; 3†	4; 2†	5; 3†	*p* = 0.74
	Weekday	5; 2†	4; 3†	4; 3†	5; 3†	*p* = 0.82
	Weekend	4; 2†	4; 3†	4; 3†	4; 3†	*p* = 0.60
Total Vigorous (mins)	Weekly	91; 84†	78; 123†	104; 98†	141; 131†	*p* = 0.06
	Weekday	74; 73†	62; 102†	71; 84†	94; 110	*p* = 0.19
	Weekend	17; 27†	18; 24†	18; 29†	36; 42†	*p*=0.03*
Daily Vigorous (mins)	Weekly	13; 16†	15; 19†	17; 15†	21; 23†	*p* = 0.12
	Weekday	18; 18†	14; 21†	20; 17†	20; 23†	*p* = 0.29
	Weekend	9; 17†	11; 16†	12; 22†	21; 25†	*p* = 0.08
Percentage of Time in Vigorous (%)	Weekly	2; 2†	2; 2†	2; 2†	3; 3†	*p* = 0.13
	Weekday	2; 2†	2; 3†	3; 2†	3; 3†	*p* = 0.32
	Weekend	1; 2†	1; 2†	2; 3†	3; 4†	*p*=0.03*
Total MVPA (mins)	Weekly	285; 187†	297; 209†	311; 208†	388; 234†	*p* = 0.15
	Weekday	239; 140†	225; 210†	234; 150†	279; 222†	*p* = 0.28
	Weekend	75; 65†	77; 51†	82; 61†	97; 93†	*p* = 0.23
Daily MVPA (mins)	Weekly	51; 29†	53; 33†	54; 30†	58; 39†	*p* = 0.33
	Weekday	50; 34†	56; 37†	53; 36†	60; 39†	*p* = 0.46
	Weekend	42; 32†	47; 36†	42; 45†	55; 41†	*p* = 0.31
Percentage of Time in MVPA (%)	Weekly	7; 4†	7; 4†	7; 4†	8; 5†	*p* = 0.29
	Weekday	7; 5†	7; 4†	7; 5†	8; 4†	*p* = 0.50
	Weekend	6; 4†	7; 5†	6; 6†	8; 5†	*p* = 0.08

†= median; interquartile range (nonparametric data) * = significant difference between groups
p = one-way ANOVA p-value, *p* = Kruskal Wallis p-value with significance at alpha ≤ 0.05

Weekday and weekend sedentary behaviour and MVPA were compared in the total sample and in each group. Related-Samples Wilcoxon Signed Rank Tests revealed significant differences in sedentary behaviour during weekdays and weekend days in the whole sample. Analysis of the overall sample revealed that children spent more time in sedentary behaviours during the weekend than on weekdays ($p<0.0005$). Analysis of individual groups indicated no significant differences were found between weekday and weekend sedentary behaviour in the ASD, ASD&DCD or DCD groups. The TD group were significantly more sedentary during the weekend than they were on weekdays ($p=0.004$). PA levels on weekdays were compared to weekend days and no significant differences were found in any of the groups.

As data was collected on subjects throughout the year, analysis was carried out to investigate whether there was a difference in PA levels during term time in school and during the summer holidays. Data were analysed using Mann-Whitney U Tests for non-parametric data. No significant differences were observed between subjects measured during term-time compared with those measured during the summer holidays in any of the categories of activity. These analyses did not differ between groups or in the overall sample.

A higher proportion of children in the TD group were meeting recommended guidelines of at least 60 minutes a day than children in any of the other groups, but this was not statistically significant ($p>0.05$). However, it was notable that less than half of children in all groups were meeting these guidelines. Table 5.5 shows the percentage of children meeting these guidelines by group.

Table 5.5: Proportions of Subjects Meeting PA/Exercise Guidelines by Group

Group	ASD (n=27)	ASD&DCD (n=25)	DCD (n=36)	TD (n=31)
Meeting guidelines	9	9	12	17
% Of Group	33.3%	36%	38.7%	47.2%

A significantly smaller proportion of females were meeting exercise guidelines than males in the overall sample (p=0.009). However, this trend was not apparent when the groups were analysed individually. Similar proportions of children classified as underweight, healthy weight, overweight and obese were meeting exercise guidelines in the overall sample and in each of the four groups.

5.3.2 Fitness

A Kruskal Wallis H test for non-parametric data was used to investigate between group differences in fitness test scores (see Table 5.6). Significant between-group differences in fitness test scores were observed (p<0.0001). Post-hoc pairwise comparisons revealed that the TD group achieved higher scores than the ASD, the ASD&DCD and the DCD groups (p<0.0001, p=0.001 and p=0.003, respectively). No significant differences were found in any other combination of groups.

Table 5.6: Results for Fitness Test by Group

Group	ASD (median; IQR) (n=27)	ASD&DCD (median; IQR) (n=28)	DCD (median; IQR) (n=43)	TD (median; IQR) (n=41)	Significance
Fitness Test Level Reached	2.1; 1.1	2.3; 1.0	2.2; 0.8	3.3; 2.0	$p < 0.0001$*

p = Kruskal Wallis p-value with significance at alpha ≤ 0.05
* = significant difference between groups

Similar to MVPA, a gender effect was found for fitness (p<0.0001). Males were fitter than females in all four groups. No group by gender effect was observed (p=0.31).

When the fitness of children who were meeting exercise guidelines was compared across the groups a significant difference was revealed (p=0.006). Those in the ASD group who were meeting guidelines scored lower on the fitness test than TD children who were also meeting the guidelines (p=0.023). (Note: unadjusted p-values for the TD-ASD&DCD and TD-DCD pairwise comparisons were also significant but did not survive adjustment (p=0.062 and p=0.068, respectively), suggesting the study's power to detect these changes is low due to the complex four group study design). When the sample who were not sufficiently active to meet recommended guidelines was analysed for fitness, a significant between-group difference was noted (p=0.042). However, post-hoc testing did not reveal significant differences in any pairwise comparisons, suggesting that differences did not survive correction for multiple comparisons.

5.3.4 Energy Expenditure

Energy expenditure in kilocalories per minute (Kcals/min) and oxygen consumption per kilogram of body weight (VO_2/kg) were analysed at rest, during gait and during the fitness test. These results are presented in Table 5.7. There were no significant differences between the groups in these variables at rest or during gait. Significant between-group differences were found in VO_2/kg during the exercise test (p=0.004). Post-hoc pairwise comparison revealed that the TD group had higher VO_2/kg values than the ASD&DCD and DCD groups (p=0.027 and p=0.007, respectively). No other combinations of pairs yielded significant results. No gender or age effects were observed (p>0.05).

Table 5.7: Results for Energy Expenditure

Group	ASD (n=21) (mean ± SD)	ASD&DCD (n=24) (mean ± SD)	DCD (n=35) (mean ± SD)	TD (n=41) (mean ± SD)	Significance
EE at Rest (kcals/min)	1.3; 0.4[†]	1.4; 0.4[†]	1.4; 0.5[†]	1.4; 0.4[†]	*p* = 0.36
VO_2 at Rest (ml/min/kg)	8.1 ± 1.8	8.2 ± 3.1	8.2 ± 2.5	9.1 ± 2.7	p = 0.22
EE Gait (kcals/min)	2.1; 1.1[†]	2.7; 0.9[†]	2.6; 0.9[†]	2.2; 0.8[†]	*p* = 0.11
VO_2 Gait (ml/min/kg)	14.1 ± 2.5	15.1 ± 4.7	15.8 ± 4.1	16.1 ± 2.7	p = 0.21
EE Fit Test (kcals/min)	6.1; 4.1[†]	8.0; 3.4[†]	7.9; 5.0[†]	7.6; 5.4[†]	*p* = 0.79
VO_2 Fit Test (ml/min/kg)	45.2; 8.7[†]	43.1; 11.8[†]	42.7; 17.1[†]	50.4; 16.5	*p* = 0.01*

[†]= median; interquartile range (nonparametric data)
p = one-way ANOVA p-value with significance at alpha ≤ 0.05
p = Kruskal Wallis p-value with significance at alpha ≤ 0.05
* = significant difference between groups

5.4 DISCUSSION

This study presented some interesting results. No significant differences in MVPA were found, but the majority of all children were not sufficiently active to meet recommended guidelines. Although no between-group differences in sedentary behaviour were apparent in the current study, all groups spent a large percentage of time in sedentary behaviour – approximately 60% of the time for all children was accounted for by sedentary behaviour. These levels of sedentary behaviour are concerning as high levels of sedentary behaviour have been found to be a risk factor for mortality independent of MVPA (Healy et al., 2016, Ekelund et al., 2006, Song et al., 2015).

The results of this study partially agree with previously published research in this area. Sedentary behaviour was not found to be significantly different between the four

groups which is in agreement with Beutum et al., (2013) but contrasts with Tyler et al., (2014) who found that children with ASD spent more time in sedentary behaviours than TD children. The results of these data were unexpected given the results reported in Chapter 4 (see Section 4.3.4, page 83). These data indicated that children with ASD and those with ASD&DCD spent more time being sedentary than their typically developing peers. That difference was not observed in this case. Like Chapter 4's, this study also observed that the TD and DCD group spent a similar amount of time in sedentary behaviours. There are a number of reasons why the data presented here may differ from those reported in Chapter 4. Data in Chapter 4 were derived from informant based questionnaires, whereas these data were derived from more objective observational data using actigraphs. Informant bias may therefore account for the discrepancies. Additionally, the survey used in Chapter 4, did not account for all sedentary behaviour. Sedentary behaviour in the questionnaire referred to leisure-time sedentary behaviour such as that spent watching television or reading a book, whereas all sedentary behaviour throughout the day, including time sitting in class, was detected by the accelerometer. This may be of greater relevance in the ASD group who have been shown to have increased risks for premature mortality. A recent study highlighted that the average age at death was 18 years younger than the general population (Hirvikoski et al., 2016). Premature mortality in this group was associated with cardiovascular risks, in addition to suicide and epilepsy. These data illustrate that behaviours associated with increased cardiovascular risks (i.e. sedentary behaviours and inactivity) are present at a high level already in young people with ASD. To date there is limited literature regarding morbidity and mortality rates in DCD.

In these data there were no differences between weekly or weekday MVPA in the whole sample or in the individual groups. This concurs with results by Bandini et al., (2013) but contrasts with results of studies by Tyler et al., (2014) and Pan et al., (2016) who found reduced weekly and weekday MVPA in children with ASD. Tyler et al., (2014) had an older sample of children (aged 9-18 years) and Pan et al., (2016)'s sample consisted of adolescents. This may account for the discrepancies in results between those studies and the current study. Analysis of weekend MVPA presented in this study showed results that were in agreement with the analysis of two other studies which have examined this variable (Pan et al., 2016; Bandini et al., 2013). In these data, vigorous PA was significantly reduced in the ASD group compared to the TD group at weekends. Vigorous PA has been shown to be the most beneficial intensity level of activity for overall health and is considered a preventative factor in the development of conditions such as cardiovascular disease and diabetes (Lee and Paffenbarger, 2000, Morris et al., 1980, Tremblay et al., 1994, Trapp et al., 2008, Boutcher, 2010). This finding in relation to vigorous PA is important as previous studies have shown that cardiovascular disease, insulin resistance syndrome and obesity are more prevalent in those with developmental disabilities, such as ASD or DCD, than those without developmental disabilities (Lang et al., 2010, Rimmer et al., 2010, Draheim, 2006, Faught et al., 2005).

Fitness was shown to be significantly reduced in all three groups with diagnoses of neurodevelopmental conditions compared to typically developing controls, which is in agreement with many studies examining this variable (Rivilis et al., 2011, Pan et al., 2016, Schott et al., 2007, Tyler et al., 2014, Cairney et al., 2007, Tsiotra et al., 2009,

Pan, 2014b). These results were therefore expected. The results also showed that children with ASD who were meeting exercise guidelines were less fit than TD children who were also meeting guidelines. The reason for this is unknown and raises questions future research may provide clarity on. One possible reason for this is that the current guidelines for children may not be sufficient for children with neurodevelopmental disorders to increase their cardiorespiratory fitness. Reduced muscular tone has been well documented in children with ASD or DCD and low muscle tone implies lower energy expenditure (Ming et al., 2007, Barnhart et al., 2003). Energy expenditure may therefore be a confounding factor. However, the results of the present study did not find significant differences in energy expenditure at rest or in the oxygen cost of walking in children with ASD or DCD. The differences in VO_2 measured at the point of completion of the fitness test were consistent with differing fitness levels rather than differing energy costs. Few studies have examined energy expenditure in children with ASD or DCD, and no published studies have examined resting metabolic rate in either condition. Therefore, this is an area of interest for future research.

A limitation in this study design was that measuring resting metabolic rate was not possible as fasting would have been necessary to achieve accurate results. Since children with neurodevelopmental disorders, especially ASD, exhibit repetitive and ritualistic behaviour and in some cases deviations from daily routines may cause considerable distress to the child. Therefore for this study, the measurement of resting metabolic rate was considered too disruptive. It is possible that disturbance to mealtime routines could have impacted the child‘s behaviour and willingness to participate in the study (Lotter, 1966, Greaves et al., 2006, Boyd et al., 2012, Boyd et

al., 2010). Furthermore, many families travelled long distances to partake in the studies and it was deemed inappropriate to request that their child fasted for a long period of time prior to exercise.

This study was novel in that it was the first to objectively analyse PA and fitness in children with ASD and comorbid DCD. However, the study design with four independent groups reduced the statistical power to detect significant between-group differences. This may have impacted the results as other research has shown differences in PA between children with ASD or DCD and TD children. Future research with a simpler two group study design and a larger sample comparing PA levels in children with comorbid ASD and DCD to TD children may have the power to detect significant differences or trends.

CHAPTER 6: MOTOR FUNCTION

6.1 INTRODUCTION

As discussed in Chapter 2, children with Autism Spectrum Disorder (ASD) may demonstrate motor stereotypies such as pacing, jumping/hopping, skipping and spinning and it has been suggested that these may be considered restricted and repetitive behaviours. The results of the literature review carried out in Chapter 2 examining studies of gait in children with ASD suggest a tendency to augment walking stability with a wider base of support and decreased range of motion, and research has shown evidence of motor impairment in children with ASD and/or DCD. Motor impairment and movement patterns may have a significant impact on a child's ability and willingness to participate in PA and exercise. The results of Chapter 4 and Chapter 5 (Chapters 4 and 5) have shown that children with ASD and/or DCD are not sufficiently active to meet PA guidelines, and have lower fitness scores than TD peers.

Gait deviations have also been reported in DCD as discussed in Chapter 1 (see Section 1.5.3, page 14). However, no study has yet reported on gait parameters in children with a dual diagnosis of ASD and DCD to the author's knowledge.

The objectives of this study were as follows:

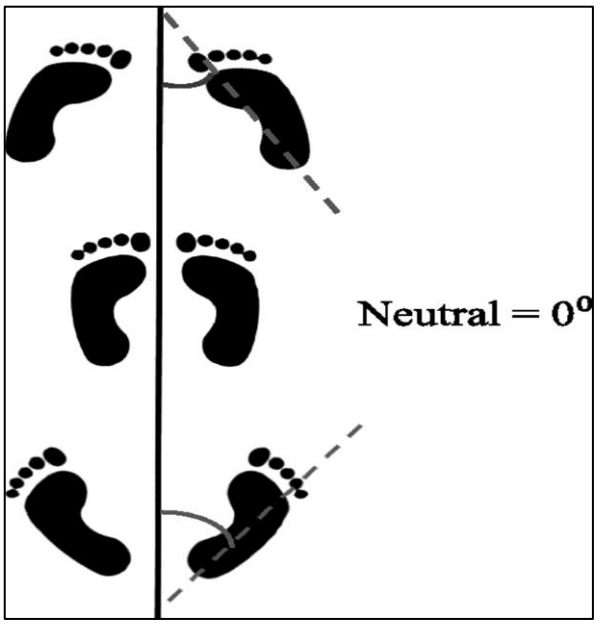
1. To determine temporal-spatial gait parameters in children with ASD, in children with DCD and in children with comorbid ASD and DCD
2. To compare temporal-spatial gait parameters in the above groups to typically developing children

3. To determine sagittal-plane kinematic gait parameters at the hip, knee and ankle in children with ASD, in children with DCD and in in children with comorbid ASD and DCD
4. To compare sagittal-plane kinematic gait parameters at the hip, knee and ankle in the above groups to typically developing children
5. To determine motor coordination in children with ASD, in children with DCD and in children with comorbid ASD and DCD.
6. To compare motor coordination in the above groups to typically developing children
7. To determine the relationship between gait patterns and motor coordination in children with ASD, in children with DCD and in children with comorbid ASD and DCD.

6.2 METHODS

Information on recruitment of participants is given in Chapter 3 (see Section 3.2, page 53) and participant demographics and anthropometric characteristics for this study are presented in Chapter 5 (see Section 5.3.1, page 100). A description of the terminology used to describe gait in this section can be found in Chapter 3 (see Section 3.5.1; page 64). Additionally, the term foot progression angle refers to the angle made by the line of progression of gait (i.e. the direction in which the person is moving) and the long axis of the foot from the heel to the 2nd metatarsal (Cibulka et al., 2016). A visual description of this angle is shown in Figure 6.1. Note: negative value corresponds to lateral deviation (i.e. toe-out) and positive values correspond to medial deviations (i.e. toe-in).

Figure 6.1: Foot Progression Angle



(Carr et al., 2016)

A description of the gait analysis system used in data collection for this study and the protocol for participant preparation for gait analysis is described in Chapter 3 (Section 3.5.1, page 64). All participants walked on a treadmill at their preferred speed. Parents were asked to confirm that the speed at which the child walked on the treadmill was approximately their usual walking speed.

A description of the Movement Assessment Battery for Children, 2nd Edition which was used in this study to assess motor coordination is available in Chapter 3 (see Section 3.5.2, page 71).

Statistical Analysis

Data was imputed into Microsoft Excel throughout data collection and imported into IBM's Statistical Package for the Social Sciences (SPSS) version 22 for statistical analysis. Between-group differences were analysed using one-way ANOVA or Kruskal-Wallis H Tests as appropriate for distributions. Post-hoc tests were carried out on

significant findings and were adjusted for multiple pairwise comparisons using the Holm-Bonferroni method (Holm, 1979). Gait parameters were plotted against coordination results to examine potential trends. Significance level was set as $p<0.05$.

6.3 RESULTS

6.3.1 Participants

As Chapter 5 and Chapter 6 share a cohort, results regarding participant characteristics as presented in Chapter 4 apply to this study (see Section 5.3, page 99). For convenience of the reader, participant characteristics are presented again below in Table 6.1.

Table 6.1: Participant Characteristics

Group	ASD (n=29) (mean ± SD)	ASD&DCD (n=30) (mean ± SD)	DCD (n=41) (mean ± SD)	TD (n=43) (mean ± SD)	Significance
Age (years)	9.42; 3.6†	9.29; 4.0†	9.42; 3.0†	9.42; 4.25†	*p*=0.407
Height (m)	1.33; 0.2†	1.35; 0.2†	1.37; 0.2†	1.35; 0.3†	*p*=0.326
Weight (kg)	30.65; 15.5†	33.85; 17.0†	34.15; 20.5†	29.70; 13.0†	*p*=0.265
BMI (kg/m^2)	17.22; 2.5†	17.53; 6.0†	16.76; 4.4†	16.27; 2.2†	*p*=0.112
BMI Percentile	64; 56†	77; 49†	50; 56†	39; 35†	*p*=0.035*
Body Fat Percentage	21.8; 5.4†	24.1; 11.2†	20.9; 6.4†	20.1; 6.9†	*p*=0.067
IQ	101 ± 15.5	99 ± 15.7	102 ± 13.1	116 ± 13.2	p<0.0001*
ADOS-2 Comparison Score	7.0 ± 2.3	7.5 ± 2.0	N/A	N/A	p=0.233

† = median; interquartile range (nonparametric data)
p = Independent Samples T-test p-value with significance at alpha ≤ 0.05.
p = Kruskal Wallis p-value with significance at alpha ≤ 0.05.
* = significant difference between groups.

6.3.2 Gait Analysis

The results presented in this section represent the analysis of gait parameters at the self-selected treadmill velocity of participants with ASD, ASD&DCD, DCD and typically developing controls. Gait analysis was not carried out on 10 participants as the application of gait wands and markers caused sensory or behavioural distress. The numbers of children in each group who completed gait analysis are as follows: ASD n=23; ASD&DCD n=28; DCD n= 39; TD n=43.

Temporal-spatial Parameters

Temporal-spatial parameters analysed were step width, step length, stride length, stride time, stance percent time, cadence and velocity. Mean and standard deviation for these parameters are presented in Table 6.2 (Note: median and interquartile range (IQR) are presented for nonparametric data). Significant between-group differences were noted in step width, step length and stride length. Post-hoc pairwise comparisons revealed that step width was significantly larger in the ASD&DCD group compared to controls ($p = 0.05$). No significant differences were found in any other pair combinations for step width. Step length was significantly shorter in the ASD&DCD group compared to TD controls ($p = 0.049$). No significant differences were found in any other pairwise comparisons for step length.

Table 6.2: Results for Temporal-spatial Parameters

Event	ASD (n=23) (mean ± SD)	ASD&DCD (n=28) (mean ± SD)	DCD (n=39) (mean ± SD)	TD (n=43) (mean ± SD)	Significance
Step Width (mm)	133 ± 27	139 ± 36	120 ± 31	119 ± 26	p = 0.025*
Step Length (m)	0.36; 0.11†	0.35; 0.10†	0.38; 0.13†	0.40; 0.09†	*p* = 0.038*
Stride Length (m)	0.76; 0.24†	0.76; 0.23†	0.84; 0.26†	0.86; 0.22†	*p* = 0.05*
Stride Time (s)	1.15; 0.16†	1.19; 0.24†	1.22; 0.16†	1.23; 0.17†	*p* = 0.138
Stance Percent Time (%)	67.63 ± 1.74	68.29 ± 2.33	67.73 ± 2.00	67.22 ± 1.75	p = 0.175
Cadence (steps/min)	214 ± 21	207 ± 28	201 ± 23	202 ± 27	p = 0.23
Velocity (m/s)	0.61; 0.21†	0.56; 0.19†	0.61; 0.24†	0.67; 0.20†	*p* = 0.123

† = median; interquartile range (nonparametric data)
p = one-way ANOVA p-value with significance at alpha ≤ 0.05.
p = Kruskal Wallis p-value with significance at alpha ≤ 0.05.
* = significant difference between groups.

Sagittal Plane Kinematics

Hip, knee and ankle joint angles in the sagittal plane were calculated at six points in the gait cycle: initial contact, opposite initial contact, mid-stance, opposite mid-stance, toe-off, opposite toe-off. The range of motion exhibited during the gait cycle by hip, knee and ankle joints were also calculated.

No significant differences were found between the groups in hip joint angles at any of the six time points or in hip range of motion during the gait cycle. These results are summarised in Table 6.3. P-values approaching significance (<0.1) were noted for hip angle at initial contact and hip angle at toe-off.

Table 6.3: Results for Hip Flexion-Extension (+/-) (all expressed in degrees)

Event	ASD (n=23) (mean ± SD)	ASD&DCD (n=28) (mean ± SD)	DCD (n=39) (mean ± SD)	TD (n=43) (mean ± SD)	Significance
Initial Contact	30; 10[†]	35; 12[†]	32; 14[†]	32; 9[†]	*p* = 0.09
Opposite Initial Contact	1; 13[†]	4; 14[†]	1; 11[†]	-3; 11[†]	*p* = 0.156
Mid-Stance	10; 13[†]	15; 12[†]	10; 11[†]	7; 11[†]	*p* = 0.138
Opposite Mid-Stance	28 ± 8	32 ± 8	30 ± 8	28 ± 6	p = 0.152
Toe-off	7; 11[†]	10; 12[†]	7; 12[†]	4; 11[†]	*p* = 0.073
Opposite Toe-off	19; 15[†]	24; 11[†]	20; 10[†]	21; 11[†]	*p* = 0.153
Range	38 ± 5	40 ± 8	41 ± 6	41 ± 7	p = 0.489

[†]= median; interquartile range (nonparametric data)
p = one-way ANOVA p-value with significance at alpha ≤ 0.05.
p = Kruskal Wallis p-value with significance at alpha ≤ 0.05.
* = significant difference between groups.

Significant differences in knee angles at mid-stance and toe-off were determined between the groups. Post-hoc pairwise comparisons revealed that the ASD&DCD group were significantly more flexed at the knee at mid-stance than TD controls (p = 0.047). No significant differences were found in any other pairwise comparisons for knee angle at mid-stance.

The ASD&DCD group also exhibited significantly more flexion at the knee at toe-off than TD controls (p = 0.036). No significant differences were found in any other pairwise comparisons for knee angle at toe-off. Kinematic findings at the knee are presented in Table 6.4. P-values approaching significance (<0.1) were noted for knee angle at initial contact and knee angle opposite initial contact.

Table 6.4: Results for Knee Flexion-Extension (+/-) (all expressed in degrees)

Event	ASD (n=23) (mean ± SD)	ASD&DCD (n=28) (mean ± SD)	DCD (n=39) (mean ± SD)	TD (n=43) (mean ± SD)	Significance
Initial Contact	14 ± 8	16 ± 10	14 ± 9	11 ± 9	p = 0.098
Opposite Initial Contact	12 ± 5	11 ± 5	11 ± 5	9 ± 4	p = 0.076
Mid-Stance	15; 7†	14; 7†	12; 9†	10; 7†	*p* = 0.023*
Opposite Mid-Stance	51; 10†	49; 11†	47; 11†	47; 8†	*p* = 0.544
Toe-off	41; 6†	42; 6†	40; 7†	40; 7†	*p* = 0.036*
Opposite Toe-off	18 ± 7	20 ± 8	18 ± 8	16 ± 8	p = 0.281
Range	53 ± 4	51 ± 5	54 ± 6	54 ± 6	p = 0.154

† = median; interquartile range (nonparametric data)
p = one-way ANOVA p-value with significance at alpha ≤ 0.05.
p = Kruskal Wallis p-value with significance at alpha ≤ 0.05.
* = significant difference between groups.

No significant differences were found between the groups in ankle joint angles at any of the six time points or in hip range of motion during the gait cycle. A non-significant trend was noted for ankle angle at toe-off ($p<0.1$). These results are presented in Table 6.5.

Table 6.5: Results for Ankle Dorsiflexion-Plantarflexion (+/-) (all expressed in degrees)

Event	ASD (n=23) (mean ± SD)	ASD&DCD (n=28) (mean ± SD)	DCD (n=39) (mean ± SD)	TD (n=43) (mean ± SD)	Significance
Initial Contact	0; 5†	1; 5†	-1; 8†	0; 7†	*p* = 0.736
Opposite Initial Contact	4; 7†	5; 4†	6; 6†	7; 6†	*p* = 0.847
Mid-Stance	9 ± 4	10 ± 5	9 ± 5	10 ± 4	p = 0.608
Opposite Mid-Stance	7; 7†	7; 5†	7; 6†	8; 5†	*p* = 0.808
Toe-off	-8; 14†	-5; 12†	-7; 14†	-12; 10†	*p* = 0.06
Opposite Toe-off	-3; 8†	0; 9†	-1; 9†	-2; 9†	*p* = 0.649
Range	40; 8†	40; 10†	40; 7†	41; 10†	*p* = 0.307

† = median; interquartile range (nonparametric data)
p = one-way ANOVA p-value with significance at alpha ≤ 0.05.
p = Kruskal Wallis p-value with significance at alpha ≤ 0.05.
* = significant difference between groups.

Foot progression angle was also analysed and significant between-group differences were observed (see Table 6.6). Post hoc tests revealed that the ASD&DCD group had a larger outward foot progression angle than the TD group (p=0.038). No other pairwise comparisons revealed significant differences.

Table 6.6: Results for Foot Progression Angle (+ implies toe-in; - implies toe-out) (degrees)

Mid-stance	ASD (n=23) (Median; IQR)	ASD&DCD (n=28) (Median; IQR)	DCD (n=39) (Median; IQR)	TD (n=43) (Median; IQR)	Significance
Foot Progression Angle	-12; 13	-17; 9	-17; 10	-13; 7	*p* = 0.01

p = one-way ANOVA p-value with significance at alpha ≤ 0.05.
p = Kruskal Wallis p-value with significance at alpha ≤ 0.05.
* = significant difference between groups.

6.3.3 Motor Coordination

One child in the ASD&DCD group refused to complete the Movement Assessment Battery for Children, 2nd Edition. All other children completed the assessment. To allow for the overall sample to be analysed together, comparable percentile scores for the Manual Dexterity, Aiming & Catching and Balance categories and the total test percentile score were analysed. A Kruskal Wallis H Test revealed significant between-group differences in the Manual Dexterity category ($p<0.0005$). Post-hoc pairwise comparisons with adjusted p-values revealed that the TD group scored significantly higher than the ASD, ASD&DCD and DCD groups ($p<0.0005$ for all three comparisons). There were no significant differences between the ASD, ASD&DCD and DCD groups ($p>0.05$).

Further significant between-group differences were observed in the Aiming & Catching category ($p<0.0005$). Post-hoc pairwise comparisons with adjusted p-values revealed that the TD group scored significantly higher than the ASD, ASD&DCD and DCD groups ($p<0.0005$ for all three comparisons). There were no significant differences between the ASD, ASD&DCD and DCD groups ($p>0.05$).

Significant between-group differences were also found for MABC-2 overall scores ($p<0.0005$). Post-hoc pairwise comparisons with adjusted p-values revealed that the TD group scored significantly higher than the ASD, ASD&DCD and DCD groups ($p<0.0005$ for all three comparisons). There were no significant differences between the ASD, ASD&DCD and DCD groups ($p>0.05$). Medians and interquartile ranges for Movement ABC-2 percentile scores are presented in Table 6.7.

Table 6.7: Results for Movement Assessment Battery for Children, 2nd Edition.

Category	ASD (n=29) (Median; IQR)	ASD&DCD (n=29) (Median; IQR)	DCD (n=41) (Median; IQR)	TD (n=43) (Median; IQR)	Significance
Manual Dexterity	2; 20†	1; 7†	2; 4†	37; 49†	$p < 0.0001$*
Aiming & Catching	9; 24	2; 8	9; 29	56.5; 59	$p < 0.0001$*
Balance	5; 31	1; 9	2; 9	37; 26	$p < 0.0001$*
Total Test Scores	5; 9	1; 2	1; 5	37; 38	$p < 0.0001$*

p = Kruskal Wallis p-value with significance at alpha ≤ 0.05.
* = significant difference between groups.

6.3.4 Relationship Between Gait Parameters and Motor Coordination

In order to investigate the relationship between gait and coordination, gait parameters which were found to be statistically significant (step width, step length, knee angle at mid-stance and knee angle at toe-off) were plotted against motor coordination test results in the whole sample and also by group. No monotonic relationships were identified from the scatter plots (see examples in Appendix X). Therefore, no correlations were run on these data under advisement from a statistician.

6.3.5 Relationship Between Motor Function and ASD Severity/IQ

ADOS-2 comparison scores were plotted against Movement ABC-2 scores to assess the relationship between ASD severity and motor impairment (see Figure 6.2). No monotonic relationship was apparent. Therefore Spearman's rho (r_s) was not calculated. Movement ABC-2 scores were also plotted against IQ (see Figure 6.3). A positive monotonic relationship was observed (i.e. as with increasing IQ, Movement ABC-2 scores increased). Spearman's rho was calculated as r_s = 0.59 indicating a moderate positive correlation between IQ and motor coordination.

Figure 6.2: ADOS-2 Comparison Score Plotted against Movement ABC-2 Score (ASD and ASD&DCD Groups)

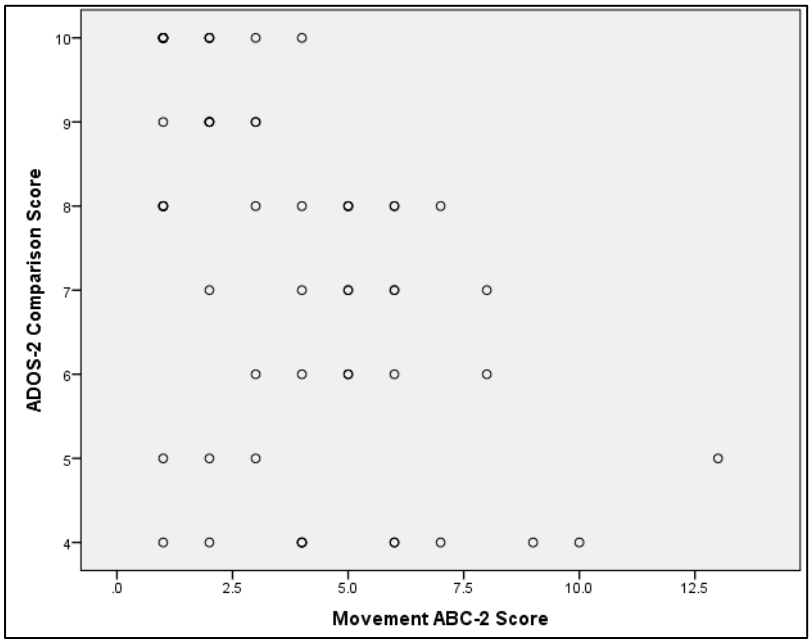
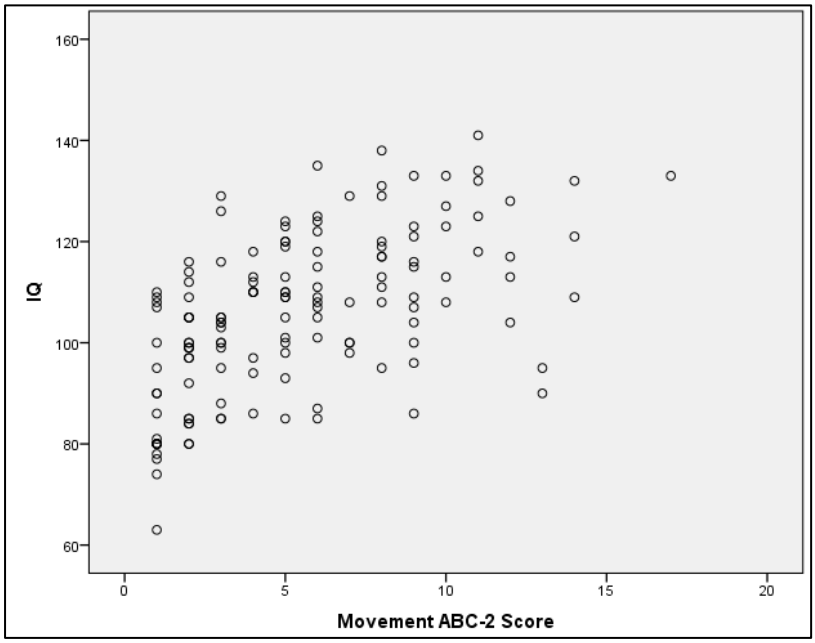


Figure 6.3: IQ Score Plotted against Movement ABC-2 Score (All Groups)



(r_s = 0.59)

6.3.4 Correlations with Physical Activity and Fitness Levels Reported in Chapter 5

In order to investigate the relationship between PA levels, fitness and motor function, daily MVPA, daily sedentary behaviour and fitness scores were plotted against motor

coordination test results and against gait parameters in the whole sample and also by group. No monotonic relationships were identified from the scatter plots (see examples in Appendix X). As before, no correlations were run on these data under advisement from a statistician.

6.4: DISCUSSION

The main aim of this study was to describe gait patterns and motor ability in children with ASD and/or DCD. The results have shown that differences exist in gait patterns in children with ASD and DCD when compared to typically developing peers.

Step width has been found to be increased in children with ASD and in children with DCD in published literature by Lim et al., (2016), Nobile et al., (2011), Shetreat-Klein et al., (2012), Wilmut et al., (2016). Although the differences in step-width were not significant in the ASD-TD and DCD-TD comparisons in this study, the ASD&DCD group was found to have a significantly wider step width which is consistent with the above-mentioned studies. Between-group differences in both step length and stride length were both found to be significant, however post-hoc tests for stride length failed to produce significant results in pairwise comparisons. Step length was found to be significantly reduced in the ASD&DCD group compared to TD controls. This is consistent with studies which have examined step length in children with ASD including Weiss et al., (2013), Nobile et al., (2011) and Vernazza-Martin et al., (2005). It is not clear whether comorbid DCD was controlled for in these studies as in this study. These two parameters combined suggest a tendency for children with ASD and comorbid DCD to augment their walking stability, perhaps due to poor core strength and stability and poor balance. This is further evident from increased flexion at the

knee at mid stance which helps lower the centre of gravity and further augment walking stability. This trend has been highlighted in studies examining gait in children with ASD or DCD, accompanied with a reduced cadence compared to TD controls (Nobile et al., 2011, Morrison et al., 2013). However, this study did not find a significant difference in cadence between the groups.

Foot progression angle was found to be significantly larger in children with ASD and comorbid DCD than the typically developing controls (i.e. children with ASD and DCD tended to have a larger toe-out angle). This angle has been studied in children with ASD previously and no significant differences were found (Nobile et al., 2011). This discrepancy is possibly due to comorbid DCD which was not studied by Nobile et al., (2011). Excessive out-toeing has been shown to be associated with increased medial foot pressure which may result in over-pronation at the subtalar joint (i.e. flat footedness) and subsequent painful conditions such as plantar fasciitis and knee pain (Cibulka et al., 2016). Therefore, this deviation may have a restrictive impact on physical activity.

Differences in some gait parameters, such as velocity or cadence, in children with ASD or DCD when compared with TD controls were not apparent in this study although they have been reported in other studies. This is perhaps in part due the four-group study design which is less powerful and requires correction for multiple comparisons. A more conservative design was implemented in this study in an attempt to probe the role of DCD-ASD comorbidity more deeply. This study may have been underpowered to detect these differences and future studies may seek to investigate larger samples to resolve the contradictions between studies.

The ASD, ASD&DCD and DCD groups all showed significant motor impairment in the Movement ABC-2. The ASD&DCD groups, however, scored consistently the lowest implying more severe impairment, although the pairwise comparisons of this group with the DCD group or the ASD group did not yield significant results. This impairment may explain the differences observed in gait parameters in the dual-diagnosis group compared to typically developing controls that were not apparent in the ASD or DCD groups.

Correlation between severity of motor impairment and abnormal gait patterns were expected in this study, however no linear relationships were observed. This may be due to relatively small sample sizes. Another explanation for this may be that the gait differences detected were relatively subtle while the motor impairment differences observed were stark so a relationship between these variables may be quite difficult to detect. Furthermore, this phenomenon suggests that there may be other explanatory factors, such as differences in cerebellar development as suggested in the literature review (see Chapter 2). This has not been directly assessed in this study and would be an area of interest for future research, with an aim to determine the relationship between gait, motor function and brain structure and function in children with ASD and comorbid DCD using neuroimaging.

There were a number of strengths in the current study. The study aimed to investigate the specific role of comorbid ASD and DCD on gait and motor functioning, which has not been studied to date. This was the basis for the four diagnostic group study design. Unfortunately this was also a limitation as discussed previously due to the impact on power. A further strength was the inclusion only of subjects not on current treatment

with any psychoactive medications. Psychoactive medications, particularly anti-psychotic medications, are commonly used in ASD for management of irritability and associated behavioural challenges. These are active in the basal ganglia and extrapyramidal parkinsonian-like side effects may impact patterns and smoothness of movement and may have confounded the data. Although sample size was on par with published research in the area, a further limitation of this study was a relatively small sample size, particularly in the context of the complex four group study design.

A further limitation of this study is that it was necessary to perform gait analysis using a treadmill due to reflection interference issues in the gait laboratory with the optical based Codamotion system. Treadmill gait has been shown to be qualitatively and quantitatively similar to overground gait, with similar gait mechanics in both methods (Riley et al., 2007). Furthermore, although the children were given time to acclimatise to the treadmill, many of them had not been on a treadmill before and many reported anxiety while others reported excitement. Although little is known about the impact different emotional states have on gait patterns, it has been suggested that individuals' gait patterns may change with changes in their emotions (Janssen et al., 2008).

The motor impairments and altered movement patterns suggested by the findings of this study are consistent with other studies done in the area, although this study is novel in that the differences were observed between TD children and children with comorbid ASD and DCD. The severe motor impairment in children with ASD and comorbid DCD was expected but had not been documented previously. There is a tendency to augment walking stability, which is perhaps due to balance deficits. These

patterns may impact on the PA of children with these neurodevelopmental disorders. As these disorders commonly co-occur with other neurodevelopmental disorders, such as ADHD or SPD, identifying barriers of and benefits and motivators for PA may be key to encouraging more participation and activity in these children.

CHAPTER 7: BARRIERS AND BENEFITS OF EXERCISE IN CHILDREN WITH NEURODEVELOPMENTAL DISORDERS

7.1: INTRODUCTION

Children with neurodevelopmental disorders are more likely to be overweight or obese than typically developing peers, have altered movement patterns and difficulty with social communication and language skills. These factors suggest that children with neurodevelopmental disorders may face greater challenges and barriers to physical activity (PA) and participation than typically developing children. Children with neurodevelopmental disorders have been shown to be less physically active than peers, and this may have implications on health which then become additional barriers to PA.

Few studies have examined the barriers to and motivators for exercise in children with ASD and other neurodevelopmental disorders. One study reported the main barriers to PA in children with special needs to be the child's lack of interest, the scarcity of PA programmes appropriate for children with special needs, behavioural challenges and parents' lack of time (Yazdani et al., 2013). Another study examined barriers to PA and the relationship to PA and screen time in children specifically with ASD (Must et al., 2015). They reported that parents of children with ASD reported significantly more barriers to PA than parents of typically developing children, and that the number of reported barriers to PA was inversely correlated with the hours spent in PA per year. The more barriers reported by parents, the fewer the hours spent in PA by their children.

With low levels of PA reported in children with ASD and other neurodevelopmental disorders, as shown in previous chapters, it is becoming clear that there is a need for exercise intervention in this group. Therefore, it is essential to have an understanding of the barriers to exercise faced by these children as well as the benefits and motivators in order to allow appropriate treatment planning and exercise prescription.

7.1.1 Aims and Objectives

The aim of this study was to qualitatively describe barriers, benefits and motivators associated with children with neurodevelopmental disorders. The objectives were as follows:

1. To determine perspectives on benefits, motivators and barriers to exercise in children with neurodevelopmental disorders of:
    a. Professionals working with children diagnosed with neurodevelopmental disorders
    b. Parents of children diagnosed with neurodevelopmental disorders
2. To describe emerging themes and concepts
3. To make recommendations for clinical practice based on the results

7.2: METHODS

A qualitative descriptive design was chosen for this study as very little is known about benefits of and barriers to exercise in children with neurodevelopmental disorders, both in Ireland and throughout the world. As many children with neurodevelopmental disorders, particularly those with ASD have difficulty with social interaction and communication, it was deemed beyond the skill set of the author to conduct interviews or focus groups directly with these children. To get an overall view of factors affecting physical activity participation in children with neurodevelopmental

disorder, the target population was professionals who work with these children and also parents of children with these conditions.

7.2.1 Recruitment

The study sample was comprised of those who met the following inclusion criteria:

- Professionals who work specifically with children with neurodevelopmental disorders.

  or

- Parents of children aged six to 18 years with neurodevelopmental disorders without comorbid non-neurodevelopmental conditions which would restrict the child's ability to exercise (e.g. cardiovascular disease, poorly-controlled asthma or neurological conditions such as cerebral palsy).

Potential participants were contacted by the TCD Autism Research Group via the central email address 'autism@tcd.ie'. Ana McLaughlin, who is a member of the Autism Research Group, acted as a gatekeeper. Contact details of administrative officers and clinical managers from private practice physiotherapy and occupational therapy clinics, HSE Assessment and Intervention teams and special needs services (e.g. Enable Ireland) were obtained in a web search. Emails were then sent inviting staff members to the focus groups with the information leaflet attached. Parents of child participants from past research studies undertaken by the research group who had given consent to be contacted about further research were also invited. Those whom expressed an interest in participating were contacted directly by the lead investigator by phone. During this phone call potential participants were screened for inclusion and exclusion criteria and given the opportunity to ask any questions they

may have had. Those who met inclusion criteria above who were available and willing to attend the Trinity Centre for Health Sciences on one evening were recruited into the study and were emailed directions and the relevant focus group agenda by the lead investigator. Recruitment materials and consent forms are presented in Appendix XI.

7.2.2 Data Collection

Five focus groups (A-E) were conducted in March/April 2016 by the lead investigator. Each one lasted approximately one hour. Group A was comprised of eight multi-disciplinary professionals who specialise in treatment of children with neurodevelopmental disorders – one physiotherapist, one occupational therapist, two speech and language therapists, two psychologists, one psychiatrist and one personal trainer. Groups B-E consisted of between eight and ten parents of children with neurodevelopmental disorders who had participated in previous research studies with the TCD Autism Research Group. Participants were given I.D. code numbers beginning with the letter of the group they took part in for anonymity and these codes are used throughout this chapter when referring to quotes (for example, quotes by professionals will be labelled A1-A8).

The location of the focus groups was room 1.54A in the Trinity Centre for Health Sciences. The classroom was arranged with chairs in a circle to encourage discussion between participants. A registration period of 30 minutes was scheduled prior to each focus group. During this time refreshments were served and written informed consent was obtained from each participant. The lead investigator introduced herself to each participant and they were encouraged to ask any questions they had. The rationale behind the focus groups and an overview of the lead investigator's qualifications and research interests were explained to each group. The participants were encouraged to

talk amongst themselves so a rapport within the group could be developed prior to commencement. No non-participants were present during the focus groups and no written notes were taken during the focus groups.

The main questions for the focus groups were as follows:

1. What might discourage or prevent children with neurodevelopmental disorders from being physically active? Please give examples from your own experiences with children.
2. What might encourage/motivate children with neurodevelopmental disorders to be physically active? Please give examples from your own experiences with children.
3. What benefits might children with neurodevelopmental disorders get from being physically active? Please give examples from your own experiences with children.

Warm-up exercises including general questions about physical activity and exercise and the participants' own experiences with regard to barriers, benefits and motivators were used to get the participants thinking about the subject matter. An agenda for the focus groups are presented in Appendix XII.

7.2.3 Data Analysis

All focus groups were recorded anonymously using a dictaphone (*Sony IC Recorder Model ICD-P520*). The recordings were transcribed verbatim and a thematic analysis was carried out as recommended by Braun and Clarke (2006). A codebook was developed for coding the focus group transcriptions. Codes and themes were derived from the data. The researchers independently read the transcriptions several times

and coded the data line-by-line to identify emerging concepts. The lead investigator coded all five transcriptions and two transcriptions were coded by Emer McGowan, a fellow PhD candidate in the Physiotherapy department who was trained in qualitative research methods and had carried out several qualitative research studies. The lead investigator's experience in qualitative research consisted of attending a seminar on qualitative research methods in 2013. There was high agreement between the two coders with the first codebook, and full agreement was reached with the addition of three new codes and minor changes to five original codes. Data saturation was reached (i.e. no new codes or themes were identified from the transcription of focus group E). Please see Appendix XII for the final version of the codebook.

Trustworthiness

Two main strategies were employed to optimise the trustworthiness of the data. Upon completion of the initial analysis, the list of themes generated was sent to five randomly selected participants to verify the interpretation of the data. This process is known as member checking. Two researchers independently coding and interpreting the data provided enhanced data credibility, a platform for reflection and discussion and, therefore, a more thorough understanding.

Throughout the results section which follows, quotes taken from the focus group transcripts which were representative of commonly reported themes are given in support of findings, and to give the reader insight into the perspectives of focus group participants.

7.3: RESULTS

Forty three individuals took part in the focus groups – eight professionals and 35 parents. The majority of participants (72%) were female. Participant characteristics for each focus group are presented in Table 7.1. All participants were Irish and currently living in Dublin. All professionals and 28 parents were in formal employment at the time of the focus groups. Each of the professionals had at least three years' experience working with children with neurodevelopmental disorders. Seven parental focus group participants were stay-at-home parents.

Table 7.1: Focus Group Participant Characteristics

Group	n (female: male)	Age (mean ± SD)	Age Range
Professionals (A)	8 (7:1)	35.25 ± 8.3	26-47
Parents (B)	8 (6:2)	43.38 ± 4.4	37-50
Parents (C)	9 (6:3)	40.67 ± 3.1	36-46
Parents (D)	8 (6:2)	42.25 ± 3.7	35-46
Parents (E)	10 (6:4)	38.40 ± 3.1	35-43

All participants had formal education at least up to the age of 18 years. Each of the professionals had gone on to higher or further education, as had 25 of the parents. All the parents had at least one child aged between six and 14 years with a diagnosis of one or more neurodevelopmental disorders (although 18 years of age was used as the upper limit for the child's age in the inclusion criteria, none of the participants had children with neurodevelopmental disorders over the age of 14). Characteristics of the children of parental focus groups participants are presented in Table 7.2.

Table 7.2: Characteristics of Children of Parental Focus Groups Participants

Group	Age Range of Children (years)	Gender of Children (F:M)	Diagnosis of Children
Parents (B)*	6-12	6:3	ASD (n=4) DCD (n=3) ASD, SPD, ID (n=1) DCD, ADHD, SPD (n=1)
Parents (C)	6-14	6:3	ASD (n=3) ASD, DCD (n=3) ASD, ADHD (n=1) ASD, DCD, SPD (n=1) DCD, Dyslexia, Hypermobility (n=1)
Parents (D)	6-14	7:1	ASD (n=1) DCD (n=3) ASD, DCD (n=2) ASD, DCD, Dyslexia (n=1) ASD, DCD, SPD (n=1)
Parents (E)*	6-14	7:4	ASD (n=4) DCD (n=6) ASD, ADHD, SPD (n=1)

Abbreviations: ASD Autism Spectrum Disorders; DCD Developmental Coordination Disorder; SPD Sensory Processing Disorder; ADHD Attention Deficit Hyperactivity Disorder

*denotes focus group with participant with more than one child with neurodevelopmental diagnoses

The majority of participants were either unaware that guidelines for childhood PA existed or were aware that guidelines existed but did not know what they were. Interestingly, none of the professionals' focus group participants could state the correct recommended guidelines.

Barriers to and benefits/motivators for PA reported in the focus groups are listed in Table 7.3 in four levels of influence: intrinsic (i.e. intrapersonal), environmental, education and perceptions of others, and family. Factors within each level are listed in order from the most reported factors to the least reported factors.

Table 7.3: Overview of the Barriers to and Benefits/motivators for PA in Children with Neurodevelopmental Disorders as Reported by Parents (1) and Professionals (2)

Barriers	Benefits/Motivators
Intrinsic Factors	
Self-consciousness/confidence (1, 2)	Mood regulation (1, 2)
Sensory issues (1)	Reduction of anxiety/stress (1)
Physical deficits (1, 2)	Fun/enjoyment (1)
Lack of interest (1)	Sensory regulation (1, 2)
Difficulty with social interaction (1, 2)	Physical function (1, 2)
Anxiety (1, 2)	Social engagement (1, 2)
Child's awareness of their diagnosis (1, 2)	Sleep improvement (1)
	Sense of achievement (1)
	Weight management/appetite improvement (1)
	Improvement in attention/concentration (1, 2)
	Improvement in self-confidence (1)
Environmental Factors	
The competitive nature of sport (1)	Supportive environment (1, 2)
Lack of exposure (1, 2)	Availability of mixed-ability clubs/teams (1, 2)
Lack of resources (1, 2)	Small group sizes (1)
Screen-time (1,2)	Exposure/widening of interests (2)
Exclusion (1, 2)	
Attitudes of others (1, 2)	
Weather (1, 2)	
Education and Perceptions of Others	
Perceptions and awareness re: conditions (outside family) (1, 2)	Education of coaches/teachers (1, 2)
Lack of prioritisation of PA in school (1, 2)	Appropriate instructions (1)
PA education of teachers/coaches (1, 2)	
Family Factors	
Family routine (1, 2)	Encouragement by family members (1)
Academic prioritisation (1, 2)	Exercising as a family (1)
Parent perceptions about PA (2)	
Parent perceptions about disability (2)	
Cost (1)	

Abbreviations: PA: physical activity

Intrinsic Factors

The most common intrinsic barrier reported was self-consciousness with both professionals and parents reporting that children are less likely to participate in PA if they feel they are being compared to other children who may have a higher skill level:

> "*The awareness of not being good at something as comes with age can be a huge barrier to PA in these children. I've seen it in clinics. If they fail at a movement task they're reluctant to try it again*" (Participant A2, Professionals' Focus Group, 02/03/2016)

> "*My child worries what people think of him, feelings of being worse than peers. He's quite self-conscious*." (Participant D7, Parents' Focus Group, 30/03/2016)

The next most commonly reported intrinsic factors were sensory issues and other physical deficits (e.g. poor motor coordination, fatiguing easily), followed by lack of interest:

> "*The echoes in that environment [indoor sports halls] can be so loud. The sensation is too much. And the sensation of outfits, he's picky about the clothes he can wear*." (Participant B2, Parents' Focus Group, 09/03/2016)

> "*My daughter gets so tired very easily. She also suffers from pains in her joints. Possibly sensory-related like some of the others*." (Participant C4, Parents' Focus Group, 23/03/2016)

> "*No interest – he just dislikes exercise. No energy for it all. He's also very clumsy and I think then because he's not good at it he doesn't want to [participate]*." (Participant C8, Parents' Focus Group, 23/03/2016)

The most commonly reported intrinsic benefits/motivators were mood regulation and a reduction in anxiety and stress. Mood regulation was reported by one professional

and by many of the parents. PA was reported to reduce the anxiety and stress faced by their children by parent participants.

> "*trying to get that arousal level [necessary for productive speech and language therapy] and because you've got the low arousal or the high arousal and you need two different types of movements – you either need the really calming movements or the alerting ones*." (Participant A1, Professionals' Focus Group, 02/03/2016)

> "*He's never happier than when he's cycling, he really enjoys the freedom of it. He also really enjoys the group aspect of his playfit class which is for all ages and abilities*." (Participant C8, Parents' Focus Group, 23/03/2016)

> "*Joy…[being physically active] brings real happiness. Also provides an outlet for social interactions, and a way to get frustrations out and relieve some of that stress*." (Participant E3, Parents' Focus Group, 06/04/2016)

Environmental factors

The competitive nature of sport was reported as a barrier to PA by 40% of parents, and lack of exposure/resources was the next most commonly reported barrier reported by the majority of professionals and was also reported by parents.

> "*I think teachers can be difficult. Sometimes they can approach it as somebody who's teaching the sport is good at the sport and is teaching everybody to be good at a sport and doesn't understand what it might be like to be unable to catch a ball or not be good at the basic function for the sport, which is difficult for a child who is differently abled. When it's competitive, the child with different ability will be the last picked. Little things, pick your teams. You lose, you won. There's a hierarchy and the same two people are always last*." (Participant B1, Parents' Focus Group, 09/03/2016)

"*Sports by their very nature are competitive and we have had very negative experiences in mainstream clubs except where coaches have had specific training and in those clubs our children can become more involved and have a better experience*." (Participant C9, Parents' Focus Group, 23/03/2016)

"*One of the biggest things parents say to me that there's nothing out there and they come to me saying I never knew this existed so I think there's a severe lack of opportunity for them and it needs to be addressed. I've had a lot of parents saying to me "I tried him in the local club and it just didn't work out.""*
(Participant A4, Professionals' Focus Group, 02/03/2016)

"*I wish there were team sports and activities catering for ASD diagnosed children. We're currently exploring this but there is little available*." (Participant E4, Parents' Focus Group, 06/04/2016)

Environmental motivators included the availability of a supportive environment for exercise run by individuals or groups familiar with neurodevelopmental conditions and teams/clubs which welcomed children with mixed-abilities and did not prioritise talent over participation.

"*Sports by their very nature are competitive and we have had very negative experiences in mainstream clubs except where coaches have had specific training and in those clubs our children can become more involved and have a better experience*."
(Participant C9, Parents' Focus Group, 23/03/2016)

"My girls play soccer for a main stream club who have a FAI Football For All Club Programme. It is about participation rather than competitiveness. It's brilliant!"
(Participant E9, Parents' Focus Group, 06/04/2016

"*I feel like if there was a wider range of activities that were aimed at a mixed ability group instead of competitive environments. Or even ones specifically for children with these conditions. I think small group sizes really work too*."

(Participant B5, Parents' Focus Group, 09/03/2016)

Education and perceptions of others

Negative perceptions and poor awareness of the conditions was reported as a barrier whereas education of teachers/coaches about the conditions and the positive effects of exercise was a motivator.

"*And all you want them to do is get involved in physical activity so, I think a lot of the clubs are mainstream, and it's not their fault, they don't have the resources but you know, I think maybe if they try to become a little bit more educated. If they got maybe somebody in the clubs to say we're going to train this guy up to learn more about kids with needs then maybe we could start something but there is none*."

(Participant A4, Professionals' Focus Group, 02/03/2016)

"*Definitely. I think the instructions can be confusing too. If more coaches etc. were a bit more aware of what ASD even is and what works for the kids it would help. Like picture charts and letting him know what's happening next. That can ease stress and anxiety about it too*." (Participant B6, Parents' Focus Group, 09/03/2016)

"*Being in an inclusive environment with coaches experienced in DCD & coordination difficulties so she doesn't feel left behind would make such a difference*."

(Participant E2, Parents' Focus Group, 06/04/2016)

Family Factors

Family routine and the subsequent lack of time were reported to be the biggest barriers on the family level. Encouragement by family members and exercising as a family was reported to be a motivator by parents.

> "*if you're doing sport or anything extra-curricular it's exactly that so again if you don't have a family who are going to bring you or it's not convenient or whatever, you're not going to do it*."
> (Participant A3, Professionals' Focus Group, 02/03/2016)

> "*I find that too, my husband works shift so time is a definite barrier. And money, some of the sports are so expensive. I mean we find one that benefits her and helps her progress physically and socially but then we can't afford to keep it up or don't have the time to bring her. Heartbreaking! I wish there were more free clubs I get that they cost a lot to run*."
> (Participant B3, Parents' Focus Group, 09/03/2016)

7.4: DISCUSSION

Few studies have examined barriers to and benefits/motivators for PA in children with neurodevelopmental disorders and this study was the first to do so in an Irish population. Globally, several studies have determined barriers and motivators for PA in children with ASD or DCD using questionnaires or parental interviews (Stanish et al., 2015, Must et al., 2015, Obrusnikova and Miccinello, 2012, Obrusnikova and Cavalier, 2011, Cairney et al., 2005). To the best of the author's knowledge, no other studies have utilised a focus group design with professionals across a variety of disciplines and parents of children with neurodevelopmental disorders. The overarching theme that emerged from the results of this study was access. Absence or shortage of access to supportive environments in which children with neurodevelopmental disorders are

encouraged to participate fully in PA was a major barrier to which parents attributed their child's inactivity. Negative experiences in clubs or teams in the past created a reluctance/fear to attend for both the parent and the child. The results of this study are in agreement with previous research examining perceived benefits and barriers affecting PA participation in children with neurodevelopmental disorders. Yazdani et al. (2013), Pan et al. (2011a) and Shields and Synnot (2016) all reported that an influential barrier to PA in children was a lack of PA opportunities appropriate to children with neurodevelopmental disorders.

Education of adults in authoritative roles, both in schools and sports clubs, was viewed as both a barrier and a motivator. In parents' experiences, those who were familiar with neurodevelopmental disorders and how they affect children were more encouraging and inclusive and the children reaped greater benefits. This suggests that increasing awareness of these conditions could have a positive effect on participation. Cost-effective policies could be put in place to allow easier dissemination of information about neurodevelopmental disorders and to educate individuals on the potential for using exercise as an intervention for many challenging behavioural problems associated with neurodevelopmental disorders. Mandatory training of personnel in authoritative roles in sports clubs regarding practical supports for children with ASD (e.g. the use of visual aids to support understanding of instructions etc.).

Studies assessing barriers to exercise in typically developing children and adolescents reported that the main barriers were lack of easy-to-access facilities, time constraints associated with academic work, low self-confidence or self-consciousness and

unsupportive family (Kelishadi et al., 2010, Allison et al., 1999, Allender et al., 2006, Fernandez et al., 2017). All of these factors were reported by parents in this study but were not the most influential barriers, with the exception of self-consciousness. This implies that children and adolescents with neurodevelopmental face the same challenges as typically developing children and are also burdened by factors which affect them more severely. This is in agreement with research carried out by Shields and Synnot, (2016). One of themes identified in their study was 'similarities and differences' which described barriers common to children with and without disability, and the additional barriers faced by children with neurodevelopmental disorders.

The most commonly reported motivators for PA in typically developing children were fun and enjoyment, weight management and social interaction (Brockman et al., 2011, Allender et al., 2006). As was highlighted for barriers to PA, these factors were also reported to affect children with neurodevelopmental disorders in this study. However, social engagement was also perceived to be a barrier in children with neurodevelopmental disorders as they have difficulty with social communication. For these children, PA provides the opportunity for children to engage with peers and build social skills but may also be a cause of social anxiety, creating a paradoxical phenomenon which can be challenging to overcome. This increases the importance of a supportive and encouraging environment in which these children can be physically active.

A study reviewing strategies for success in physical education in children with ASD recommended the use of social stories and visual forms of communication such as the picture exchange communication system to prepare children and adolescents with

neurodevelopmental disorders for physical activity (Menear and Neumeier, 2015). Results of Chapter 5 (Chapter 6) revealed that children with ASD or DCD who were meeting exercise guidelines were still less fit than typically developing children who were meeting exercise guidelines. This, combined with findings of reduced activity and fitness from this study and from Chapter 4 and Chapter 5 (chapters 4 and 5), highlight the need for individualised exercise prescription in this cohort. This is backed up by the 'one size does not fit all' theme which emerged from Shields and Synnot (2016), and further confirmed with recommendations for the adaptation of the curriculum and teaching practices to suit the child's developmental level and ability to understand (Menear and Neumeier, 2015).

Strengths of this study include the participation of multi-disciplinary professionals who work with children with neurodevelopmental disorders and parents of children with neurodevelopmental disorders. This study design provided insight from a number of different perspectives, and allows for recommendations in both the family and clinical settings. The use of two researchers independently coding the focus group transcripts improved the credibility of the data and allowed a more in-depth analysis and understanding of findings. A limitation of the study was that the sample of professionals was relatively small compared to the parental sample. The author (via the gatekeeper) invited approximately 40 professionals. It is regrettable that just eight were available and willing to attend. A further limitation is that children with neurodevelopmental disorders were not part of the sample due to communication difficulties faced by children with ASD. Future studies may include such a sample with the support and expertise of professionals with experience in supporting

communication in people with ASD, for example speech and language therapists. It is unfortunate that this resource was not available to the author.

More barriers to PA are faced by children with neurodevelopmental disorders than their typically developing peers (Must et al., 2015). Self-consciousness, a lack of resources, insufficient knowledge of neurodevelopmental disorders and unavailability of mixed-ability sports and activities were the most commonly reported barriers to exercise in neurodevelopmental disorders. Increasing awareness of the conditions and educating parents regarding the positive effects of PA may have a mediating effect. However, the need for individual exercise prescription may be the most influential factor on PA in neurodevelopmental disorders.

CHAPTER 8: DISCUSSION

This chapter gives an overview of the main findings of this thesis, including implications for clinical practice, policy and future research. The work presented in this thesis set out to investigate movement and exercise in children with neurodevelopmental disorders. Specifically the focus was on the impact of two neurodevelopmental disorders, ASD and DCD on gait, physical activity and fitness. Additionally barriers to exercise and physical activity were explored.

8.1 SUMMARY OF THE MAIN FINDINGS OF THIS RESEARCH

The review of the literature concerning physical activity in children with neurodevelopmental disorders (see Section 2.2, page 39) indicated debate regarding the PA levels of children with ASD or DCD compared to typically developing children. Indeed, the two studies examining PA levels in children with ASD and/or DCD in this thesis reported conflicting results. Chapter 4 (page 76) utilised an online survey to measure PA in a large sample of children and found significant differences in the levels of both PA and sedentary behaviour between the groups. Typically developing children were more active and less sedentary than their counterparts with ASD or comorbid ASD and DCD. Children with DCD spent a similar amount of time in sedentary behaviour as typically developing children, but were less active. However, when objectively measured in Chapter 5 (page 96), PA levels were similar across the groups with the exception of vigorous-intensity activity at the weekends. Children with ASD spent significantly less time in vigorous-intensity PA at weekends than typically developing children. The discrepancies in findings of Chapter 4 and Chapter 5 may be

explained by the difficulty of describing unstructured PA with questionnaires and surveys, and further by inadequate power in Chapter 5.

Cardiorespiratory fitness of children with ASD and/or DCD was shown to be significantly lower than that of typically developing peers. As discussed in Chapter 1 (see Section 1.5.1, page 13), cardiorespiratory fitness is a health-related component of fitness. This suggests that children with these conditions are at a higher risk of poor health outcomes in comparison to their fitter typically developing counterparts. Individuals with ASD have been shown to be at a higher risk for premature mortality (Hirvikoski et al., 2016). Risk factors for several causes of premature mortality in ASD documented in the study by Hirvikoski et al., (2016) have been shown to be reduced by moderate to vigorous PA. Although few differences were observed in PA levels between the four groups as discussed in the previous paragraph, it is worth reiterating that a high proportion of all children were not sufficiently active to meet PA guidelines for health as recommend by the ACSM, (2013).

In Chapter 2 (see Section 2.1, page 22), a literature review of gait in children with ASD is described. Similar to the literature on PA in neurodevelopmental disorders, the results of this review were inconclusive and highlighted contradictory results between studies examining gait in children with ASD. The general findings of this review were that children with ASD tended to augment their walking stability with wider and shorter steps, and with increased flexion which lowers the body's centre of gravity. This aligned with the results of Chapter 6 where reduced step width, step length and increased flexion were noted at the knee at both mid-stance and toe-off. However, these deviations were observed in children with ASD and comorbid DCD rather than in

the group with ASD and no diagnosis of DCD. The previous studies reviewed did not control for the presence of DCD comorbid to ASD which may partially explain some of the conflicting results reported. These findings are relevant as it has been suggested that gait analysis may provide early indications of ASD and therefore aid early diagnosis (Esposito and Venuti, 2008).

Further motor impairments were noted in children with ASD, DCD and comorbid ASD and DCD during the tasks of the Movement ABC-2. It is interesting to note that no significant differences were found between these three groups, although the ASD and DCD group scored significantly lower in all tasks suggesting more severe impairment. It is possible that there was insufficient power in this study to detect significant differences. It is unclear whether this is a result of insufficient statistical power. The results of Chapter 7 (page 127) suggest that children with neurodevelopmental disorders face similar barriers to PA and exercise as the barriers reported in relation to typically developing children, however they encounter additional barriers and require additional support and encouragement in order to participate in PA and exercise. For example, a barrier common to all children was a lack of access to facilities whereas typically developing children did not face the same issues with communication and exclusion experienced by children with ASD and other neurodevelopmental disorders.

The studies showed that children with neurodevelopmental disorders were less fit and engaged in less PA. Motor impairment and deviations from normal movement patterns were noted. It was also shown, not surprisingly, that these children experience greater barriers to engaging in physical exercise compared with typically developing peers. This has implications for policy and practice.

8.2 IMPLICATIONS FOR CLINICAL PRACTICE

To the best of the author's knowledge, there is no data available regarding the referral rates to services such as physiotherapy or occupational therapy within the HSE for children with neurodevelopmental disorders, or indeed subsequent attendance rates. These data might be useful to understand what the reasons for referrals are and to quantify service needs. Furthermore, this knowledge may also highlight whether these clinical services provide a suitable environment in which to encourage children to be more active and improve awareness of the importance of PA. As discussed in Section 8.1, the findings of the three studies in this thesis which examined PA or factors directly influencing PA, in children with neurodevelopmental disorders suggest a need for individually-tailored PA and exercise programmes for these children. The majority of all children in Ireland are not meeting recommended guidelines of accumulating at least 60 minutes of MVPA every day. Furthermore, those with neurodevelopmental disorders who managed to reach this target still had significantly poorer fitness than their typically developing counterparts. This suggests that these guidelines are not sufficient for these children to achieve health-related benefits of PA and fitness. It is unclear what causes this. Potential factors include the prevalence of overweight and obesity or an underlying physiological factor which may require longer duration or higher intensity PA in order to attain these benefits.

As overweight and obesity increases throughout the population in this country, and indeed in other western societies, children who are predisposed to sedentary lifestyles and overweight and obesity such as children with ASD and/or DCD are at an even greater risk. Moreover they are more likely to be prescribed medications that cause them to have increased appetite, weight and derangements in blood triglycerides and

glucose metabolism such as second generation anti-psychotics. Improved assessment of the barriers to PA in children presenting to physiotherapists or assessment and intervention teams might help to better tailor exercise interventions to increase PA and improve compliance. Furthermore, there is a need to improve the awareness of parents regarding the importance of physical activity. Chapter 7 showed that the majority of parents were unaware of PA guidelines for children. The discrepancy between the questionnaire reported data on activity and the measured activity objectively possibly reflects that parents have limited understanding of the different kinds of activity which might also contribute to poor awareness of their child's exercise needs.

Many children with ASD are prescribed anti-psychotic medications as part of treatment for a variety of symptoms and challenging behaviours (Posey et al., 2008). These medications have many therapeutic uses including reducing irritability and aggression. However there are significant metabolic side-effects associated with the more commonly prescribed second generation antipsychotic medications. For example, derangements in blood triglycerides and rapid weight gain are commonly observed and this is associated with an increased risk of insulin resistance (Pringsheim et al., 2011). The use of these medications in the absence of better alternatives is currently unavoidable, however the implementation of careful dietary management and exercise interventions would help to offset some of these risks (Poulin et al., 2007).

8.3 IMPLICATIONS FOR POLICY

Mission statements for governing bodies in sport such as the Football Association of Ireland (FAI) and the Gaelic Athletic Association (GAA) all state that they strive to provide an inclusive environment for all. However, the GAA define their inclusiveness with no specific mention of special needs or disability. The FAI run a "Football For All" programme which welcomes children with various disabilities in over 30 clubs nationwide. However, evidence from Chapter 7 suggests that children with neurodevelopmental disorders are often excluded from participation in mainstream clubs. Disruptive behaviour has been suggested as the cause for this exclusion. This presents a paradoxical situation as PA and exercise have been shown to alleviate challenging and disruptive behaviours in ASD (Liu et al., 2016), yet the opportunity to do so is limited as a result of these behaviours.

The results of studies presented in this thesis suggest the need for policies to be put in place, or for better implementation of existing policies, in order to increase the availability of supportive and inclusive environments in which children with neurodevelopmental disorders are encouraged to be physically active. This could involve mandatory training of personnel from sports clubs in facilitating communication with children with conditions such as ASD. For example, training in the use of visual aids and social stories to explain rules of sports and activities may improve participation and nurture skills. Visual aids have been shown to improve performance in motor tasks in children with ASD (Liu and Breslin, 2013). Training in these areas may also facilitate a change in mindset of sports clubs with regards to ASD and other neurodevelopmental disorders. Initiatives such as the Sports Inclusion Disability Programme by Sport Ireland have increased the number of people trained in

facilitating sports participation in individuals with disability and this number continues to grow.

8.4 CRITICAL ANALYSIS OF THIS WORK

As discussed throughout this thesis, inadequate power may have impacted on the results observed. Increased sample size in studies 2 and 3 would have enhanced the power of these studies. The conservative four group study design was chosen in an attempt to probe the role of DCD-ASD comorbidity more deeply. Although sample sizes were on par with existing literature in the area, the studies had relatively low sample sizes. This has also been stated as a limitation in much of the research published in the area (Ambrosini et al., 1998; Nayate et al., 2005; Calhoun et al., 2011; Chester & Calhoun et al., 2012). Extensive and time consuming efforts were made to recruit a larger sample in order to increase the ability to detect significant changes. Approximately 200 letters were printed and sent to relevant clinic lists from Beechpark Services (a HSE Disability Service in South Dublin). No participants were recruited in this effort. Similarly, approximately 250 emails were sent to school principals in an attempt to recruit controls and subjects with ASD and/or DCD. The author received no replies from this endeavour. The author also attended neurodevelopmental clinics in AMNCH on many occasions to speak to and disseminate information leaflets to parents. This also was unsuccessful in recruiting subjects. The most fruitful method of recruitment proved to be the use of social media to disseminate information about the studies and subsequent word of mouth. A further 25 participants enrolled in the study and made appointments but did not attend. Many of these did not return the author's attempts to contact them to reschedule the appointment, while approximately ten parents reported that their child had initially agreed to participate but had changed

their mind and refused to attend or reschedule. In unpublished data from the Autism and Neurodevelopmental Disorders Research Group, we have identified that research participation in children with ASD and other neurodevelopmental disorders is likely to be impacted by lack of accessibility, inconvenience and concern that research methodologies might impact on the child negatively. This is possibly a factor in this study.

The sample size and four group design also contributed to difficulties encountered during statistical analysis of the data. Much of the data were not normally distributed. This further reduced the power to detect significant between-group differences as more rigorous non-parametric tests were required. When running statistical tests with significance set at alpha ≤ 0.05 there is a 5% chance of obtaining a Type 1 error. Type 1 error refers to a false positive, i.e. a false statistically significant result. When multiple tests are run, the probability of obtaining a Type 1 error increases. Throughout this thesis, approximately 100 statistical tests were run. A possible solution to this is the use of a Bonferroni correction (Perneger, 1998). A Bonferroni correction is applied by dividing the significance level by the total number of statistical tests run. In this thesis, that would reduce the significance level to 0.05/100 which equates to 0.0005. Bonferroni corrections are suitable for use when variables of interest are independent of each other. In this thesis, many of the variables are related either directly or indirectly. Furthermore, with the complex study design employed in Chapter 4, Chapter 5 and Chapter 6 where post hoc corrections for multiple comparisons were implemented, the use of Bonferroni corrections would have been overly conservative and would have over-corrected, therefore increasing Type 2 error where the null

hypothesis is falsely accepted. It was therefore deemed inappropriate to adopt a Bonferroni correction. The author acknowledges that omitting the use of a Bonferroni correction increased the probability of Type 1 error in this thesis.

Variation in Chapter 5 and Chapter 6 may be categorised into the following three types: subject, rater and system (Monaghan et al., 2007). Potential sources of subject variation were identified as natural variation in movement, velocity and PA, footwear and real differences due to pathological change (Monaghan et al., 2007). As footwear is worn for the Movement ABC-2, this may have been a source of variation which was unavoidable. Variations in movement and PA due to pathological change, independent of neurodevelopmental disorders (e.g. lower limb injury), were addressed via the inclusion and exclusion criteria. Participants were barefoot for gait analysis reducing the effect of footwear on variation in gait variables. Participants were encouraged to walk on the treadmill for at least five minutes to acclimatise and to allow for the selection of natural walking velocity with the aid of their parents to encourage a true self-selected velocity. Velocity has been shown to have a significant influence on kinematic parameters in the sagittal plane in healthy children (Schwartz et al., 2008). It is not clear whether this is applicable to children with neurodevelopmental disorders. There were no significant differences in velocity between the groups. Therefore velocity was not controlled for in the analysis.

Due to a limitation in the number of available markers and driveboxes, trunk, upper limb and foot kinematics were not assessed during gait analysis. Children with ASD have been shown to exhibit reduced arm swing (Rinehart et al., 2006a). It would have been of interest to examine this phenomenon in children with DCD and in children

with comorbid ASD and DCD as it is an area which has not been well studied. This may have provided further evidence of a rigid gait pattern with augmented stability. The use of the treadmill for assessing gait had several implications. For one, kinetic (i.e. force) data could not be obtained. The literature review on studies investigating gait in children with ASD (see Section 2.1, page 22) showed that few studies have examined kinetic gait parameters in children with ASD. There is little evidence to show the differences between treadmill gait and overground gait in healthy children and none to show these differences in children with neurodevelopmental disorders. However, due to reflection issues in the gait laboratory, the use of the treadmill was necessary to improve the quality of gait data obtained. It is regrettable that Chapter 6 could not add to the literature and further examine kinetic parameters in ASD.

Sources of possible rater variation include anthropometric measurements and identification of anatomical landmarks. Other sources include gait marker placement and wand alignment, instructions given during Movement ABC tasks and the fitness test. To minimise the impact of rater variation, one rater completed all test sessions and data processing. The order of the physical tests was the same for all participants. The data collection period for Chapter 5 and Chapter 6 was prolonged over approximately three years. This was in part due to damage to equipment which had to be sent for repair lasting approximately 3 months. Difficulty in recruiting this cohort as discussed at the beginning of this section also contributed to the longevity of the data collection period. The author acknowledges that with equipment familiarisation over time, rater variation may have been reduced. However, for the reasons discussed above, this was unavoidable.

System variation was addressed with regular servicing of the Codamotion gait analysis system and the Cosmed K4b^2 gas analysis system. The gait system alignment procedure was carried out at the beginning of each testing day. Calibration of the K4b^2 was carried out prior to each individual test participant. Firmware of Actigraph Gt3X+ monitors were updated regularly.

As there was a lack of exploratory research conducted in PA and movement patterns of children with ASD and comorbid DCD, an observational and comparative descriptive design was required. Therefore, a cross-sectional study design was implemented for the studies discussed in chapters 4-7 in this thesis. This type of study design is useful for describing phenomena as they occur in their natural setting and to explore the association between variables (Levin, 2006). There are several advantages to employing a cross-sectional design including cost–effectiveness as data is collected on one occasion only and there is no loss of participants in the time to follow up assessments (Brink and Wood, 1998). Additionally, a large amount of data can be collected and analysed. However there are also limitations to this type of study design. The most applicable limitations to the research presented in this thesis is the inability to infer causality and the requirement of a very large sample size to allow generalisation of phenomena described to the whole population (Levin, 2006).

8.5 IMPLICATIONS FOR FUTURE RESEARCH

The studies presented in this thesis have provided some interesting insight into the PA and fitness levels of children with neurodevelopmental disorders. Upon reflection, it has also highlighted areas of interest for future research. Hypotonia has been reported as the most common motor impairment in ASD and has also been well documented in

DCD (Ming et al., 2007, Barnhart et al., 2003). It has been suggested that hypotonia may account for reduced balance capacity in individuals with Prader-Willi or Ehlers-Danlos syndromes, two rare genetic conditions characterised by muscular hypotonia (Galli et al., 2011). To date, this effect has not been studied in ASD or DCD but may be an area of interest for future study as balance deficits have been well documented in both conditions. As tone has an impact postural control, its presence may impact movement patterns and PA (Massion, 1998). The effect of hypotonia on PA, fitness and movement patterns in children with ASD and/or DCD has not yet been studied. The confounding results of Chapter 5 which show that children with ASD and/or DCD were less physically fit but similarly physically active compared to typically developing controls suggests that another factor is influencing fitness. It is possible that hypotonia may reduce the effectiveness of PA in improving health-related components of fitness, although this is speculative as little research has been done to investigate this.

Longitudinal studies examining the effects over time of variables such as age on PA and fitness levels would add depth to the base of knowledge provided by the studies in this thesis. Randomised control trials determining the efficacy of interventions on PA may be the key to unlock the problem of reduced PA and fitness in children with ASD and DCD. Another area which could be improved upon in future research is sample size, as previously discussed. Larger studies in the future could address this limitation. The overall findings of this thesis suggest altered PA levels and patterns in neurodevelopmental disorders. However, differences in MVPA were not objectively detected. Another method of increasing the power of future studies to detect significant differences would be an alternative design. This thesis has indicated that

there are differences in PA, fitness and motor function between children with comorbid ASD and DCD and typically developing peers. On several occasions, significant differences were noted but with correction for multiple comparisons necessary in the four group design, these differences were no longer significant. Therefore, future studies examining the differences observed solely between the two groups may yield significant and interesting findings that were not apparent in this research.

8.6 CONCLUSION

This thesis highlighted deficits in physical activity and fitness in children with neurodevelopmental disorders that may place them at future risks of health outcomes. Some deficits in gait were detected that appear to be more pronounced in the dual diagnosis group and that this study added to the literature by looking at the impact of comorbidity. Significant additional barriers to exercise exist for individuals with neurodevelopmental disorders that are clearly impacting on the ability of people with neurodevelopmental disorders to meet exercise guidelines.

This research has investigated a relatively understudied topic in relation to neurodevelopmental disorders that has the potential to increase risks for significant negative health outcomes, e.g. cardiovascular risks. Further studies would help to clarify how best to promote engagement with exercise in order to promote physical health and wellbeing.

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APPENDICES

Appendix I – Ethical Approval Letters

Ethical Approval – Chapter 4, 2 and 3

THIS NOTEPAPER MUST NOT BE USED FOR PUBLICATIONS OR INDEXING PURPOSES

SJH/AMNCH Research Ethics Committee Secretariat
Ursula Ryan Ph: 4142342 email: Ursula.Ryan@amnch.ie
Secretariat Fax 4142371

THE ADELAIDE & MEATH HOSPITAL, DUBLIN
INCORPORATING
THE NATIONAL CHILDREN'S HOSPITAL

TALLAGHT, DUBLIN 24, IRELAND
TELEPHONE +353 1 4142000

Mr. Deirdre Kindregan
Physiotherapy Department
Trinity Centre for Health Sciences
St. James's Hospital
Dublin 8

January 25th 2013

Re: Gait analysis in children with Autistic Spectrum Disorders and Developmental Coordination Disorder

Please quote this reference in any follow up to this letter: 2013/01/08 Chairman's Action

Dear Ms. Kindregan,

Thank you for your recent submission of the above proposal to the SJH/AMNCH Research Ethics Committee.

The Chairman, having reviewed the proposal on behalf of the SJH/AMNCH Research Ethics Committee has given ethical approval on behalf of the Committee.

Yours sincerely

Ms. Ursula Ryan
Secretary,
SJH/AMNCH Research Ethics Committee

Feidhmeannacht na Seirbhíse Sláinte
Health Service Executive

Beechpark Services for Children with an Autistic Spectrum Disorder
Linn Dara Child & Adolescent Mental Health Services
Bridge House, Cherry Orchard Hospital, Ballyfermot, Dublin 10.

Name: Deirdre Kindregan

Date: 14/01/2014

Re: Gait analysis, energy expenditure and physical activity in children with ASD and dyspraxia

Dear Ms Kindregan,

Thank you for applying to the HSE Linn Dara / Beechpark research ethics committee, which was discussed on 08/01/2014. In addition, thank you for sending on your amendments as requested by the committee. I am pleased to report that the study (including the amendments) has been granted approval by the Committee.

If during the course of the research project, any significant amendments or alterations to the proposed research are required, approval must again be sought from this committee.

Wishing you every success with the study,

Yours sincerely,

_____
Ms Sarah O'Sullivan
Secretary of the Linn Dara / Beechpark Ethics Committee,
Ph: 045 8445020 Email: Sarah.osullivan1@hse.ie

Members present when decision was taken:

Ms Paula Dillon	Chair / Clinical Nurse Specialist Linn Dara Sevices
Ms Sarah O'Sullivan	Occupational Therapist Beechpark Services
Ms Niamh Quivlivan	Speech and Language Therapist, Beech Park Services.
Ms Roberta Mulligan	Senior Social Worker Linn Dara CAMHS
Ms. Lisa Clarke	Social Worker

Coláiste na Tríonóide, Baile Átha Cliath
Trinity College Dublin
Ollscoil Átha Cliath | The University of Dublin

Deirdre Kindregan
Discipline of Physiotherapy
Trinity Centre for Health Sciences
St James's Hospital
Dublin 8

26th January 2016

Ref: 20151207

Title of Study: Barriers to sport and exercise in children with Neurodevelopment conditions.

Dear Ms Kindregan,

Further to a meeting of the School of Medicine Research Ethics Committee held in December 2015, we are pleased to inform you that the above project has been approved.

Yours sincerely,

Professor Thomas Rogers
Chairperson
School of Medicine Research Ethics Committee

An tOllamh Paul Browne MB, FRCPI, FRCPsych
Ceann Scoil an Leighis

Professor Paul Browne MB, FRCPI, FRCPsych
Head of School of Medicine

Ms. Orla Bannon
Riarthóir na Scoile

Ms. Orla Bannon
School Administrator

Scoil an Leighis
An Institiúid Eolaíochtaí Bithleighis
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iption purposes

Ospidéal Mhuire na Leanaí, Cromghlinn
Our Lady's Children's Hospital, Crumlin

ETHICS (MEDICAL RESEARCH) COMMITTEE OFFICE

Tel: + 353 (01) 409 6307/6243

Ms Deirdre Kindregan
PhD Candidate
Physiotherapy – School of Medicine – TCD
Physiotherapy Department
Trinity Centre for Health Sciences
James's Street
Dublin 8

29th January 2016

REC Reference: GEN/362/14

Gait analysis, energy expenditure and physical activity in children with Autism and other neurodevelopmental disorders
Principal Investigators: Dr. David Webb, Ms. Deirdre Kindregan.

Dear Ms Kindregan

At a meeting which was held on, 19th January 2016, the Ethics (Medical Research) Committee of this hospital reviewed the resubmitted application in relation to the above study.

The Committee noted that the invitation letters to Parent/Guardian and School Principal states that contact details were acquired from the school/School Days Website. The Committee advised that consent would be required for contact details to be used for this Study and that Dr. Gormley and Dr. Webb should, in the first instance, request consent from the Parent/Guardian to pass contact details onto the Investigator. The Committee requested that the invitation letters be re-worded to reflect same.

The Committee requested clarification in relation to whether or not Trinity's Clinical Indemnity covers child participants on the Trinity site at St. James's Hospital. The Committee advised that if there is an issue with insurance, the Primary Consultant in OLCHC, would have to be informed and should seek clarification on this issue.

The Committee advised that the Children First Guidelines must be adhered to and must comply with Our Lady's Children's Hospital, Crumlin, Policy.

Cont'd/

The Committee noted that the Information Leaflets may be challenging for children with an intellectual difficulty.

The Committee also noted also that participation in this Study will require two appointments – one at this hospital and one in the Trinity Centre, St. James's Hospital. The Committee requested that the Information Leaflets be amended to reflect this fact.

The Committee requested that data be encrypted.

The Study was approved, subject to the above conditions.

Please forward a copy of the updated documentation to this Office to complete our records.

The Committee would like to thank you, Professor Louise Gallagher and Dr. John Gormley for being present at the meeting.

Yours sincerely

Claire Rice
Secretary
Ethics (Medical Research) Committee

CC: Dr. David Webb, Consultant, E.E.G. Department (Neuroscience Centre –Waves) OLCHC.

Trinity College Dublin
Coláiste na Tríonóide, Baile Átha Cliath
The University of Dublin

Parents' Information Leaflet – Study Group

Title of study:
Gait analysis, energy expenditure and physical activity in children with Autism and/or Dyspraxia.

Introduction:

- This study aims to see if Autism has an effect on the way in which the legs move during walking in children, and to explore whether there is a different pattern of walking compared to that in children without Autism. Very few previous studies have examined the effects of Autism on walking characteristics.
- Many children diagnosed with Autism have a secondary diagnosis of Dyspraxia or Developmental Coordination Disorder. This study will try to determine whether it is Autism or Dyspraxia that causes any altered walking patterns found.
- Fitness levels and energy expenditure will also be measured.
- This study will also attempt to assess any link between the above and physical activity levels and participation in sport in children with Autism and/or Dyspraxia.

Groups:
This study is focused on children with a diagnosis of Autism Spectrum Disorders OR Dyspraxia/DCD OR a dual-diagnosis of both conditions. Children with typical development will also be included to act as controls for comparison.

Procedures:
Should you wish to participate in this study to help answer these important questions, you and your child would be asked to attend the Trinity Centre for Health Sciences on one occasion for testing, and possibly a second occasion for certain tests to be carried out if they have not previously done for your child (only for children with ASD). At this visit, your child's walking characteristics will be measured and recorded.

DAY OF TESTING

- An appointment will be made over the phone for your child to attend the Gait Laboratory (walking measurement room) of the Trinity Centre for Health Sciences, St. James's Hospital seven days later.
- For this day, your child will be asked to bring shorts and running shoes.
- The lead investigator will be present at this session.
- You are asked to stay for the testing which will last around 2 hours.
- On arrival at the centre, the lead investigator will carry out a clinical assessment of your child. During this assessment, measurements of your child's legs will be taken, for example- knee width, leg length, and ankle movement.
- You and your child will then be asked to fill in a questionnaire on your child's activity levels and participation in sport.
- Your child will be asked to put on a backpack and mask to measure how much energy they are using at rest and during a walking task.
- Lightweight markers will be attached to your child's legs with double sided tape and Velcro strapping. These markers will allow a computer to measure exactly the way in which your child's legs move when they are walking.
- Your child will then be asked to walk on a treadmill at two different speeds.
- A brief fitness test will then be carried out.
- You will then be given a small device for your child to wear on a belt every day for one week to measure your child's activity levels. The investigator will tell you about the device and how it works, and will give you a pre-paid envelope in which to send the device back to the centre.

Benefits:

A report of your child's results will be given after the test, and it is hoped the results of this study will help future medical practice to treat walking problems in children. Should abnormalities which require medical attention be noted when his/her walking results are analysed, you and your GP will be notified to enable remedial action to be taken.

Risks:

As there is a fitness test, there are certain risks involved in taking part in this study. Your child may experience pain, fatigue, dizziness or difficulty breathing during the exercise test and may wish to stop the test. If so, the test will be stopped immediately. In the unlikely case of an emergency, the investigator is first-aid trained and there is an AED on site. Your child will be brought to Our Lady's Children's Hospital, Crumlin for treatment if necessary.

Exclusion from participation:

Your child cannot participate in this study if they have any of the following-

- History of cardiac or respiratory disease.
- Poorly controlled asthma.
- Currently taking prescribed anti-psychotic medication.
- Under the age of 6 or older than 18 years.
- Pregnancy.
- Any injury to their legs in the last three months.

Confidentiality:
Your child's identity will remain confidential. Your child's name will not be published and will not be disclosed to anyone outside the study group. Data which will identify your child will be kept after the study is completed, however it will not be used in future unrelated studies without your specific permission being obtained.

The only circumstance in which confidentiality may be broken is in the unlikely event that it is suspected that your child is at risk. If this is the case, confidential details may be given to a child protection officer. This protocol is put in place solely for the protection of your child.

Voluntary participation:
If you decide you would like your child to participate in this study, please contact Deirdre Kindregan at (01) 8963613 or via e-mail kindregd@tcd.ie.

Stopping the study:
If you decide you would like your child to take part in the study, you may withdraw your child at any time.

You understand the investigators may withdraw your child's participation in the study at any time without your consent.

Permission:
This project has Research Ethics Committee approval from the AMNCH/SJH, NCRC and Linn Dara Ethics Committees.

Further information:
You can get more information or answers to your questions about the study, your child's participation in the study and their rights, from Deirdre Kindregan at (01) 896 3613 or via e-mail to kindregd@tcd.ie. If the study team learns of important new information that might affect your desire to remain in the study, you will be informed at once.

Trinity College Dublin
Coláiste na Tríonóide, Baile Átha Cliath
The University of Dublin

Parents' Information Leaflet – Control Group

Title of study:
Movement Patterns and Physical Activity in Children with Neurodevelopmental Disorders

Introduction:

- This study aims to see if neurodevelopmental disorders have an effect on the way in which the legs move during walking in children, and to explore whether there is a different pattern of walking compared to that in typically developing control children.
- Fitness levels and energy expenditure will also be measured.
- This study will also attempt to assess any link between the above and physical activity levels and participation in sport in children with neurodevelopmental disorders.

Groups:
This study is focused on children with a diagnosis of Autism Spectrum Disorders OR Dyspraxia/DCD OR a dual-diagnosis of both conditions. Children with typical development will also be included to act as controls for comparison.

Procedures:
Should you wish to participate in this study to help answer these important questions, you and your child would be asked to attend the Trinity Centre for Health Sciences on one occasion. At this visit, your child's walking characteristics, coordination and fitness will be measured and recorded.

DAY OF TESTING

- An appointment will be made by email or over the phone for your child to attend the Gait Laboratory (walking measurement room) of the Trinity Centre for Health Sciences, St. James's Hospital seven days later.
- For this day, your child will be asked to bring shorts and running shoes.
- The lead investigator will be present at this session.
- You are asked to stay for the testing which will last between 90 minutes and 2 hours.
- On arrival at the centre, the lead investigator will carry out a clinical assessment of your child, including an IQ test. During this assessment, measurements of your child's legs will be taken, for example- knee width, leg length, and ankle movement.

- You will be asked to fill in several questionnaires about your child.
- Your child will be asked to put on a backpack and mask to measure how much energy they are using at rest and during a walking task.
- Lightweight markers will be attached to your child's legs with double sided tape and Velcro strapping. These markers will allow a computer to measure exactly the way in which your child's legs move when they are walking.
- Your child will then be asked to walk on a treadmill at two different speeds.
- A brief fitness test will then be carried out.
- You will then be given a small device for your child to wear on a belt every day for one week to measure your child's activity levels. The investigator will tell you about the device and how it works, and will give you a pre-paid envelope in which to send the device back to the centre.

Benefits:

A report of your child's results will be given a few weeks after the appointment, and it is hoped the results of this study will help future medical practice to treat walking problems in children and promote physical activity for health. Should abnormalities which require medical attention be noted when his/her walking results are analysed, you will be notified to enable remedial action to be taken.

Risks:

As there is a fitness test, there are certain risks involved in taking part in this study. Your child may experience pain, fatigue, dizziness or difficulty breathing during the exercise test and may wish to stop the test. If so, the test will be stopped immediately. In the unlikely case of an emergency, the investigator is first-aid trained and there is an AED on site. Your child will be brought to Our Lady's Children's Hospital, Crumlin for treatment if necessary.

Exclusion from participation:

Your child cannot participate in this study if they have any of the following-

- History of cardiac or respiratory disease.
- Poorly controlled asthma.
- Currently taking prescribed anti-psychotic medication.
- Under the age of 6 or older than 18 years.
- Pregnancy.
- Any injury to their legs in the last three months.

Confidentiality:

Your child's identity will remain confidential. Your child's name will not be published and will not be disclosed to anyone outside the study group. Data which will identify your child will be kept after the study is completed, however it will not be used in future unrelated studies without your specific permission being obtained.

The only circumstance in which confidentiality may be broken is in the unlikely event that it is suspected that your child is at risk. If this is the case, confidential details may be given to a child protection officer. This protocol is put in place solely for the protection of your child.

Voluntary participation:
If you decide you would like your child to participate in this study, please contact Deirdre Kindregan at (01) 8963613 or via e-mail kindregd@tcd.ie.

Stopping the study:
If you decide you would like your child to take part in the study, you may withdraw your child at any time.

You understand the investigators may withdraw your child's participation in the study at any time without your consent.

Permission:
This project has Research Ethics Committee approval from the AMNCH/SJH, National Children's Research Centre and Linn Dara Ethics Committees.

Further information:
You can get more information or answers to your questions about the study, your child's participation in the study and their rights, from Deirdre Kindregan at (01) 896 3613 or via e-mail to kindregd@tcd.ie. If the study team learns of important new information that might affect your desire to remain in the study, you will be informed at once.

Children's Information Leaflet – Aged 6-8 years

Walking study

What does the study want to see?

My name is Deirdre Kindregan and I am a physiotherapist.

I would like to see the way children walk.

What would you do?

If you would like to be in my study, you can come to see me at St. James's Hospital.

When you get there I will tell you everything that we will do. You can ask me any questions before we start. If you want to help me, I will ask you to write your name on a page which says you would like to be in the study.

Your Mammy or Daddy will be there the whole time.

On this day I will ask you some questions about your legs and then I will check how tall you are and how heavy you are.

I will ask you to answer some questions on a sheet about sports you like and how much exercise you do. Your Mammy or Daddy can help you.

First I will ask you to wear a backpack and a mask and sit for a little while.

We will then see how you walk. You will be asked to wear swimming togs or a vest and shorts. Your Mammy or Daddy can help you put them on. Before this test, I will measure some parts of your legs. I will then stick some tiny pads onto your legs using sticky tape and straps.

You will then walk for a few minutes, and then you'll run up and down for a few minutes.

After this, I will show you a small gadget for you to wear on your belt every day for one week. This gadget will tell me how much you move about during the day.

What will you find out from all these tests?

After the test you will get to see yourself as a stick figure walking on the computer screen.

Is this study safe for you?

This study is very safe and will not make you feel sick or hurt.

Will people know I am doing the tests?

The only people who will know you are doing the study are your parents and the people doing the study.

What do I do if I would like to do the study?

If you would like to do the study, you should tell your parents and they can tell me.

What do I do if I change my mind?

If you change your mind and do not want to be in the study, you can tell your parents and they can tell me, and that's OK.

What do you do if you have any questions?

If you have any questions, you can tell your parents and they can telephone me to ask me. You can also ask me when you see me at the hospital.

Children's Information Leaflet – Aged 8-12 years

Walking study

What does the study want to see?

My name is Deirdre Kindregan and I am a physiotherapist.

I would like to see the way different young people walk.

What would you do?

If you think you would like to be in my study, you will visit me at St. James's Hospital.

When you get there I will tell you everything that we will do. You can then ask me questions before we start. If you still want to be in the study, I will ask you to write your name on a page which says you would like to be in the study.

Your Mam or Dad will be there the whole time.

On this day I will ask you some questions about your legs and then I will check how tall you are and how much you weigh.

I will ask you to answer some questions on a sheet about sports you like and how much exercise you do.

I will ask you to wear a backpack and a mask and sit for a little while.

We will then do the walking test.

For the walking test you will be asked to wear swimming togs or a vest and shorts. Before this test, I will measure some parts of your legs. I will then stick some tiny pads onto your legs using sticky tape and straps.

You will then walk for a little while. Then I'll ask you to run between two cones for a few minutes.

After this, I will show you a small gadget for you to wear on your belt every day for one week. This gadget will tell me how much you move about during the day.

What will you find out from all these tests?

After the test you will get to see yourself as a stick figure walking on the computer screen.

Is this study safe for you?

This study is very safe and will not make you feel sick.

Will people know I am doing the tests?

The only people who will know you are doing the study are your parents and the people doing the study.

What do I do if I would like to do the study?

If you would like to do the study, you should tell your parents and they can tell me.

What do I do if I change my mind?

If you change your mind and do not want to be in the study, you can tell your parents and they can tell me, and that's OK.

What do you do if you have any questions?

If you have any questions, you can tell your parents and they can telephone me to ask me. You can also ask me when you see me at the hospital.

Children's Information Leaflet – Aged 12-14 years

Walking study

What does the study want to see?

My name is Deirdre Kindregan and I am a physiotherapist.

I would like to see the way different young people walk.

What would you do?

If you think you would like to be in my study, you will visit me at St. James's Hospital with your parents.

When you get there I will tell you everything that we will do. You can then ask me questions before we start. If you still want to be in the study, I will ask you to write your name on a page which says you would like to be in the study.

On this day I will ask you some questions about your legs and then I will check your height and your weight. I will ask you to answer some questions on a sheet about sports you like and how much exercise you do.

I will ask you to wear a backpack and a mask and sit for a little while. We will then do the walking test. For the walking test you will be asked to wear swimming togs or a vest and shorts. Before this test, I will measure some parts of your legs. I will then stick some pads onto your legs using sticky tape and straps.

When the measurements are done, I'll ask you to walk on a treadmill for a few minutes. After this I'll ask you to run between two cones – like the bleep test you might have done in P.E. in school.

Finally, I will show you a small gadget for you to wear on your belt every day for one week. This gadget will tell me how much you move around during the day.

What will you find out from all these tests?

After the test you will get to see yourself as a stick figure walking on the computer screen.

Is this study safe for you?

This study is very safe and will not hurt you in any way.

Will people know I am doing the tests?

The only people who will know you are doing the study are your parents and the people carrying out the study.

What do I do if I would like to do the study?

If you would like to do the study, you should tell your parents and they can tell me.

What do I do if I change my mind?

If you change your mind and do not want to be in the study that's OK, you can tell your parents and they can tell me.

What do you do if you have any questions?

If you have any questions, you can tell your parents and they can telephone me to ask me. You can also ask me when you see me at the hospital.

Children's Information Leaflet – Aged 14-16 years

Walking study

What does the study want to see?

My name is Deirdre Kindregan and I am a physiotherapist.

I would like to see how different young people walk and how fit they are.

What would you do?

If you think you would like to be in my study, you will visit me at St. James's Hospital with your parents.

When you get there I will tell you everything about the study. You can ask me any questions you have before we start. If you still want to be in the study, I will ask you to sign a form which says that you agree to be in the study.

On this day I will ask you some questions about your legs, for example if you've had any injuries, and then I will check your height and your weight. I will ask you to fill out a questionnaire about sports you like and how much exercise you do.

I will ask you to wear a backpack and a mask and sit still for about fifteen minutes. We will then do the walking test. For the walking test you will be asked to wear swimming togs or a vest and shorts. Before this test, I will measure the length of your legs and feet. I will then stick some markers onto your legs using sticky tape and Velcro straps.

When the measurements are done, I'll ask you to walk on a treadmill for a few minutes. After this I'll ask you to run between two cones – like the bleep test you might have done in P.E. in school.

Finally, I will show you a small gadget, called an accelerometer, for you to wear on your belt every day for one week. This will tell me how much you move around during the day.

What will you find out from all these tests?

After the test you will get to see yourself as a stick figure walking on the computer screen and see how your walk.

Is this study safe for you?

This study is very safe and will not hurt you in any way.

Will people know I am doing the tests?

The only people who will know you are doing the study are your parents and the people carrying out the study.

What do I do if I would like to do the study?

If you would like to do the study, you should tell your parents and they can tell me.

What do I do if I change my mind?

If you change your mind and do not want to be in the study that's OK, you can tell your parents and they can tell me.

What do you do if you have any questions?

If you have any questions, you can tell your parents and they can telephone me to ask me or you can telephone me with your parents. You can also ask me when you see me at the hospital.

Trinity College Dublin
Coláiste na Tríonóide, Baile Átha Cliath
The University of Dublin

Participant Information Leaflet (16-18 years)

Title of study:

The effects of Autism and Dyspraxia on gait parameters in children.

Introduction:

- This study aims to see if Autism has an effect on the way in which the legs move during walking in children and young adults, and to explore whether there is a different pattern of walking compared to that in children without Autism. Very few previous studies have examined the effects of Autism on walking characteristics.
- Many children diagnosed with Autism have a secondary diagnosis of Dyspraxia or Developmental Coordination Disorder. This study will try to determine whether it is Autism or Dyspraxia that causes any altered walking patterns found.
- Fitness levels and energy expenditure will also be measured.
- This study will also attempt to assess any link between the above and physical activity levels and participation in sport in children with Autism and/or Dyspraxia.
- Finally, this study will see if the results found are the same for boys and girls.
- To do this, a number of children who do not have Autism are also being asked to volunteer their time.

Procedures:

Should you wish to participate with us in this study to help answer these important questions, you would be asked to attend the Trinity Centre for Health Sciences on one occasion for testing, and possibly one occasion for certain tests to be carried out if they have not previously done. At this visit your walking characteristics will be measured and recorded.

DAY OF TESTING

- An appointment will be made over the phone for you to attend the Gait Laboratory (walking measurement room) of the Trinity Centre for Health Sciences, St. James's Hospital seven days later.
- You are asked to bring a vest and a pair of shorts to wear for the assessment.

- The lead investigator will be present at this session.
- On arrival at the centre, the lead investigator will carry out a clinical assessment. During this assessment, measurements of your legs will be taken, for example- knee width, leg length, and ankle movement.
- You will then be asked to fill in a questionnaire on your physical activity levels and participation in sport.
- You will be asked to put on a backpack and mask to measure how much energy you use at rest and during a walking task.
- Lightweight markers will be attached to your legs with double sided tape and Velcro strapping. These markers will allow a computer to measure exactly the way in which your legs move when they are walking.
- You will then be asked to walk on a treadmill for a few minutes.
- Before you leave, you will be given a small device to wear on a belt every day for one week to measure your activity levels. The investigator will tell you about the device and how it works, and will give you a pre-paid envelope in which to send the device back to the centre.

Benefits:

An analysis of your walking pattern will not be given after the test, however it is hoped the results of this study will help future medical practice to treat walking problems in children. Should abnormalities in your walking pattern be noted when the results are analysed, you will be notified to enable remedial action to be taken.

Risks:

As there is a fitness test, there are certain risks involved in taking part in this study. You may experience pain, fatigue, dizziness or difficulty breathing during the exercise test and may wish to stop the test. If so, the test will be stopped immediately. In the unlikely case of an emergency, the investigator is first-aid trained and there is an AED on site. You will be brought to Our Lady's Children's Hospital, Crumlin for treatment if necessary.

Exclusion from participation:

You cannot participate in this study if you have any of the following:

- ADHD
- History of cardiac or respiratory disease
- Poorly controlled asthma
- Diagnosis of diabetes
- Currently taking prescribed medication*

  *that may affect your movement patterns
- Under the age of 6 or older than 18 years
- Pregnancy
- Any injury to their legs in the last three months
- Sensory integration issues that would not allow you to tolerate Velcro straps around your legs

Confidentiality:

Your identity will remain confidential. Your child's name will not be published and will not be disclosed to anyone outside the study group. Data which will identify you will be kept after the study is completed, however it will not be used in future unrelated studies without your specific permission being obtained.

Voluntary participation:

If you decide you would like to participate in this study, please contact Deirdre Kindregan at (01) 8963613 or via e-mail kindregd@tcd.ie. Alternatively you may fill out the enclosed expression of interest form, place it in the envelope provided and send it by post, the lead investigator will then contact you to arrange an appointment.

Stopping the study:

You understand the investigators may withdraw your participation in the study at any time without your consent. If the study team learns of important new information that might affect your desire to remain in the study, you will be informed at once.

Permission:

This project has Research Ethics Committee approval from the AMNCH/SJH, NCRC and Linn Dara Ethics Committees.

Further information:

You can get more information or answers to your questions about the study, your participation in the study and your rights, from Deirdre Kindregan at (01) 896 3613 or via e-mail to kindregd@tcd.ie.

TRINITY COLLEGE DUBLIN

INFORMED PARENTAL CONSENT FORM

PROJECT: Gait analysis, energy expenditure and physical activity in children with Autism and/or Dyspraxia.

LEAD INVESTIGATORS: Prof. Louise Gallagher, Dr. John Gormley, Ms Deirdre Kindregan.

BACKGROUND:

This study aims to determine the effects of Autism on walking characteristics of young people.

DECLARATION:

I have read, or had read to me, the information leaflet for this project and I understand the contents. My daughter/son and I have had the opportunity to ask questions and all my questions have been answered to my satisfaction. As his/her guardian, I freely and voluntarily agree for my child to be part of this research study, though without prejudice to my or my child's legal and ethical rights. I understand that I may withdraw my child from the study at any time and I have received a copy of this agreement.

I understand data and consent forms will be retained after the study reaches completion and that data will only be used in related studies.

PARTICIPANT'S NAME:
…….

PARTICIPANT'S SIGNATURE:
……...

NAME OF CONSENTER, PARENT or GUARDIAN:
…….....

SIGNATURE:
…….

CONTACT DETAILS:
……

Date: ……

ASSENT FORM

Children aged 6-8 years

PROJECT: Walking study.

LEAD INVESTIGATORS: Prof. Louise Gallagher, Dr. John Gormley, Deirdre Kindregan.

DECLARATION:

I have read the letter from Deirdre. I would like to take part in the study to see how I walk. I got to ask any questions and am happy to do it.

Nobody is making me take part in this study and I know that I can stop at any time I like.

I know that Deirdre will keep some information about me in this study but that she will not give it to anyone else unless I say that it's OK.

YOUR NAME: ……

YOUR SIGNATURE: ……

Date: ……

ASSENT FORM

Children aged 8-12 years

PROJECT: Walking study.

LEAD INVESTIGATORS: Prof. Louise Gallagher, Dr. John Gormley, Deirdre Kindregan.

DECLARATION:

I have read the letter from Deirdre. I would like to take part in the study to see how I walk. I got to ask any questions and I understand the answers I got. I am happy to be a part of the study.

Nobody is making me take part in this study and I know that I can stop at any time I like.

I know that Deirdre will keep some information about me in this study but that she will not give it to anyone else unless I say that it's OK. I understand that this study is to help other children.

YOUR NAME: …….

YOUR SIGNATURE: ……

Date: ……

ASSENT FORM

Children aged 12-14

PROJECT: Walking, energy use and exercise in children.

LEAD INVESTIGATORS: Prof. Louise Gallagher, Dr. John Gormley, Deirdre Kindregan.

BACKGROUND:

This study is trying to find out how different young people walk, how active they are and how much energy they use.

DECLARATION:

I have read, or had read to me, the information leaflet for this project and I understand what it says. I was able to ask questions and all my questions have been answered to my satisfaction.

I am volunteering to be part of this research study and I understand that I can change my mind and decide not to take part in the study at any time I like.

I understand that information and forms will be kept after the study is finished but that it will not be used in anything other than this study without my permission.

YOUR NAME: …….

YOUR SIGNATURE: ……

Date: ……

ASSENT FORM

Children aged 14-16

PROJECT: Walking, energy use and physical activity in children.

LEAD INVESTIGATORS: Prof. Louise Gallagher, Dr. John Gormley, Deirdre Kindregan.

BACKGROUND:

This study is trying to find out how different young people walk, how much energy they use and how physically active they are.

DECLARATION:

I have read, or had read to me, the information leaflet for this project and I understand what it says. I was given the opportunity to ask questions and all my questions have been answered to my satisfaction.

I am volunteering to be part of this research study and I understand that I can change my mind and decide not to take part in the study at any time.

I understand that information and consent forms will be retained after the study reaches completion and that data will not be used in unrelated studies without my permission.

YOUR NAME: …….

YOUR SIGNATURE: ……

Date: ……

TRINITY COLLEGE DUBLIN

CONSENT FORM

(Aged 16-18)

PROJECT: Gait analysis, energy expenditure and physical activity in children with Autism and/or Dyspraxia.

LEAD INVESTIGATORS: Prof. Louise Gallagher, Dr. John Gormley, Deirdre Kindregan.

BACKGROUND:

This study aims to determine the effects of Autism on walking characteristics of young people and to determine fitness levels and participation in sport.

DECLARATION:

I have read, or had read to me, the information leaflet for this project and I understand the contents. I have had the opportunity to ask questions and all my questions have been answered to my satisfaction.

I freely and voluntarily agree to be part of this research study, though without prejudice to my legal and ethical rights.

I understand that I may decide not to take part in the study at any time and I have received a copy of this form.

I understand that information and consent forms will be retained after the study reaches completion and that data will not be used in unrelated studies without my permission.

PARTICIPANT'S NAME: …….

PARTICIPANT'S SIGNATURE: ……

CONTACT DETAILS: ……

Date: ……

Cosmed K4b^2 User Guide

Deirdre Kindregan

Version 1.4 – January 2016

Physiotherapy Dept., TCD

Warm-up & Setup

1. Plug battery into unit – wire with green dot into battery, red connector into unit
2. Plug other batteries into charger (see instructions for timer at on last page)
3. Press power button – screen should come on, if not try a different battery and plug this one in to charge
4. "Optimum warm up" will come up on screen, press enter
5. Leave to warm up for 45 minutes
6. Plug turbine and sampling line into unit
7. Plug in heart rate monitor probe
8. Attach antenna to unit

Fig. 1

9. Plug battery into receiver unit and attach antenna
10. Insert USB from receiver unit into USB port on right side of laptop*NB*
11. Open K4b^2 program on laptop

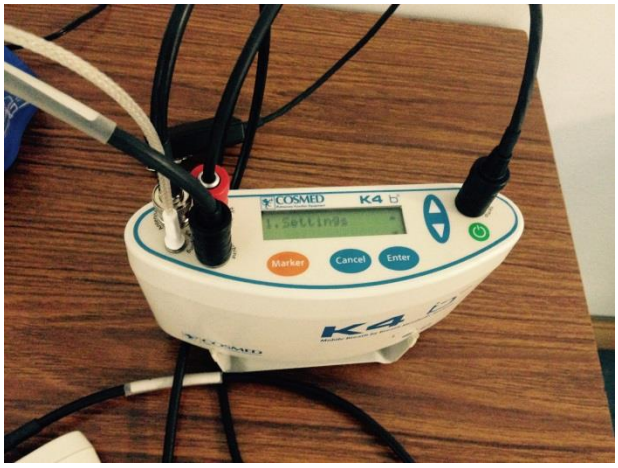


Figure 1 - Attachments

Calibration:

Scroll down using the arrow buttons to '4. Calibration' and press enter

1. Room air calibration

- Press enter
- Do not breathe near sampling line
- When calibration is done, press cancel

2. Reference Gas Calibration
   - Scroll down and press enter
   - Plug O_2 tank line into reference gas calibration box (See Fig. 2)

Figure 2 - Reference Gas Calibration Box

   - Open valves (one on side of O_2 tank with wrench (anticlockwise to open, clockwise to close); one in front of dial)
   - Note: valves are perpendicular to O^2 line when closed, parallel to line when open (See Fig. 3 – valves are open)

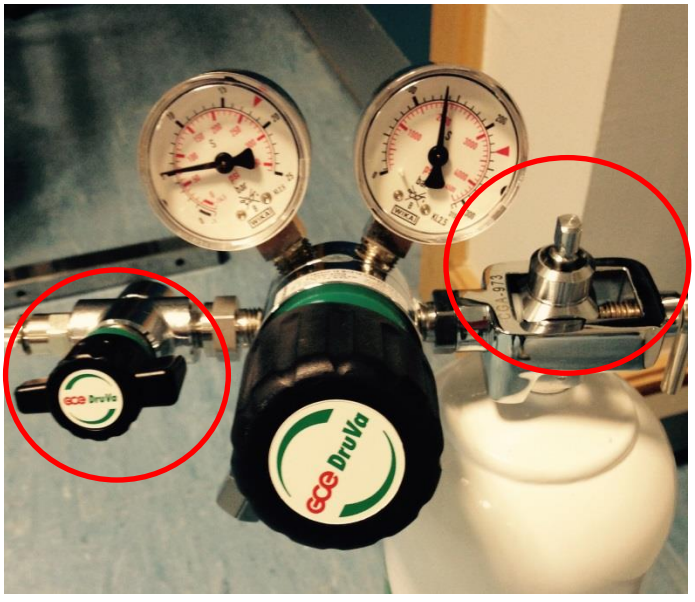


Figure 3 – Valves (open)

   - Press enter
   - Room air calibration will take place as above

- When prompted, insert sampling line into reference gas calibration box and open grey valve
- When calibration is done, press cancel
- Close all three valves, take care when removing O_2 line from box

3. Turbine calibration
    - Attach turbine connectors
    - Insert sampling line into turbine
    - Scroll down to 'Turbine Calibration' and press enter
    - Fit turbine to calibration syringe and operate syringe by smoothly pulling handle fully out and pushing it fully in
    - When calibration is done, press cancel and return syringe to press
4. O_2/CO_2 Delay Calibration
    - Unplug sampling line from turbine
    - Scroll down and press enter
    - Room air calibration will take place as above
    - When prompted, insert sampling line into turbine
    - Place turbine in mouth and press enter
    - Inhale and exhale in sync with the beeps
    - When calibration is done, press cancel
    - Failed – error warning may come up on screen if breaths not synced properly
    - Repeat calibration in this case from number 4. above

<u>Preparation of participant</u>

- Apply some gel to heart rate monitor and fit onto participant with strap
- Fit harness to participant and adjust to fit snugly
- Attach face mask to participant and check for leaks by pressing your hand against the hole at the front – if they can breathe out with your hand blocking the hole you will need to adjust the straps
- Remove face mask to attach turbine
- Insert unit into slot at front of harness – make sure it clicks
- Insert battery into slot at back of harness – again make sure it clicks
- Ensure lines and cables are not tangled*
- Fit face mask onto participant
- Ensure participant is comfortable and can breathe normally

Collecting data

- On laptop click file>patients and click “New”
- Insert participant code, name, DOB and gender and click OK
- Click test and a dialogue box will come up
- Insert participant’s height and weight and press OK
- Ensure Telemetry box is checked and click OK
- Click OK when prompted
- On unit, scroll down to “3. Test”
- “1. Patient’s data” will come up
- Insert patient details (I.D., Height, Weight, Age and Gender) using arrows and press enter
- Scroll down to Start Test
- ‘GPS…’ on screen – press cancel twice
- ‘Set humidity…’ on screen – press enter
- Press enter to start test
- Ensure numbers are going up on screen (not flashing)
- Press marker when starting test
- Data should appear on laptop screen
- N.B. for exercise testing – ensure heart rate is coming up on screen as this will be important for your Max HR cut-off, if not use polar watch to monitor and make note of time when HR cut-off is reached
- Press cancel to end test
- Save (Data can be transferred from the K4b2 unit to the laptop by attaching the ned of the USB cable into the bottom of the unit, selecting your subject’s file from the ‘Patients’ menu on the laptop, and Test>Receive Test>*scroll to appropriate file>Download
- Export to excel and back up data (only you are responsible for what happens to your data so make sure you have it backed up on USBs)

Leaving the lab

- All equipment must be washed and left ready for next user
- Wash turbine, facemask and plastic attachment by soaking for 15 minutes in a jug with one spoon of disinfectant (ensure it is fully dissolved before putting turbine in)
- DO NOT LET SAMPLING LINE GET WET
- Leave to dry on the draining board
- Put $K4b^2$ and all attachments back in the case (Fig. 4)
- Battery units should be left charging if depleted – if not depleted leave the wire from the unit attached to the battery so next user knows it was used

- *N.B* do not leave charger on overnight – use timer if you are going to leave while they are charging (see Fig. 5 for instructions)

Figure 4 – Case layout

- Shut down laptop and store in case
- Put briefcase and box back on shelf
- Leave laptop under desk
- Leave lab neat and tidy
- Switch off lights and lock door

7-Day Digital Timer

Code:**N21JU**

IMPORTANT

Don't plug in an appliance where the load exceeds 13 Amp. Always ensure the plug of any appliance is fully inserted into the timer outlet. If cleaning of the timer is required, remove from mains power and wipe timer with a dry cloth.
DO NOT IMMERSE TIMER IN WATER OR ANY OTHER LIQUID.
Heaters and similar appliances should never be left unattended during operation. The manufacturer recommends such appliances not be connected to timers.

BRIEF INTRODUCTION

1.The Timer has a total of 8 ON/OFF programs.
2.MANUAL ON/AUTO/MANUAL OFF can make settings as required very easily.
3.12/24 hour mode is switchable.
4.Summertime function is very useful for areas with summertime system.
5.Random function to make the settings executed with a random delay of 0-32 minutes.
6.16 combinations of day or blocks of days are available for this Timer as follows:
Mo
Tu
We
Th
Fr
Sa
Su
Mo,Tu,We,Th,Fr,Sa,Su
Mo,Tu,We,Th,Fr
Sa,Su
Mo,Tu,We,Th,Fr,Sa
Mo,We,Fr
Tu,Th,Sa
Mo,Tu,We
Th,Fr,Sa
Mo,We,Fr,Su

INITIAL OPERATION

1.Plug the Timer into a regular 220-240 Volt household power point and turn the power on.
2.Leave for approximately 12 hours to charge the Memory Back Up batteries.
3.After charging clear all current information by pressing the MASTER CLEAR button with a sharp object such as a pen or pencil.
4.The timer is now ready to be set up for use.

SETTING CURRENT TIME

1.Press the CLOCK button and hold, simultaneously press the WEEK button until the actual day is displayed. Continue by pressing HOUR or MINUTE button until the current hour or minute is displayed.When setting, the buttons WEEK, HOURorMINUTE can be held down for rapid forward counting.
2.Release both buttons. The week and time will be set.
3.To reset incorrect time, repeat previous steps.

SETTING PROGRAMS

1.Press the TIMER button and release. The first ON setting can be made now.
2.Press WEEK button to set the day or blocks of days. Set the time by pressing the HOUR button and then MINUTE button.
3.Press the TIMER button again to finish the first ON setting and enter into the first OFF setting. By repeating 2 to make the first OFF setting.
4.Press the Timer button again to finish the first OFF setting and enter into the 2 ON setting. Repeat 2 and 3 to program remaining settings.
5.After completing settings, press the CLOCK button and the TIMER is ready to operate.

EXAMPLE: TIMER ON at 18:15 and TIMER OFF at 22:15 everyday
a. Press TIMER and LCD displays 1_ON
b. Press WEEK till LCD displays"MO,TU,WE,TH,FR,SA,SU"
c. Press HOUR till LCD displays 6:00PM or 18:00
d. Press MINUTE till LCD displays 6:15PM or 18:15
e. Press TIMER again and LCD displays 1_OFF
f. Repeat the above c. and d. till LCD displays 10:15PM or 22:15
j. Press the button "RES/RCL" to stop some programs, press the button "RES/RCL" again to recall the selective programs.
TIP: When verifying your programs ensure that the settings do not overlap, especially when using the block option.

MANUAL ON/AUTO/MANUAL OFF SETTING

1.Press ON/AUTO/OFF button to revert the three mode in turn.
2.In MANUAL ON or MANUAL OFF mode the Timer doesn't operate as settings in AUTO mode.
3.When the mode is turned from MANUAL to AUTO, the Timer will keep the setting of MANUAL until to the next timer setting.

RANDOM FUNCTION

1.Press the RANDOM button in the AUTO mode. The LCD will display RANDOM
2.When this function is ON , the settings will be executed with a random advance of 0-32 minutes botween 6:00PM and 6:00AM.
3.Press the RANDOM button again to cancel this function.

12/24 HOUR MODE

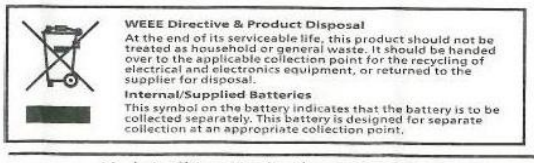
Press CLOCK and TIMER simultaneously to turn between 12 and 24 Hour mode.

SUMMERTIME FUNCTION

1.Press the buttons CLOCK and ON/AUTO/OFF simultaneously in TheAUTO mode. The LCD will show SUMMER.
2.The clock will set back one hour.
3.Press the two buttons again to revert to Wintertime.

SPECIFICATIONS

Voltage	220-240V AC 50Hz
Max Load	13A, 3000W
Min Setting Time	1 Minute
Operating temperature	- 10 to +40°C
Accuracy	2 minute per month
Battery backup	Ni-Mh1.2V>100hours



WEEE Directive & Product Disposal
At the end of its serviceable life, this product should not be treated as household or general waste. It should be handed over to the applicable collection point for the recycling of electrical and electronics equipment, or returned to the supplier for disposal.

Internal/Supplied Batteries
This symbol on the battery indicates that the battery is to be collected separately. This battery is designed for separate collection at an appropriate collection point.

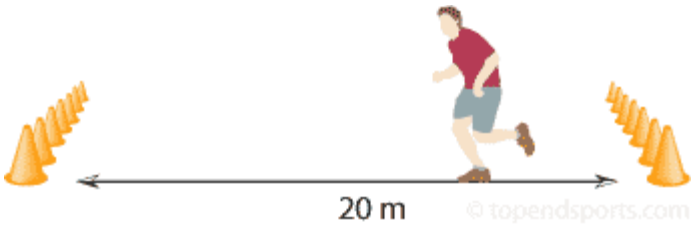
CE

Made in China, Maplin Electronics, S63 5DL, UK. www.Maplin.co.uk

20m Multistage Fitness Test (Beep Test) Instructions

The 20m multistage fitness test (MSFT) is a commonly used maximal running aerobic fitness test. It is also known as the 20 meter shuttle run test, beep or bleep test among other names.

- **Equipment:** Flat, non-slip surface, marking cones, 20m measuring tape, beep test cd, cd player, recording sheets.
- **Procedure:** This test involves continuous running between two lines 20m apart in time to recorded beeps. For this reason the test is also often called the 'beep' or 'bleep' test. The subjects stand behind one of the lines facing the second line, and begin running when instructed by the recording. The speed at the start is quite slow. The subject continues running between the two lines, turning when signalled by the recorded beeps. After about one minute, a sound indicates an increase in speed, and the beeps will be closer together. This continues each minute (level). If the line is reached before the beep sounds, the subject must wait until the beep sounds before continuing. If the line is not reached before the beep sounds, the subject is given a warning and must continue to run to the line, then turn and try to catch up with the pace within two more 'beeps'. The test is stopped if the subject fails to reach the line (within 2 meters) for two consecutive ends after a warning.



- **Scoring:** The participant's score is the level and number of shuttles (20m) reached before they were unable to keep up with the recording. Record the last level completed (not necessarily the level stopped at).

Beep Test Recording Sheet

Date: _____ Time: _____ Conditions: _____

Level 1	1 2 3 4 5 6 7
Level 2	1 2 3 4 5 6 7 8
Level 3	1 2 3 4 5 6 7 8
Level 4	1 2 3 4 5 6 7 8 9
Level 5	1 2 3 4 5 6 7 8 9
Level 6	1 2 3 4 5 6 7 8 9 10
Level 7	1 2 3 4 5 6 7 8 9 10
Level 8	1 2 3 4 5 6 7 8 9 10 11
Level 9	1 2 3 4 5 6 7 8 9 10 11
Level 10	1 2 3 4 5 6 7 8 9 10 11
Level 11	1 2 3 4 5 6 7 8 9 10 11 12
Level 12	1 2 3 4 5 6 7 8 9 10 11 12
Level 13	1 2 3 4 5 6 7 8 9 10 11 12 13
Level 14	1 2 3 4 5 6 7 8 9 10 11 12 13
Level 15	1 2 3 4 5 6 7 8 9 10 11 12 13
Level 16	1 2 3 4 5 6 7 8 9 10 11 12 13 14
Level 17	1 2 3 4 5 6 7 8 9 10 11 12 13 14
Level 18	1 2 3 4 5 6 7 8 9 10 11 12 13 14 15
Level 19	1 2 3 4 5 6 7 8 9 10 11 12 13 14 15
Level 20	1 2 3 4 5 6 7 8 9 10 11 12 13 14 15 16
Level 21	1 2 3 4 5 6 7 8 9 10 11 12 13 14 15 16

* circle the level reached for each participant, and write their name next to that line.

 for detailed instructions for conducting the beep test, see http://www.topendsports.com/testing/tests/20mshuttle.htm

Table 1. The 20 m shuttle run test: prediction of $\dot{V}O_2$max from maximal shuttle run speed and age.

Stage (min)	Max speed (km h^{-1})	Predicted $\dot{V}O_2$max (ml kg^{-1} min^{-1}) according to speed (km h^{-1}) and age (year)												
		6	7	8	9	10	11	12	13	14	15	16	17	⩾18
1	8.5	46.9*	45.0*	43.0*	41.1*	39.1*	37.2*	35.2*	33.3*	31.4*	29.4*	27.5*	25.5*	23.6*
2	9.0	49.0	47.1	45.2	43.4	41.5*	39.6*	37.8*	35.9*	34.1*	32.2*	30.3*	28.5*	26.6*
3	9.5	51.1	49.3	47.5	45.7	43.9	42.1	40.3*	38.5*	36.7*	35.0*	33.2*	31.4	29.6
4	10.0	53.1	51.4	49.7	48.0	46.3	44.6	42.9	41.2	39.4*	37.7*	36.0	34.3	32.6
5	10.5	55.2	53.6	51.9	50.3	48.7	47.0	45.4	43.8	42.1	40.5	38.9	37.2	35.6
6	11.0	57.3	55.7	54.2	52.6	51.1	49.5	47.9	46.4	44.8	43.3	41.7	40.2	38.6
7	11.5	59.4	57.9	56.4	54.9	53.4	52.0	50.5	49.0	47.5	46.0	44.6	43.1	41.6
8	12.0	61.5	60.1	58.6	57.2	55.8	54.4	53.0	51.6	50.2	48.8	47.4	46.0	44.6
9	12.5	63.5	62.2	60.9	59.6	58.2	56.9	55.6	54.2	52.9	51.6	50.3	48.9	47.6
10	13.0	65.6*	64.4*	63.1*	61.9*	60.6*	59.4*	58.1	56.9	55.6	54.4	53.1	51.9	50.6
11	13.5	67.7*	66.5*	65.3*	64.2*	63.0*	61.8*	60.6*	59.5*	58.3	57.1	56.0	54.8	53.6
12	14.0	69.8*	68.7*	67.6*	66.5*	65.4*	64.3*	63.2*	62.1*	61.0	59.9	58.8	57.7	56.6
13	14.5	71.9*	70.8*	69.8*	68.8*	67.8*	66.8*	65.7*	64.7*	63.7*	62.7*	61.6	60.6	59.6
14	15.0	73.9*	73.0*	72.0*	71.1*	70.2*	69.2*	68.3*	67.3*	66.4*	65.4*	64.5	63.6	62.6*
15	15.5	76.0*	75.1*	74.3*	73.4*	72.5*	71.7*	70.8*	69.9*	69.1*	68.2*	67.3*	66.5*	65.6*
16	16.0	78.1*	77.3*	76.5*	75.7*	74.9*	74.1*	73.4*	72.6*	71.8*	71.0*	70.2*	69.4*	68.6*
17	16.5	80.2*	79.5*	78.7*	78.0*	77.3*	76.6*	75.9*	75.2*	74.5*	73.8*	73.0*	72.3*	71.6*
18	17.0	82.3*	81.6*	81.0*	80.3*	79.7*	79.1*	78.4*	77.8*	77.2*	76.5*	75.9*	75.3*	74.6*
19	17.5	84.3*	83.8*	83.2*	82.7*	82.1*	81.5*	81.0*	80.4*	79.9*	79.3*	78.7*	78.2*	77.6*
20	18.0	86.4*	85.9*	85.4*	85.0*	84.5*	84.0*	83.5*	83.0*	82.5*	82.1*	81.6*	81.1*	80.6*

*Values extrapolated beyond the ±2 s experimental range of data.

Note 1. Predicted $\dot{V}O_2$max (Y, ml kg^{-1} min^{-1}) is obtained from maximal shuttle run speed (X_1, km h^{-1}) and age (X_2, year rounded to the lower integer) from the following regressions: 6–18 years old: $Y = 31.025 + 3.238X_1 - 3.248X_2 + 0.1536X_1X_2$; 18 years old and over: $Y = -24.4 + 6.0X_1$.

Note 2. Stage numbers are announced every 30 s on the tape. Predicted $\dot{V}O_2$max could thus be obtained immediately upon termination of the test from this table.

Typographic error: -27.4, not -24.4

Clinical Assessment

Subject Code:_____

Subject DOB:_____

Gender:_____

Ax Date:_____

Researcher:_____

Actigraph #:_____ BF%_____

GAIT

Trial No.	Speed

ANTHROPOMETRIC DATA (all in mm except height and weight):

Height		
Weight		
Pelvic width		
Pelvic depth		
Ankle joint width	L	R
Knee joint width	L	R
Thigh length	L	R
Shank length	L	R
Foot length	L	R
Leg length	L	R

PASSIVE RANGE OF MOTION DATA (in degrees):

Joint	Movement	Passive ROM
Ankle	Dorsiflexion	
	Plantarflexion	
Knee	Extension	
	Flexion	
Hip	Extension	
	Flexion	
	Abduction	
	Internal rotation	
	External rotation	

SKELETAL OBSERVATIONS:

Genu Valgum	L	R
Genu Varum	L	R
Femoral Torsion	L	R

Appendix VII – Active Video Game Questionnaire
(Cuisle Forde, née O'Donovan, PhD 2013)

Confidential questionnaire on leisure time activities and active computer game use among children in 6th class.

We are asking you to fill in this questionnaire because we want to know how active you are, and how much time you spend playing computer games and watching the television. We will be asking children in lots of different schools to complete this questionnaire and hope that the answers will help us understand how children in 6th class spend their time.

Please read each question carefully before you answer.

Write in or tick (√) the answers.

School Name:_____

Today's Date: (dd/mm/yyyy) _____

Your date of birth: (dd/mm/yyyy) _____

Your age: _____

Gender: ( ) Male ( ) Female

1. Do you walk to and from school?

( ) No

( ) Yes

( ) Sometimes

( ) One way only

2. How long does it take you to walk to school (one way)?

_____ minutes.

( ) I never walk to school.

3. Do you cycle to and from school?

( ) Yes

( ) No

( ) Sometimes

4. How long does it take you to cycle to school (one way)?

_____ minutes

( ) I never cycle to school

5. Tick all activities that you did at least 10 times in the **PAST YEAR**. Do **not** include time spent in school physical education classes. Make sure you include all sport teams that you participated in during the last year.

( ) Basketball	( ) Gymnastics	( ) Swimming (laps)
( ) Bicycling	( ) Hurling	( ) Swimming (free swim)
( ) Ballet	( ) Rollerblading	( ) Camogie
( ) Dance classes	( ) Skateboarding	( ) Running for exercise
( ) Gaelic Football	( ) Soccer/Football	

If there is one or more activities you did at least 10 times in the last year that are **not listed above** please write them on the lines provided below:

_____

_____

_____

_____

6.

- List each activity that you have ticked or written in question 5 in the table below.
- Write down as best as you can amount of time spent in each activity.
- The first line is filled in as an example.

Write in the **activities** you do in the boxes below.↓	How many **months in the last year** did you do this activity? (Write in a number between 1 and 12)	How many **weeks in a month** did you do this activity? (Write in a number between 1 and 4)	How many **days in a week** did you do this activity? (Write in a number between 1 and 7)	How many **minutes a day** would you spend doing this activity?
Soccer	9	4	2	60

7. In an **average school week** how much time do you spend in PE class?

( ) None

( ) Up to 30 minutes a week

( ) Up to 1 hour a week

( ) Between 1 and 2 hours a week

( ) Between 2 and 3 hours a week

( ) Over 3 hours a week

8. In the **last 7 days** (don't include today), how much time did you spend watching television? (Include times before and after school. Include time spent watching movies at home, e.g. watching a DVD).

( ) None, I do not have access to a television.

( ) None, I have access to a television but I did not watch any in the last 7 days.

( ) In the last 7 days I watched over 20 hours of television.

( ) In the last 7 days I watched between 14 and 20 hours of television.

( ) In the last 7 days I watched between 10 and 14 hours of television.

( ) In the last 7 days I watched between 5 and 10 hours of television.

( ) In the last 7 days I watched less than 5 hour of television.

9. What **electronic game consoles** do you have access to at home? Tick as many as required.

( ) I do not have any electronic games at home

( ) Nintendo Wii	( ) Nintendo DS
( ) X-Box	( ) Game Cube
( ) Play-Station 2	( ) Sony Eye-Toy
( ) Play-Station 3	( ) Applications/Games on iTouch/Mobile phone
( ) Lap-top/PC	( ) Dance Dance Revolution

( ) X-Box Kinect

( ) Other, please specify in space provided: _____

10. In the **last 7 days** (don't include today), how much time did you spend playing electronic games? (Include computer games. Include portable games like the Nintendo DS or applications on an iTouch or a mobile phone. Do **not** include social networking sites like Facebook.)

( ) None, I do not have any electronic games

( ) None, I have access to electronic games but I did not play them in the last 7 days

( ) In the last 7 days I spent over 14 hours playing electronic games

( ) In the last 7 days I spent between 10 and 14 hours playing electronic games

( ) In the last 7 days I spent between 6 and 10 hours playing electronic games

( ) In the last 7 days I spent between 4 and 6 hours playing electronic games

( ) In the last 7 days I spent between 2 and 4 hours playing electronic games

( ) In the last 7 days I spent between 1 and 2 hours playing electronic games

( ) In the last 7 days I spent less than 1 hour playing electronic games

11. Do you have access to social networking sites? (This includes sites such as Bebo, Facebook, Myspace, and StarDoll).

( ) Yes ( ) No

12. How much time in the **last 7 days** (don't include today), did you spend on social networking sites?

( ) None, I do not have access to social networking sites

( ) None, I have access to but do not use social networking sites

( ) None, I have access to and use social networking sites, but have not used them in the last 7 days

( ) In the last 7 days I spent less than 1 hour on social networking sites

( ) In the last 7 days I spent between 1 and 2 hours on social networking sites

( ) In the last 7 days I spent between 2 and 4 hours on social networking sites

( ) In the last 7 days I spent between 4 and 6 hours on social networking sites

( ) In the last 7 days I spent between 6 and 10 hours on social networking sites

( ) In the last 7 days I spent between 10 and 14 hours on social networking sites

( ) In the last 7 days I spent over 14 hours on social networking sites

13. How much time have you spent using a computer in the **last 7 days,** (don't include today)? (Include time spent on the internet, or typing. Do **not** include time spent doing school work on a computer. Do **not** include playing computer games, or time on social networking sites like facebook.)

( ) None, I do not have access to a computer

( ) None I have access to a computer but did not use it for these reasons in the last 7 days

( ) In the last 7 days I spent over 14 hours using a computer

( ) In the last 7 days I spent between 10 and 14 hours using a computer

( ) In the last 7 days I spent between 6 and 10 hours using a computer

( ) In the last 7 days I spent between 4 and 6 hours using a computer

( ) In the last 7 days I spent between 2 and 4 hours using a computer

( ) In the last 7 days I spent between 1 and 2 hours using a computer

( ) In the last 7 days I spent less than 1 hour using a computer

This part of the questionnaire is about

<u>activity video game consoles ONLY.</u>

- **Activity video games are video games that ask you to move your body to play them.**
  For example Nintendo Wii Sports and Xbox Kinect Adventures.
- **If you stand up and move your arms or legs to play a game, it is called an activity video game.**
- **Games where you don't have to move your body much can also be played on activity video game consoles.**
  For example Wii Mario Kart.

14. For each **activity video game console** listed below, please tick whether or not you have ever played it (even if you only played it once), if you have access to it at home, and if so how long you have had access to it at home.

Activity video game	**Have you ever played this console?**		**Have you access to this console at home?**		**How long have you had this video game console at home? (Tick one box below)**						
	Yes	No	Yes	No	I don't have this at home	Less than 1 month	1 to 2 months	2 to 6 months	6 to 12 months	1 to 2 years	Over 2 years
Nintendo Wii											
Playstation Move											
Xbox Kinect											
Dance Dance Revolution, home version											
Sony EyeToy											
Other, please specify game/console:_____											

15. For each **activity video game console** listed below, please tick how much time you spent playing it in **the first 7 days** you had access to it at home. For example, if you got a console for Christmas last year, write down how much time you spent playing it in the week following Christmas.

Activity video game	How much time did you spend playing this activity video game **in the first 7 days you had access to it at home?**									
	None, I have never had this console	None, but I have, (or used to have) this console	Less than 10 minutes	10 to 30 minutes	30 to 60 minutes	1 to 2 hours	2 to 4 hours	4 to 6 hours	6 to 8 hours	Over 8 hours
Nintendo Wii										
Playstation Move										
Xbox Kinect										
Dance Dance Revolution, home version										
Sony EyeToy										
Other, please specify game/console:_____										

15. In the last 7 days how much time did you spend playing **activity games** on an activity video game console. This means you were **standing up** while playing and moving either your arms or legs or both (e.g. Wii Sports, Xbox Kinect Adventures, Dance games).

( ) None, I do not have access to activity video games.
( ) None, but I do have access to activity video games.
( ) Less than 10 minutes in the last 7 days
( ) Between 10 and 30 minutes in the last 7 days
( ) Between 30 and 60 minutes in the last 7 days
( ) Between 1 and 2 hours in the last 7 days
( ) Between 2 and 4 hours in the last 7 days
( ) Between 4 and 7 hours in the last 7 days
( ) Between 7 and 14 hours in the last 7 days
( ) Over 14 hours in the last 7 days

16. In the last 7 days how much time did you spend playing **non-active games on an activity video game console**, where you were **sitting down** while playing (e.g. Nintendo Wii Mario Brothers).

( ) None, I do not have access to activity video games.
( ) None, but I do have access to activity video games.
( ) Less than 10 minutes in the last 7 days
( ) Between 10 and 30 minutes in the last 7 days
( ) Between 30 and 60 minutes in the last 7 days
( ) Between 1 and 2 hours in the last 7 days
( ) Between 2 and 4 hours in the last 7 days
( ) Between 4 and 7 hours in the last 7 days
( ) Between 7 and 14 hours in the last 7 days
( ) Over 14 hours in the last 7 days

Thank you for taking the time to complete this questionnaire.

Physical Activity in School-age Children with ASD and/or Dyspraxia

General Information

1. Please state your child's gender.

- ◯ Male
- ◯ Female

2. What age is your child? Please give age in years and months.

3. Please tick your child's diagnosis.

- ☐ Autism Spectrum Disorder
- ☐ Dyspraxia/Developmental Coordination Disorder
- ☐ Typical Development - no diagnosis of any condition

Physical Activity in School-age Children with ASD and/or Dyspraxia

School-based Physical Activity

4. How does your child get to school?

- ◯ Walk
- ◯ Cycle
- ◯ Car
- ◯ Public Transport
- ◯ Other (please specify)

5. In the box below, please give details of how long (in minutes) it takes for your child to walk or cycle to school. If your child does not walk or cycle to school, please insert 'N/A' into the box below.

6. In an average school week, how much time does your child spend in P.E. class?

- ○ None
- ○ Up to 30 minutes a week
- ○ Up to an hour a week
- ○ 1-2 hours a week
- ○ 2-3 hours a week
- ○ More than 3 hours a week

Physical Activity in School-age Children with ASD and/or Dyspraxia

Physical Activity

7. Please tick all the activites that your child did at least 10 times in the past year. Do not include time spent in P.E. class. Make sure to include all sports teams your child took part in at least 10 times in the past year.

- ☐ Athletics
- ☐ Basketball
- ☐ Camogie
- ☐ Cycling
- ☐ Dance
- ☐ Gaelic Football
- ☐ Gymnastics
- ☐ Hockey
- ☐ Hurling
- ☐ Martial Arts
- ☐ Rollerblading
- ☐ Rugby
- ☐ Skateboarding
- ☐ Soccer
- ☐ Swimming (laps)
- ☐ Swimming (free swim)
- ☐ Other (please specify)

8. In an average week, how much time, in total, would your child spend taking part in the activities from question 7?

- ◯ None
- ◯ Less than 1 hour a week
- ◯ 1-2 hours a week
- ◯ 2-4 hours a week
- ◯ 4-6 hours a week
- ◯ 6-10 hours a week
- ◯ 10-14 hours a week
- ◯ Over 14 hours a week

Physical Activity in School-age Children with ASD and/or Dyspraxia

Sedentary Behaviours

9. In the last 7 days (not including today), which of the following activites has your child taken part in?

- ☐ Watched television (including movies/DVDs)
- ☐ Used a computer or tablet
- ☐ Played electronic games consoles (e.g. PlayStation, Xbox etc.)
- ☐ Played handheld electronic games consoles (e.g. DS, PSP etc.)
- ☐ Read a book
- ☐ None of the above
- ☐ If your child did more than one activity listed above, please state the activity he/she spent the most amount of time doing in the box below.

10. In the last 7 days, how much time, in total, did your child spend in the activities listed in question 9?

- ◯ None
- ◯ Less than 1 hour
- ◯ 1-2 hours
- ◯ 2-4 hours
- ◯ 4-6 hours
- ◯ 6-10 hours
- ◯ 10-14 hours
- ◯ More than 14 hours

Physical Activity in School-age Children with ASD and/or Dyspraxia

Thank you for taking the time to complete this survey...

11. If you would like more information about our research, please put your contact details in the boxes below.

Name	
Email address	
Phone number	

Participant Information Leaflet

ActiGraph Activity Monitor

Thank you for agreeing to wear the ActiGraph Activity Monitor. The ActiGraph measures your physical activity levels and provides us with information on the about of time you spend engaging in different intensities of activity. The following information leaflet addresses some frequently asked questions. Should you have any queries please contact the Physiotherapy Postgraduate and Research Room at the Trinity Centre for Health Sciences, St. James's Hospital on 01-8963613.

1. **How many days do I wear the monitor?**

   You are requested to wear the activity monitor for one week (7 days) during waking hours.

2. **Do I wear the monitor to bed?**

   No. You put the monitor on first thing in the morning and take it off last thing at night. You are requested to record the time you put the monitor on in the morning and the time you take if off at night in the activity diary provided.

3. **Do I wear the monitor in the shower?**

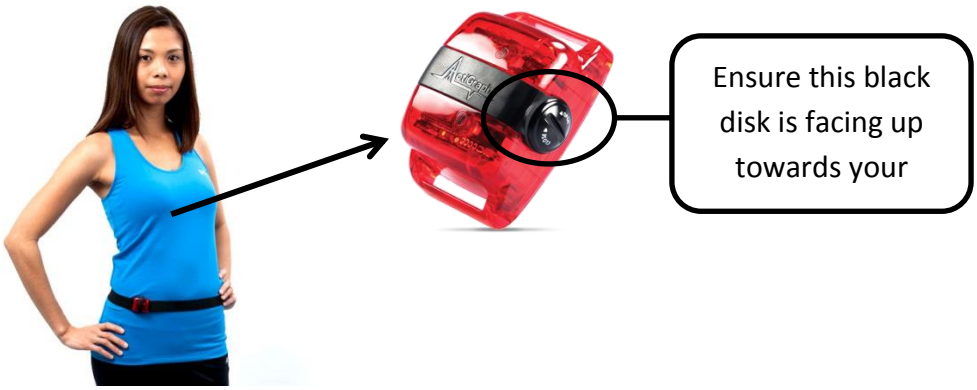
   No. You should remove the monitor during any water-based activity such as showering, bathing or swimming. You are requested to record these activities, including the times your take the monitor on and off in the activity diary provided.

4. **Do I need to press any button to start / finish the monitor?**

   No. The monitor is set-up by the researcher leading your study. You do not have to press any button to activate or stop the monitor.

5. **Where on my body is the monitor worn?**

The monitor is connected to a flexible strap with a clip. The strap should be worn like a belt around your waist with the monitor sitting at hip level on the right side of your body (see picture). Ensure the black disk on the side of the monitor is pointing towards your head. The strap should not be too tight or too loose. You can adjust the strap size if necessary. You may wear the monitor under or over your clothes.



6. **Do I need to charge the monitor during the week?**

No. Do not plug the monitor into any power source or connect to any USB cable during the week and this may wipe the data collected.

7. **I forgot to wear the monitor – what should I do?**

If you forget to wear the activity monitor on a particular day don't worry. Please write down clearly in the activity diary which day you forgot to wear the monitor and just carry on wearing it as normal the following day.

8. **What should I do when I finish wearing the activity monitor?**

When you finish wearing the monitor please return it to us in the stamped addressed envelope provided. Please return the monitor to us as soon as possible to ensure that the battery does not die before we receive it.

Try not to change your activity levels while wearing the monitor as our aim is to get an idea of <u>normal</u> activity patterns

Thank you very much for recording your physical activity

Physical Activity Diary

You are requested to wear your ActiGraph Activity Monitor during **all waking hours**. You will have to remove the activity monitor when you are going to bed or during water-based activities such as showering or swimming. Please record the time you put the activity monitor and the time you take it off in the following activity diary. If you forget to wear the monitor for a day please record this clearly in the activity diary. This record will help us to analyse your physical activity data as accurately as possible.

Should you have any queries please contact the Physiotherapy Postgraduate and Research Room at the Trinity Centre for Health Sciences, St. James's Hospital on 01-8963613. The following example outlines the details required.

Example:

On Date	On Time	Off Date	Off Time	Activity completed while not wearing the monitor
04.10.2013	*8.20am*	*04.10.2013*	*7.10pm*	*Shower*
04.10.2013	*7.30pm*	*04.10.2013*	*10.30pm*	*Sleeping in bed*
05.10.2013	*8.10am*	*05.10.2013*	*10.50pm*	*Sleeping in bed*

Log

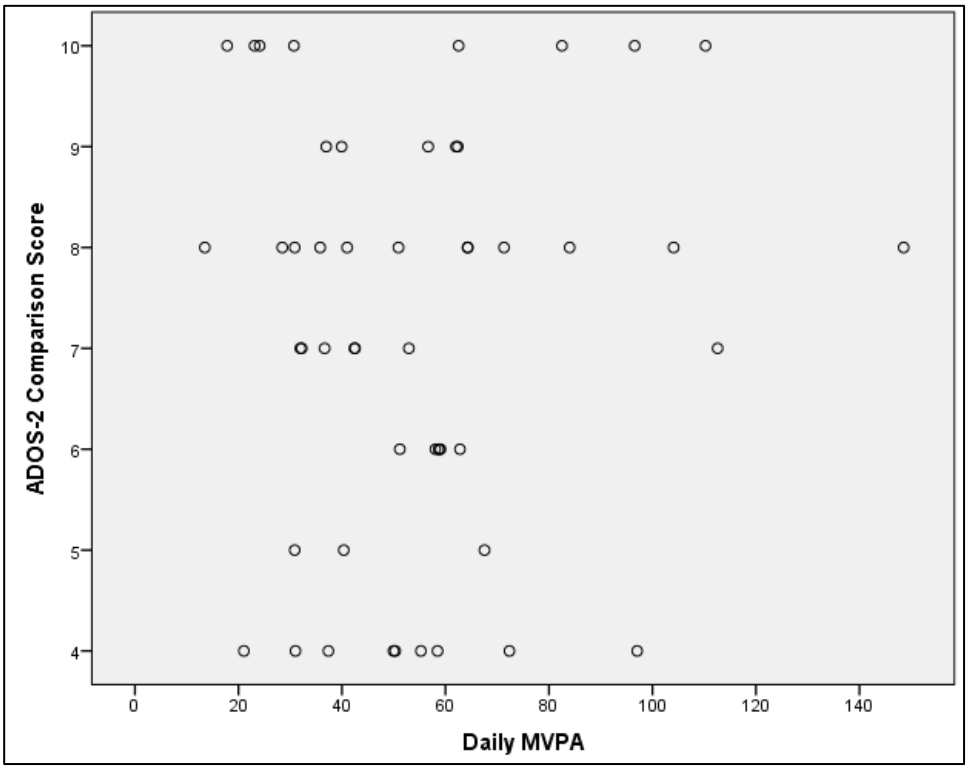
Participants Name/Study ID: _____

On Date	On Time	Off Date	Off Time	Activity completed while not wearing the monitor

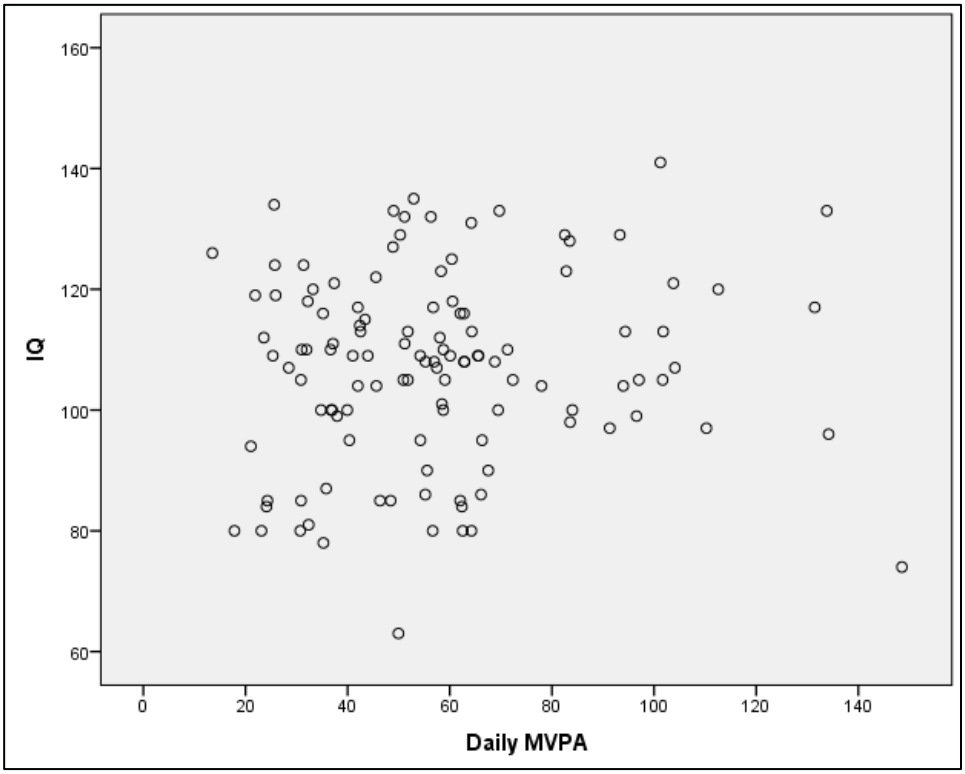
Thank you for taking the time to record your physical activity.

Appendix X – Examples of Scatter Plots from Chapter 5 and Chapter 6

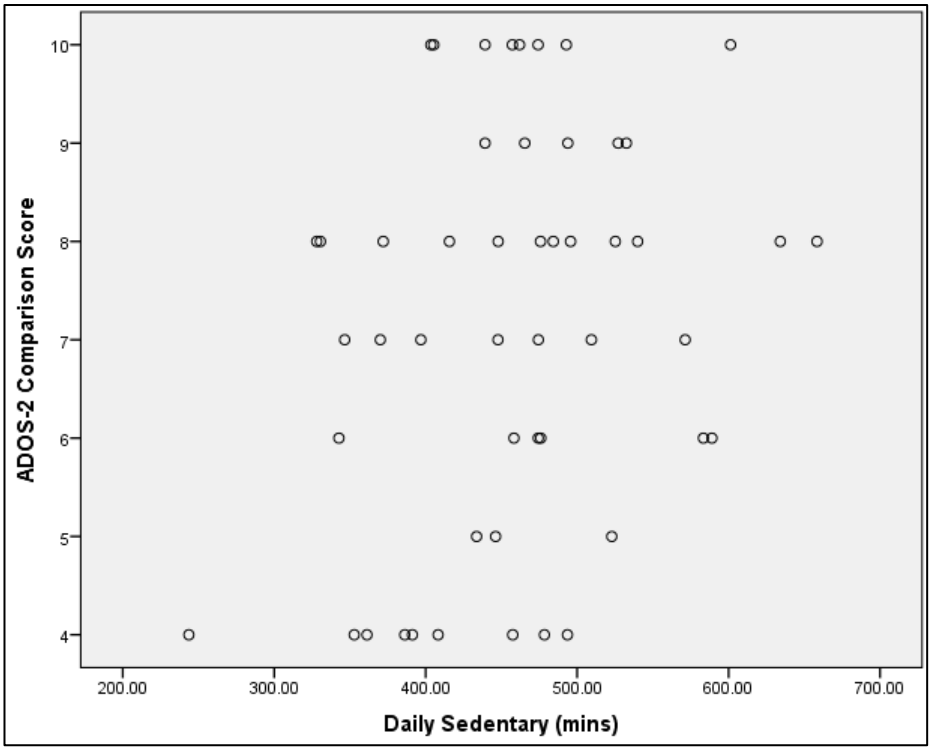
Example Plot 1: ADOS-2 Comparison Score V Daily MVPA



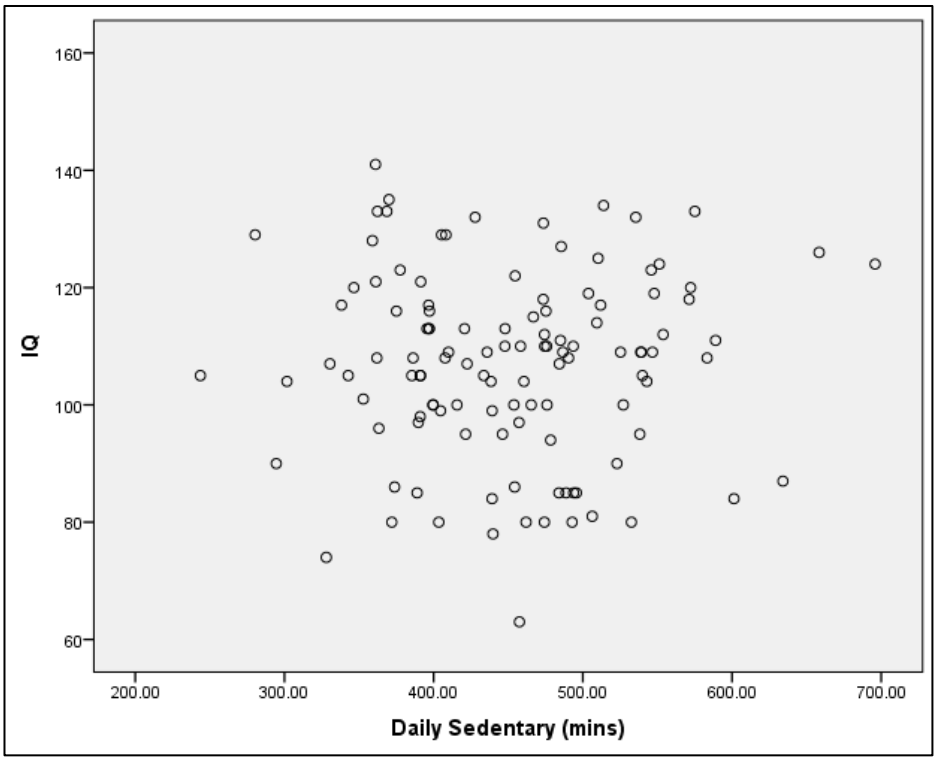
Example Plot 2: IQ Score V Daily MVPA



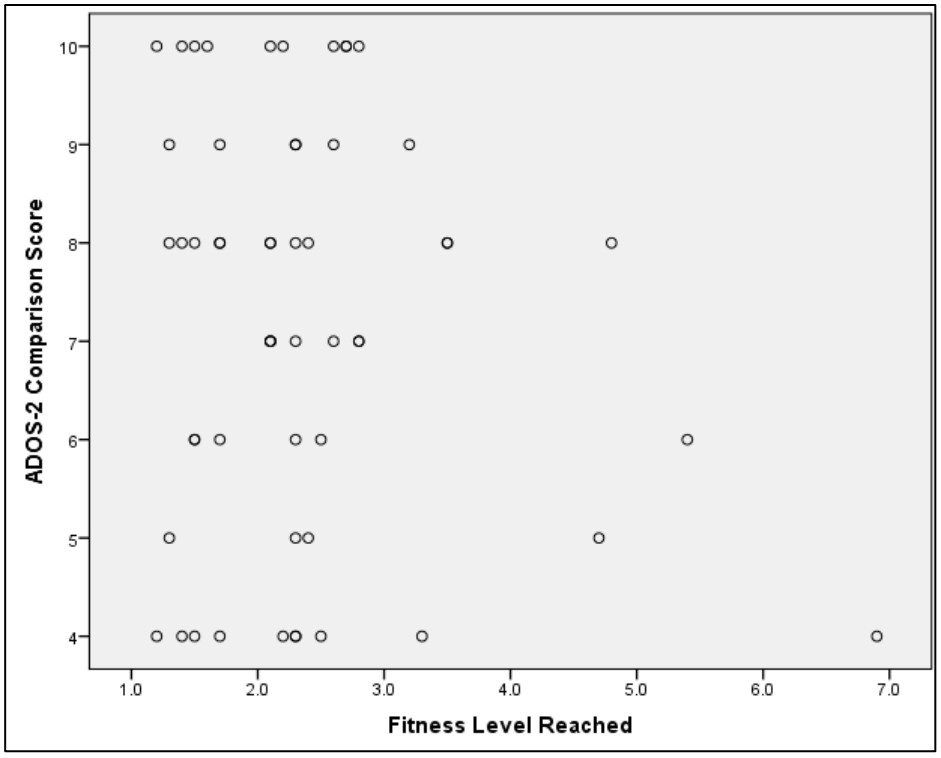
Example Plot 3: ADOS-2 Comparison Score V Daily Sedentary



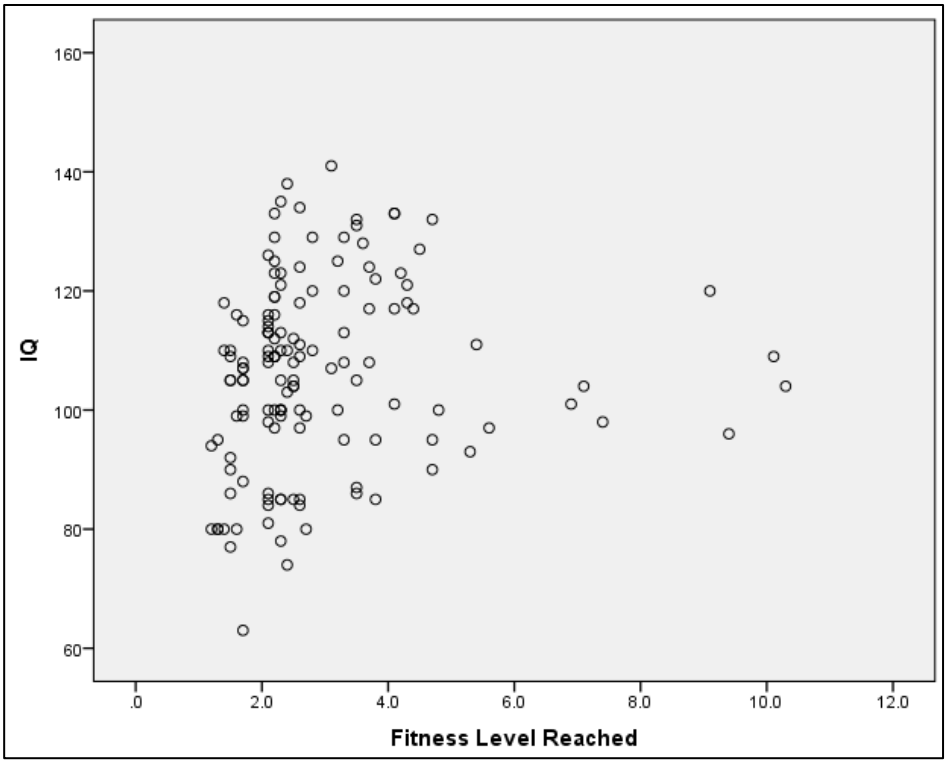
Example Plot 4: IQ Score V Daily Sedentary



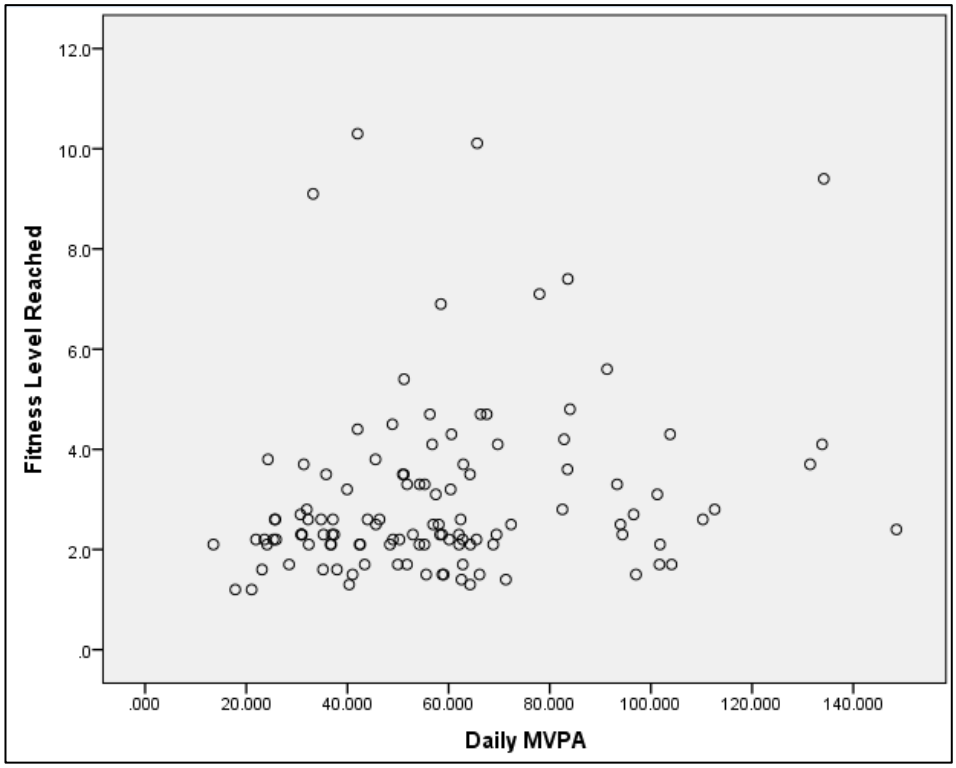
Example Plot 5: ADOS-2 Comparison Score V Fitness Test Score



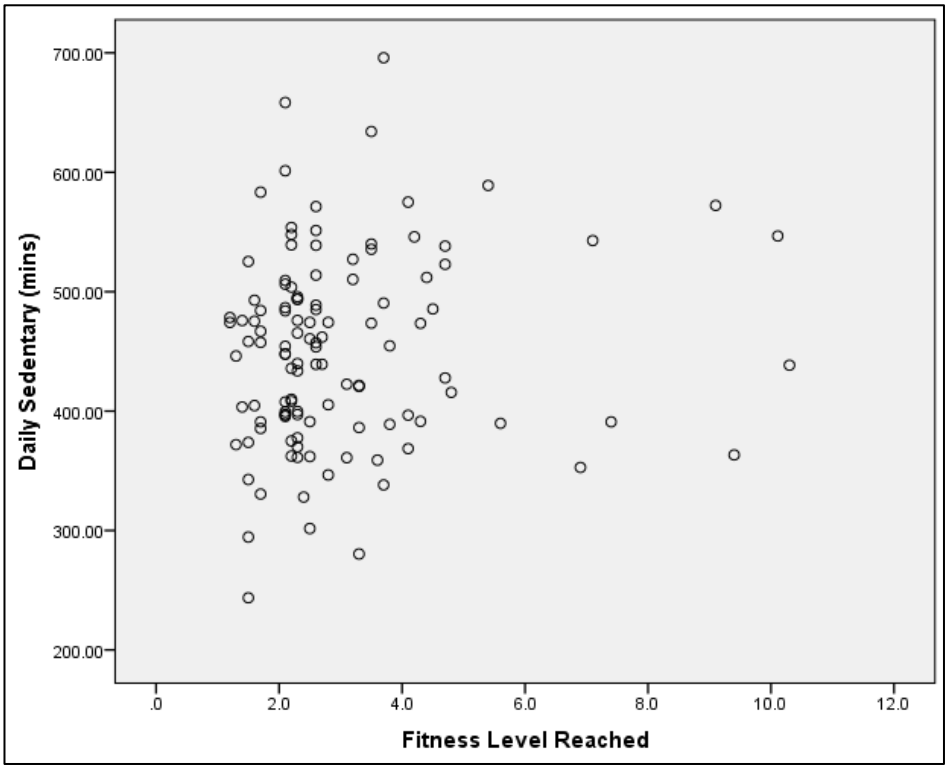
Example Plot 6: IQ Score V Fitness Test Score



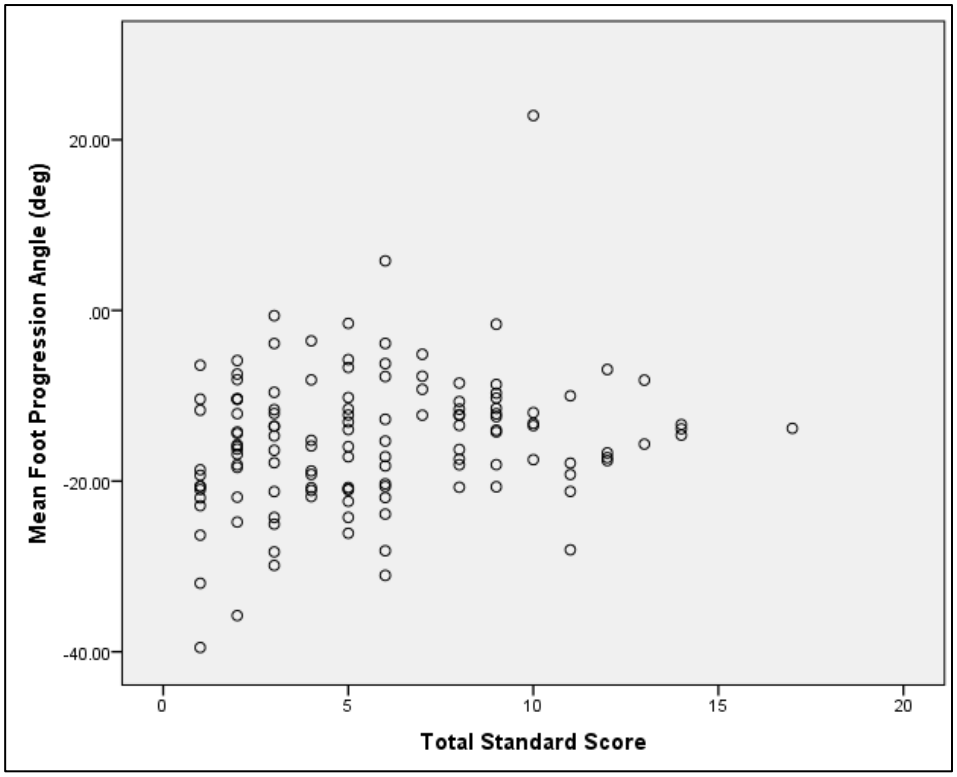
Example Plot 5: Fitness Test Score V Daily MVPA



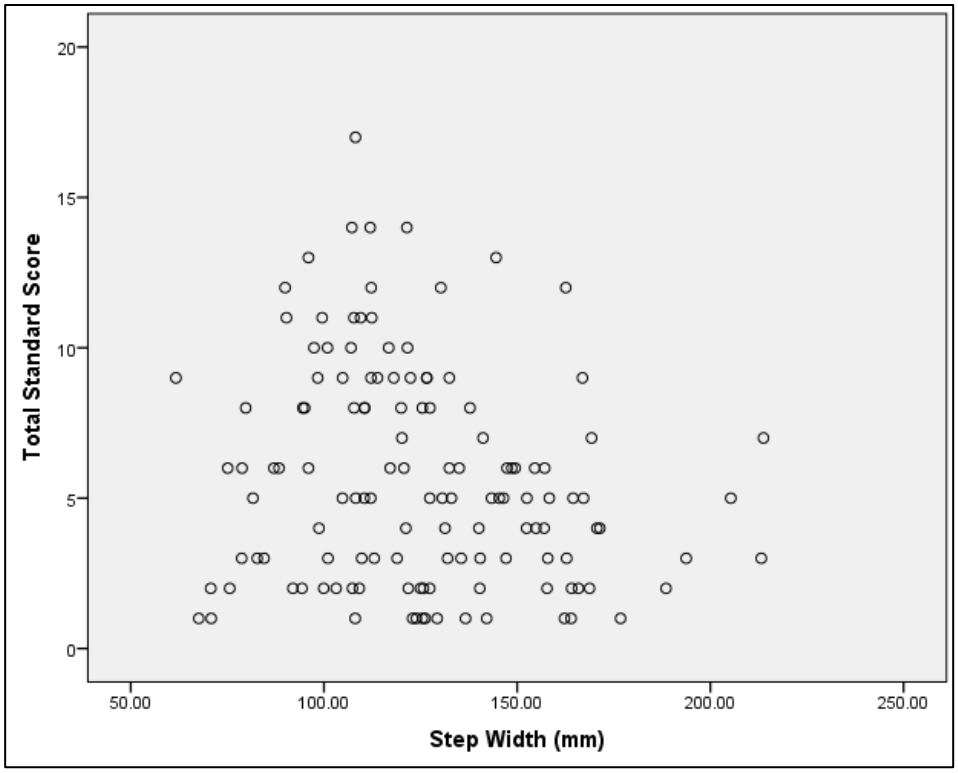
Example Plot 6: Daily Sedentary V Fitness Test Score



Example Plot 7: Foot Progression V Movement ABC-2 Scores



Example Plot 8: Movement ABC-2 Score V Step Width



Appendix XI – Recruitment Materials for Chapter 7

EMAIL TO PARTICIPANTS - TEMPLATE

Dear Sir/Madam,

I am writing to you on behalf of Deirdre Kindregan, who is carrying out research in our group.

Deirdre is a chartered physiotherapist who is beginning the process of developing a questionnaire to examine the perceived benefits and barriers to exercise in children with neurodevelopmental disorders. As part of this process, she will be running focus groups for the purpose of gaining insight into the opinions of professionals who work with children with neurodevelopmental disorders, parents of children with neurodevelopmental disorders and adolescents with diagnoses of one or more neurodevelopmental disorder.

She would like to invite you/your child to take part in these focus groups. Participation would involve one visit to the Trinity Centre for Health Sciences for approximately two hours. The information leaflet is attached. Participation is on a voluntary basis and you/your child may withdraw at any time.

If you would like more information, please contact Deirdre directly by email at kindregd@tcd.ie or telephone on (01) 896 3613.

Best regards,

Ana McLaughlin
Autism Research Group

INFORMATION LEAFLET: PROFESSIONALS

Trinity College
The University of Dublin

Project Title:

Development of a questionnaire on benefits and barriers to exercise in children with neurodevelopmental disorders.

Aims and Objectives:

This study aims to determine perspectives of professionals, parents and adolescents on benefits and barriers to exercise in children with neurodevelopmental disorders, and to use the information obtained during the focus groups to develop a parent-reported questionnaire.

Participation:

If you would like to participate, you would be asked to attend the Trinity Centre for Health Sciences, on the grounds of St James's Hospital, on one occasion. You would take part in a focus group of peers and colleagues and you would be asked to give your opinion on benefits of and barriers to exercise in children with neurodevelopmental disorders, based on your clinical experience.

The session will last approximately two hours.

Any opinion you express during the focus group will be respected.

You will be given the opportunity to ask any questions you may have, and you have the right to withdraw from participation at any stage.

Data and consent forms will be retained after the study reaches completion but will only be used in related studies and will be fully anonymous.

Contact:

If you would like to participate, or would like more information, please contact Deirdre at kindregd@tcd.ie or on (01) 896 3613.

INFORMATION LEAFLET: PARENTS

Trinity College
The University of Dublin

Project Title:

Development of a questionnaire on benefits and barriers to exercise in children with neurodevelopmental disorders.

Aims and Objectives:

This study aims to determine perspectives of professionals, parents and adolescents on benefits and barriers to exercise in children with neurodevelopmental disorders, and to use the information obtained during the focus groups to develop a parent-reported questionnaire.

Participation:

If you would like to participate, you would be asked to attend the Trinity Centre for Health Sciences, on the grounds of St James's Hospital, on one occasion. You would take part in a focus group of other parents and you would be asked to give your opinion on benefits of and barriers to exercise in children with neurodevelopmental disorders, based on your own experience.

The session will last approximately two hours.

Any opinion you express during the focus group will be respected.

You will be given the opportunity to ask any questions you may have, and you have the right to withdraw from participation at any stage.

Data and consent forms will be retained after the study reaches completion but will only be used in related studies and will be fully anonymous.

Contact:

If you would like to participate, or would like more information, please contact Deirdre at kindregd@tcd.ie or on (01) 896 3613.

Trinity College Dublin
Coláiste na Tríonóide, Baile Átha Cliath
The University of Dublin

INFORMED CONSENT FORM

PROJECT: Focus groups on benefits and barriers to exercise in children with neurodevelopmental disorders.

LEAD INVESTIGATORS: Ms Deirdre Kindregan

ACADEMIC SUPERVISORS: Dr John Gormley, Prof Louise Gallagher

BACKGROUND:

This study aims to determine perspectives of professionals, parents and adults with neurodevelopmental disorders on benefits and barriers to exercise in children with neurodevelopmental disorders, and to use the information obtained during the focus groups to develop a parent-reported questionnaire. The focus groups will be audio-recorded and you may request a copy of the transcript by contacting Deirdre after the focus group.

DECLARATION:

I have read, or had read to me, the information leaflet for this project and I understand the contents. I have had the opportunity to ask questions and all my questions have been answered to my satisfaction. I freely and voluntarily agree to be part of this research study, though without prejudice to my legal and ethical rights. I understand that I may withdraw from the study at any time and I have received a copy of this agreement.

I understand data, including audio recordings, and consent forms will be retained after the study reaches completion and that data will only be used in related studies.

PARTICIPANT'S NAME: ……

PARTICIPANT'S SIGNATURE: ……

Date: ……

Identifying Benefits of and Barriers to Exercise in Children with Neurodevelopmental Disorders

Purpose of this focus group exercise: To better understand factors that may promote or prevent physical activity and exercise for children with neurodevelopmental disorders.

Time required: 90 minutes

Items required: Poster boards and markers

Outline:

1. **Registration and Consent (5 minutes)**
2. **Introductions and establishing ground rules (5 minutes)**
   i.e. everyone should feel free to share, avoid interrupting, etc.

3. **Appreciative Inquiry Interview (5 to 10 minutes)**
   - Find a partner (not a friend or colleague)
   - Share a story about hobbies, those involving physical activity or not
   - Share opinions on the benefits and barriers to exercise in own life
   - Have a few people report to get examples

4. **Brainstorming (30 minutes)**
   Participants will first be asked to answer in relation to adults, then repeat exercise for children, and again for children with neurodevelopmental disorders
   - What are the recommended guidelines for exercise?
   - What benefits can exercise have?
   - What stops people exercising?

5. **Scenarios (20 minutes)**

   Focus group members are asked to discuss the following scenarios and identify benefits and barriers to physical activity in each one.

   - 8 year old boy loves sports but isn't very good at them. He gets a diagnosis of dyspraxia and then doesn't want to play anything anymore. Discuss.
   - 13 year old girl with ASD and anxiety has done ballet since she was 6. The other people in her group start socialising outside of ballet but she is not comfortable doing that. Discuss.
   - 15 year old boy with ASD does not want to leave the house after school and spends all his time playing computer games. Discuss
   - 10 year old boy with ADHD has started acting aggressively at school and at home. Discuss.

6. **Reflections**

   Focus group members are asked to reflect on how they feel about the topic after the discussions

   - What points were most interesting?
   - Has your opinion changed on the subject as a result of the discussions?

7. **Closing and Feedback**

   - Have participants fill in an anonymous feedback form in which they write down the three concepts that were the most important to them
   - Thank participants for taking part

Appendix XIII – Final Version of Codebook for Thematic Analysis (Chapter 7)

Barriers: Intrinsic

Name	Description	Code
Physical deficit	Physical factors which lead to poor performance e.g. poor coordination or lack of skill	BI1
Lack of interest	No desire to take part in physical activity	BI2
Anxiety	The feeling of dread or fear associated with physical activity or accompanying socialisation	BI3
Self-consciousness	Awareness of ones strengths and weaknesses, lack of self-confidence	BI4
Diagnosis	The mental or emotional effect of the knowledge of one's own diagnosis of ASD/DCD	BI5
Sensory issues	Sensitivity of a child to environmental factors such as noise, light, texture of clothing	BI6
Difficulty with social interaction	Social impairment of children with neurodevelopmental disorders leading to isolation	BI7

Barriers: Environmental:

Name	Description	Code
Lack of Exposure	Lack of opportunity or experience of a variety of different exercise activities	BE1
Weather	The effect of bad weather on the availability of or desire to take part in physical activities	BE2
Screen time	The distraction of or preference to use devices such as tablets, computers, television	BE3
Lack of resources	Unavailability of or limited access to appropriate facilities for exercise/services	BE4
Exclusion	No allowance of children with these conditions in clubs or areas	BE5
Attitudes of others	Treatment of children with conditions by people outside the family e.g. other parents or other children	BE6

The competitive nature of sport	The focus of many exercise/PA groups being on winning/losing or how good one is	BE7

Barriers: Education (of others)

Name	**Descriptions**	**Codes**
Education of parents	Lack of awareness regarding the importance of physical activity by parents	BEd1
PA education of teachers/coaches	Lack of awareness regarding the importance of physical activity by authoritative figures	BEd2
Perceptions re: conditions (outside family)	Education/awareness of authoritative figures re: conditions and physical effects of same	BEd3
Lack of prioritisation of exercise in school	P.E not prioritised/no teaching of importance of exercise/no designated teacher etc.	BEd4

Barriers: Family

Name	**Descriptions**	**Codes**
Family Routine	Inability to fit exercise in due to family commitments or other children etc.	BFam1
Parent perceptions about exercise	The opinions of parents regarding exercise and its benefits/drawbacks. Including fear/anxiety of parents	BFam2
Parent perceptions about disability	The opinions of parents regarding diagnosis of a conditions including fear/anxiety	BFam3
Cost	The financial cost of participation in sports clubs or activities	BFam4
Academic Prioritisation	Prioritisation of homework or study-related extra-curricular activities over exercise	BFam5

Benefits:

Name	**Descriptions**	**Codes**
Social engagement	Interaction with peers and use or practice of social skills	Ben1
Mood regulation	The calming effects of exercise which may lead to mood improvement/happiness	Ben2
Reduction of anxiety	The improvement of anxiety and stress levels with exercise	Ben3

Confidence	Improvement in self-confidence related to exercise	Ben4
Sensory Regulation	The awareness of one's body and ability to control sensory processes	Ben5
Function	General improvement in bodily functions secondary to exercise	Ben6
Attention	Improvement in child's ability to concentrate or focus on a task	Ben7
Exposure	Experience of new things and widening of interests	Ben8
Weight management/Appetite	The effect of exercise on weight loss and maintenance, including eating patterns	Ben9
Sleep	The improvement in sleep patterns post-exercise	Ben10
Strength	An increase in strength as a result of exercise, including core strength	Ben11

Motivators:

Name	**Descriptions**	**Codes**
Fun/Enjoyment	Experiencing emotional pleasure during physical activity or exercise	M1
Supportive environment	Encouragement by authoritative figures such as teachers or coaches	M2
Availability of mixed-ability activities	A non-competitive environment where participation is encouraged for all children	M3
Small groups	Availability of activities with a smaller ratio of teacher/coach to number of children	M4
Appropriate instructions	The use of clear and easy-to-understand directions appropriate for children with ASD e.g. visual cues/picture charts	M5
Sense of achievement	Attainment of goals or being awarded for participation	M6