EFFECTIVE INTERVENTIONS FOR CHILDREN AND ADOLESCENTS WITH AUTISM SPECTRUM DISORDERS AND OTHER SPECIAL EDUCATIONAL NEEDS

by

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ABSTRACT

The need for cost-effective interventions for individuals with Autism Spectrum Disorders (ASD) and related conditions is growing rapidly. Recent research suggests that only a limited number of individuals who are eligible for intervention are actually receiving such services. This thesis first discusses the current evidence-base for interventions aimed at young children with ASD. It then outlines a frequent oversight in the literature regarding motor development and resonance difficulties in this population, which forms the basis for developing motor-based interventions for these individuals. Two large-scale experimental school-based studies are then presented which examine the effects of an ABA-based high-intensity physical exercise as an effective intervention for children with ASD and other Special Educational Needs (SEN). In one study, this intervention is compared with a low-intensity exercise program. Results indicate that high-intensity physical exercise results in significant short-term (0-to-90 minute), but not long-term (24 hours+), improvements in cognitive/behavioral flexibility (executive functions) in students with ASD and students with SEN. Furthermore, both high-intensity and low-intensity exercise resulted in significant reductions in stress, in both the short-term and long-term in students with ASD and students with SEN. These findings provide direct evidence for the effectiveness of physical exercise as a school-based intervention.

To my Grandpa and Dad who we lost during this process, and to my Mum who has supported me through everything: this is for you.

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STATEMENT OF AUTHORSHIP

Chapter 2 of this thesis has been prepared for submission to an academic journal, and is largely presented in manuscript format. However, revisions and omissions have been made to avoid repetitions and/or topic diversions in this thesis. I am the primary author of this paper and conducted the systematic review alone, as well as the interpretations. The other contributors are Supriya Malik, who provided insight into intervention session descriptions, the real-world translation topic, and Table 1.1, and my supervisor, Dr. Joseph McCleery, who provided feedback on the systematic review.

Chapter 3 of this thesis has been published as an open-access journal by Frontiers. My supervisor, Dr. Joseph McCleery, was invited to submit a paper on this topic, and initiated and supervised my lead on the content and detail of this paper. Dr. McCleery and I shaped the overall article together, and I then completed the literature review, write-up and summaries. Dr. Chysi Stefanidou contributed to the early motor development section, and Dr. Dimitrios Sampanis contributed to the Transcranial Direct Current Stimulation (tDCS) section. Final edits were made by myself, with feedback from Dr. McCleery, who was responsible for the order of content, final summary conclusions, and criteria for inclusion. The content presented is my intellectual content as much as the presented empirical chapters.

CHAPTER 1: THESIS INTRODUCTION

Autism spectrum disorder (ASD) is a developmental condition that is characterised by deficits in social interaction, communication and behaviour (APA, 2013; Smith, Reichow & Volkmar, 2015). Often surfacing around two years of age, ASD is a lifelong disorder that is now estimated to affect 1 in 88 children in the USA (Centre for Disease Control: CDC, 2012; Baio, 2012). According to a recent report by Knapp, Romeo and Beecham (2007), autism costs the UK almost £28 billion, with costs growing as each individual enters adulthood. Of the total amount, a significant 59% is dedicated to health and social care provision. It is crucial, therefore, that cost-effective interventions are developed for this population. Since the lifetime cost estimate is over £4.5 million for a single low-functioning individual, such interventions must be accessible and appropriate throughout the lifespan.

An aim of this thesis is to systematically review the current state of the evidence-base for ASD interventions (Chapter 2), before outlining a frequent oversight in this field- namely, the prominence of motor development and resonance difficulties within this population (Chapter 3). Research suggests that a sub-set of children with autism experience notable motor-related deficiencies from a very early age, which are likely to negatively affect social-communicative, language and cognitive development in this population. We outline the necessity for well-controlled motor-based intervention studies that build upon the discussed methodological weaknesses of the current intervention research literature.

Given this need, we present physical exercise as a viable motor-based intervention for children and adolescents with ASD and other Special Educational Needs (SEN). As demonstrated in multiple populations, the effects of moderate- to high-intensity physical activity extend far beyond the physical health benefits that it is perhaps most associated with. Both acute and chronic exercise engagement have been associated with improvements to emotional well-being (Ströhle, 2009), academic performance (Hillman et al., 2009), adaptive behaviour (Kern, Koegel, Dyer, Blew and Fenton, 1982) and cognitive functioning (Tomporowski, 2003; Pan et al., 2015). Importantly, Trudeau and Shephard (2008) found that academic achievement is unhindered after increasing the time spent engaged in physical activity at a cost to the time spent in other subjects. Conversely, increasing the time spent in academic subjects at a cost to physical activity does not improve grades and may in fact lead to significant health risks.

However, due to the complex and socially-demanding nature of many exercise opportunities throughout the school years, many with ASD find it difficult to participate in physical activity (Finkelstein, Nickel, Barnes and Suma, 2010). Indeed, as established by Sallis (1993) and Nicholson et al. (2011), physical activity rapidly decreases in adolescence, with an overwhelming number at risk of being overweight (Getchell et al., 2012). A further aim of the present empirical research is therefore to establish an effective framework to promote the participation of this population in physical exercise, based on effective techniques currently used by other types of ASD intervention.

Our novel empirical research will be presented in Chapters 4 and 5. Chapter 4 explores the effects of aerobic exercise on reported stress following acute (Study 1) and chronic (Study 2) school-based programmes for children with ASD and other SEN. Chapter 5 then presents the executive function results from these programmes, with insight into both switching and inhibition performance. Finally, Chapter 6 will summarise and discuss these findings, highlighting the extent to which the results from these novel programmes both interact with and extend the current literature.

CHAPTER 2: A SYSTEMATIC REVIEW OF EVIDENCE SUPPORTING EARLY INTERVENTION FOR CHILDREN WITH AUTISM SPECTRUM DISORDERS

2.1. Introduction

Autism is a pervasive developmental disorder that impairs social interaction and communication, and gives rise to stereotypical and/or repetitive motor behaviours (ICD-10: WHO, 1993; DSM-5: APA, 2013). Reports suggest that the average age of ASD diagnosis in community-based settings is 3- to 4-years in the UK (Baird et al., 2006), and 3- to 7-years in the USA (Mandell et al., 2005). Although there is no known singular cause, there is consensus that its development is strongly associated with genetic factors (Brian et al., 2008; Ozonoff et al., 2011). Despite financial and societal pressure to intervene at the earliest stage possible, there is surprisingly little information available regarding which particular approaches to early intervention have an evidence-base for this population, or at which ages.

In the current study, we conduct a comprehensive review of group design experimental studies of interventions for children diagnosed with autism, from birth to 6 years of age. First, we describe three major approaches taken in early intervention for this population. Next, we systematically review and describe the level and nature of the evidence in support of each approach. Following from this, we address several critical limitations to our current understanding of the effectiveness and relative effectiveness of early intervention for this population. We particularly focus on limitations to experimental designs and comparisons, as well as systematic differences in how the approach to intervention appears to have affected the selection of experimental approaches, targets and, thereby, the data and results. Finally, we conclude the review by describing current needs and future directions for research on early interventions for infants and young children with autism.

2.1.1. Evidence-Base for Early Intervention for Autism Spectrum Disorders

Given the increase in the number of children receiving a diagnosis of autism, the demand for effective intervention services in the community has also increased dramatically (Odom et al, 2003). However, even when children are identified early on, the availability of early intervention services is limited (Shattuck et al., 2009). For example, a study conducted by Rosenberg et al., (2009), found that only 10% of those under the age of 2 who were eligible to receive early intervention for developmental delays actually received those services. Given the limited resources available to children, families, and service providers in the community, it is critically important to ensure that any services provided to this population ultimately have an established evidence-base.

The goal of this section is to present a comprehensive review and description of the level and nature of the current evidence supporting early interventions for Autism Spectrum Disorders. In particular, we consider three major types of intervention: Behavioural, Developmental, and Developmental-Behavioural. In reality, there is somewhat of a continuum of relationship between these interventions, and by no means do interventions each fall into a set category. Nevertheless, for the purpose of review, we present these categories based upon differences in the nature and degree of their respective incorporation versus lack of incorporation of several components of operant teaching methods (see Table 1 for intervention ingredients). Behavioural interventions are placed in this category on the basis of entailing structured applied behaviour analysis (ABA), whilst Developmental interventions were classified as those with a primary focus on relationship-building in natural environments, rooted more solidly in Piagetian theory (Piaget, 1952). Developmental-Behavioural interventions bridge this gap, incorporating key aspects of both approaches (Schreibman et al., 2015).

In this section, we also provide descriptions and evaluation of the information provided in Table 2 and Table 3. Collectively, these tables reflect and represent detailed information on the experimental methods, measures, and results of all experimental group design studies conducted on an early intervention for children with autism spectrum disorders until December, 2015. In order to obtain this information, a systematic literature review was conducted across multiple databases, including PsycINFO, PubMed, and Web of Science. The selection criteria for qualifying studies was that the mean age of participants was 6-years of age or below; that the research involved a comparison of two groups of at least 10 participants in each group, and that the outcome of the intervention could be segregated for ASD participants from non-ASD participants in cases where other populations have been included in the sample. Retrospective studies were not included. In essence, these studies reflect all randomized controlled trials and pilot randomized controlled trials (RCTs), as well as all group comparison studies with pre-intervention and post-intervention measures but which did not meet the full experimental standards of RCT designs. For example, a number of studies that were included in this review did not effectively randomize participants to intervention conditions, and others did not require assessors to be blind to the intervention condition or status of the participants. Critically, these and other violations to experimental control are identified in Table 1 and Table 2, and also considered and discussed in some detail below.

2.1.1.1. Behavioural Interventions

Behavioural interventions for children with autism are based on operant learning theory, using the principles of applied behaviour analysis (ABA) by including three strategies: (A) antecedent factors before a target behaviour is likely to occur, (B) behaviourally defined and measured intervention targets, and (C) consequences following the occurrence of a target or non-target behaviour. Behavioural interventions are the most widely known approaches for children with autism, aiming to teach new skills as well as alternative, adaptive behaviours to reduce the frequency and severity of maladaptive behaviours (Cohen & Volkmar, 1997). Initial behavioural interventions were highly structured and adult-directed (e.g., Lovaas, Koegel, Simmons, & Long, 1973), and typically incorporated a variety of explicit prompting, punishment, and reinforcement strategies to increase or decrease target skills and behaviours.

One of the most widely used teaching methods of ABA is Discrete Trial Training (DTT). DTT is often used to teach children with autism a broad range of behaviours, ranging from academic tasks to activities of daily living and social-communicative behaviours. The method emphasizes the importance of one-to-one teaching in a highly structured setting. The child is typically made to sit at a small table, often in a distraction-free room, with the therapist sitting across the table. Specific and individualized behaviourally-defined goals are identified and systematically taught to each child. Each 'discrete trial' broadly follows a similar sequence, with the target behaviour changing based on child progress. The therapist typically brings out a task for the child and provides an instruction. If the child successfully performs the target behaviour in response to the instruction or task, then this behaviour is *reinforced* with praise, food, and/or access to a preferred toy. If the child does not complete the task, access to the reinforcer is denied and a verbal cue is typically given (e.g., "no," "try again"). Once one task is completed, the child is cued for another trial or for the next task. Over time, if the child does not respond to the instruction, then he/she is *prompted* by the therapist to respond appropriately. Success and progress are typically defined by the learner performing accurately on 80% of trials. DTT is often carried out with the child for 30 to 40 hours per week. Some of the most crucial components of DTT include that it is highly structured, therapist led, and that each successful target behavior is reinforced by items which are motivating but not necessarily linked directly to the natural outcome of the behaviour.

Overall, positive evidence for behaviour-based interventions is prominent in the ASD literature. Commonly referred to as Intensive Behavioural Intervention (IBI), research in this area has frequently been described as well-established (Rogers & Vismara, 2008; Eikeseth, 2009; Eldevik et al., 2010). The most pioneering study is that of Lovaas (1987), which initially demonstrated the efficacy of IBI. In this study, 38 children (mean age 37.8 months) were randomly assigned to an intensive (40 hours per week) treatment group, or to a control group that received fewer than 10 hours of therapy per week. A further control group of 21 children was taken from an existing study by Freeman and Skarda (1985). Standardised measures were taken of participant IQ at pre-treatment and follow-up, and information about each child's school placement was collected. After two years, 47 percent of those in the intensive treatment condition were assessed as having normal intellectual and educational functioning, as well as successful inclusion in mainstream schools. Alternatively, for the 40 control participants, only two percent achieved this same standard.

Given the impressive nature of these initial findings, a number studies have been focused on replicating these positive effects. Many subsequent studies explored efficacy using case studies and multiple-baseline designs (Birnbrauer & Leach, 1993; Perry, Cohen, & De Carlo, 1995; Green, Brennan, & Fein, 2002), but some large-scale studies and RCTs have also been published over the past decade. For example, Sallows and Graupner (2005) randomly assigned 24 children to either a Lovaas-style EIBI treatment group, or to a less-costly, intensive parent-directed condition. Following four years of intervention, 48 percent of all participants demonstrated significant gains in their cognitive, social, and academic abilities. Unexpectedly, no group differences were found between the clinic-directed and parent-directed groups, which the authors suggest may have been due to the intuitive nature of parents seeking to fill the senior-therapist role. However, such reduced supervision effects are not consistently upheld, and the benefits of a high-intensity, high-structured treatment programme are frequently highlighted (Smith, Groen and Wynn, 2000). For example, Smith and colleagues conducted a similar randomised controlled trial comparing an early Lovaas-based treatment with parent-implemented therapy in a group of 15 children. Upon completion of treatment, the intensive behavioural therapy group demonstrated improvements in intelligence, language, visual-spatial skills, and academic performance when compared to the parent-administered intervention group (Smith, Groen, & Wynn, 2000).

In addition to these comparisons to treatment as usual (TAU) and to parent-implemented low intensity EIBI, there is some evidence that the beneficial effects of EIBI are upheld when compared to other therapist-implemented programs. For example, Eikeseth, Smith, Jahr, and Eldevik (2002) conducted a large-scale quasi-random experimental study comparing the effects of EIBI with an 'eclectic' treatment similar to those readily available in the community (including sensory integration therapy, TEACCH-based procedures, and discrete trial training). Both programs were of equal intensity (around 28 hours per week) and were provided to participants aged between 4 and 7 years. Measurements of intellectual functioning, visual-spatial skills, adaptive functioning, and language were taken using standardized assessments at pre-treatment and following 1 year of intervention. At follow-up, it was found that those who had received EIBI treatment achieved notable gains in most of the standardized measures when compared to control children. No significant group differences were observed for the Vineland Socialization and Daily Living subscales, however. The authors concluded that, despite several limitations of the research – including small sample size and non-random group allocation – the results evidence superior effects of IBI from the age of 4 years. Further research is required to explore these effects in younger children, particularly given that this study involved school-based implementation which may not be transferable to the younger population.

The majority of IBI studies provide approximately 30 hours per week of intervention, with follow-up measures typically obtained between one to two years after the intervention commences. A recent study by Eldevik, Hastings, Jahr, and Hughes (2012) demonstrates the importance of increased intensity when implementing behavioural intervention programmes. Investigating the effects of EIBI in 31 children aged between 2-and 6-years, the authors compared gains in children receiving EIBI treatment with those from children who continued to receive treatment as usual (TAU). Measures of adaptive behaviour and full scale intellectual functioning were taken at pre-treatment and after two years. In line with previous research, the intervention group outperformed the TAU group in all assessments with the exception of the VABS Daily Living subscale. However, the intensity of this treatment averaged at 13.6 hours per week – an intensity level below half of the recommended rate (30 hours; Virues-Ortega 2010). Not surprisingly, then, the effect size estimations were smaller in this study than in previous studies of EIBI, prompting a debate about intensity of intervention (see Eldevik et al., 2010).

Although we have presented and discussed several studies that have produced positive findings for EIBI, there are notable inconsistencies in the amount of child progression across the multiple studies. As ascertained by Howard et al (2005), this is likely due to the variations in methodology, particularly those which deviate from the original Lovaas (1987) study methods. The authors highlight reduced intensity across all studies, as well as lower pre-treatment language and IQ scores, and length of the treatment programs when compared to the original study. Since previous studies had not wholly replicated the findings of Lovaas (1987), Howard and colleagues sought to achieve closer replication, with eligible participants receiving up to 40 hours of treatment. In this large-scale experimental study of participants with a mean age of 34 months, significant gains were discovered after 14 months of EIBI intervention when compared to an 'eclectic' intervention (a combination of TEACCH, sensory integration, and applied behaviour analysis), or a non-intensive control treatment. Those children who received intensive intervention made significant gains over the other participant groups in all standardised measures, with the exception of motor skills. Specifically, performance on assessments of cognition, language, and adaptive skills were significantly greater in the intensive

(EIBI) group compared with both comparison groups, who in turn did not significantly differ from one other. However, it is critical to note two serious limitations to this study, which are that the experimental measures were not conducted by blind assessors, and that participant group allocation was parent-determined. Nevertheless, the scale of this study (61 children) certainly makes it noteworthy in the field.

Considering the research to date, the effects of EIBI tend to be rooted most solidly and consistently in IQ and academic gains (see Tables 2 and 3). This being said, very few studies of EIBI have directly measured impacts of intervention on social skills. Those studies that have, such as Eikeseth, Smith, Jahr, and Eldevik (2002), however, have not observed consistent group differences in these behaviours. Furthermore, there appears to be some level of inconsistency across all types of gains in this literature, as well. For example, Magiati, Charman, and Howlin (2007) conducted a two-year prospective outcome study to compare children who received community-based EIBI with those receiving services from autism-specific nurseries. Measures of language, play, cognition, adaptive behaviour, and autism symptom severity were administered at intake and at follow-up. No group differences were observed in language, cognition, play skills, or autism symptom severity. However, contrasting the findings of Eikeseth, Smith, Jahr, and Eldevik (2002), the EIBI group did score somewhat higher in follow-up VABS Daily Living Skills. Despite this failure to replicate previous findings, overall the findings of studies of EIBI do generally suggest consistent IQ and academic gains as a result of early intensive behavioural intervention (Eikeseth, Smith, Jahr & Eldevik, 2002; Sallows and Graupner, 2005). Further research is required, however, particularly into whether or not intensive behavioural intervention has positive effects on social versus academic and language skills.

Taken together, findings from behavioural intervention studies demonstrate benefits of intensive interventions for young children with autism. While many studies in this area typically focus on young school-aged samples, several studies now evidence efficacy in preschoolers, including children as young as 18 months of age (see Table 1). Further research is clearly required in order to further explore and solidify the existence and nature of positive effects in children of preschool age and younger, but the initial evidence is very promising, at least in targeted domains. Therefore, as described in several previous reviews (Warren et al, 2011; Reichow et al, 2012; Dawson, 2013), although the current research base is moderately strong, the field is lacking in optimally controlled studies and relies heavily on non-randomised designs (see Tables 2 and 3). Further research is therefore required before conclusive judgments of efficacy and effectiveness can be confidently confirmed for younger populations, and for domains other than IQ, academic performance, and language skills.

2.1.1.2. Developmental Interventions

Developmental, or relationship-based, interventions focus on each child's ability to develop and engage in healthy relationships with other people in the close environment. As Atchison et al. (1997) indicate, the aim of these programs is to assist children to develop skills in thinking and relating different events in an organized and logical manner. The developmental model of early intervention has its primary bases in Piagetian theory of development as well as the psycho-social model of language-learning (Ingersoll, 2010). The premise is that the developmental sequence in which children gain skills is the same and therefore, although delayed, the pattern of skill development should remain the same for children on the autism spectrum. There are many programs that have been developed using this model, including the DIR/Floortime (Developmental, Individual Differences, Relationship) model, RDI (Relationship Development Intervention), SCERTS (Social, communication, emotional regulation, and transactional support), and Hanen's More than Words (HMTW). All of these programs focus primarily on improving the quality of interaction between child and caregiver/therapist. Adult responsiveness is the most important facilitative strategy in this intervention approach, with a core belief that children are active learners in their development and that it is through interactions with significant others that they learn and shape their development (Dawson, 2008; Rogers & Dawson, 2010). Therefore, social and communicative competence is targeted during reciprocal interaction with others. All of these approaches require the parent to learn intervention techniques and administer them in the child's natural environment.

A typical example of a developmental intervention can be seen in Relationship Development Intervention (RDI), which uses parent-child interaction and play as the basis for teaching various social, emotional, and communication skills (Relationship Development Assessment, RDA; Larkin et al 2013). The parent is taught specific interaction patterns with the child, including pacing, pausing, and environmental manipulation. Unlike DTT/IBI, RDI therapy is administered in the child's natural settings using everyday activities, such as clearing away toys, as the background for learning. Parents are often expected to report back to a therapist with videos and journals, and to engage in weekly feedback and guidance consultations. In clear contrast to traditional behavioural interventions, RDI and other developmental interventions are child-directed, with the child playing an active and primary role in deciding the nature and direction of activity during the intervention.

Both pilot and full-scale RCTs have evidenced positive effects for developmental intervention approaches. For example, Aldred, Green, and Adams (2004) randomised 28 children with a mean age of 49.5 months into a parent-mediated social-communication intervention (the PACT project) group or a treatment as usual group. Upon completion of 12 months of participation, those in the target intervention group demonstrated significant reductions in total Autism Diagnostic Observation Schedule (ADOS) scores, as well as increases expressive language and communication initiations when compared with controls. These findings are relatively unique in documenting direct reductions in core autism symptom severity measured on a standardized diagnostic assessment as a result of early intervention. Furthermore, Casenhiser, Shanker, and Stieben (2013) conducted an RCT of DIR/Floortime-based intervention with 51 children from as young as 24 months of age. The authors reported significant gains in social interaction skills in the DIR group relative to participant receiving community-based treatment as usual, after 12 months of intervention. However, no differences were observed between the two groups on standardised assessments of language. In fact, to date, no assessor-rated language gains have ever been reported in any study of a developmental intervention, with all reported findings of language gains for this type of intervention exclusive to un-blinded parent-report measures.

Despite these positive effects, well-controlled studies in this area have also produced null results when considering the direct impact of the intervention on primary outcome measures. For example, Carter et al (2011) randomly assigned sixty-two children (mean age: 20 months) with ASD into a 3-month HMTW intervention group, a parent implemented intervention group, or a treatment as usual group. No main effects of the HMTW intervention were found, although it was discovered that those in the HMTW group with lower levels of object interest at pre-intervention derived the greatest communication benefits when compared to those with higher scores at this time point. Aside from these exploratory analyses, the reported null findings warrant caution when considering this intervention for very young children with ASD. In a follow-up study of the PACT (Preschool Autism Communication Trial) intervention described above, Green and colleagues also reported no significant group differences between those receiving PACT intervention versus control participants (Green and Raygorodetsky, 2010).

Interestingly, the developmental intervention studies meeting criteria vary greatly in both the intensity and the length of the intervention prior to data collection at follow-up. With regards to intensity, there is in fact very little control for amount of intervention hours implemented across many developmental studies. Furthermore, since most studies involve parent-implementation in the home, researchers must rely on parent reports/logs to ascertain how many hours the child received intervention. Indeed, studies have often reported a poor/inaccurate response rate (e.g. Solomon et al., 2007), or have not used such logs at all (e.g. Pajareya and Nopmaneejumruslers, 2011). There is also significant variability regarding the time lapse between measures at baseline and follow-up across studies on developmental interventions. For example, whilst the study by Casenhiser, Shanker, and Stieben (2013) involved 3.9 hours of DIR/Floortime per week for 12 months, the same intervention was implemented by Pajareya and

Nopmaneejumruslers (2011) in their RCT for only 3 months and at a rate of approximately 15 hours per week.

With regards to targets for behavioural change measures, developmental intervention studies have generally placed greater emphasis on measuring play-based outcomes for this population (e.g. Schertz et al., 2010), with less emphasis on measuring language skills (e.g., Eikeseth et al., 2002; Migiati, Charman, & Howlin, 2007). As reported by Ingersoll and Shreibman (2006), the developmental intervention field has demonstrated a shift of focus towards the teaching of non-verbal social-communication skills, which typically develop earlier in life. This shift of focus appears to be rooted in evidence suggesting a positive relationship between early non-verbal social skills, such as joint-attention, and the later development of language skills (Ingersoll and Shreibman, 2006). Those studies of developmental interventions that have measured language at this earlier stage, however, have yielded mixed results. For example, a study by Drew et al., (2002), found that following 12 months of joint attention-based intervention, children (mean age: 35 months) randomly assigned to the parent training group demonstrated greater gains in language on the MacArthur Communicative Development Inventory (CDI) when compared to a control group receiving local community-based intervention services. Despite a strong experimental design, the small sample size (12 per group) and low level of control over implementation prompted Oosterling et al (2010) to attempt to replicate the observed effects in a larger group (67 children) of the same age. However, after 12 months of intervention, no significant effects were observed in either the primary (language) or secondary (clinical global improvement) outcome measures utilized. Again, the level and intensity of parent-implementation was not formally assessed, further pointing to a clear and current limitation across a vast majority of parent-based intervention research studies.

Given the play-based and parent-implemented nature of developmental interventions, it is unsurprising that many of the studies on this intervention type include children from around 2 years of age, which is younger than the majority of studies of DTT/IBI. Of the intervention studies meeting criteria for inclusion in Table 2 and Table 3, the youngest samples are observed in the developmental intervention studies. For example, the HMTW study by Carter et al. (2011) includes a sample range starting at the age of 15 months. The same cannot be said for any other types of intervention, potentially making it the most toddler-focused category of intervention research to date.

Although efforts to experimentally and systematically examine the effectiveness of developmental interventions are certainly increasing, and several high quality studies have already been conducted and reported, it is difficult to make judgments regarding the effectiveness of this type of intervention based on the mixed results that have been observed in the studies conducted to date. There is certainly promising evidence for gains in social-communication skills in children with ASD as young as 20 months of age as a result of this type of intervention. However, the majority of the existing studies of developmental interventions do not control for likely sampling and/or participant group assignment biases (see Table 3). Furthermore, the current developmental intervention literature is heavily reliant upon un-blinded parent-report measures (see Table 3), and the few well-controlled studies that have been conducted to date have produced a mix of positive effects and convincing null findings (see Table 2). This being said, the positive effect reported by Aldred and colleagues is relatively unique in evidencing positive effects on core autism symptom severity (Aldred, Green, & Adams, 2004, but see also Green and Raygorodetsky, 2010). It is further worth noting that developmental intervention studies may be at a position of disadvantage in comparisons with other intervention types due to their reliance on parents as interventionists, which is a factor that will be discussed in some detail below.

2.1.1.3. Developmental-Behavioural Interventions

In an effort to bridge the gap between behavioural and developmental interventions, and to potentially gain the benefits of each approach, a new set of early intervention programmes has emerged which take into account the prominent core ingredients from each. Examples of developmental-behavioural interventions include the Early Start Denver Model (ESDM; Rogers & Dawson, 2009), JASPER (Joint Attention, Symbolic Play, Engagement, Regulation; Kasari, Freeman and Paparella, 2006), Reciprocal Imitation Training (RIT; Ingersoll 2010), and Early Achievements (EA; Landa et al., 2011). Interventions that also fall under this category include naturalistic ABA interventions, such as Pivotal Response Treatment (PRT), which were initially developed to improve generalization of skills when compared with traditional ABA interventions such as DTT (Koegel, Carter, & Koegel, 1998; Schreibman and Koegel, 1996). The substantial overlaps with DTT/IBI concern intervention tools and principles, such as systematic physical prompting procedures, and reinforcement that is both temporally sensitive and contingent upon particular target behaviours. However, unlike DTT/IBI, and similar to pure developmental approaches, these interventions place emphasis on adult sensitivity and responsiveness to the motivations of the child, and environmental manipulation/modelling of developmentally appropriate behaviours. Combining the approaches, emphasis is also placed on shared control; a balance between the adult-direct nature of traditional behavioural interventions and the child-directed nature of traditional developmental interventions. Developmental-behavioural interventions also focus on the use of natural reinforcers for target behaviours (e.g., child says "ball," child receives the ball) versus reinforcers unrelated to the target behaviours that are commonly used in traditional behavioural interventions (e.g., child says "ball," child receives a small piece of candy).

Several well-designed and well-controlled RCTs have provided evidence for positive impacts of developmental-behavioural interventions. In 2010, Dawson and colleagues investigated the efficacy of the Early Start Denver Model (ESDM) in a randomised controlled trial of 48 toddlers with ASD (Dawson et al., 2010). Children were assigned to receive the ESDM or referred to community-based intervention. After 24 months of treatment, those who received the ESDM demonstrated significant gains in blind-experimenter administered IQ (15.7 points), as well as receptive and expressive language, relative to the control group. Participants receiving the ESDM also exhibited significant positive changes in autism diagnostic classification (e.g., reduced from Autistic Disorder to PDD-NOS), relative to the control group. Finally, ESDM participants exhibited significantly better performance on standardised measures of adaptive behaviour post-treatment, relative to the control group, who demonstrated significant losses in adaptive behaviour.

The positive findings of this well-controlled study are among the strongest and most convincing in the early intervention literature at this time. Following from this large-scale therapist-implemented RCT, Rogers and colleagues sought to replicate the findings in the context of a parent-mediated implementation of the ESDM (Rogers et al., 2012). However, no group differences were observed between the parent-mediated ESDM group and community services group (Rogers et al, 2012). Therefore, although the evidence suggests that the ESDM may be highly effective as a comprehensive early intervention program, even this protocol does not appear to be robust enough to overcome known challenges to parent-mediated intervention. For example, in a recent review of the literature, Dawson and Burner (2011) determined that parent-mediated intervention is only effective for improving child behaviours approximately one-third of the time (Dawson & Burner, 2011).

The results of experimental studies have provided further evidence for positive effects of less comprehensive, more targeted developmental-behavioural interventions. For example, Ingersoll (2010) conducted a pilot RCT into the effects of RIT on elicited and spontaneous imitation in twenty-one young autistic children, relative to a group of wait-list control participants. Significantly larger imitation gains were reported in the treatment group across experimental assessments of both structured and unstructured imitation skills. However, perhaps due to the small sample size and despite true randomization, the RIT intervention group had slightly but significantly higher verbal abilities than the control group at intake. Regarding the developmental-behavioral intervention JASPER, Kasari and colleagues (2006; 2008; 2010) have reported multiple randomized controlled trials designed to investigate the effects of a joint attention (JA) intervention upon the joint engagement (JE) of young children with ASD, with positive effects. For example, in their initial study (Kasari et al, 2006; 2008), 58 preschoolers were randomly assigned to JA intervention, Symbolic Play (SP) intervention, or a non-treatment control group. Intervention was implemented for 30 minutes per day, for up to 6 weeks. JA, SP, language, and blinded mother-child interaction measures were administered at several time-points throughout the course of the study. After 12 months, both the JA and SP groups demonstrated greater gains in expressive language when compared to the non-treatment group. Furthermore, those with the lowest language levels at intake benefitted most from the JA intervention with regards to gains in language skills.

Intervention duration for developmental-behavioural research studies meeting criteria for the current review range from 6 weeks to 24 months. Intensity also varies somewhat, with some studies reporting as few as 3 hours per week of intervention (e.g. Ingersoll, 2010) and others implementing therapy for as many as 4 hours per day for 5 days per week (e.g., Dawson et al, 2010). Further, participant samples in several of the developmental-behavioural intervention studies reviewed here range from as young as 18 months old up to 4 years of age.

Across studies, developmental-behavioural intervention studies have been designed to measure a wide range of gains, including cognition, language, social skills types, and adaptive behaviour. However, the most consistent and most prominent measures and gains have been observed in language and IQ. For example, although studies of the ESDM use social measures, such as the VABS and the ADOS, the results appear relatively much weaker or, sometimes, non-existent relative to the IQ and language gains. This being said, studies of both RIT and JA interventions have provided evidence for positive effects of developmental-behavioural interventions on targeted measures of social behaviour (e.g., Ingersoll, 2010; Kasari 2006, 2008, 2010). Therefore, it is possible that developmental-behavioural interventions reliably generate positive effects on social skills, but that these effects are not consistently sufficient to measurably alter core aspects of autism social symptom severity. Alternatively, or additionally, it is possible that developmental-behavioral interventions that are focused intently on particular skills, such as the focus of Reciprocal Imitation Training on social imitation and JASPER on joint attention in particular, drives the observed effects.

In sum, there is a moderate level of evidence to suggest that developmental-behavioural interventions have positive impacts upon cognitive and language skills between 1 ½ and 4 years of age in children with autism spectrum disorders. This level of evidence is based on RCTs or pilot RCTs evidencing positive effects on child behaviour for several different developmental-behavioural interventions (see Table 2 and 3). Effects on cognitive and language skills appear to be the most consistent positive findings, with effects on social functioning relatively consistent when sensitive measures of targeted skills (e.g., imitation, joint attention) are used. When coarser, more global measures such as standardized diagnostic assessments (i.e., ADOS) and the social domain of standardized measures of adaptive functioning (i.e., VABS) are used, results are notably less consistent (See Tables 2 and 3).

2.2. Limitations of the Current Autism Early Intervention Literature

Although the number and quality of experimental group design studies for early intervention for children with autism is growing, the literature continues to suffer from serious limitations. Here, we discuss the nature and extent of several of the key limitations.

Randomization of Participants to Treatment Conditions. The majority of the large-scale experimental studies reviewed in this paper fail to provide sufficient control for likely biases in the sampling and allocation of participants to a target intervention versus a comparison intervention or other comparison/control group (see Table 1). The failure to effectively randomize participants associated with the majority of studies reviewed in this paper is a serious experimental violation, which can be assumed to allow for participant selection or self-selection and/or confounding of the experimental manipulation for comparing the intervention conditions. The results of studies with insufficient controls for potential sampling and group allocation biases must be interpreted with extreme caution. There is a significant and pressing need for more truly randomized controlled intervention trials on this topic and population.

Experimental Blinding of Measures. A large number of the studies reviewed in the current report suffered from an absence of experimental "blinding" of the individuals who administered the pre-intervention and post-intervention dependent measures (see Table 3). Furthermore, in several of these studies, the only dependent measures exhibiting positive effects of the intervention are those that were not experimentally blinded. Examples include language-learning effects observed in parent report measures, such as the MacArthur-Bates Communicative Development Inventory (CDI), in several studies. While extensive research has established the CDI as one of the most reliable measures of natural language comprehension and production across numerous populations (Heilmann, Weismer, Evans & Hollar, 2005; Thal, DesJardin & Eisenberg, 2007), it is extremely difficult to impossible to blind parents to the intervention status of their child in a study. There is, therefore, a significant and pressing need for studies use experimenter-blinded direct behavioural measures of natural language comprehension and production. Experiments employing novel, experimenter-blind behavioural measures of other skills (e.g., Structured Imitation Assessment, Unstructured Imitation Assessment, Early Social Communication Scales), have played a critical role in furthering our understanding of the nature of intervention effects. To date, however, there have been serious limits to the availability of effective naturalistic measures of other skills, which likely underlies failures to employ experimentally blinded measures, or to observe effects in them, in a large number of studies.

Small Sample Sizes. Approximately one quarter of the studies reviewed in the current report utilized relatively small participant samples (i.e., n<30; see Table 2). Furthermore, of those studies that have utilized significant samples, a large number have suffered from notable violations to experimental control (see Tables 2 and 3). This practice, which is likely related to limits on research funding and other resources, may contribute to increased risk for both Type I and Type II statistical errors, ultimately contributing to unreliable and/or inconsistent results being in the published literature. There is, therefore, a significant and pressing need for more and better support for well-controlled intervention trials with appropriate and sufficient sample sizes.

Effects Above and Beyond a Comparison Intervention. A significant number of the studies reviewed in this report failed to provide evidence for effects above and beyond a comparison treatment. This is likely due to ethical constraints that require that all children in a study receive some level or degree of intervention. Despite this reasonable concern, however, the very limited number of studies that have directly compared interventions and produced positive effects of one intervention over the other seriously limits our current understanding of the effectiveness of early intervention for this population. In particular, we have only somewhat limited information regarding whether or not any/all positive effects observed in direct comparisons with null findings are simply due to developmental effects unrelated to the intervention in these studies. There is, therefore, significant and pressing need for more high quality studies directly comparing one or more interventions to community-based treatment as usual and/or to other active control conditions. Optimal intervention designs may include direct comparisons of two interventions targeting different aspects of development (e.g., joint attention, imitation) both to one another and to a treatment as usual control condition. The use of targeted pre-intervention and post-intervention measures (e.g., joint attention measures, imitation measures) in this design increases the likelihood of observing positive effects of one intervention of the other, as well as each intervention over the treatment as usual condition (e.g., Kasari et al, 2006; 2008).

Exploratory Predictor Analyses and Models. Another limitation to the existing literature is the relative absence of strong, a priori theoretical models and hypotheses for use in predictor analyses. A number of studies have conducted analyses designed to identify participant characteristics associated with differential response to intervention. However, the researchers do not always provide clear a priori hypotheses to support these analyses. This increases the likelihood that the analyses may be exploratory in nature, which in turn increases the likelihood that the results of these analyses may not hold up to attempts to replicate. Regardless, it is evident that the field is in need of clear theoretical and conceptual models for systematically examining differential child responsivity to different types of existing and future autism early intervention (see e.g., Sherer & Schreibman, 2005; Schreibman et al., 2009; Stahmer, Schreibman, & Cunningham, 2011). The development and a priory model testing of stronger theoretical and conceptual models for this purpose would not only increase the validity and reliability of specific predictor analyses but also have the potential to serve as over-arching frameworks for guiding and supporting future development in early intervention. For RCTs, these a priori models and hypotheses should be included in the initial trial description during trial pre-registration.

2.3. Parent-Mediated Versus Therapist-Mediated Intervention

The rising number of children receiving a diagnosis of ASD creates pressure to establish methods for disseminating interventions both more broadly and with reduced expenses (Baio, 2012). One model that has been suggested to serve this purpose is parent-mediated intervention, where the parent is trained or supported to provide direct intervention for their child. Benefits in the generalization of skills to other environments have also been found to be associated with parent-mediated interventions, which have further supported this approach as a low-cost model with significant potential for decreasing the strain on the professional (Steiner, Koegel, Koegel, & Ence, 2012; Vismara et al, 2013).

A significant number of interventions and studies have utilized a parent-mediated intervention model over the last 30 years. Most prominent among these have been the developmental interventions, which rely heavily on parent-mediated intervention as a result of a theoretical model that suggests a critical role for the parent-child relationship in infant and child development and learning. However, studies utilizing parent-mediated intervention have produced mixed results and have been relatively unimpressive in their attempt to demonstrate changes in child behaviour (see Dawson & Burner, 2011, for review). In 2007, McConachie and Diggle conducted a review of the literature on parent-mediated intervention for children with autism. They concluded that experimental designs of these studies were limited by small sample sizes, insufficient intervention length, varying methodology used, and a reliance on parent report measures, among others (McConachie & Diggle, 2007). Since this time, however, several high-quality, larger-scale studies have been conducted on parent-mediated intervention, with effects on child behaviour continuing to be inconsistent and unconvincing (Dawson & Burner, 2011). Indeed, there have even been studies of the same intervention showing different results seemingly dependent upon the therapist-mediated versus parent-mediated delivery of the therapy. For example, a large and high quality experimental study of a parent-mediated version of the ESDM did not produce any differences in the children in the intervention group relative to children randomized to a community based intervention (Rogers et al, 2012); however, the same intervention was shown to produce relatively strong and convincing positive effects over a treatment as usual control group when delivered via a therapist-mediated approach (Dawson et al., 2010).

As described earlier, studies of developmental interventions have produced mixed results. Furthermore, the variability in whether or not effects are observed in studies of developmental interventions appears to be pronounced in the context of high quality, well-controlled experimental trials in some cases. Given the fact that all studies of developmental early interventions to date have used a parent-mediated intervention approach, it is distinctly possible that the observed variability is due to inconsistent effects associated with parent-mediated interventions of all types with this population (see Dawson & Burner, 2011 for review). If so then developmental intervention studies may be at a disadvantage relative to the majority of behavioural and developmental-behavioural intervention studies, which typically use a therapist-mediated intervention in an isolated laboratory setting and with direct supervision from highly experienced clinicians. As a result, it is critically important that this factor be carefully considered when comparing and contrasting relative evidence across intervention types. Furthermore, future research should aim to account for, control for, and experimentally manipulate this aspect of intervention dissemination (expert vs parent) within and across intervention types, in an effort to disentangle the impact of intervention type from the impact of expert versus parent implementation of the intervention.

2.4. Real-World Translation

There has recently been an increased focus on evidence-based practice in order to improve psychological services provided in real-world, community settings. Thus far we have compared and contrasted the evidence base for different intervention programs and approaches, and identified some with more robust findings and others with relatively less support. However, according to Hunsley (2007), evidence-based practice relies equally on clinician expertise and robust scientific evidence. Although focus on evidence-based practice has greatly increased in the ASD literature, there is still very little research examining clinician characteristics and levels or types of expertise, with most discussion focusing on the delivery and maintenance of intervention methods and approaches (Allen and Warzak, 2000). Few studies acknowledge the highly trained and experienced nature of therapists used in intervention studies, nor their supervision by world-leading clinical experts, except with regards to experimental control and basic indices of fidelity of implementation, for example. Furthermore, to date relatively few study reports have acknowledged or addressed the fact that the effects observed in their studies may not be replicated outside of ideal, controlled laboratory settings.

2.5. Clinician Characteristics

To date, the majority of the ASD intervention research literature is based on findings in University-based settings, implemented by experienced graduate-level students who are closely supervised to achieve specified levels of fidelity of intervention implementation. A potentially critical factor in these studies is the fact that these students have other, less tangible clinical skills and characteristics, which may include timing sensitivity for prompting, selecting appropriate goals based on child progress, and relatively rapidly assessing and understanding the individual character and motivations of each child. Additionally, interpersonal skills such as the ability to build rapport and the ability to effectively engage in creative play likely enhance the quality, interactive flow, and interpersonal engagement during each session. It is therefore not unwarranted to suggest that variance in these or other characteristics may greatly impact upon treatment efficacy, even within the same intervention protocol. Indeed, Ebert and Kohnert (2010) have maintained that clinical expertise may be the most prominent and important features for clinician success. Moreover, therapist and environmental characteristics drastically change when practices are transferred from the laboratory to the community. For example, field-based therapists often have far greater caseloads and varied training backgrounds, and child characteristics are much broader than those included in current research studies. Specifically, whilst many lab-based studies select only those with no other existing conditions (see Table 3), children with ASD often have co-morbid conditions and disorders, perhaps which presumably alter therapeutic needs and impacts.

When evaluating autism interventions, it is important to consider the various factors that may be contributing to its success. These include, but are not limited to, clinician characteristics, the setting in which the intervention is implemented, and individual child characteristics. These will not only aid in developing best practices, but also shed light on the differential effects of interventions. Future research on effectiveness of interventions should most definitely consider multi-site research administered by therapists with varying educational and training backgrounds. Published reports of these studies should then aim to ensure sufficient documentation of such factors, including therapist characteristics and levels of setting control.

2.6. Summary

We have conducted a comprehensive review of the literature reporting experimental group design studies aimed at examining the effects of interventions for children diagnosed with autism spectrum disorders from birth to six years of age. In doing so, we have taken particular focus on the level and nature of the evidence base for the three major approaches to intervention: behavioural, developmental, and developmental-behavioural. We find that there exists a limited number of high quality, well-controlled studies for each of these types of intervention. However, we also find that the nature and consistency of the positive effects observed varies according to intervention type. Specifically, the evidence for positive effects of early intervention is stronger and more consistent for both behavioural and developmental-behavioural approaches than for the developmental approach to intervention. Furthermore, this pattern is particularly apparent for improvements on experimental measures of cognitive and language, versus measures of autism symptom severity. However, we also determine that developmental intervention studies are at a relatively critical disadvantage when it comes to generating/observing intervention effects, due to consistent reliance on parent-mediated intervention; whereas studies of behavioural and developmental-behavioural interventions have more commonly examined therapist-mediated intervention. Based upon this review, we highlight a need for more well-controlled studies of early interventions for this population. In particular, we highlight common violations to participant selection and randomization, as well as failure to utilize experimenter-blind pre-intervention and post-intervention measures in many studies. We also note the need for more direct comparisons of two or more early interventions, and suggest study designs and measures that can be expected to maximize the likelihood of observing reliable effects if and when they exist. We highlight the need for the development and testing of theoretical and conceptual models that can be used to generate a priori predictions for child characteristics associated with response to intervention. Finally, we suggest that future research will need to focus on the development and examination of parent-training strategies, which might then also facilitate more direct and more equivalent comparisons of the various early intervention approaches with one another.

Having identified the strengths and weaknesses of existent literature and explored future directions, we are optimistic that this novel and highly detailed systematic review lights a path forward toward research that will enable direct and positive impacts on the lives of individuals with autism.

Table 1. Intervention Ingredients for Behavioural, Developmental-Behavioural, and Developmental Interventions.

Intervention Technique & Definition	Interventions that include the technique	Other names for the technique	Interventions that use different names	Other Definitions	Kind of Intervention
Prompting – Assisting the child to respond correctly to cue. Interventions differ on the stress on type of prompt acceptable as well the staging of prompts.	All known interventions	Physical Prompt	DTT, PRT, Milieu teaching, Incidental teaching, RIT, Interpersonal Synchrony, JAML	The therapist physically manipulates the child's body in order to produce desired action/behaviour	Behavioural (B), Developmental-Behavioural (DB) & Developmental (D)
		Verbal prompt	DTT, PRT, Milieu teaching, Incidental teaching, RIT, Interpersonal Synchrony, JAML, ESDM	The therapist uses only verbal commands/instructions (such as 'Say ball') in order to produce desired action/behaviour	
		Indirect Prompting	RDI, DIR/Floortime	The therapist uses indirect words or manipulations of the environment in order to produce a desired behaviour (e.g. 'I think it's cold outside' to help	

				child take an action to wear a jacket)	
		Scaffolding	RDI, DIR/Floortime, ESDM, SCERTS, Hanen more than Words	The therapist provides accommodations and supports in order to help child achieve desired behaviour/action	
Reinforcement - Given in order to strengthen a response. Can be positive (child has access to object) or negative (object is taken away), verbal, object-based or social in nature. Behavioural approaches rely heavily on object-based reinforcers (e.g. toys, food) as well as both positive & negative reinforcement. DSP approaches rely primarily on social reinforcement.	All known interventions	Natural Reinforcement	PRT,ESDM, Milieu Teaching, Hanen More than Words, Interpersonal Synchrony	The activity is directly related to reinforcement such that it is a natural consequence of the activity/behaviour (e.g. child requests for water & is given a glass) rather than providing an unrelated object. Tangible and social rewards are provided contingent upon specific target behaviors that are defined by the teacher a priori.	B, DB, D

		Contingent Reinforcement	PRT, DTT, Milieu teaching, Incidental teaching	Small, behaviourally-defined attempts by the child are reinforced through access to an immediate tangible or social reward	B & DB
		Praise/Social Reinforcement	RIT, RDI, DIR/Floortime, SCERTS	Child is reinforced by verbal praise or approval or through physical touch, e.g., hug, pat, hi-five. Praise is not necessarily contingent upon or temporally linked to pre-defined target behaviors, and child is not denied access to tangible items or activities for reinforcement purposes.	D, DB
		Encouraging	JAML	Child is reinforced through verbal praise or approval or through physical touch e.g. hug, pat, hi-five. Praise is	DB

				not necessarily contingent upon or temporally linked to pre-defined target behaviors, and child is not denied access to tangible items or activities for reinforcement purposes.	
Time Delay - Creating a brief delay in the action/instruction/comment in order to increase independence in initiation and spontaneity of response.	All known interventions	Waiting time	JAML, SCERTS,	-	DB, D
		Pause/Slowing down	RDI, Hanen More than Words, DIR/Floortime,	-	D
Environmental Manipulation – Manipulating the environment in order to help child produce a desired response. Includes concepts like structuring environment, removing distractions, keeping the	All known interventions	Structured environment	DTT	stresses table top structuring	B
		Structured environment	Hanen More than Words	stresses natural environment to be structured and distraction free	D

environment natural, and motivating operations.		environmental arrangement	Interpersonal Synchrony	-	DB
		controlling access	PRT, Milieu Teaching	The therapist controls access to desired objects in order to produce a response	DB
		playful obstruction	DIR	Obstructing child from gaining access to desired object/goal to produce a response	D
		Framing	RDI	Structuring the environment to enhance response rate	D
		communicative temptations	Incidental teaching	Creating situations which reinforce child to communicate (can include blocking access, tempting etc.)	DB
Cueing – Giving precise instructions to the child.	DTT, PRT, Incidental Teaching, RDI, RIT, SCERTS, Interpersonal	Commenting on joint focus	RDI, DIR, Hannen More than Words, ESDM	Therapist verbalises/comments to direct child's attention to object of interest	DB & D

Behavioural –focus is on generating a response ("What is this?") DSP – focus is on directing attention ("Watch me")	Synchrony, JAML, ESDM	Focusing	JAML	Directing attention using verbal/non-verbal means to a joint focus	DB
Fading – Guiding the individual through less and less of prompt with the ultimate goal of eliminating it	DTT, Milieu Teaching, ESDM, Incidental teaching, JA mediated learning, PRT, RIT, Theory of Mind Training, RDI	-	-	-	B, DB, D
Shaping – Gradually prompting the child towards closer approximations to the correct response/action	DTT, ESDM, Incidental teaching, JA mediated learning, PRT, RIT	-	-	-	B & DB
Modelling – It involves an adult or peer providing a demonstration of the target behaviour or language	DTT, Milieu Teaching, RDI, SCERTS, Interpersonal Synchrony,	-	-	-	B, DB & D

response that should result in an imitation of the target behaviour.	JAML, ESDM, PRT, RIT				
'Following the child's lead' – Child initiates action/play and teaching is based around the child's interest	PRT, Milieu Teaching, Hanen more than Words, RDI, DIR/Floortime, SCERTS, RIT, ESDM, Interpersonal Synchrony, JAML	-	-	-	DB, D
Adult/Caregiver Responsiveness – Includes techniques like heightened responsiveness, increased emotional affect & emotional exchanges, indirect language stimulation.	DIR/Floortime, RDI, Hanen more than Words, RIT, ESDM, SCERTS, JAML	-	-	-	D, DB
Scaffolding – Giving the child support (could be environmental or verbal) in order to achieve a desired behaviour (e.g. decreased distance between child & therapist).	RDI, DIR/Floortime, ESDM, SCERTS, Hanen more than Words	-	-	-	D, DB

Could be used as a prompting technique or shaping technique					
Contingent Imitation – Imitating child's behaviour/actions/verbalizations	RIT, ESDM, DIR/Floortime, JAML, SCERTS	-	-	-	DB, D
Reading the child's intentions – Observing the child carefully and understanding the meaning of the behaviour undertaken.	RDI, DIR/Floortime, JAML, ESDM	-	-	-	D, DB
Linguistic Mapping – Using language to describe child action/behaviour the child is undertaking. Depends on child's level of language.	RIT, DIR/Floortime, RDI, PRT, ESDM	Labelling/ commenting	RDI, DIR/Floortime, JAML	-	D, DB
Joint activity routines – Therapist and child are equal partners engaged in the same cooperative activity.	ESDM, DIR/Floortime, RDI, Hanen More than Words, JAML	-	-	-	D, DB
Challenging the child – Building on the child's current skills by adding a challenge to move him/her up to the next level of development	SCERTS, RDI, PRT, ESDM	Zone of proximal development	SCERTS, RDI	-	D, DB
Activity Variations –	Incidental teaching, RDI,	-	-	-	D, DB

Adding changes in activities and routines both within activities and across tasks	DIR/Floortime, SCERTS				
Visual supports – are mostly picture tools that enable a learner to independently track events and activities	SCERTS, ESDM, Hanen more than Words	-	-	-	D, DB
Chaining – Tying together two desirable behaviours in order to produce a behavioural sequence	DTT, Incidental Teaching, ESDM	-	-	-	B, DB
Pacing – Frequency with which a prompt is made/behaviour is expected	DTT, RIT, PRT	Task timing	PRT	Interspersing maintenance task (tasks already mastered) with acquisition task (tasks to be mastered)	B, DB
Mand- Asking question or specific instructions in order to elicit a response	Milieu Teaching, DTT	-	-	-	B, DB
Extinction – A strategy that withdraws or terminates the reinforcement of a behaviour/action to reduce or eliminate the behaviour/action.	DTT	-	-	-	B

Table 2: Evidence-base for ASD-targeted interventions

	Paper	Design	Sample size	Age	Intensity	Comparison group	Skill measured	Gains above control
EIBI	Itzchak et al. (2008)	LES	81 (44 tx, 37 TAU)	19-35m	tx: 35-40 h/week	TAU/ Eclectic	IQ, ASD diagnosis	IQ
	Cohen, Amerine-Dickens & Smith (2006)	LES	42 (21 EIBI, 21 cont.)	24-45m	tx: 32-40 h/week	Community Int.	IQ, language, NV skills, adaptive behaviour	IQ, adaptive behaviour
	Eikeseth et al (2002). EIBI vs Eclectic	LES	25 (13 tx, 12 cont.)	4-7y	tx: 28 h/week for first year	TAU/ Eclectic	IQ, visuo-spatial skills, language, adaptive functioning	IQ, receptive language, expressive language, total language, VABS Communication, VABS Composite.
	Eikeseth et al (2007). EIBI vs Eclectic. Follow-up of participants in Eikeseth et al., 2002, at an average of 8 years of age.	LES	25 (13 tx, 12 cont.)	4-7y	tx: 28 h/week for first year	TAU/ Eclectic	IQ, visual-spatial skills, language, adaptive functioning (VABS). All blinded. Behavioral problems - teacher report (not blinded).	VABS Communication, VABS Daily Living, VABS Socialization, VABS Composite, VABS Maladaptive Behavior. Achenback CBCL-TRF Social (not blinded), Achenbach CBCL-TRF Aggressive (not blinded).
	Eikeseth et al (2012)	LES	59 (35 tx, 24 cont.)	24m -7y	tx: 23 h/week for first year	TAU/ Eclectic	VABS adaptive behavior subscales; VABS maladaptiv	VABS Adaptive Behaviour - Total. VABS Adaptive

							e behavior subscale; CARS (autism severity). All non-blinded.	Behavior - all sub-scales (non-blinded).
	Eldevik et al (2012)	LES	43 (31 tx, 12 cont.)	42-46m	tx: 13.6 h/week	TAU/ Eclectic	IQ, adaptive functionin g	IQ, adaptive behaviour
	Fava et al. 2011	LES	22 (12 tx, 10 cont.)	26-81m	tx: 14 h/week, cont: 12 h/week	TAU/ Eclectic	ADOS, mental developme ntal standard scores, CDI, frequency of challengin g behaviours	autism severity, developme ntal and language skills, challengin g behaviours
	Howard et al. (2005)	RCT	61 (29 EIBI, 16 HI eclectic, 16 LI eclectic)	31-37m	tx: 25-40 h/week, HI: 25-30 h/week, LI: 15 h/week	HI vs LI eclectic	IQ, visual-spatial skills, language, adaptive functionin g	outcomes did no differ for two eclectic groups, Tx group performed sign better on all measures, except motor skills
	Lovaas (1987)	LES	40 (19 tx, 21 cont.)	34-42m	tx: 40 h/week	TAU	IQ, academic placement, ASD diagnosis.	IQ, ASD diagnosis
	Magiati, Charman & Howlin (2007)	LES	44 (tx: 28 EIBI, cont: 16)	23-53m	tx: 32.4 h/week, cont: 25.6 h/week for first year	Communit y Int.	ADOS, language, play, cognition, VABS	VABS daily living only
	Reed, Osborne & Corness (2007a)	LES	27 (14 tx, 13 tx)	41-43m	HI tx: 30 h/week, LI cont.: 12 h/week	Low Intensity int.	Autism symptoma tology, Developm ental	IQ, educationa l functionin g

							functionin g, IQ, adaptive functionin g	
	Reed, Osborne & Corness (2007b)	LES	48 (12 tx, 20 eclectic cont, 16 portage)	38-43m	tx: 30.4 h/week, eclectic cont: 12.7 h/week, portage: 8.5 h/week	Communit y Int and Portage (parent-administra ted)	Autism symptoma tology, Developm ental functionin g, IQ, adaptive functionin g, comorbid problems	IQ, educationa l functionin g
	Remingt on et al. (2007)	RCT	44 (tx: 23, cont: 21)	30-42m	tx: 25.6 h/week, cont: 15.3 h/week	TAU	IQ, language, adaptive functionin g, behaviour functionin g, Nonverbal social communic ation, parental wellbeing	Increase mental age, IQ, language, adaptive functionin g, positive social behaviours .
	Sallows and Graupne r (2005)	RCT	23 (tx: 13, cont: 10)	24-42m	tx: 37.6 h/week, cont: 31.6 h/week	Parent-directed EIBI	IQ, language, adaptive functionin g, social functionin g, academic functionin g	Similar performan ce on both groups on all outcome measures.
	Smith, Groen & Wynn (2000)	RCT	28 (tx: 15, cont: 13)	18-42m	tx: 24.5 h/week, cont: 15-20 h/week	Parent training	IQ, Language, VABS, Socio emotional functionin g (Achenbac h checklist), academic skills, class placement,	IQ, visual-spatial ability, language, academic achieveme nt

							progress in treatment, parent evaluation	
	Strauss et al. (2012)	LES	44 (tx: 24, cont: 20)	26-81m	tx: 35 h/week, cont: 12 h/week	TAU/ Eclectic	ADOS, VABS, CDI, mental developmental standard scores, frequency of challenging behaviours	Autism symptom severity, mental developmental state and early language production
RDI								
Floortime/DIR/PLAY Project	Casenhiser, Shanker & Stieben, (2013)	RCT	51 (tx: 25, cont: 26)	24-48m	tx: 3.9 h/week	TAU	Language, social	Social, not language
	Jocelyn, et al. (1998)	Community-based RCT	35 (tx: 16, cont: 19)	24-72m	tx: 21.4 h/week, cont: 19.9 h/week	Community Int.	TRE-ADD Autism Quiz, Autism Behaviour Checklist, Developmental Measures (EIDP/PSDP), Stress Arousal Checklist, The Family Assessment Measure, Client Satisfaction Questionnaire	Language in EIDP/PSDP, TRE-ADD Autism Quiz; Client Satisfaction Questionnaire
	Pajareya and Nopmaneejumruslers (2011)	Pilot Community-based RCT	32 (tx: 16, cont: 16)	24-72m	tx: 15.2 h/week	TAU	FEAS/ CARS/ FEQ	Gains in all 3 measures
	Solomon et al (2014)	RCT	128 (tx: 64, cont: 64)	2.8-5.11 y	tx: 14 h/week	Community Int.	ADOS, SCQ, parent-child	ADOS, parent-child interaction

							interactions (MBRS, CBRS), developmental (Mullen, FEAS), language (MCDI)	s, trends in developmental
Hanen's More Than Words	Carter et al (2011)	RCT	62 (tx: 32, cont: 30)	15–25m	Not stated	TAU	Parental responsivity Communication	Communication dependent on baseline
	McConachie et al. (2005)	RCT	51 (tx: 26, cont: 25)	24-48m	tx: 20 h/week	TAU	ADOS, BSQ, JAFA, MCDI – vocab	CDI – vocab, JAFA
PACT project	Aldred, Green & Adams (2004)	RCT	28 (tx: 14, cont: 14)	24m -6y	tx: 3.5 h/week	TAU/ TEACCH	ADI, ADOS, VABS, Language, parent-child interaction, parenting stress	ADI, ADOS, VABS, Language, parent-child interaction
	Green et al (2010)	RCT	146 (tx: 74, cont: 72)	2.4-11y	tx: 3.5 h/week	TAU	ADOS-G social communication, parent-child interaction, language, adaptive behaviour	Parent-rated language, assessor-rated parent-child interactions, restricted/ repetitive behaviours
Project imPACT								
'Preschoolers with Autism' (PEBM)	Tonge et al (2014)	RCT	105 (tx: 70, cont: 35)	30-61m	tx: 1.25 h/week, cont: 1.25 h/week	Parent ed and counselling	VABS, emotional/ behavioural problems, developmental level, language	VABS communication, socialisation (dependent on baseline), daily living skills, some motor

Building Blocks	Roberts et al (2011)	RCT	95 (CB tx: 28, HB tx: 28, WL: 29	26-60m	CB: 2 h/week, HB: 1 h/week	HB, WL	Communication, social skills, adaptive functioning and psychopathology	VABS social scale, language comprehension (Reynell) for centre-based over home-based
SCERTS								
Denver Model								
RPMT (Yoder /Stone) Responsivity Prelinguistic Milieu Teaching	McDuffie, Lieberman & Yoder (2012) VS PECS	RCT	32 (RPMT: 16, PECS: 16)	18-60m	RPMT: 1 h/week, PECS: 1 h/week	PECS	Object interest	Object interest
Sonrise								
JA Int. / JASPER	Drew et al (2002)	Pilot RCT	24 (tx: 12, cont: 12)	18-40m	JA: 7 h/week, cont: 11.2 h/week	Community int.	Language, Nonverbal IQ, ADI-R	Language, Nonverbal IQ
	Goods et al (2013)	Pilot RCT	15 (tx: 7, cont: 8)	3-5y	tx: 30 h/week, cont: 30 h/week	Community int.	SP, ESCS, engagement states and spontaneous communicative gestures	Diverse play, time spent engaged, requesting gestures
	Kaale, Smith, & Sponheim (2012)	RCT	61 (tx: 34, cont: 27)	24-60m	tx: 3.3 h/week	TAU	MSEL, ESCS, JA, JE	JA and JE initiation in play with teachers
	Kasari, Freeman, & Paparella (2006)	RCT	58 (tx: 20, play: 21, cont: 17)	3-4 years	tx: 30 h/week, cont: 30 h/week	symbolic play or TAU control	ESCS, Structured play assessment, caregiver-child interactions	Child-initiated JA, Mother–child interactions (Symbolic types,

								Level of play)
	Kasari et al (2008)	RCT	58 (tx: 20, play: 21, cont: 17)	3-4y	tx: 30 h/week, cont: 30 h/week	symbolic play or TAU control	JA skills, SP skills, mother-child interactions, and language development	Expressive language, language in JA int. for ppts with lowest language levels over SP or controls
	Kasari et al (2010)	RCT	38 (tx: 19, cont: 19)	21-36m	tx: 2.25 h/week	WL	Functional play, symbolic play, joint attention	Responsivity to JA, functional play (not symbolic)
	Kasari et al (2014)	RCT	112 (CMM tx: 60, CEM tx: 52)	2-5y	CMM tx: 2 h/week, CEM tx: 2 h/week	CMM vs CEM	ADOS, MSEL, Caregiver-child interaction (JA), ESCS, symbolic play	JE, JA, symbolic play
	Kasari et al. (2015)	RCT	86 (tx: 43, PEI cont: 43)	22-36m	Tx: 30 h/week classroom-based; 1 h/week experimental protocol, PEI cont: 30 h/week classroom-based; 1 h/week experimental protocol,	Parent-only psychoeducational intervention (PEI)	MSEL, language, PSI, Caregiver Involvement Scale	JE, play diversity, highest play level achieved, and generalization to the child's classroom for child-initiated joint engagement.
	Lawton and Kasari (2012)	Pilot RCT	16 (tx: 9, cont: 7)	3-5y	tx: 12 h/week	WL	ESCS, IJA, engagement states	IJA
	Oosterling et al (2010)	RCT	67 (tx: 36, cont: 31)	26-40m	tx: 7 h/week	TAU	Language, general improvement (Clinical Global Impression	None

							- Improvement scale), engagement, early precursors of social communication	
	Schertz et al (2013)	RCT	23 (tx: 11, cont: 12)	24-48m	tx: 3.5 h/week	Community int.	Turn-Taking, Initiating Joint Attention, Expressive Communication (MSEL), parent-child interaction, VABS	Moderate effect sizes, although not significant, in primary measures
	Wong (2013)	LES	33 (SP tx: 10, JA tx: 14, cont: 9)	3-6y	tx: 8 h/week	WL	Joint attention/ engagement, play	Joint attention/ engagement, play
STAR								
LEAP	Boyd et al (2014)	Quasi-experimental	196 (LEAP tx: 54, TEACCH tx: 85, NMS: 59)	36-60m	Not specified	LEAP and TEACCH vs NMS	Problem behaviour, Repetitive behaviour, ADOS, CARS, LIPS-R, MSEL, PICS, PLS-4, SCQ, SRS, RBS-R, VABS.	LEAP: Autism Characteristics (ACS), communication, Reciprocal Social Interaction (RSI) TEACCH: ACS, Comm, RSI NMS: ACS, Comm
	Strain & Bovey (2011)	RCT	294 (tx: 177, cont: 117)	5-7y	tx: 21 h/week	Training manuals only	CARS, Mullen, PLS-4, SSRS, Social validity	Gains in all measures (cognitive, language, autism symptoms, problem behaviour and social skills)

ESDM	Dawson et al (2010)	RCT	45 (tx: 24, cont: 21)	18-30m	tx: 20 h/week	Community int.	IQ, adaptive behaviour, diagnosis, socialisation, motor skills, repetitive behaviour, daily living skills, language	IQ, adaptive behaviour, communication, daily living skills, language and motor skills. Not ADOS, socialisation or RB.
	Rogers et al (2012)	RCT	98 (tx: 49, cont: 49)	12-24m	tx: 1-15 h/week	TAU	Core ASD and developmental measures, imitation, JA (ESAT (12-15m), ITC, M-CHAT (15-24m), ADOS, Mullen, MCDI, VABS, Imitation, Social engagement (Dawson, 2004))	No group differences in gains between the tx and cont groups. Higher parent fidelity scores were significantly related to milder ASD symptoms and higher developmental scores.
iBASIS-VIPP (Video Interaction to Promote Positive Parenting)	Green et al. (2015)	RCT	54 (tx: 28, cont: 26)	7-10m	tx: 6-12 sessions/ 5 months	TAU	Primary: infant attentiveness to parent (MACI); Secondary: atypical infant behaviour, parent-child interaction, direct and parent-reported language, event-related	Gains in infant attentiveness, atypical infant behaviour, parent-child interaction and adaptive functioning.

							potentials to vowel change, attention disengagement, and adaptive function (AOSI; MSEL; VABS-II, MCDI).	
Father-mediated Intervention, VIT University	Louis and Kumar (2015)	RCT	30 (tx: 15, cont: 15)	30-60m	tx: 3 training sessions /6 months	TAU	The Play Based Observation (PBO), The Griffiths Mental Developmental Scales (GMDS), The Vineland Social Maturity Scale (VSMS) and the Rendel Shorts Questionnaire (RSQ; autism features)	Play, language acquisition, social engagement and behavior
RIT	Ingersoll (2010)	RCT	21 (tx: 11, cont: 10)	27-47m	tx: 3 h/week	TAU	IQ, language, adaptive behaviour	IQ, language, adaptive behaviour
Landa's Early Achievements intervention	Landa, Holman, O'Neill and Stuart (2011)	RCT	48 (IS tx: 24, Non-IS cont: 24)	21-33m	tx: 11.5 h/week	IS vs Non-IS	JA, shared positive affect, imitation, language	Imitation, social, language, cognitive
PECS	Carr and Felce, 2007b	RCT	41 (tx: 24, cont: 17)	36m-7y	tx: 15 h/week	TAU	Speech	Speech
	Gordon et al. (2011)	RCT	84 (tx: 26, WL: 30, cont: 28	4-10y	Unspecified	WL and non-treatment cont	Communication	Communication

	Howlin et al (2007)	RCT	85 (tx: 26, WL: 30, cont: 28	4-10y	Unspecified	WL and non-treatment cont	Language, ADOS, PECS use	PECS use, initiations
	Lerna et al (2012)	RCT	18 (relative group numbers unspecified	18-60m	tx: 1.5 h/week	Conventional Language Therapy	GMDS, ADOS, VABS, Unstructured Free-Play with Examiner	VABS social domain, JA requests and initiation, and duration of cooperative play in the PECS vs CLT group.
	Schreibman and Stahmer (2014)	RCT	39 (PECS tx: 19, PRT tx: 20)	20-45m	tx: 20 h/week	PECS vs PRT	MSEL, Spoken vocab, Adaptive communication, Augmentative communication	No differences in gains between groups
	Yoder & Lieberman (2010)	RCT	36 (PECS tx: 19, RPMT tx: 17)	18-60m	tx: 15 h/week of parent training	PECS vs RPMT	Communication (ESCS)	Communication (ESCS)
	Yoder and Stone (2006a,b)	RCT	36 (PECS tx: 19, RPMT tx: 17)	24-36m	tx: 15 h/week of parent training	PECS vs RPMT	Communication	Communication
PRT	Hardan et al (2015)	RCT	53 (tx: 27, cont: 26)	24m -6y	tx: 4 h/week, cont: 2 h/week	Psycho education	Language, VABS	Language, VABS
Sensory Integration	Schaaf et al (2014)	RCT	32 (tx: 17, cont: 15	56-83m	tx: 3 h/week	TAU	Self-care, GAS (individual goal attainment), PDDBI, PEDI, VABS	Self-care, GAS, PEDI
Scottish Centre for Autism	Salt et al (2002)	Pre-Post	17 (tx: 12, cont: 5)	34-50m	tx: 4 h/week	WL	Bayley Scale of Infant Development, BPVS, VABS,	Socialization, daily living skills, motor and adaptive behaviour

							PVCS, MCDI, Symbolic Play Test-2, ESCS, PSI-3 (not blinded).	composite scales of the VABS. Imitation scales of the PVCS. JA, social interaction
Early Social Interaction (ESI) project	Wetherby et al (2014)	RCT	80 (IESI tx: 40, GIESI cont: 40	18-24m	tx: 3 h/week, cont: 1 h/week	Individual vs group ESI	SCS, ADOS, VABS, Mullen	SCS; VABS Communication, Daily Living, Socialisation; Mullen Receptive Language

Table terms

LES= Large experimental study, **RCT**= randomised-controlled trial, **tx**= treatment group, **cont**= control group, **HB**= home-based, **CB**= centre-based, **WL**= wait list, **NMS**= Non-model specific, **IS**= Interpersonal Synchrony, **NIS**= Non-interpersonal Synchrony, **IESI**= Individual ESI, **GESI**= Group ESI, **JA =** Joint Attention, **JE =** Joint Engagement, **NV =** Non-verbal**, VABS =** Vineland Adaptive Behaviour Scale**, CARS =** Childhood Autism Rating Scale, **ADOS =** Autism Diagnostic Observation Schedule, **ADI-R =** Autism Diagnostic Interview - Revised, MSEL = Mullen Scales of Early Learning, **FEAS =** Functional Emotional Assessment Scale, **EIDP =** Early Intervention Developmental Profile, **PSDP =** Preschool Developmental Profile, **SCQ =** Social Communication Questionnaire, **MACI** = Manchester Assessment of Caregiver–Infant; **MCDI =** MacArthur Communicative Development Inventory, **BSQ =** Behavioural Style Questionnaire, **JAFA =** Joy and Fun Assessment, **SRS =** Social Responsiveness Scale, **ESCS =** Early Social Communication Scales, **ACS =** autism characteristics, **RSI =** reciprocal social interaction, **RB =** repetitive behaviours, **RBS-R =** Repetitive Behaviour Scale – Revised, **ESAT =** Early Screening for Autistic Traits, **GMDS =** Griffiths Mental Development Scales, **PDDBI =** Pervasive Developmental Disorder Behavior Inventory, **BPVS =** British Picture Vocabulary Scale, **PVCS =** Pre-verbal Communication Schedule, **LIPS-R =** Leiter International Performance Scale-Revised, **PLS-R =** Preschool Language Scales – Revised, **PSI =** Parenting Stress Index, **PEDI =** Pediatric Evaluation of Disability Inventory, **AOSI** = Autism Observation Scale for Infants

Table 3: Evidence-base for ASD-targeted interventions - additional information

	Paper	Participant inclusion/exclusion criteria	Blinded?	Random assignment?	Duration of int.	Design issues/limitations
EIBI	Itzchak et al. (2008)	ASD diagnosis. Aged 16- to 35-months.	No	No	1 year	No randomisation – grouped based on diagnoses. Etiologies and neurological pathogenesis need further investigation.

	Cohen, Amerine-Dickens & Smith. (2006)	ASD or PDD-NOS diagnosis. IQ above 35 on BSIDR. Aged 18- to 42-months. No additional medical diagnosis. Geographical availability. No >400 hours of behavioural intervention prior. Parental willingness.	Yes	No	>3 years	No randomisation. Quasi-experimental design.
	Eikeseth et al. (2002)	Aged 4- to 7-years. IQ greater than 50. Registered in particular community catchment area.	Yes	No	1 year	No randomisation. Clinical supervisor overlap.
	Eikeseth et al. (2007)	Aged 4- to 7-years. IQ greater than 50. Registered in particular community catchment area.	Partial	No	1 year	No randomisation. Non-blinded measures. Clinical supervisors overlap.
	Eikeseth et al. (2012)	Aged 2- to 7-years. Txt group enrolled in particular centre's services. No prior receipt of EIBI.	No	No	1 year	Non-blinded measures. Comparison group not assessed using VABS maladaptive behaviour subscale or CARS.
	Eldevik et al. (2012)	ASD or PDD-NOS diagnosis. Aged 2- to 6-years. IQ test and measure of adaptive behaviour at intake and after 2 years of intervention. Min. 5 hours/week intervention.	Yes	No	2 years	No randomisation. Bias in the referral process. No data on the factors affecting referral decisions. Only IQ and adaptive behaviour outcome data were available.
	Fava et al. (2011)	ASD or PDD-NOS diagnosis. No additional medical diagnosis.	No	No – parent choice	6 months	Assignment to treatment groups was parent-

						determined rather than random.
	Howard et al. (2005)	ASD or PDD-NOS diagnosis. Aged <48 months. Primary language: English. No additional medical diagnosis. No previous treatment >100hours.	No	No	7 months	Non-blinded measures.
	Lovaas (1987)	ASD diagnosis. Aged <40 months. Mental age >11 months.	Yes	No	2 years	No randomisation.
	Magiati, Charman & Howlin. (2007)	ASD diagnosis. Aged 22- to 54-months. No additional medical diagnosis. Primary language: English. Geographical availability. No other intensive intervention allowed.	No	No	2 years	Recruiting difficulties. No randomisation.
	Reed, Osborne & Corness (2007a)	ASD diagnosis. Aged 2- to 4-years. No other major intervention allowed.	Yes	No	9-10 months	Low statistical power. Beneficial to have a finer grained analysis of the day-to-day tasks and activities.
	Reed, Osborne & Corness (2007b)	ASD diagnosis. Aged 2- to 6-years. No other major intervention allowed.	No	No	10 months	Fidelity of the treatment regimes. Lack of direct observation.
	Remington et al. (2007)	ASD diagnosis. Aged 30- to 42-months. No additional medical diagnosis. Living in family home.	Yes – Cues may have revealed condition	No	2 years	No randomisation. Small sample size may have been the reason that some effects (such as parental adjustment to EIBI) were not found, as in larger-scale studies.

	Sallows & Graupner. (2005)	ASD diagnosis. Aged 24- to 42-months.	No	Yes	4 years	Although matched on age and IQ, other pre-treatment variables, such as imitation, correlated even more strongly with outcome and were not equal in the two groups.
	Smith, Groen & Wynn. (2000)	ASD or PDD-NOS diagnosis. Aged 18- to 42-moths. Geographical availability. IQ ratio between 35 and 75. No additional medical diagnosis.	No	Yes	1 year	Only one measure used to social skills – VABS social domain, based on parent report. Small sample size with heavily-tailed, skewed distribution of scores.
	Strauss et al. (2012)	ASD or PDD-NOS diagnosis. No additional medical diagnosis. Completed 6 months of treatment. Re-evaluated after 6 months.	No	No	6 months	Additional measures on treatment and parental factors were not available for both study groups – only for EIBI.
DIR	Casenhiser, Shanker & Stieben. (2013)	ASD diagnosis only. Aged 2- to 5-years. Completed 12 months intervention. Parent willingness.	Yes	Yes	12 months	Treatment group is neither a no-treatment group nor a group receiving a uniform type or dosage of treatment. Unavoidable self-selection bias.
	Jocelyn et al. (1998)	ASD or PDD diagnosis only. Aged 24- to 72-months. Geographical availability. Physically-able to participate. Attend day care or school.	Partial – Blinded: EIDP/PSDP	Yes	12 weeks	Heavy reliance on non-blinded parent report measures.

	Pajareya & Nopmaneejumruslers. (2011)	ASD diagnosis only. Geographical availability. Literate, healthy parents.	Yes – FEAS	Yes – based on age and symptom severity.	3 months	Limited DIR-specific measures. No documentation of number of hours of parent-led therapy.
PLAY project	Solomon et al. (2014)	ASD only. Aged 3- to 5-years. Primary language: English.	Partial – non blinded parent report (MCDI).	Yes	12 months	Both groups received similar community services, making it difficult to assess PLAY in isolation.
Hanen's 'More Than Words'	Carter et al. (2011)	ASD only. Aged <24months. No genetic disorder.	Partial – majority blind.	Yes	3.5 months	Non-norm referenced instruments.
	McConachie et al. (2005)	Suspicions of ASD. Aged 24- to 48-months.	Partial – Joy and Fun Assessment ratings blinded.	No – dependant on availability.	3 months	Participant bias - the research group was significantly more economically advantaged than the ''refusers''.
PACT/ JA	Aldred, Green & Adams. (2004)	No severe global developmental delay. Primary language: English. Not hearing or visually impaired. Literate and healthy parents.	Yes	Yes	12 months	No experimental control for whether the gains were the result of extra non-specific therapist attention, or reduced parental anxiety.
	Green et al. (2010)	No ASD twins. Non-verbal age: 12 months or younger. Primary language: English. Not hearing or visually impaired.	Partial – Blinded Preschool Language Scales and video-rated interaction.	Yes	18 months	No range of the autism spectrum that was often included in previous trials.
Preschoolers with Autism (PEBM)	Tonge et al. (2014)	No Asperger's and PDD-NOS allowed. Primary language: English.	Partial – non-blinded parent reports.	Yes	20 weeks	Parent reports (non-blinded). Significant differences between the

		No ABA programme followers.				control group and the treatment groups for age, developmental level, level of language development, symptoms of autism and adaptive behaviour. In general, the control group was older and higher functioning at initial assessment. No fidelity scoring.
Building Blocks	Roberts et al. (2011)	ASD only.	No	Yes	40 weeks	Parent report (non-blinded).
RPMT	McDuffie, Lieberman & Yoder (2012) vs PECS	Aged 18- to 60-months Not hearing impaired. No severe sensory or motor deficits. Less than 10 words.	Yes	Yes	6 months	Children in both conditions received an active treatment; maturation cannot be easily separated from intervention effects.
JA	Drew et al. (2002)	ASD via CHAT	No	Yes	12 months	Parent report. Non-matched groups at initial assessment for NVIQ. Lack of systematic checking regarding the hours and fidelity of the parent training intervention. Some in control group began receiving other intervention during the study.
	Goods et al. (2013)	ASD diagnosis. Aged 3- to 5-years. Less than 10 words.	Yes	Yes	12 weeks	The small sample may not have been at the

		Attending non-public school.				appropriate developmental level to gain joint attention gestures, which was a focus of this study.
	Kaale, Smith, & Sponheim (2012)	ASD diagnosis. Aged 24- to 60-months. Attending preschool. Primary language: Norwegian.	Yes	Yes	8 weeks	Absence of confirmation for participant clinical diagnoses.
	Kasari, Freeman, & Paparella (2006)	ASD Diagnosis. Aged 3- to 4-years. No additional medical diagnoses. Geographical availability.	Yes	Yes	5-6 weeks	Unmeasured engagement in other interventions.
	Kasari et al (2008)	ASD diagnosis. Aged 3- to 4-years. No additional medical diagnoses. Geographical availability. Min. 4 weeks EIP.	Partial – Observations blinded.	Yes	5-6 weeks	Engagement in other interventions – difficult to control for sole effects of intervention.
	Kasari et al. (2010)	ASD diagnosis. Aged >36 months.	Yes	Yes	8 weeks	Services may not have been consistent as service-providers were not coordinated. Some parents may have been receiving either structured behavioural guidance, while others were encouraged to use a more relationship-based approach.
	Kasari et al. (2014)	No genetic comorbidities.	Yes	Yes	12 weeks	Difficulty with recruitment and retention – 28% final dropout.
	Kasari et al. (2015)	Clinical diagnosis of ASD confirmed by independent testers (ADI-R and ADOS).	Yes	Yes	10 weeks	Study designed to isolate active ingredient of intervention – only compared

		No significant physical disabilities. Parent and child were available for follow-up assessments.				to 1 other intervention. Further work required to expand types of intervention.
	Lawton & Kasari. (2012)	ASD diagnosis. Aged 3- to 5-years. Attending public preschool. Teacher willing to take part in study. No additional medical diagnosis.	Yes	Yes	6 weeks	Limitations in the school calendar affecting experimental control.
	Oosterling et al. (2010)	ASD or PDD-NOS only. Aged 12- to 42-months. Primary language: Dutch	Partial	Partially – Based on location however.	2 years	Biased randomisation. No formal checking of treatment fidelity.
	Schertz et al. (2013)	ASD only. Absence of JA. Aged <30 months.	Partial – Blinded observations	Yes	7 months	Non-blinded standardised measures.
	Wong. (2013)	ASD only.	Blind coding of behaviour	Yes	2 months	Small sample size did not allow for analysis of how the implementation of the intervention was related to children's outcomes.
LEAP, TEECCH	Boyd et al. (2014)	ASD or developmental delay diagnosis. Aged 3- to 5-years. No previous exposure to CTM. Min. 6 months exposure to treatment or control condition.	No	No	3 years	Selection bias Assessors were not blind to children's group assignment. NMS (non-model-specific) group, not TAU.
	Strain & Bovey. (2011)	Unspecified.	Partial	Yes	2 years	No direct observational measures of child behaviour - information from schools only.

ESDM	Dawson et al. (2010)	ASD diagnosis.	No – Parent report.	Yes	2 years	Not blinded.
	Rogers et al. (2012)	Literate, healthy parents. No other intervention >10 hours/ week. No additional medical diagnosis.	Yes	Yes	12 weeks	The community group received almost double the amount of intervention than the P-ESDM group. P-ESDM group were not receiving the "full dosage" of the intervention until the very end of the measurement period.
iBASIS-VIPP (Video Interaction to Promote Positive Parenting)	Green et al. (2015)	No substantial medical disorder in the infant No twins No prematurity of less than 34 weeks No birthweight of less than 5 lbs (2·27 kg).	Yes	Yes	5 months	Assessors blinded, but families not blinded. Selection bias – family/clinic referral. Low generalisability – sample included restricted sample of high income/qualification families.
Father-mediated Intervention, VIT University	Louis and Kumar (2015)	No comorbid conditions such as seizure disorder Only children who fulfilled the criteria on DSM-IV for autism were included	No	Yes	6 months	TAU
RIT	Ingersoll. (2010)	Not outlined.	Yes	Yes after matching	10 weeks	Groups were not equivalent on elicited imitation performance at pre-treatment.
Early Achievements Intervention	Landa et al. (2011)	ASD diagnosis. Aged 21- to 33-months. Non-verbal mental age >8 months. No ASD diagnosed siblings.	Yes	Yes	6 months	Children in both conditions received an active treatment; maturation cannot be easily separated from

		Primary language: English.				intervention effects.
PECS	Carr & Felce. (2007)	ASD diagnosis. Aged 3- to 7-years. No previous PECS teaching.	No	No – location-based	4-5 weeks	Additional staffing resources required.
	Gordon et al. (2011)	ASD diagnosis. Aged 4- to 11-years. Little or no functional language. No sensory impairment. No previous PECS teaching.	No	Yes	20 months	Response predictor analysis based on subgroups (communication / symptom severity). Study not powered for this - chance of Type II errors. Limited resources – reliance on classroom observations only. Few generalizable requesting opportunities.
	Howlin et al. (2007)	ASD diagnosis. Aged 4- to 11-years. Little or no functional language. No sensory impairment. No previous PECS teaching.	No	Yes	5 months	No follow-up of teacher fidelity. Limited resources – reliance on classroom observations only. Few generalizable requesting opportunities. Assessors not blinded to group allocation.
	Lerna et al. (2012)	ASD diagnosis. Aged 18- to 60-months Little or no functional language. No PECS or other ACC systems. No additional medical, motor or sensory diagnosis.	Yes	No	6 months	No randomisation - convenience sample.
	Schreibman &	ASD diagnosis. Aged <48 months.	Yes – In-home treatme	Yes	23 weeks	Children in both conditions received an

	Stahmer. (2014)	No more than 9 words. No additional sensory and medical diagnosis No previous PECS or PRT. Parental willingness.	nt and ADOS			active treatment; maturation cannot be easily separated from intervention effects.
	Yoder & Lieberman. (2010)	Aged 18- to 60-months. Less than 10 words. Pass a hearing screening.	Yes	Yes	6 months	Measures different to real-life PECS - children did not have to make any discrimination among symbols before making the picture exchange.
PECS / Milieu	Yoder & Stone. (2006a,b)	ASD or PDD-NOS diagnosis. Aged 18- to 60-months. Less than 10 words.	Partial – Blinded to hypotheses only.	Yes	6 months	Pre- and post-assessments were also the primary data coders and could not be kept blind to the children's treatment assignment.
PRT	Hardan et al. (2015).	ASD diagnosis. Aged 2- to 6-years. Communication-Delayed on Preschool Language Scale. Participation >60 min weekly individual speech therapy. No additional medical diagnosis.	Partial	Yes	12 weeks	Varying doses of PRT depending on consistency of parent implementation. Necessary to allow continuation of community treatments. Differences between PRTG and PEG. No data regarding fidelity of treatment implementation. Parent report (non-blinded)
Sensory Integration	Schaaf et al. (2014)	ASD diagnosis. Aged 4- to 7-years. Non-verbal cognitive level. Difficulty processing	Yes	Yes	10 weeks	Although participants were randomised based on autism severity and

		and integrating sensory information. Parent willing to attend.				cognition, unable to include strata in analysis due to sample size.
SCA	Salt et al. (2002)	ASD only.	No	No	10 months	No randomisation – non-random assignment of participants resulted in the control group possessing a significantly higher mean IQ at treatment onset.
Early Social Interaction (ESI)	Wetherby et al. (2014)	ASD only. Aged 16- to 20-months. Geographical availability.	Yes	Yes	9 months	Children in both conditions received an active treatment; maturation cannot be easily separated from intervention effects. Non-blinded parent reports - effects observed on adaptive behaviour may be confounded by higher parent expectation in individual-ESI, compared to group-ESI.

CHAPTER 3: MOTOR DEVELOPMENT AND MOTOR RESONANCE DIFFICULTIES IN AUTISM: IMPLICATIONS FOR EARLY INTERVENTION FOR LANGUAGE AND COMMUNICATION SKILLS

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3.1. Introduction

Autism is a pervasive developmental disorder that is diagnosed based upon behavioural criteria for impairments in social skills, communication and language skills, and restricted interests and repetitive behaviours. Autism is currently considered to be a “spectrum” disorder, with three Pervasive Developmental Disorders now being termed Autism Spectrum Disorders (ASDs): Autistic Disorder, Aspergers Disorder, and Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS). Individuals with these three different ASDs differ somewhat in regards to the nature and/or severity of their early language and intellectual difficulties. However, individuals with these three ASDs are similar in that they share impairments in social and communication skills, and that the onset of their difficulties begins by three years of age (American Psychiatric Association, 2000).

The only motor abnormalities currently included in the diagnostic criteria for ASDs are stereotypical repetitive behaviours (American Psychiatric Association, 2000; see also Lord and Jones, 2012). These repetitive behaviours include motor stereotypies, such as hand and finger mannerisms, body rocking, and arm flapping (Lord et al., 1994; Loftin et al., 2008). However, impairments in motor development commonly observed in children and adults with ASDs are not limited to motor stereotypies (Kopp et al., 2010; Linkenauger et al., 2012). Early motor delays, gait abnormalities, and difficulties with gross and fine motor coordination, postural control, and imitation have been found to constitute significant neurological co-morbid conditions in this population (Provost et al., 2007; Bhat et al., 2011; Maski et al., 2011). For example, Nobile et al. (2011) examined motor dysfunction in ASDs and found that children diagnosed with Autistic Disorder presented with stiffer gait, difficulties maintaining a straight line while walking, and postural abnormalities. Similarly, other studies have reported an "ataxic" gait in adults with autism (Hallett et al., 1993), and reduced postural stability, especially when somatosensory input was disrupted (Minshew et al., 2004). Deficits in postural stability and motor coordination in individuals with ASDs were confirmed through a recent meta-analysis conducted by Fournier and colleagues (Fournier et al., 2010). Children and adults with autism have also been found to exhibit praxis and imitation difficulties, including manual, postural, and orofacial imitation (Rogers et al., 1996, 2003; Stone et al., 1997; Stone and Yoder, 2001; Williams et al., 2004; Mostofsky et al., 2006; Dziuk et al., 2007; Vanvuchelen et al., 2007, 2010; Stieglitz Ham et al., 2008; Dowell et al., 2009). Critically, evidence suggests that deficits in motor skills, coordination, and balance are not limited to individuals with ASD experiencing cognitive delays (Jansiewicz et al., 2006). A variety of mechanisms have been proposed to account for the motor functioning differences observed in individuals with ASDs, including abnormalities in the cerebellum (Fatemi et al., 2012), impairments in frontal-striatal connections (Fournier et al., 2010), difficulties in self-other mapping (Williams et al., 2001), impaired sensory input (Gowen and Hamilton, 2013), and impaired multisensory integration (Gowen and Hamilton, 2013).

The aim of the current review is to outline the evidence for ASD-related motor development and motor resonance difficulties, and to examine current research on interventions that attempt to apply motor-related approaches to improve speech/language and social communication skills in children with autism. Similar to recent reviews by others (e.g., Iverson, 2010; Bhat et al., 2011), we first describe the existing evidence for early delayed, impaired, and atypical motor development in autism. In this review, we place particular emphasis on research related to several motor development mechanisms and milestones believed to be associated with concurrent and later speech/language and social communicative functioning. Next, we address current evidence for impairments in motor resonance (i.e., "mirror neuron") functioning in individuals with autism, which has implications for social engagement during communication interactions. After this, we carefully examine and evaluate the existing motor-related autism intervention research that targets speech/language and social-communication skills. This includes augmentative and alternative communication (AAC) interventions, more directly motor-based behavioural interventions, electromagnetic brain stimulation interventions, and interventions that utilize synchronous motor activities to increase speech/language and social communication skills. The current review differs distinctly from previous reviews, which have focused primarily on interventions for sensorimotor skills themselves (e.g., Baranek et al., 2008; Bhat et al., 2011), as opposed to motor-related attempts to specifically target speech/language and communication skills. We conclude our review by describing research needs and future directions for research on early interventions for speech/language and social-communication skills from a motor-related perspective.

3.2. Early Motor Development in Autism

Evidence suggests that autism is caused by a complex combination of multiple genetic and environmental factors. Twin studies examining the concordance of autism in monozygotic versus dizygotic twins provide evidence that genetics play a key role (Folstein and Rutter, 1977; Ritvo et al., 1989; see also Hallmayer et al., 2011). In addition to strong genetic influence on the development of autism itself, milder versions of the social, communication, and other difficulties experienced by individuals with ASD have also been documented in unaffected first-degree relatives (i.e., siblings, parents) of those with ASDs (Landa et al., 1991; Bolton et al., 1994; Hughes et al., 1997; Piven and Palmer, 1997; Piven et al., 1997; Folstein et al., 1999; Murphy et al., 2000; Pickles et al., 2000; Bishop et al., 2004; Adolphs et al., 2008; Smith et al., 2009). These results provide evidence that the complex genetic mechanisms that contribute to the development of autism also impact upon other members of families affected by autism. This, then, creates an opportunity to explore the effects of familial/genetic risk factors on various brain and behavioural mechanisms early in life in ASD, through the study of infant siblings of children already diagnosed with ASDs (Rogers, 2009; Yirmiya and Charman, 2010).

Extensive research has been conducted on motor behaviours and motor-related skills in infants who are at high risk for developing autism, with solid implications for our understanding of motor development associated with autism (Iverson and Wozniak, 2007; Rogers, 2009). In a comprehensive review of the autism high-risk infant literature, Rogers (2009) concludes that delays in motor development have been a consistent finding in this population. Of particular note is her conclusion that some important, albeit subtle, repetitive movements, and unusual sensory behaviours appear to emerge earlier in development than impairments in social and communication skills in this population (Rogers, 2009). In this section of the review, we focus on the key findings of the autism early motor development literature, with an emphasis on those motor and motor-related behaviours that are believed to be most relevant to successful communication and language development.

One of the earliest developing motor-related behaviours having associations with language development is the vocal-motor and facial-motor coordination that emerges during face-to-face interactions in the first half of the first year of life (Iverson and Fagan, 2004). During this time, infants begin to engage in coordinated vocal and facial motor activity routines (such as reciprocal vocalizations, imitation of mouth opening, positive/negative facial expressions, and gaze) on a second-by-second timing scale, with both familiar and unfamiliar communicative partners. This motor synchrony reflects interpersonal coordination of listening to and producing vocal-motor activity, which can be considered developmental precursors to the timing pragmatics of interpersonal interaction during conversation (Colonnesi et al., 2012). Existing evidence suggests that the nature and degree of this early infant coordination and tuning of motor activity with others predicts later infant social-emotional and cognitive development in typically developing infants (Feldman et al., 1996).

Yirmiya et al. (2006) measured communicative synchrony in 4-month-old infant siblings of children diagnosed with autism and low-risk infants without a family history of autism during mother-infant interactions. They uncovered evidence for weaker synchrony for infant-led interactions in the high-risk group (see also Brisson et al., 2011). Furthermore, the authors reported that these infants at risk for autism displayed fewer non-verbal requesting behaviours (such as pointing), and performed worse than low-risk infants on the language scales of the Bayley Scales of Infant Development, in follow-up at 14 months of age (Yirmiya et al., 2006). These findings support the hypothesis that risk for autism is associated with impaired vocal-motor coordination synchrony at 4-months of age, and that this has relevance to the later development of linguistic and pre-linguistic behaviours.

Another major stage of links between motor activity and language development occurs during the second half of the first year of life (Bates et al., 1999; Bates and Dick, 2002). Studies have shown that sharp increases in coordinated and repetitive arm movement and hand banging co-occur with the onset of reduplicative babble (i.e., canonical babble; e.g., "baba") between 6- and 11-months of age in typically developing infants, likely reflecting entrainment of the vocal and manual motor systems (Locke et al., 1995; Iverson et al., 2007; see also Petitto and Marentette, 1991; Petitto et al., 2004). This relationship is robust across typical infants of widely varying age of reduplicative babble/hand banging onset (Eilers et al., 1993; Iverson et al., 2007), as well as children with delayed language, including those with Down Syndrome and those with Williams Syndrome (Cobo-Lewis et al., 1996; Masataka, 2001). Finally, delayed onset of reduplicative babble has been found to be a marker for delays in speech and language in the general population of infants (Oller et al., 1998).

In 2007, Iverson and Wozniak examined the rate of rhythmic arm movements during pre-babble and babble onset sessions in high-risk and low-risk infants. Rates of rhythmic arm movements increased from the pre-babble sessions to the babble-onset sessions in both high-risk and low-risk infants; however, this increase was lower in the high-risk group (Iverson and Wozniak, 2007). In addition, the high-risk infants exhibited delays in reduplicative babble onset and first word use between 5 and 14 months of age, as well as delays in language development at 18 months of age (Iverson and Wozniak, 2007). A related study by Gernsbacher et al. (2008) found that scores on oral-motor (e.g., blowing bubbles) and manual-motor skills (e.g., pointing to request) during home videos distinguished infants who later developed autism from those who were typically developing, as well as infants who were later minimally and highly fluent. Together, these findings suggest that oral-motor and manual-motor skills may contribute to both social-communication and speech/language skills deficits in this population.

Another major stage of links between motor, speech, and language development occurs from approximately 10- to 20-months of age. There is evidence to suggest that typically developing infants learn to understand word-object relationships through repeated episodes of shared joint visual attention to an object (e.g., following a point to look at the ball together) paired with adults verbally labelling the objects (e.g., “ball”) during this period (Baldwin, 1995). This represents a complexity of emerging skills in following and comprehending the motor actions of others in relation to increasingly specific distal targets, and in increasingly dynamic activities and contexts (e.g., Tomasello and Farrar, 1986; Baldwin et al., 1996; Flom et al., 2004).

There is extensive evidence that both young children diagnosed with autism and young toddlers at risk for autism exhibit pervasive impairments in joint attention behaviours. In 2005, Goldberg and colleagues identified deficits in social-communicative behaviours, including responding to joint attention bids, in both 17-month-old high-risk infants and 2-year-old children already diagnosed with autism, compared with typically developing infants and children (Goldberg et al., 2005). In another study, involving 20-month olds diagnosed with autism, Charman (2003) found that declarative, triadic gaze switching was correlated with both language ability and autism symptom severity outcomes at 42 months of age (see also Yoder et al., 2009). Together, these results provide evidence to support the hypothesis that early deficits in the understanding of the gestures and actions of others are present from early in life in this population, and that these deficits are predictive of later social-communication and language deficits in children with autism (see also Rogers, 2009). Given the evidence from typical development, it will also be important to examine potential relationships between early exploratory and locomotor activity and later joint attention and language skills in infants at high risk for autism (see e.g., Campos et al., 2002).

Alongside the development of these social coordination and social-communication aspects of action perception and understanding, there is extensive evidence for more direct, in vivo links between gesture and language development in infants and children. Specifically, once infants have mastered the basic understanding of the gestures and actions of other people, they begin to regularly produce and employ increasingly complex communicative and symbolic gestures of their own, furthering their own communications and their language development (Bates and Dick, 2002). For example, the onset of recognitory gesture production, such as putting a cup to one's mouth and pretending to drink, correlates with the onset of vocal naming, both within and across infants between 11- and 16-months of age (Volterra et al., 1979; Shore et al., 1990). Between 18- and 20-months of age, gestures with one meaning are used in combination with words with other meanings, in order for the child to begin to be able to produce longer communications (e.g., point to chair and say “mom” to request that mom sits down; see Bates and Dick, 2002, for discussion). Impairments in the production of recognitory gestures as well as the coordination of speech and gesture during communication are core diagnostic measures of early childhood autism, which are included in the Autism Diagnostic Observation Schedule and the Autism Diagnostic Interview (Lord et al., 1994, 2000).

In this section, we have reviewed evidence that suggests that infants and young children with autism exhibit deficits and/or delays in a number of motor-related milestones that are believed to reflect critical stages in speech/language and communication development. Indeed, several of these deficits and delays have been found to be concurrently and/or predictively associated with important speech/language and social communication abilities in these infants and children. These motor and motor coordination milestones are likely to be supported by the core motor system and its mediators, including the primary motor cortex, cerebellum, motor-related frontal-striatal connections, visual regions involved in action perception, and a distributed system for sensorimotor integration (see Figure 1 and Table 4 for more information). These findings have clear implications for how motor-related interventions might be used to facilitate and support speech/language and communication development in this population, which is the focus of this review. Before we address this, however, we discuss the evidence for deficits in the motor resonance (i.e., "mirror neuron") system in individuals with autism. This system, which is involved in "mirroring" the actions of others within our own motor planning (i.e., premotor cortex) system, has been proposed to impact upon language development directly (Oberman et al., 2005), or to index social engagement with relevance for speech/language and social communication development in ASD.

3.3. Motor Resonance Deficits in Individuals with Autism

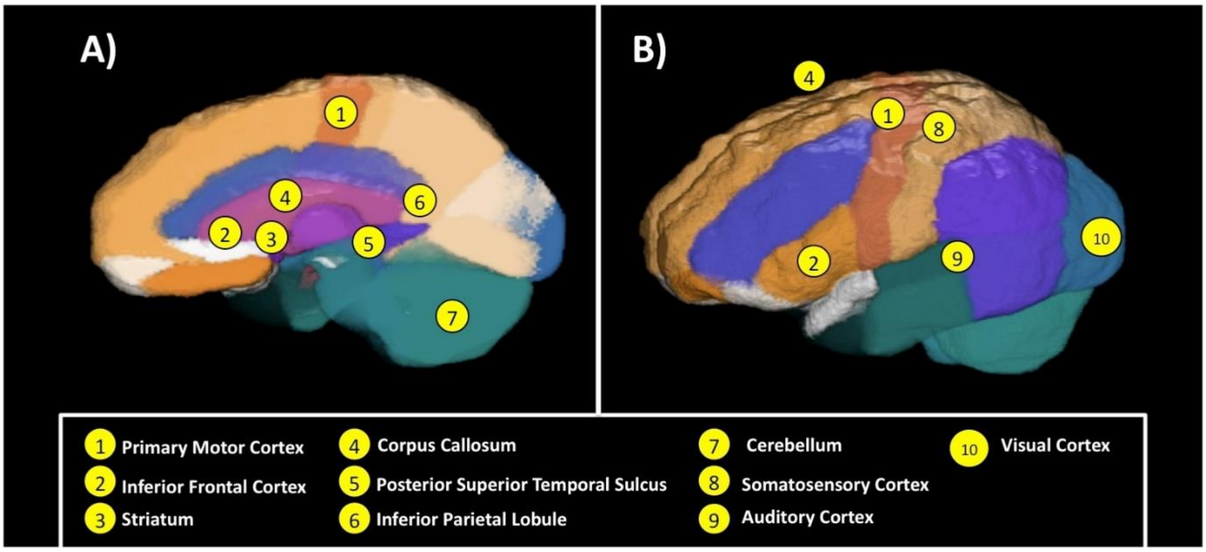
Extensive research, particularly over the past 15 years, has provided convincing evidence that our motor system "resonates" the actions of others that we view, hear, or view and hear (di Pellegrino et al., 1992; Rizzolatti et al., 1996; Iacoboni et al., 1999; Kohler et al., 2002; Gazzola et al., 2006). That is, our motor planning and related action production systems in pre-motor and other regions of the cortex appear to "mirror" the actions of observed others onto our own action/motor planning system (e.g., Inferior Frontal Gyrus, Inferior Parietal Lobule, Superior Temporal Sulcus; see Figure 1 and Table 4), presumably allowing us to better represent and understand the nature and details of the actions and activities of others (Rizzolatti and Craighero, 2004). This "mirror neuron" system (MNS) has been proposed to underlie a number of critical social-interactive and social-communicative skills, including imitation, language development, empathy, and understanding the social perspectives and intentions of others (Iacoboni and Dapretto, 2006). Following an initial suggestion that impairments in mirror neuron functioning may play an important role in the behavioural deficits observed in individuals with autism in 2001 (Williams et al., 2001), behavioural and neuroimaging research has sought to test this hypothesis. Although the findings are somewhat mixed, and there is particular debate about behavioural data on MNS functioning and its proposed relationship to imitation functioning in the literature (Southgate and Hamilton, 2008; see also Hamilton, 2009), the hypothesis of impaired motor resonance in individuals with ASD has generally been supported in the experimental behavioural and brain imaging literatures (Oberman and Ramachandran, 2007; Becchio and Castiello, 2012; Enticott et al., 2012; Oberman et al., 2012).

Table 4: Brain regions and mechanisms associated with motor aspects of language development

Number (see Fig 1.)	Brain Region or Mechanism	Description
1	Primary Motor Cortex	Primary cortical generator of motor activity, both simple and complex.
2	Inferior Frontal Cortex	Motor planning region, and key region of the frontal mirror neuron system; also includes Broca's area. Includes representations of hand and mouth actions, and has been implicated in links between

		hand and mouth actions that facilitate speech/language production and development.
3	Striatum	Portion of the subcortical basal ganglia system, involved in the modulation of movement; affected by inputs from motivational systems.
4	Corpus Callosum	Bundle of neural fibers that connect the left and right hemispheres of the brain, facilitating inter-hemispheric communication and coordination.
5	Posterior Superior Temporal Sulcus	Cortical region involved in biological motion perception. Key region of the posterior mirror neuron system, which has been specifically implicated in perceptual aspects of action encoding and understanding.
6	Inferior Parietal Lobule	Cortical region involved in the association and integration of sensory information. Key portion of the posterior mirror neuron system, which has been specifically implicated in goal-related aspects of action understanding.
7	Cerebellum	Neural region involved in the coordination, precision, and timing of movement, motor learning, and motor integration.
4, 8, 9, 10	Neural integration and connectivity	Both motor and language functioning require coordination and integration across multiple sensory modalities and hemispheres. For example, motor planning and motor coordination require integration of information from visual and motor cortices. Similarly, speech perception requires visual-motor integration (e.g., mouth movement, speech sounds), and meaningful/iconic language involves the integration of multiple real-world experiences with objects that are encoded within and across the visual, somatosensory, motor, and auditory cortices.

Figure 1: Neural Regions and Mechanisms. Cortical and subcortical regions involved in motor functioning, action perception, and neural coordination and connectivity for sensorimotor and speech/language functioning. See Table 4 for brief descriptions of these regions and associated mechanisms. Images of an average of 6-year-old child brain generated via the Magnetic Resonance Image database of Sanchez, Richards, and Almli (see Sanchez, Richards, & Almli, 2012).



Despite extensive evidence for reduced visuomotor resonance in individuals with autism, it is clear that the MNS is not entirely "broken" in this population. For example, individuals with ASD have been found to exhibit normal motor interference during simultaneous execution-observation of meaningless arm movements (e.g., Gowen et al., 2008; see Becchio and Castiello, 2012, for review). Most relevant to the current review, Oberman et al. (2008) used electroencephalography (EEG) mu suppression to uncover evidence for normal MNS activation during the observation of the actions of familiar people, but reduced MNS activation during the observation of the actions of unfamiliar people, in children with autism. These data provide direct evidence that the MNS of children with autism is, in fact, capable of responding normally to the actions of others. Along these same lines, a study by Pierce and Redcay (2008) used functional Magnetic Resonance Imaging (fMRI) to uncover evidence that the Fusiform Face Area (FFA) is also activated normally in response to familiar faces, but not in response to unfamiliar faces, in children with autism.

Like the MNS, evidence had generally supported the hypothesis of impaired FFA functioning in individuals with autism prior to this. Together, these findings on familiarity effects in social processing (i.e., MNS, FFA) are consistent with the hypothesis that lack of social and/or emotional familiarity with, or interest in, unfamiliar others may be driving reduced activation of social brain networks, including the MNS, in children with autism. One distinct possibility is that children with autism exhibit reduced social interest and/or social-cognitive attention for strangers, relative to other children. This hypothesis receives support from event-related potentials (ERPs) EEG evidence that very young children with autism exhibit reduced late frontal cortex activity in response to unfamiliar faces (Dawson et al., 2002). More specifically, Dawson et al. (2002) found that both typically developing children and children with developmental delays without autism showed larger amplitude ERPs in response to unfamiliar relative to familiar faces, suggesting increased neural activity for the processing of unfamiliar people. However, children with autism did not exhibit this "interest in strangers" effect. In the same study, all three groups of children did exhibit differential brain responses to familiar versus unfamiliar toys, suggesting that this difference in children with autism reflected a lack of neural activity and cognitive processing specifically for unfamiliar people (see also Oberman et al., 2008; Pierce and Redcay, 2008; Becchio and Castiello, 2012; Dawson et al., 2012).

In summary, evidence suggests that individuals with autism exhibit reduced or absent motor resonance activity during the observation of the actions of unfamiliar others. While it was initially suggested that this reduced/absent activity reflects a “broken” MNS (Williams et al., 2001; Oberman and Ramachandran, 2007), more recent results and analysis suggests that reduced/absent mirror neuron activity may reflect reduced social engagement in this population (Oberman et al., 2008; Becchio and Castiello, 2012). Taking the latter view, in the current review, we consider early behavioural interventions that teach speech/language and social communication skills in the specific context of socially engaging synchronous motor activities as a potential motor-related pathway to increasing social-communication and language skills in this population.

3.4. Interventions

Delays and impairments in motor and motor-related development in infants and children with autism have implications for early intervention in this population. Whereas previous reviews have focused on interventions aimed at improving sensory and motor functioning (Baranek et al., 2008) and other ASD-related behaviours (Sowa and Meulenbroek, 2012), here we review and discuss existing and emerging motor interventions that are more directly relevant for increasing social-communication and language skills in toddlers and children with autism. We focus particular attention on their theoretical and practical relationships to motor theories of social-communication and language development, as well as to their existing evidence base. In examining the evidence base, we consider several types, or levels, of evidence (see Table 5). These include case study reports, which can involve descriptions of multiple children but without experimental controls. Next, we consider experimental single subject designs, which exert experimental control through the use of baseline recordings of varying lengths across multiple children, thus more reliably attributing intervention effects to intervention onset. Along with these, we include small-scale pseudo-experimental research designs, whereby children are assessed pre- and post-intervention, but without a comparison control group to account for potential naturally occurring developmental improvements in the target behaviours. Finally, we consider large-scale experimental group studies, Randomized Controlled Trials (RCTs; efficacy trials), and RCTs conducted in community settings (effectiveness trials). As ASDs are a unique class of developmental disorders, we focus our review specifically on the evidence-base for the efficacy and effectiveness of each intervention for children with ASDs. Finally, we focus exclusively on interventions for non-verbal and minimally verbal children, because there are existing evidence-based interventions that are effective for more verbally able children with autism (Koegel, 2000). We start with sign language intervention, which has previously been proposed to be a mechanism for linking motor-based gesture and speech and language development in these children.

Table 5: Levels of evidence for each intervention

Intervention	Brief Description	Case Reports	Experimental Single-Subject Designs (multiple baseline designs, reversal designs) and Small-Scale Pseudo-Experimental Group Designs	Large Experimental Study	Randomized Controlled Trial	Community-Based Randomized Controlled Trial	Summary of Evidence
Augmentative and Alternative Communication (AAC) Interventions							

Sign Language (SLT)	**Teaches child to use hand, arm, facial, and other actions to create symbolic communications**	**5+** 1- Fulwiler & Fouts (1976) 2- Brady & Smouse (1978)	**15+** 1- Carr, Kologinsky & Leff-Simon (1987) 2- Barerra & Sulzer-Azaroff (1983) 3- Sundberg, Endicott & Eigenheer (2000)	**1** 1. Layton (1988) 2. Yoder & Layton (1988)	**0**	**0**	Extensive research base. Weak but mixed evidence for learning of sign language. Weak evidence for learning of speech. Weak evidence for learning of speech via sign plus speech training. See Schwartz and Nye (2006).
Picture Exchange Communication System (PECS)	Teaches child to exchange pictures with others, in order to make requests and comment	**5+** **Speech:** 1- Webb (2000) 2- Bondy & Frost (1994) **Communication:** 3- Anderson, Moore & Bourne (2007) 4- Malandraki and Okalidou (2007)	**25+** **Speech:** 1- Charlop-Christy et al. (2002) 2- Carr and Felce (2007a) **Communication:** 3- Travis & Geiger (2010) 4- Greenberg, Tomaino & Charlop (2012)	**1** **Communication:** 1- Lerna et al (2009)	**3+** **Speech:** 1- Yoder & Stone (2006a) **Communication:** 4- Yoder & Lieberman (2010)	**1** **Speech:** 1- Gordon et al (2011) **Communication:** 2- Howlin et al (2007)	Extensive research base. Moderate evidence for both picture-based and verbal communication gains. See Sulzer-Azaroff et al. (2009).
Motor-Based Behavioural Interventions							
Prompts for Restructuring Oral Muscular Phonetic Targets (PROMPT)	Uses physical prompts to the vocal apparatus, as well as social, kinesthetic, and proprioceptive awareness, to increase speech and language.	**0**	**1** 1- Rogers et al (2006)	**0**	**0**	**0**	Limited evidence in ASD.

Auditory Motor Mapping Treatment	Teaches the pairing of sounds with motor actions during picture-based word teaching in order to facilitate vocalisations	**0**	**1** 1- Wan et al (2011)	**0**	**0**	**0**	Limited evidence in ASD.
Electromagnetic Brain Stimulation Interventions							
Transcranial Direct Current Stimulation (TdCS) / Transcranial Magnetic Stimulation (TMS)	Electromagnetic brain stimulation procedures.	**0**	**1** 1. Schneider and Hopp (2011)	**0**	**0**	**0**	Limited evidence in ASD
Interventions Targeting Synchronous Motor Activities							
Early Start Denver Model (ESDM)	Integrative model of play-based behaviorist/operant teaching methods within a comprehensive developmental framework.	**5** 1. Voos et al (2012) 2. Vismara & Rogers (2008)	**20+ (includes PRT evidence)** 1. Vismara, Colombi & 2. Vismara & Lyons (2007) 3. Pierce & Schreibman (1995) 4. Stahmer (1995)	**1** 1. Baker-Ericzén, Stahmer and Burns (2007)	**1+** 1. Dawson et al (2010)	**1** 1. Rogers et al (2012)	Extensive research base (includes PRT evidence). Moderate evidence for verbal and non-verbal communication gains. See Warren et al. (2011).
Reciprocal Imitation Training (RIT)	Uses reciprocal imitation and behaviorist principles to teach the child to imitate the motor actions and gestures of others in a play context.	**0**	**3+** 1- Ingersoll & Schreibman (2006) 2- Cardon & Wilcox (2011)	**1** 1- Ingersoll (2010)	**0**	**0**	Moderate evidence for non-verbal communication and imitation gains.

3.4.1. Augmentative and Alternative Communication (AAC) Interventions

3.4.1.1. Sign language training

For non-verbal autistic children, training in augmentative and alternative communication (AAC) offers a route via which these individuals can begin to communicate. The two most widely accepted AAC strategies are Sign Language Training (SLT; Carr et al., 1978) and the Picture Exchange Communication System (PECS; Bondy and Frost, 1994; Frost and Bondy, 2002; see Figure 2). Research suggests that educators believe that both of these strategies are viable options for teaching communication skills to children with autism displaying severe deficits in communication skills (Stahmer et al., 2005).

Given the strong links between gesture and verbal communication in typically developing infants, including those described in the sections above, the use of SLT to facilitate speech in developmentally delayed populations has a logical theoretical basis. Indeed, early studies investigating the impact of SLT on children with autism yielded promising results, in both the communicative and social domains (Miller and Miller, 1973; Bonvillian and Nelson, 1976; Fulwiler and Fouts, 1976; Brady and Smouse, 1978; Konstantareas, 1984). Contrary to expectations, however, these marked improvements in communication did not include speech development. Furthermore, the effectiveness of sign language alone as a means to facilitate speech in non-vocal autistic children was quickly called into doubt; as was the degree of experimental control employed by early research in this area (Carr et al., 1978; Carr, 1979; see Table 5).

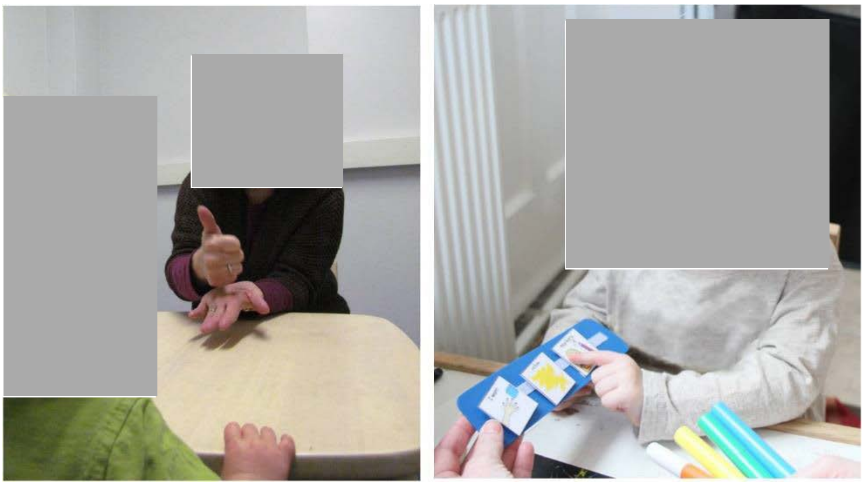
Following the recognition that SLT did not lead to meaningful increases in speech in children with autism, studies utilizing training sessions that focused on coupling sign language with other forms of training (e.g., speech intervention plus SLT) were conducted. This combined intervention approach proved to be more effective than sign language alone for eliciting spoken vocabulary in nominally verbal autistic children (Brady and Smouse, 1978; Layton and Baker, 1981; Konstantareas, 1984; Yoder and Layton, 1988). However, when considering this research, it is important to note that the participants in these studies had existing verbal skills. Therefore, it has yet to be examined whether SLT in any form can elicit verbal communication gains in non-vocal autistic children. Moreover, outcomes following SLT are extremely and unusually variable. For example, although a small number of individuals with autism adopt sign language as their primary mode of communication and appear to readily learn signs (Barrera et al., 1980; Stull et al., 1980), others are unable to attain even the most basic signing skills (Webster et al., 1973; Brady and Smouse, 1978; Carr et al., 1978).

Despite decades of research into SLT as an effective tool for teaching those with ASD, the evidence that it leads to novel and/or increased functional uses of communication, speech, and language in this population is weak. Those who suggest that sign language, or total communication (sign plus speech), may serve to increase such skills in autistic individuals often base their arguments on single-subject research (Carr et al., 1978, 1987; Casey, 1978; Cohen, 1979; Schepis et al., 1982). Although rich in detail, the majority of these more promising SLT studies provide no measure of fidelity of implementation, few explored generalizability, and many fail to disclose sufficient detail for either clinical application or experimental replication (Millar et al., 2000; Schwartz and Nye, 2006). In their review of

SLT in this population, Layton and Watson (1995) maintain that, despite extensive training, the majority of nonverbal children fail to develop any form of vocalization and, at most, learn a few basic signs, as a result of SLT. In a more recent review of sign language and communication gains in children with autism, Schwartz and Nye (2006) conclude that teaching communication through signing does not serve as an effective intervention to improve either sign or oral language communication in children on the autism spectrum (see also Millar et al., 2000).

While the poor results of SLT have often been overlooked in the literature, some attempt has been made to explain these findings. One proposed explanation for the relative failure of SLT is that the successful acquisition and use of sign language as a communicative tool is dependent on the ability to form a variety of manual-motor signs and there are many individuals with ASD who do not possess the fine motor skills required (Bonvillian and Blackburn, 1991; Seal and Bonvillian, 1997; National Research Council, 2001). Similarly, Mirenda and Erickson (2000) outline “the three I's” that contribute to successful sign language acquisition: imitation, iconicity, and intelligibility. They maintain that children with autism demonstrate a lack of imitation, symbolic representation, and motor coordination/planning skills, while the successful acquisition and use of sign language relies largely on the possession of these abilities (see Figure 1 and Table 4 for relevant neural mechanisms). In each of these proposed explanations, deficits and delays in motor and motor-related skills are key to explaining why children with autism generally fail to develop both sign language-based communication and speech and language skills as a result of SLT.

Figure 2: Augmentative and Alternative Communication (AAC) Interventions. Child and therapist engaged in Sign Language Training (left) vs. Picture Exchange Communication System (PECS) training (right). Sign Language Training (SLT) uses behaviorist imitation and prompting methods to teach children to use hand, arm, facial, and other body actions to produce symbolic communications. The Picture Exchange Communication System (PECS) uses behaviorist methods to teach children to hand one or more pictures to a variety of communicative partners, in order to request items/activities, respond to simple questions, and comment.



3.4.1.2. Picture exchange communication system (PECS)

Given the lack of meaningful progress as a result of SLT, it is unsurprising that the field has turned its attention to other AAC training practices. The PECS is a form of AAC that utilizes pictures as its primary medium of communication and, like SLT, has foundations in behaviourist principles. The primary goal of PECS is to establish and increase spontaneous communication within social contexts, which is initiated through picture-based communication (Bondy and Frost, 1998). PECS is a structured and manualised intervention program that is designed to teach children to communicate via a book containing detachable pictures (see Figure 2).

The PECS protocol is divided into six phases, each designed to expand upon the child's development during the previous phase. In Phase I, the child is taught to hand a single picture to another person, in exchange for a desired item or activity (e.g., a ball). In Phase II, the child is taught to exchange pictures with multiple people in multiple environments. Phase III teaches the child to discriminate and select among pictures for a number of desired items. Phase IV teaches the child to produce simple sentence structures (e.g., “I want ____.”) using pictures, which are then handed to communicative partners using a sentence strip (see Figure 2). Finally, Phases V and VI teach responding to simple questions and commenting, using pictures. The child typically progresses from basic picture-based requesting, to more advanced picture-based responding and spontaneous commenting (Bondy and Frost, 1998). The surface appeal of PECS over sign language is understandable given that it does not rely on the communicator possessing complex fine motor skills, nor does it burden the communicator with learning a completely new language (Bondy and Frost, 1994). Furthermore, the gains facilitated by PECS do not appear dependent upon the child possessing pre-existing skills (Bondy and Frost, 2002; Yoder and Stone, 2006a,b), and PECS appears to be readily learned by children with autism as well as other developmental disorders (Schwartz et al., 1998; Mirenda and Erickson, 2000; Charlop-Christy et al., 2002; Ganz and Simpson, 2004; Preston and Carter, 2009).

Although not initially developed to teach spoken language, a large and growing body of evidence demonstrates that PECS can assist with spoken language development in children with autism with existing, albeit limited, verbal skills (Bondy and Frost, 1994; Liddle, 2001; Charlop-Christy et al., 2002; Kravits et al., 2002; Magiati and Howlin, 2003; Anderson et al., 2007; Carr and Felce, 2007a; Carré et al., 2009; Jurgens et al., 2009; Preston and Carter, 2009; Sulzer-Azaroff et al., 2009; Greenberg et al., 2012). Early non-experimental, retrospective research by Bondy and Frost (1994) suggested that after one year of PECS usage, 76 percent of 66 young children developed speech either as their sole means of communication or alongside picture communication. Following a series of experimental single-subject design studies suggesting positive effects on both communication and speech as a result of PECS intervention, several large scale experimental studies have provided further strong and convincing evidence that PECS increases both social-communication and speech/language skills in children with autism. Indeed, increases in spoken and socio-communication skills through PECS training appear to be as prominent as in speech-based interventions (Yoder and Stone, 2006a,b; Lerna et al., 2012). For example, Yoder and Stone (2006a) compared the effects of PECS and Responsive Education and Prelinguistic Milieu Teaching (RPMT) in 36 toddlers and young children with autism. Both interventions were implemented for the same length of time, and at the same intensity. After six months of training, it was found that PECS training resulted in increased verbalisations, both in terms of frequency and range of words. Although children in both treatment groups were found to have made similar speech-related improvements by their six-month follow-up, the authors highlight that these results provide evidence that PECS leads to more swift speech development when compared to RPMT. Similarly, recent research by Lerna et al. (2012) compared the efficacy of PECS with Conventional Language Therapy (CLT) in a group of preschool children with ASD. Following six months of treatment, those receiving PECS demonstrated significant improvements in their joint attention, requesting, and imitation skills.

Although RCT's are severely lacking in the field of autism education research (Carter and Wheldall, 2008; Preston and Carter, 2009), the few large-scale examinations that have involved such advantageous designs have also replicated the promising data on PECS (Table 5). For example, a recent school-based RCT of PECS versus Treatment As Usual (TAU) by Howlin and colleagues highlighted gains in spontaneous requesting through picture use, speech, or both (Howlin et al., 2007). Gordon and colleagues (2011) examined these same data from 84 autistic children across 15 British schools, observing changes in spontaneous communication following immediate, delayed, or no PECS training. They found that children who had received immediate treatment demonstrated significant increases in both spontaneous speech/vocalisations, and in their usage of PECS. Furthermore, Carr and Felce (2007b) compared a PECS training group (n = 24) with a no treatment control group (n = 17), and uncovered evidence for significant increases in linguistic communicative initiations that included the use of spoken words within the PECS treatment group, and no improvements in such skills within the no-treatment control group. This, again, demonstrates the efficacy of PECS in eliciting both verbal and non-verbal communicative behaviours in children with autism.

It is worth noting that children with autism typically exhibit increases in speech during Phases IV and V of PECS training (Charlop-Christy et al., 2002; Ganz and Simpson, 2004). During these Phases, they are learning to use a larger number of pictures, and have also started to point rhythmically to sentences, often syllable by syllable (Frost and Bondy, 2002). Prior to Phase V, children are taught to (a) communicate with pictures (Phase I), (b) travel and seek their communication partner (Phase II), (c) discriminate individual pictures and what they each represent (Phase III), and (d) structure sentences through the use of a string of picture cards (Phases IV and V; Frost and Bondy, 2002). Phase IV is also the period during which a time delay procedure is used by the therapist, whereby she or he pauses after speaking the first portion of the picture-phrase (e.g., says “I want … ”) and waits 3–5 s for the non-verbal or minimally verbal child to verbalise the label for the item they have requested (e.g., “ball”) before providing the item to the child. In this instance, the child's rhythmic pointing to the pictures (e.g., I-want-BALL) continues as the therapist stops speaking, potentially facilitating the child's verbalisation of the target item (e.g., “ball”). As mentioned, a plethora of research has demonstrated the link between the onset of speech, and the development of coordinated hand banging gestures. It is possible that the speech gains observed in many children during this phase of PECS are a reflection of this link, with implications for the potential importance and validation of hand-mouth motor plans, as described in relation to auditory motor mapping intervention below.

In sum, although there are strong links between motor-based symbolic gesture and speech development in typical infants and children, extensive research suggests that there is no robust link between SLT and increased speech in children diagnosed with autism. Although many children with autism do not readily learn the use of signs, a large body of evidence demonstrates the ease with which they acquire picture-based communication via PECS, suggesting that it is not an inability to learn that is attributable to their difficulties in sign language learning in this population. Furthermore, as outlined, research also suggests stronger links between speech development and PECS training vs. SLT, in children with autism. Some have proposed that difficulties in sign language learning are due to impairments in fine motor skills (Bonvillian and Blackburn, 1991; Seal and Bonvillian, 1997; National Research Council, 2001), whereas others have argued that it is a combination of imitation skills, iconicity, and intelligibility that present challenges to this population (Mirenda and Erickson, 2000). Next, we examine several more directly motor-based interventions that are currently under development to address social-communication and speech/language skills for this population.

3.4.2. Motor-Based Behavioural Interventions

While sign language is a gesture and motor-based intervention, there are other behavioural interventions that take an even more direct approach to addressing motor aspects of speech production. These include interventions that involve direct manipulations of the mouth and other sound-producing structures, and those that make more direct low-level links between hand and oral motor activity. Here, we describe research on the two interventions of this type that have been studied in relation to children with autism.

3.4.2.1. Prompts for restructuring oral muscular phonetic targets (PROMPT)

One intervention targeting the neuromotor underpinnings of speech production is the Prompts for Restructuring Oral Muscular Phonetic Targets (PROMPT; Chumpelik, 1984) model. PROMPT goes beyond auditory and visual input, integrating neuromotor principles with social, kinaesthetic, and proprioceptive awareness to facilitate the production of clear sounds, speech, and language (Hayden, 2002). In addition to manipulating sound-producing structures, PROMPT places importance on body movement and stability. A typical PROMPT session involves play-based or naturally occurring activities that are likely to encourage interaction initiations from the child. Using these initiations or motivators as a therapeutic opportunity, the clinician then uses vocal modelling and physical manipulations of the child's speech mechanisms as they attempt verbalisation. Such manipulations include touch, pressure, positioning, and movement to promote structural integration within the child's vocal apparatus (Hayden and Square, 1994; see Figure 3).

A PROMPT is available for every vowel or consonant in the English language, as well as for every single or combined speech-sound utterance. Specifically, therapists may use parameter prompts to provide support to the jaw and facial muscles; surface prompts to aid the formation of speech sounds and their associated timings and transitions; syllable prompts to teach the critical combination of jaw support and lip positioning required to produce legible syllables; and finally, complex prompts may be administered when teaching the formation of single sounds (Hayden, 2006) Due to these multiple types of prompts, the PROMPT model can be used to build upon the motor skills of children at all stages of speech production, from first-word attempts to the production of more intelligible speech. Throughout the course of intervention, manual prompts are gradually faded as the child demonstrates heightened oral awareness and control.

The PROMPT intervention method has been examined in a number of studies, although most report on individual case studies. For example, Square et al. (2000) examined six young children with language and phonological disabilities and, following PROMPT intervention, discovered increased accuracy of target word production, and generalization of abilities to untrained words. Gains were also noted in overall communication, social interaction, and intelligibility. Furthermore, Square et al. (1986) noted the efficacy of PROMPT training in three patients with acquired apraxia, whilst a recent study by Ward et al. (2009a,b) found gains in intelligibility, consonant accuracy, and generalized vocal improvements in children with cerebral palsy and speech impairments. In a case study of a severely apractic-aphasic male, PROMPT training for 41 weeks was associated with maintained articulation accuracy in a set of core functional words and phrases (Freed et al., 1997). Finally, although Dodd and Bradford (2000) found no effect of PROMPT intervention in three boys with phonological impairment without articulation disorders, Grigos et al. (2010) discovered increased articulation accuracy in a single subject with severe articulation impairment.

To date, only one published study has explored the effects of the PROMPT method in children with ASD. Rogers et al. (2006) randomly assigned 10 non-verbal children with autism to receive one of two interventions: the Denver Model (a play-based program based on reciprocal communication and social engagement; Rogers et al., 2000), or PROMPT. All participants received 12 weeks of treatment and were assessed for their use of novel words and phrases throughout the intervention, as well as for the maintenance of such functional communication at three weeks post-treatment. Assessments throughout and following intervention revealed that 80% of participants exhibited increases in spontaneous, functional words. In light of the small sample sizes, and in the absence of group comparisons, this study can only be considered a series of non-experimental case studies. Nevertheless, these preliminary findings do suggest potential promise for the use of the PROMPT model with autistic children, and future research should endeavour to examine a larger sample of autistic children in a RCT or other experimental assessment of the PROMPT intervention.

3.4.2.2. Auditory motor mapping training

Auditory-Motor Mapping Training (AMMT; Wan et al., 2009) is a recently developed multi-component intervention targeting the development of speech output through singing, motor activity, and imitation (Wan et al., 2010a). Based upon the hypothesis that individuals with autism have a deficient MNS, AMMT was designed to train sound-articulation associations by engaging multiple neural networks (Wan et al., 2010b). In essence, the goal of AMMT is to teach the pairing of sounds with motor actions in order to facilitate vocalizations.

During a typical AMMT session, a target word or phrase is introduced, and the therapist repeatedly intones the word or phrase while simultaneously tapping a pair of drums tuned to different pitches. The child is then encouraged or gently guided to imitate these actions, while being presented with images of the target object, action, or person. These three components are believed to work together to promote increased interactions between the auditory and motor systems, strengthening the likelihood of intelligible and functional speech production. For example, the use of intonation as opposed to simply speaking is designed to heighten bilateral fronto-temporal network activation—an area associated with components of the MNS (Brown et al., 2004; Ozdemir et al., 2006). Similarly, the engaging use of percussion has been implicated in the activation of a sensorimotor network responsible for articulatory and oro-facial movements, as well as stimulating the mapping of sounds to actions through increased bilateral activation in the fronto-parietal motor-related network (Meister et al., 2003, 2009; Lahab et al., 2007). The third component, imitation, is designed to encourage learning, and is argued to alter the responses in the MNS (Catmur et al., 2007).

One small-scale study describing several cases has been reported on AMMT as an intervention for children with autism. Wan et al. (2011) examined 6 non-verbal children with autism who each received five AMMT sessions per week throughout an eight-week period. All children were assessed on their vocal production at baseline, during the therapy, and following completion of treatment. The authors report that word and phrase articulation improved notably in all of the children, with improvements including verbalisations of both trained and untrained words. Although promising, the results from this case study series must be interpreted with caution, particularly in regards to whether or not the intervention was driving the observed effects. To date, there has yet to be an experimental study examining the efficacy of AMMT for treating children with ASD. On the other hand, the results from these initial case studies serve as a promising starting point to initiate larger-scale and experimental studies of AMMT.

3.4.3. Electromagnetic Brain Stimulation Interventions

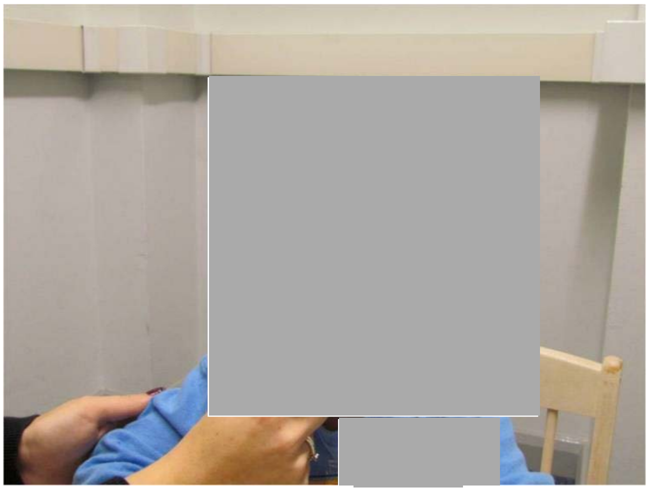
3.4.3.1. Transcranial direct current stimulation and transcranial magnetic stimulation

Transcranial Direct Current Stimulation (tDCS) and Transcranial Magnetic Stimulation (TMS) are relatively new methods via which low intensity intracranial electrical current is applied to the cerebral cortex (see Figure 3). The current is the result of a fluctuating magnetic field that comes from external resources, and tDCS and TMS are considered non-invasive brain stimulation procedures (Pascual-Leone and Walsh, 2002; Gandiga et al., 2006). In tDCS, a relatively weaker direct current is applied constantly through electrodes attached to the scalp above a brain region of interest. This current alternates the neuronal excitability in either a positive or a negative manner, leading to changes in brain function

(Nitsche et al., 2008). A combination of tDCS and other rehabilitative treatments has been studied in relation to motor training protocols (Hummel and Cohen, 2005). TMS has been successfully used to alleviate, or attempt to alleviate, neurological symptoms associated with stroke (Oliveri et al., 1999), epilepsy (Fregni et al., 2006), and a variety of psychiatric disorders (Lisanby et al., 2002). Most relevant to the current review, repetitive TMS (rTMS) has been shown to improve naming abilities in adults with chronic aphasia resulting from stroke (Martin et al., 2009; see also Mimura et al., 1998; Winhuisen et al., 2007).

In 2011, Schneider and Hopp applied tDCS to the left dorsolateral prefrontal cortex in a group of 10 minimally verbal children with autism, in order to examine the possibility of syntax acquisition as a result of tDCS (Schneider and Hopp, 2011). They found significant improvements in behavioural performance on a basic subject-verb-object sentence sub-test of the Bilingual Aphasia Test. Based on these promising group case study findings, the authors have proposed that additional research should be conducted in this area (see also Sokhadze et al., 2009). Furthermore, the results of a recent small-scale experimental study of adults with Asperger's Syndrome further suggest that the application of rTMS may, indeed, prove useful for improving language skills in those with ASD (Fecteau et al., 2011). It is important to note, however, that there are notable risks associated with both tDCS and TMS, some of which have particular practical, medical, and ethical implications for the application of these technologies to individuals with ASDs (see below for further information).

Figure 3: Motor-Based Behavioural Intervention and Electromagnetic Brain Stimulation Intervention. Child and therapist engaged in Prompts for Restructuring Oral Muscular Phonetic Targets (PROMPT) intervention. The child is also wearing a Transcranial Direct Current Stimulation (tDCS) electrode band. These two techniques are typically implemented separately. Here, the PROMPT therapist administers a physical prompt to the child's vocal-motor system, in order to facilitate production of a speech target, while the tDCS electrode applies a direct current to the left inferior frontal cortex.



In sum, there are at least three relatively new strongly motor-related interventions for potentially treating speech and language skills in young non-verbal and minimally verbal children with ASD. Interestingly, each of these interventions has precisely one published paper on their usefulness in treating this population. Also of interest, is that the results of these studies all provide promising results. This being the case, however, none of these studies were experimental in nature and, instead, took the form of a small-scale pseudo-experimental design in each case. It is clear that experimental research is now warranted in order to examine the potential efficacy and effectiveness of these novel interventions. However, the application of one of these interventions, tDCS/TMS, presents some practical, medical, and ethical challenges in relation to children with autism (see Discussion and Future Directions, below, for further information).

3.4.4. Interventions Targeting Synchronous Motor Activities

Play-based intervention methods based upon the application of behaviour analytic procedures are well-established and commonly used techniques for teaching children with autism difficulties to engage in new social, communication, play, language, and other behaviours. These interventions utilize operant teaching methods, including behaviourally-defined targets, contingent reinforcement (e.g., access to items and activities, descriptive praise), physical and verbal prompts, and shaping and fading procedures, to target skill development, while allowing the child a great deal of choice in play activities. Extensive and large-scale experimental research studies have shown that these interventions can increase generalized and spontaneous language and communication skills (Koegel and Koegel, 2006), improve social and play skills (Pierce and Schreibman, 1995; Stahmer, 1995), decrease inappropriate behaviour (Koegel et al., 2005), and improve academic motivation and performance (Koegel et al., 2010).

More recently, researchers have worked to combine developmental and behavioural intervention approaches, whereby operant teaching methods are utilized to target skills within a strong developmental framework in a play-based context. Most relevant to the current review, two of these developmental-behavioural interventions specifically target social-reciprocity and social engagement in the context of synchronous motor activities, which may represent a potential motor-related pathway to increasing social-communication and language skills in this population.

3.4.4.1. Early Start Denver Model (ESDM)

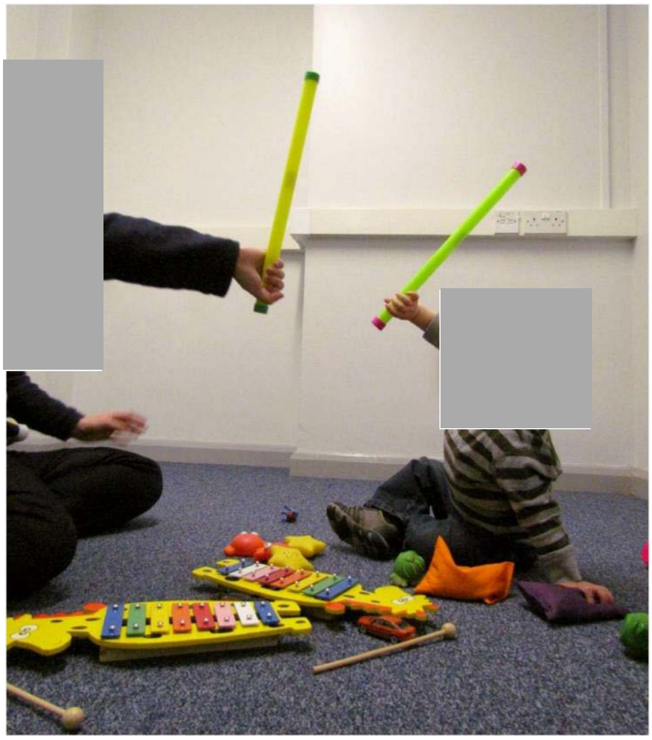
The Early Start Denver Model (ESDM) is an integration of a particular play-based behaviour analytic approach, Pivotal Response Treatment (PRT), with developmental intervention methods designed to increase reciprocal social relationships and social engagement in young children with autism (Rogers and Dawson, 2009a). As with other play-based behaviour analytic interventions, ESDM places a major focus on child motivation. Unique to the ESDM, however, is that the course of intervention for each child is based on a structured Curriculum Checklist, specifically targeting developmentally-based social-interactive skills, social communicative skills, cognitive skills, language, imitation, fine and gross motor skills, self-help skills, and adaptive behaviours (Rogers and Dawson, 2009b). The ESDM has an experimental evidence base, including an impressive and extensive set of previous experimental research studies on PRT and a large-scale RCT of the efficacy of the ESDM itself in toddlers on the autism spectrum (Dawson et al., 2010). In this study, 48 toddlers between 18 and 30 months of age were randomly assigning to either the ESDM intervention group, or to a group referred for community-provided intervention. Across the two-year training period, those in the ESDM intervention group demonstrated significant improvements in scores of adaptive behaviour and IQ (including Verbal IQ/Language) when compared to both baseline scores and the community-referral group. These toddlers also exhibited more positive changes in the severity of their autism diagnosis. That is, in comparison with community intervention, ESDM intervention led to more children experiencing changes in their diagnosis from Autism to PDD-NOS.

3.4.4.2. Reciprocal imitation training (RIT)

Reciprocal Imitation Training (RIT) is a recently developed intervention that primarily targets object and gesture-based action imitation in children with autism (Ingersoll and Schreibman, 2006). Following the same basic principles as PRT and the ESDM, RIT is child-directed and incorporates motivational strategies to facilitate engagement and learning. However, RIT was developed on the grounds that naturalistic action imitation is a critical social learning tool that contributes to rapid advances in social and cognitive development in infants and children (Meltzoff and Moore, 1977; Bates et al., 1979; Fiese, 1990; Uzgiris, 1991; Carpenter et al., 1998; Charman et al., 2000, 2003; Stone and Yoder, 2001), and is significantly impaired in children with autism (Curcio, 1978; Dawson and Adams, 1984; Stone et al., 1997; Williams et al., 2004). In essence, the RIT intervention sessions are designed to create ongoing turn-taking situations whereby the therapist and child reciprocate imitation of each other's actions (see Figure 4). The RIT therapist imitates the child's actions with objects, gestures, movements, and vocalizations, and strategically incorporates the modelling of new developmentally-appropriate actions or gestures approximately once every one to two minutes. The child is provided with up to three actions to imitate in a naturalistic play context, before being physically prompted to imitate the fourth action if and when he or she does not engage in any imitation. As the child learns to reciprocate this imitation, and in turns becomes more attentive and socially engaged with the therapist, the need for prompting decreases until child-therapist imitation is a natural part of the play routine. The ultimate goal of RIT is to increase the generalized use of spontaneous imitation of both actions with objects and gestures, while facilitating gains in other social-communicative domains (Ingersoll and Schreibman, 2006).

The efficacy of RIT as an intervention for children with ASD is evidenced by multiple well-controlled research studies. Several experimental single-subject design experiments have demonstrated increases in object and gesture imitation, as well as highlighting gains in language and social skills as a result of RIT. For example, adopting a multiple-baseline design, Ingersoll and Schreibman (2006) found that after completion of the intervention phases, all five young children with ASD exhibited considerable improvements in object imitation, pretend play, joint attention and language. Importantly, such gains in imitation were found to generalize across materials, settings, and therapists (Ingersoll and Schreibman, 2006). Ingersoll and Gergans (2007) replicated these findings in a study investigating the effectiveness of parent-implemented RIT. Again, a multiple-baseline design across three families evidenced increased spontaneous object imitation in young children with autism, with effects exceeding the teaching period (Ingersoll and Gergans, 2007). Furthermore, in addition to object imitation, gains in gesture imitation have been demonstrated in a single-subject study by Ingersoll et al. (2007). In 2010, Ingersoll attempted to further validate these findings by conducting a pilot RCT into the effects of RIT on elicited and spontaneous imitation in autistic children (Ingersoll, 2010). Randomizing 21 young children into either RIT intervention or a control group, Ingersoll found larger imitation gains in the treatment group across all primary assessments, replicating previous single-subject findings. Thus, the large evidence-base for RIT as an effective intervention tool for autistic children is promising and, unlike other forms of ASD treatment, consists of multiple designs all demonstrating the same imitation, language, and social gains in this population.

Figure 4: Interventions Targeting Synchronous Motor Activity. Child and therapist engaged in Reciprocal Imitation Training (RIT). RIT involves the therapist imitating the child's actions and gestures, and also modelling developmentally-appropriate actions and gestures for the child to imitate, in a play context. The child is encouraged and prompted to imitate, until regular spontaneous reciprocal imitation is established.



Given the dynamic and effective nature of these play-based, reciprocal action and synchrony-oriented interventions, the ESDM and RIT appear to increase child-therapist social-motor synchrony (i.e., temporal coordination of movements) and social engagement (see also Landa et al., 2010). This increase in social-motor coordination and engagement may also increase social attention and motor resonance mechanisms in these children. Recall that there is evidence that activation of the MNS, FFA, and other social brain mechanisms may be limited in response to those individuals with whom children with autism are social-emotionally disconnected (e.g., unfamiliar people). Given that the ESDM and RIT increase social-communicative and language skills, one potential mechanistic pathway facilitating some of these behavioural changes is increased motor resonance through repeated social engagement with unfamiliar people. Evidence from a recent EEG/ERP study of face processing in toddlers with autism who received the ESDM vs. community-based services provides indirect support for this hypothesis. Specifically, the ESDM intervention increased late frontal activity in response to unfamiliar faces, relative to children who received TAU (Dawson et al., 2012). Because this was a study of static face processing, as opposed to human action processing, we cannot generalize these findings to the MNS without further research. However, direct experimental examinations of this hypothesis in the future, particularly experimental studies including measures of motor resonance, will be very informative in this regard.

3.5. Discussion and Future Directions

We have described several interventions aimed at increasing social-communication and language skills in young children with autism that have theoretical and/or practical roots in relationships of these skills to motor development. In doing so, we have given serious consideration to the intervention methods as well as the existing or emerging evidence-base for each such intervention. As outlined in this review, neither practical nor theoretical links between motor and communication/language development are sufficient to predict the efficacy of an intervention for children on the autism spectrum. For example, despite very strong practical and theoretical links between early symbolic gestures, such as the iconic manual and motor signs of sign language, and speech and language development in typically developing children, extensive research suggests that SLT is not a very effective way to teach either communication or speech/language skills to children on the autism spectrum. On the other hand, evidence suggests that these children can learn a picture-based social-communication system, PECS, rapidly and effectively. Furthermore, research suggests that PECS is a relatively more effective path to speech development in these children. There are multiple potential reasons for this seemingly contradictory finding, including the possibility that impairments in motor skills (e.g., fine motor skills), motor imitation, and/or iconicity make learning and producing the manual and motor signs of sign language particularly challenging for children with ASDs (see also Figure 1 and Table 4).

We also reviewed and described several emerging intervention methods that take a more direct approach to motor aspects of speech production. These included PROMPT, which involves direct manipulations of the mouth and other sound-producing structures; AMMT, which aims to generate strong and direct temporal links between the child's auditory, motor, and speech production; and tDCS (and TMS), which involve directly stimulating motor and motor planning regions involved in speech production and other aspects of language. Although there is not yet existing experimental evidence for any of these interventions, reasonable pseudo-experimental/group case study reports on relatively well-characterized groups of children provide promising information to suggest that each of these interventions might prove effective for increase speech/language skills in this population. Therefore, experimental research is warranted on PROMPT, AMMT, and tDCS/TMS as potentially effective interventions for children with autism.

While the case study report on the group of minimally verbal children with autism receiving tDCS intervention is promising, there is also a need for caution in the pursuit of both research and practice involving the application of this technology to non-verbal or minimally verbal children with autism. While tDCS and TMS are generally believed to be safe procedures, there are also known risks (Wassermann, 1998; see also Loo et al., 2008; Rossi et al., 2009). For example, incorrect setting of electrical current or other parameters can trigger adverse events such as seizures, toxicity, headache, nausea, tissue damage, or burns. Furthermore, common adverse reactions include mild pain or sensitivity on the scalp, and headaches. It is, therefore, critically important to consider the ramifications involved with testing or treating non-verbal and minimally verbal children with autism with these technologies, given that they can neither provide informed consent nor effectively communicate injury or discomfort.

A risk of potentially even greater concern with the application of tDCS and TMS to children with autism is the potential for directly or indirectly causing seizure activity, or the onset of epilepsy. As characterized by Maski et al. (2011; see also Myers and Johnson, 2007), the prevalence of epilepsy is typically quoted in the literature as 30%. Identification of epilepsy in ASD is also challenging, due to the impact of ASD symptoms and behaviours on measurement/testing. As a result, assessing seizure risk would be very difficult to impossible for large numbers of non-verbal and minimally verbal children.

Despite the risks, tDCS and TMS have already been used to study children from a number of populations, including children who have experienced brain injury as a result of stroke (Frye et al., 2008; Kirton et al., 2010), children with language-learning disorders (Pugh et al., 2001), and children/adolescents with psychiatric disorders (Walter et al., 2001). Indeed, a clear strength of these technologies, and particularly tDCS, is that they are sufficiently streamlined and flexible in their application to be used with relatively young and relatively less able individuals. These techniques can even be used in conjunction with existing behavioural interventions (see Figure 3), potentially facilitating or enhancing their positive effects on speech and language development.

The possibility that the application of motor-related interventions might initiate the onset of even small to medium sized gains in speech development could have major long-term implications for quality of life. The results of several recent studies examining predictors of speech/language outcomes following early behavioural intervention suggest that a child producing even a few words prior to the start of intervention can play a key role in whether or not that child makes speech and language gains during the intervention (Gordon et al., 2011; Nahmias et al., 2012). Other research suggests that language abilities at 5- to 7-years of age are one of the key predictors of cognitive and adaptive skills outcomes in adulthood in this population (e.g., Gillespie-Lynch et al., 2012). At the same time, evidence suggests that relatively large percentages of autistic children who are completely non-verbal at 2-, 3-, and even 4-years of age develop speech and language skills fairly rapidly as a result of intensive early intervention (Koegel, 2000). Unfortunately, it is not currently possible to predict which non-verbal and minimally verbal young children will respond to any given early behavioural intervention (Stahmer et al., 2011).

As alluded to above, early intervention that targets speech and language skills by 2- to 4-years of age appear to be much more effective than those same interventions implemented after 5-years of age (Koegel, 2000), perhaps due to the existence of sensitive periods for speech/language and related skills (Fox et al., 2010; Windsor et al., 2011). Given the developmental complexity, and in some cases the seemingly strong biological nature, of motor development in relation to speech/language development, similar sensitive periods may exist in the relationships of motor and language/communication skills development. Therefore, the motor-related intervention pathways to language that have been discussed in this article, or others, may be most effective when intervention occurs in an ideal time window. Dependent upon the particular mechanism being targeted, this time window may be a sensitive biological/chronological age or developmental age period. For example, interventions that incorporate repetitive and coordinated hand banging may only be effective at facilitating speech when they occur during or shortly after the chronologically appropriate age of 7- to 12-months. Alternatively, intervening to increase these links may, as suggested by AMMT, still be effective at facilitating speech for any child below eight years of age who is in the pre-verbal or minimally verbal stage of development, for example. These are interesting clinical and empirical developmental questions, which can be directly examined in experimental studies.

3.6. Conclusion

In this article, we have reviewed the research on aspects of early motor development that are believed to be specifically relevant to speech/language and social communication in infants and children with autism. We have also reviewed motor-related interventions designed to increase speech/language and social-communication skills in young non-verbal and minimally verbal children with autism. This field is at an exciting time in this area of research and development. We now know from extensive research that SLT is not a very effective intervention for facilitating speech and language development in this population. Potential reasons for this include that children with autism exhibit specific difficulties in iconicity, imitation of the actions of others, and/or fine motor skills, which make it difficult for them to become effective signers. On the other hand, these children appear to learn a picture-based social-communication program relatively rapidly, and extensive evidence suggests that this type of communication training does facilitate the development of basic speech skills in many of these children. At the same time as this, small-scale pseudo-experimental studies on at least three types of recently developed motor-based speech/language interventions (PROMPT, AMMT, tDCS/TMS) have each produced very promising results. This provides an exciting opportunity for important new experimental research studies designed to directly examine the efficacy of these interventions with this population, for whom effective speech/language interventions have been challenging to identify and develop. Finally, researchers with expertise in traditional applied behaviour analytic and developmental interventions have begun working together to develop interventions that combine these two approaches. The result is a combined intervention strategy that uses highly effective operant teaching methods with a socially and motorically interactive play-based approach to enhancing speech/language and social-communication skills. The effects of these interventions on the children appear to extend beyond simple skill learning, and to enhance social attention and social engagement in ways that may facilitate the activation of social brain networks, including the motor-resonance system. We are optimistic that the field is approaching a turning point, with potentially dramatic breakthroughs to come in both our treatment and our understanding of the speech/language and social-communication difficulties in this population, as well as their relationship to motor mechanisms and development.

CHAPTER 4: THE EFFECT OF STRUCTURED EXERCISE INTERVENTIONS ON THE REPORTED STRESS OF SCHOOL-AGED CHILDREN WITH AUTISM SPECTRUM DISORDERS AND OTHER SPECIAL EDUCATIONAL NEEDS

4.0.1. Introduction

It is unsurprising that the early years are a time of increased predisposition to stress. As asserted by Siddique and D'Arcy (1984), growing up involves “dramatic transitions in the physical, social, sexual and intellectual spheres” which “must be stressful” (Siddique and D’Arcy, 1984, p. 460). Indeed, a wealth of literature from recent decades supports this assumption (Kashani & Orvaschel, 1990; Kessler, Avenevoli & Merikangas, 2001; Roza et al, 2003). Relevantly, given the nature of their disorder, individuals with ASD are believed to be highly vulnerable to stress and anxiety (Bellini, 2006; White et al, 2009). In fact, it has previously been suggested that the cause of ASD in a subset of individuals with autism is rooted in a dysfunction of the pineal–hypothalamic–pituitary–adrenal axis, an area involved in the regulation of stress (Chamberlain and Herman, 1990; Heinrichs and Koob 2004).

Research has highlighted the effects of stress on ASD-related characteristics and behaviours. In other words, stress may in fact enhance some of the negative behaviours observed in ASD. For example, a recent study by Gillott and Standen (2007) found that not only were autistic adults around three times more likely to report feelings of anxiety when compared to controls, but their reports were also significantly correlated with stress, which in turn was found to mediate the ability to adapt to change and cope with unpleasant events. Furthermore, Lytle and Todd (2009) maintained that stress produces ASD-related symptoms, such as repetitive movements, deficits in attention and cognition, and sleep disturbances. Indeed, Morgan (2006) questioned whether autism is in fact a “stress disorder”, given the vast overlap with stress-induced mechanisms and behaviours observed in the typically developing population. Finally, Groden et al (1994) highlighted that the maladaptive behaviours observed in individuals with ASD are often precipitated by stressful incidences.

In addition to the target outcomes for children with ASD presented in Chapter 2, it is apparent that interventions should also target stress and anxiety throughout childhood in this population. A variety of techniques have been adopted to reduce such tension in other populations, including cognitive restructuring (Hains and Szyjakowski, 1990; Britton et al., 2011), meditation (Barnes, Bauza & Treiber, 2003; Beauchemin, Hutchins, & Patterson, 2008) and assertiveness training (Kiselica et al., 1994; Paeezy, Shahraray, & Abdi, 2010; Hijazi et al., 2011). One of the most prominent and robust interventions for this purpose, however, is physical exercise (Salmon, 2001).

This chapter will first outline various research studies which have sought to investigate the relationship between physical exercise and emotional wellbeing. Although there is currently no clear consensus on the mechanisms driving this relationship, this chapter will then describe and discuss the explanations put forward by multiple researchers, from both psychological and physiological perspectives. The prominence of stress in the ASD population will then be highlighted, followed by a description of experimental studies conducted on this topic as part of this thesis. Study 1 will examine the effects of acute bouts of exercise on feelings of stress and anxiety, before presenting novel empirical research into this effect in the ASD and other SEN population. Study 2 then explores the effects of a longer term (6 week) exercise intervention programme in a different set of participants from the same populations. Interpretations and implications of both studies will be discussed following the results of each, with a broader discussion of the combined results presented in Chapter 6.

4.0.1.1. Behavioural Evidence

Evidence from multiple fields suggests that engaging in moderate- to high-intensity exercise leads to improvements in emotional wellbeing. In fact, Carraro and Gobbi (2012) maintain that only pharmacological treatment has a slightly superior effect over exercise, with physical activity more effective than meditation and relaxation, stretching and yoga, and group therapy in the treatment of emotional wellbeing.

Given this belief, the effects of exercise following both acute and chronic bouts of activity have been widely researched in both clinical and non-clinical populations. Interestingly, a plethora of studies have reported positive effects after just one session of exercise. For example, Roth (1989) examined the effects of a 20-minute exercise session on self-reported mood and cardiovascular response of typical active and inactive students. Although there were no cardiovascular effects, those in the exercise condition reported reductions in anxiety and tension, regardless of typical activity levels. Furthermore, a study by Steptoe, Kimbell, & Basford (1998) discovered that positive moods were more highly reported on days in which participants engaged in exercise when compared to non-exercise days. Participants also reported less daily stressors on exercise days, as well as higher depression levels on non-exercise comparison days.

A further study conducted by Berger and Owen (1988) compared the effects of 14 weeks of swimming, body conditioning, yoga and fencing on stress reduction and mood enhancement. The authors hypothesised that swimming would most effectively reduce stress, and that the effects would be short term rather than long lasting. They also predicted that all exercise modes would be effective at reducing stress when compared to a lecture-based control condition. Intriguingly, participants in the swimming condition only reported feelings of reduced tension after the first 40-minute session. Reductions in short-term state anxiety were only evident in the yoga group, who also reported reductions in tension, depression, anger, fatigue, and confusion. As predicted, there were no long-term effects of any exercise condition. However, some limitations must be considered, including the lack of randomisation to groups. Participants were able to select the exercise in which they partook, confounding the experimental nature of the observed effects and potentially accounting for any positive mood effects through enjoyment (Baumeister and MacLean, 1984).

Despite the predictions of Berger and Owen (1988), effects of longer-term engagement in exercise have been discovered by numerous research groups. For example, Brown and Lawton (1986) investigated the relationship between regular physical exercise and the prominence of stress-induced disturbance in 220 adolescents. Upon correlational analysis of self-reported physical and emotional wellbeing, stressful life events and exercise patterns, it was discovered that the effects of stress on well-being were mitigated in those exercising regularly. The authors concluded that such results implicate the efficacy of physical activity as part of a prevention programme. However, great caution must be taken when interpreting findings based upon correlations between exercise and self-reported change where no experimental intervention manipulations were conducted. Improving on this methodology, research conducted by Norris, Carroll and Cochrane (1991) validated such effects. Not only did the self-reports of 147 adolescents compliment the positive exercise-stress association found by Brown and Lawton, but a 10-week training period also demonstrated similar positive effects of high-intensity aerobic exercise on physical fitness and psychological wellbeing.

Given the encouraging results indicating positive effects of exercise activity in reducing psychological stress, more recent research has broadened its focus to explore practical real-world implications. Notably, Throne, Bartholomew, Craig and Farrar (2000) investigated the effects of a 16-week exercise programme on reactivity to stress in 41 current firefighters. Participants were randomly assigned to receive a target rowing exercise, which gradually increased in intensity, or to continue their regular regime, with experimental exposure to psychological stress both before and after the training. Importantly, the authors noted that previous studies typically focused on research-specific outcomes, such as the effects on performance on computerised cognitive tasks. To ensure that the target stressor situation was transferable to the lives of firefighters, Throne and colleagues adopted a video-based Strategy and Tactics Drill (STD), which required participants to watch a fire scene and make decisions regarding their actions. An Austin Fire Department official experimentally blinded to the intervention status of the participants sat with each participants and prompted their responses. Participating fire fighters were told that the official was evaluating their responses to the video, which displayed a burning building in which a child might be trapped. Heart rate and blood pressure were recorded, as well as responses on stress-related questionnaires. Results revealed significant fitness increases in the rowing group, and significant improvements in cardiovascular reactivity to psychological stress, with the rowing intervention group demonstrating decreased mean arterial pressure during stress when compared to controls.

Rowers also reported significantly less negative affect and stress-related anxiety following the 16-week programme, leading the researchers to recommend structured exercise training to target industry-related psychological stress.

Mechanisms Underpinning Stress and Physical Exercise

The interest in physical exercise as an intervention tool is unsurprising given that the various psychological and biological mechanisms impacted by engaging in such activity are considered to reduce feelings of stress and other maladaptive states (Girodo & Pellegrini, 1976; Long, 1993; King & Brassington, 1997; McNamara, 2000). Although there does not appear to be one singularly defined mechanism to explain the links between physical exercise and stress, various meta-analyses evidence the vast interest in understanding this relationship. Explanations suggested and proposed have included body temperature regulation (Hori et al., 1978), nervous system alterations (Solomon, 1980), psychological theories of distraction (Bahrke and Morgan, 1978) and expectation (Petruzzello et al., 1991), and, finally, direct cardiovascular effects (Hamer, Taylor, and Steptoe, 2006).

Psychological Explanations

Various psychological explanations of physical exercise in the mediation of stress have been suggested. As proposed by Bahrke and Morgan (1978), the distraction hypothesis purports that 'time-out' from stressful situations or daily routines reduces anxiety, thus accounting for the positive effects of exercise (Petruzzello et al., 1991). In the context of the school environment, this theory purports that providing access to a break from classroom-based learning may contribute to decreased stress and anxiety levels. Evidence for this effect can be drawn from results of similar distraction therapies, such as relaxation and yoga-based sessions, which also lead to reduced state anxiety (Petruzzello et al., 1991). Importantly, this theory does not necessarily contradict physiological accounts, with Morgan and O'Connor (1988) outlining a potential add-on, rather than contrasting, effect. Furthermore, the theorists maintain that the anxiety-reducing effects of exercise last longer than such comparative techniques (Raglin and Morgan, 1987), suggesting that distraction per se cannot alone account for such improvements.

An additional psychological explanation can be drawn from cognitive dissonance theory (Festinger, 1957; Cooper, 2011) and 'self' perspectives. The theory of cognitive dissonance has produced the hypothesis that individuals possess a drive to avoid conflict between attitudes, beliefs, and behaviours in order to maintain harmony. When considering exercise, whilst participants may not enjoy physical exercise for multiple reasons (Finkelstein et al., 2010), by continuing to participate, they are driven to find reasons or rationalities to enjoy and justify engaging in it. However, given that this change to cognitive consistency is not deemed immediate, this theory may only account for trait, rather than state, stress and anxiety (Petruzzello et al., 1991). Finally, concepts of 'self' in the form of fulfilling awareness, such as self-mastery, self-confidence, self-development, and self-esteem may play a large role in the positive emotional outcomes of physical exercise (Taylor, Sallis and Needle, 1985; Sonstroem and Morgan, 1989; Petruzzello et al., 1991). For example, García-Martínez, De Paz and Márquez (2011) discovered that following a 12-week programme of physical exercise, significant improvements were found in self-esteem, self-concept, emotion, and social functioning when compared to a usual care control group. Furthermore, self-esteem and self-concept scores were positively correlated with emotion. Similarly, the 'mastery hypothesis' maintains that physical exercise engagement leads to enhanced feelings of mastery and competence, in turn improving psychological health (Gauvin & Brawley, 1993).

4.0.1.2. Physiological Explanations

Despite important psychological theories and supportive results for them, the physical effects of chronic and acute exercise cannot be overlooked. Whilst physical health benefits are most prominently established (Fentem, 1994), proposals related to numerous other physiological systems have also gained influence in the field. A recent review by Moylan et al. (2013) highlights the vast number and influence of physiological explanations provided by recent research into this association. These include, but are not limited to: (I) upregulation of emotion-related endorphins and monoamine neurotransmitters, such as serotonin, dopamine, and noradrenaline (Lautenschlager et al., 2012; Sarris et al., 2012); (II) re-regulation of the hypothalamo-pituitary-adrenal axis, resulting in the reduction of glucocorticoid stress hormones (Eyre & Baune., 2012); (III) modulation of a key mediator in mood and anxiety disorders - inflammation, oxidative and nitrogen stress (O&NS; Eyre and Baune, 2011), and (IV) increased opioid activation in areas involved in the stress response - the central and peripheral nervous systems (Harber & Sutton, 1984). Since opioid antagonists, such as naloxone, heighten negative responses to stressful events (Grossman & Moretti, 1986; Allen & Coen, 1987; McCubbin, Kaplan, Manuck, & Adams, 1993; McCubbin et al., 1996), adaptive increases in opioid activity through physical exercise may account for reductions observed in reports and feelings of stress (see Salmon, 2001 for a review).

Taking a different perspective, the thermogenic model proposes that increased body heat as a result of intense activity is a direct factor in the reduction of stress and anxiety through mitigated tension (Craft and Perna, 2004). As noted by Petruzzello et al. (1991), this association is similar to those produced by sauna bathing and heightened muscle relaxation. A critical review by DeVries (1981) outlined the relationship between core body temperature, including both external and neural regions, and reports of anxiety. However, subsequent studies that have directly tested this theory have produced opposite effects. For example, upon implementing 20 minutes of moderate-high-intensity exercise and measuring body temperature of participants, Reeves et al (1985) discovered that increased temperature was positively associated with increased anxiety. Importantly, Hori et al. (1978) noted that exercise-induced temperature increases return to baseline within 90 minutes following engagement. The thermogenic theory can therefore only account for acute exercise effects, and research to date has been unsuccessful in accounting for chronic exercise effects when considering this explanation.

In addition to neural and thermogenic accounts, physical exercise is commonly associated with modulation of stress-related BP responses in multiple stressor situations (Roy and Steptoe, 1991; Probst, Bulbulian & Knapp, 1997; Bartholomew, 2000). Importantly, the mechanisms underpinning the beneficial BP modulation effects of exercise are considered to differ depending on whether exercise exposure is acute or chronic. Specifically, acute exercise appears to positively adapt immediate post-exercise hypotension (PEH; Quinn et al, 2000), whilst chronic exercise has been found to lead to anti-hypertensive effects (Pescatello et al, 2004; Pescatello, 2005). Indeed, meta-analyses from both Crew and Landers (1987) and Hamer, Taylor, and Steptoe (2006) have support the beneficial BP effects of physical activity. However, whilst Crew and Landers reported positive physical fitness (chronic) effects with reference to stress-related BP attenuation, Hamer and colleagues concluded that physical fitness was not associated with BP responses following exercise. Interestingly, the authors posit that such inconsistent effects may be due to those in chronic exercise studies performing within the post-exercise window following acute exercise (Hamer, Taylor and Steptoe, 2006). Reports have demonstrated interaction and overlap between both types of exercise. That is, repeatedly engaging in acute bouts of exercise to lessen cardiovascular reactivity may lead to an accumulative effect on overall cardiovascular health (Hamer, Taylor and Steptoe, 2006).

In sum, numerous studies and meta-analyses have proposed and presented various explanations regarding the putative mechanisms via which emotional wellbeing is positively affected by exercise (Forcier et al., 2006; Jackson and Dishman, 2006; Alex et al., 2013; Huang, Webb, Zourdos and Acevedo, 2013). Psychological theories, such as cognitive dissonance and self-perception, have been proposed to mediate this relationship, as have a number of direct and indirect physiological mechanisms. Variations in participant demographics, levels of intensity, and post-exercise testing periods are often cited as confounding issues when researchers attempt to sort through and summarize the literature on this topic. Despite a significant degree of lack of consensus on the mechanisms underlying the stress-exercise relationship, however, there is overwhelming evidence and consensus that physical exercise is an important mediator in the treatment of stress and emotional well-being (Moylan et al., 2013).

4.0.1.3. Temporal properties

The temporal properties of exercise effects are certainly important when considering the efficacy and mechanisms of programmes. This extends to three separate components– the length of the exercise session; the length of the exercise programme (acute/single bout versus chronic/repeated engagement), and the time between testing and the preceding exercise activities. Firstly, researchers such as Petruzzello et al (1991) have maintained that exercise sessions of at least 20-30 minutes are superior for reducing anxiety, whereas sessions that fall below this duration are much less effective. Second, as previously discussed, outcomes relating to emotional wellbeing may vary depending on whether immediate arousal is measured after an acute bout of exercise, or following a longer programme. For example, whilst a single bout of exercise may lead to immediate upregulation of endorphins, explanations such as the 'mastery hypothesis' may more effectively explain longer-term engagement effects. Additionally, variations exist when considering the temporal patterns of effects after the physical activity is complete. For example, whilst some researchers suggest that target behaviours gradually decline and return to baseline within 90 minutes of exercise (Levinson and Reid, 1992), others maintain a less linear after-effect, with worsened moods following exercise in fact improving relative to baseline after an additional 30-minute wait period (Tate & Petruzzello, 1995; Raglin & Wilson, 1996). These factors are essential to consider when examining and evaluating the literature, particularly with regards to studies which report null results. Often, however, these temporal properties are not carefully considered or reported, which is a serious contributing limitation to the existing literature.

4.0.1.4. Intensity

The intensity at which the physical exercise is performed may affect the outcome of emotion-related research. For example, a study by Moses, Steptoe, Mathews and Edwards (1989) compared the effects of two physical activity programmes of varying intensity: high-intensity aerobic training, and moderate intensity aerobic training. An attention-placebo group and waitlist control group were also included. A total of 75 sedentary adults completed the programme, having been randomly assigned to a group for a 10-week training period. Results revealed positive psychological improvements only in the moderate intensity group, and not in the high-intensity exercise or attention-placebo control group. Specifically, participants reported improvements in tension/anxiety, confusion, perceived coping deficits and depression. Since this testing was more than 30 minutes following exercise, results cannot be explained by the timing model as reported above. The superior effects of moderate intensity activity were also suggested by Biddle & Mutrie (2008). However, a systematic review and meta-regression analysis by Lawlor and Hopker (2001) found no differential effects of exercise type when considering physical exercise as an intervention tool for depression management. Since the specific mechanisms for the stress-exercise relationship are not yet agreed, it is for future research to further investigate the temporal and intensity effects in relation to programme efficacy.

4.0.1.5. Summary

So far, we have explored a number of intervention studies aimed at reducing feelings of stress and anxiety in the general population. Although there is limited consensus regarding underlying causes, we discussed multiple proposed mechanisms for this relationship, including psychological theories of self-perception, and physiological theories of neural arousal and adaptation. Importantly, mixed results within the literature may be explained by differential intervention study properties, such as time of testing, and intensity of the exercise. Nevertheless, overall, the results are promising, and there is a gap in the literature to investigate these effects in a well-controlled study with ASD and other SEN participants. The following section will report specifically on the effects of acute exercise engagement on stress and anxiety in clinical populations. It will then present novel empirical research based on existing findings and shortfalls in the literature. The findings will then be discussed in relation to this current literature, with further general discussion points outlined in Chapter 6.

CHAPTER 4A: STUDY 1 - THE EFFECTS OF AN ACUTE EXERCISE INTERVENTION ON SELF-REPORTED STRESS IN SCHOOL-AGED CHILDREN WITH ASD AND OTHER SEN

4.1.1. Introduction

The acute effects of physical exercise on emotional wellbeing are well documented in both clinical and typical populations. Reports include beneficial effects on a multitude of emotional states and traits, including dysphoria and depression (Craft & Landers, 1998), stress and anxiety (Fremont and Craighead, 1987; McAuley et al., 1996), and anxiety sensitivity (Broman-Fulks, Berman, Rabian and Webster, 2004). Given that such psychological gains are found to surface after just one single bout of activity, physical exercise as an intervention for various populations is gaining a vast amount of attention. The relatively accessible and cost-effective nature of physical exercise is particularly appealing, and may serve as an immediate coping strategy for negative symptomology (Vancampfort et al., 2010).

Various experimental studies provide support for the role of physical exercise in the regulation of emotion in non-typical and clinical populations. For example, Broman-Fulks and colleagues (2004) investigated the effects of 20-minute high or low (walking) intensity treadmill activity in 54 participants with heightened anxiety. After completing six acute exercise sessions over two weeks, the authors found that anxiety sensitivity was reduced following both high and low intensity exercise. High-intensity activity appeared more successful at rapidly reducing negative scores, and was alone accountable for reductions in anxiety-related physiological sensations (Broman-Fulks et al., 2004). However, low-intensity exercise appeared to reduce global anxiety sensitivity, as supported by the findings of Sexton et al. (1989). Despite limitations, including lack of generalisability due to a somewhat unrepresentative sample, the researchers conclude that physical activity may indeed be a viable treatment option for physically-able patients with anxiety.

Such results have been replicated in other clinical populations. A study by Vancampfort et al. (2010) explored the effects of acute exercise in 40 schizophrenic participants. The authors maintained that individuals with schizophrenia often have decreased coping strategies, particularly when experiencing feelings of stress. Participants were randomised to receive either 20 minutes of aerobic exercise at a self-set intensity, 30 minutes of yoga, or no exercise as a control condition. After one session, it was found that both yoga and aerobic exercise groups demonstrated significant decreases in both state anxiety and psychological stress, as well as increases in subjective wellbeing, when compared to the control group. There were no significant differences in the outcomes between the two exercise conditions, suggesting that exercise intensity may not be as important for feelings of stress and anxiety as it is for other behavioural outcomes, such as cognitive performance (Winter et al., 2007). Vancampfort and colleagues concluded that patients with schizophrenia may select aerobic activity or yoga as an effective coping strategy for stress and anxiety.

Similarly, Bartholomew, Morrison and Ciccolo (2005) reported that a single bout of moderate-intensity exercise (50-70% $VO_{2\ max}$) for at least 10- to 15- minutes is sufficient enough to reduce negative emotions, such as feelings of depression. In their study, 40 patients diagnosed with major depressive disorder (MDD) were randomly assigned to a target 30-minute aerobic exercise condition, or a 30-minute period of quiet rest. Given previous conflictions regarding the duration of effects, an important strength of this study is the reporting of temporal properties between exercise and affective state testing. That is, participants filled out the mood-related questionnaires 5, 30, and 60 minutes following exercise or rest-period completion. Results revealed a superior effect of physical exercise in two of the nine subscales of the Subjective Exercise Experiences Scale (SEES; McAuley & Courneya, 1994) and Profile of Mood States (POMS; McNair, Lorr and Droppleman, 1992). However, with the exception of the fatigue subscale of the SEES, the remaining subscales which produced no significant differences between groups highlighted the efficacy of both rest and aerobic activity in the treatment of negative emotion. Nevertheless, exercise improved vigour (POMS) and psychological well-being (SEES) when compared to the control condition, prompting the authors to recommend physical activity as a mood-regulating intervention. Although they confirmed that such increase in positive states returned to baseline within 60 minutes, Bartholomew, Morrison and Ciccolo highlight the efficacy of such an immediate effect when compared to the expected 2-8-week delay before pharmacologic treatments demonstrate an effect for patients.

Given the promising behavioural findings in other populations, it is surprising that very few large-scale studies focus on exercise as an intervention tool for stress and anxiety in ASD. Along with the positive results found in typically developing children, physical exercise appears equally beneficial to multiple populations throughout the lifespan (Kamijo et al., 2008). Indeed, Pan and Frey (2006) maintain that interventions for the young ASD population should in fact target the increase of available physical activity resources. Nevertheless, to the authors' knowledge, there is no current research into the effects on stress in the ASD population following acute bouts of physical exercise intervention in a school-based setting. As highlighted by Biddle & Mutrie (2008), short term effects of exercise may be more prominent than those occurring as a result of chronic engagement due to novelty (Motl & Gosney, 2007) or distraction (Bahrke and Morgan, 1978). However, since experimental conditions vary significantly across studies, it is somewhat difficult to compare effects both within and between them (Salmon, 2001). Furthermore, it is highly likely that improvements in emotional well-being as a result of exercise is coupled with an enjoyment of the physical exercise (Biddle & Mutrie, 2008), which previous research fails to address and/or measure. Thus, although previous studies provide a promising basis for the continuation of research in this field, they are not without restrictions and limitations. An overriding criticism of many current studies is the lack of participant blindness, with participants providing self-reports in full awareness of the hypothesis. Furthermore, the majority of current research studies rely on voluntary participation, which often excludes those who are particularly avoidant of physical exercise. Finally, few studies employ well-controlled designs with adequate sample sizes. Further research is certainly required to build upon these limitations, utilising large-scale controlled designs that aim to increase current sample size standards and target the ASD population.

Importantly, as discussed in detail in Chapter 2, many successful ASD interventions incorporate behavioural components that facilitate treatment, including shared control, increasing motivation of the child, and verbal and/or tangible reinforcement. However, to date, there is no published research that outlines how to optimally implement a physical exercise programme for those with ASD and related conditions within the school environment. Given the reportedly low levels of participation in physical exercise in this group, this study aims to successfully integrate an acute physical exercise programme into the regular school week using an ABA-style framework (see Table 6, Appendix B). We will use behavioural techniques to facilitate engagement of participants to whom physical exercise is often a non-preferred/avoided activity. Measures of diagnoses-appropriate self-reported stress and coping abilities will be taken.

Crucially, although participants will be given the opportunity to opt-out of testing, all children will be exposed to increased physical exercise as part of the school timetable. Since careful consideration will be given to reinforcement techniques and the available choice of physical activities, participants are likely to engage in sufficient exercise and maintain appropriate intensity levels throughout the sessions. Participants will be measured following one school week of daily exercise classes, and one week of education as usual. In line with previous recommendations, participants will be measured within 90 minutes of their final session to assess short-term effects. Measurements of exercise class enjoyment will also be collected both prior to the intervention period, and immediately following completion. To build on previous limitations, a double-blind design will be adopted. Measures of executive function performance will also be collected in this study, and presented in Chapter 5.

4.1.1.1. Study aims and hypothesis

In sum, the aim of this study is to investigate the effects of physical exercise on reported stress in ASD and SEN participants. Taking the current literature into account, it is expected that engaging in physical exercise will decrease reported stress in both diagnostic groups when compared to education as usual.

4.1.2. Methods

4.1.2.1. Participants

Table 7: Participant demographics for Study 1

Diagnoses	*n*	Chron. age	Gender	Order	BAS verbal	BAS Non-Verbal
ASD	24	**M:** 13.49 **SD:** 0.77	23 males, 1 female	**EDU first:** 13 **Ex. first:** 11	**M:** 72 **SD:** 14.32	**M:** 33.25 **SD:** 12.83
SEN	29	**M:** 13.22 **SD:** 0.84	22 males, 7 females	**EDU first:** 13 **Ex. first:** 16	**M:** 73 **SD:** 15.64	**M:** 29.41 **SD:** 10.35

Fifty-eight participants were recruited from a secondary school in the United Kingdom through opt-out letters sent home to parents and carers. Opportunity sampling was used to collect participants, resulting in a limit to number and age range. All children provided written consent to participate in the research. One of these pupils was excluded from analysis due to irregular school attendance during testing phases; two did not meet minimum participation (10 consecutive minutes) during exercise sessions due to non-compliance, and one pupil was unable to partake in the intervention due to recent minor surgery. In total, fifty-four pupils participated in the study: 24 with ASD (1 female, mean age= 13.49, SD= 0.77), and 29 with SEN (15 females, mean age= 13.22, SD= 0.84). Pupils were attending the Special Educational Needs school due to possessing educational statements with Moderate Learning Difficulties (MLD), Autism Spectrum Disorders (ASD) or Behavioural, Emotional and Social Disorder (BESD) as their primary needs. Those with ASD obtained their diagnosis through an external Educational Psychologist, with additional verification obtained via administration of the Autism Diagnostic Observation Schedule –

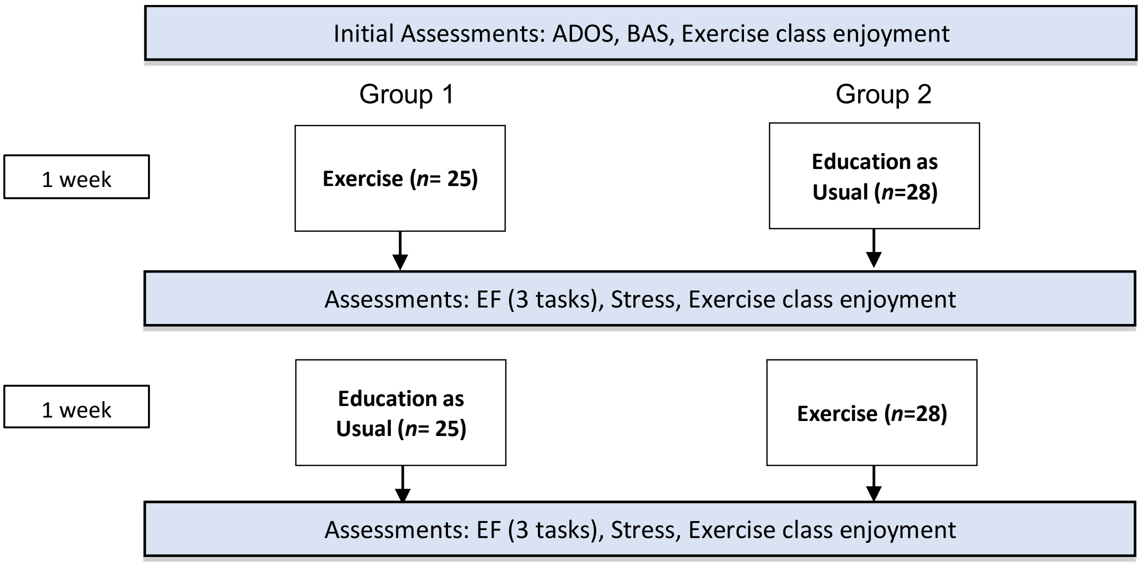
Generic (ADOS-G; Lord et al., 2000) by a research-trained examiner. Confirmation of the absence of ASD and related traits in the SEN group was also further verified by administration of the ADOS.

The British Ability Scales – Second Edition (BAS-II; Elliott, Smith & McCulloch, 1996) was administered to obtain standardised verbal (*word definition* and *verbal similarities*; ASD average= 71.43, SD= 0.77; SEN average= 73.07, SD= 15.64) and non-verbal (*matrices*; ASD average= 33.17, SD= 13.11; SEN average= 29.41, SD= 10.35) abilities. Participants were matched for chronological age ($p=0.24$), as well as verbal ($p= 0.13$) and non-verbal abilities ($p= 0.80$). All participants had normal or corrected-to-normal vision. No individual qualified for participation if they had any cardiovascular or related health problems, details of which were obtained from both the school and parents. The research was reviewed and approved by the Science, Technology, Engineering, and Mathematics Ethical Review Committee at the University of Birmingham.

4.1.2.2. Design

A large-scale within-subjects experimental design was adopted for this study (see Figure 5). True randomisation was not possible due to the nature of class-based research. Groups were predetermined by each child's class allocation, although order of treatment between classes was randomly assigned.

Figure 5: Experimental design



4.1.2.3. Measures

When reviewing the stress and anxiety-related literature, it is clear that a vast amount measures are used to determine intervention success. Although stress and anxiety are often used interchangeably, there are key differences between the two. Whilst *stress* is often brought about by a particular event or situation, such as increased workload or a change in routine, *anxiety* is the term used for a predisposition to certain events or situations, often in absence of the stressor. Although children and adolescents with ASD and other conditions are often associated with both (Groden et al., 1994), stress was chosen for this particular study through the opportunity to use two existing age-appropriate scales that focused on stressors that are likely to occur during the school week. Alternative measures of interest included analysis of cortisol change (Corbett et al., 2009), which was not within the resource limitations of this study, and the State-Trait Anxiety Inventory (STAI; Speilberger, 2010) which was deemed as less ASD-appropriate than the measures outlined below.

All measures were collected by undergraduate research assistants who were blinded to the child's experimental status at all times.

Stress

The Stress Survey Schedule for Individuals with Autism and Other Pervasive Developmental Disabilities (SSS; Groden et al, 2001)

The SSS is a 49-item questionnaire developed as a clinical tool to inform practitioners and caregivers about levels and types of patient stress. The questions are categorised into one of eight stress types: Changes, Anticipation, Unpleasant Events, Pleasant Events, Sensory/Personal Contact, Food-Related Activity, Social/Environment Interactions, and Ritual-Related Stress. Goodwin, Groden, Velicer & Diller (2007) reported validity of the SSS in both verbal and non-verbal individuals with ASD (M= 27.2 years). Although the survey primarily measures stress and susceptibility to triggers, comparison by Gillott and Standen (2007) found a significant positive correlation between scores on the SSS and levels of anxiety and coping in ASD individuals.

For the purpose of this study, given the developmental and chronological age of participants, statements with more advanced terminology were re-phrased (see Appendix D). To ensure for consistency, all research assistants were given training and printed versions of the rewritten statements. Furthermore, whilst the survey was originally designed in the format of a questionnaire, participants were read each statement verbally before being prompted for their answer. This ensured that clarification could be provided if needed. Importantly, a visual response scale was developed in collaboration with the school, incorporating the same numbers and emotion category (e.g. *None to Mild* and *Mild* *to Severe*) as the original questionnaire, as well as a colour scale similar to that used in classrooms, and representative faces (see Appendix E). Participants were reminded that their response could be in any one of these formats, indicated by pointing to the corresponding face/colour, or by saying the number/emotion category associated with the most accurate response. Since many children of this population possess social and/or communication barriers, the colour and face scales were deemed extremely practical. As recommended by the school, Statement 8 – 'being touched' – was removed, to avoid the disclosure of sensitive and confidential information.

The Pre-Post Stress Survey

The Pre-Post Stress survey was taken from a lesson plan developed by the Beacon Learning Centre (Accessed 22nd July 2012, see Appendix F). The 19 statement Pre-Post Stress Survey was designed to monitor perceived stress and coping capabilities as reported by pupils themselves. Given that this questionnaire includes statements that focus on coping strategies, such as 'I have learned skills to help me adjust to stress' and state-related statements about school, such as 'I look forward to attending school', it was deemed valuable in addition to the trait-focused SSS. Again, statements were read to each participant, to which they were instructed to simply answer with either 'Yes' or 'No'.

Exercise Enjoyment Survey

Participants were surveyed to assess enjoyment of exercise classes in school. This was important given the lack of motivation associated with exercise in this population. Participants were asked (1) whether or not they enjoy exercise in school and (2) what their reasons were for this answer. Participants could respond with either ‘Yes’, ‘No’, or ‘Don’t Know’ for the first question, and were given the opportunity to provide any reasons in the second open-ended question, which asked: ‘Why do you/don’t you like exercise in school?’ with direction depending on their previous answer. The questions were asked by blinded researchers who the children had no interaction with throughout the exercise and baseline periods. Importantly, these questions were interspersed within other measures during the testing session, and it was explained that their honest opinions will help teachers to know what the pupils do and do not like. Furthermore, they were told that all of their answers are anonymous, and nobody else could see how each individual answered. This information was deemed crucial for (a) understanding the baseline enjoyment of exercise in this ASD and SEN population, and (b) highlighting themes in the reasoning for enjoying/not enjoying exercise at school. The information also aids understanding of how future programmes can improve based on preferences of this cohort.

Heart Rate

Heart rate was measured using an ANP100 Finger Pulse Oximeter which displayed beats per minute. Children clipped the monitor onto their finger themselves to avoid any anxieties or sensory barriers. Heart rate was measured at the peak of the exercise session (around 15 minutes into the activities). The target heart rate was above 70% of participants’ maximum heart rate (Max HR). This was calculated using the equation *‘220-(age)*0.7’*. As in a study by Davis et al (2007), given their chronological age, children were verbally or tangibly (points on their chart) reinforced for averaging above 145 beats per minute when measured.

4.1.2.4. Procedure

Exercise sessions

Structured guidelines for key ABA-based intervention components were followed throughout the programme (see Appendix B). Exercise sessions were conducted by school staff who the children were comfortable and familiar with and who were blind to the experimental hypotheses. This familiarity served three purposes; (1) a level of authority and mutual respect had already been established, (2) the staff member was aware of effective motivators for each pupil, and (3) the sessions appeared less out of the ordinary for each group. For indoor activities, the school sports hall or other large rooms were used. Energy exertion was attained through jogging/lap-running activities, or aerobics. For aerobics-based sessions, a projector displayed a DVD of an instructor performing a series of high-intensity activities, such as running on the spot and star jumps.

Importantly, the class teacher as well as teaching assistants participated with the children, engaging in the same activities whilst praising the children's efforts on both individual and group levels. Outdoor exercise classes were conducted when weather conditions permitted. For outdoor activities, high-intensity exercises and games were encouraged, including jogging laps around the school field, as well as energetic running games such as 'Tag'. To reduce social demands, intensity and enjoyment were emphasised over performance, whilst competition was discouraged. A five minute warm up and cool down (stretches and brisk walking) were given before and after each 20-30-minute session respectively. Pupils were then provided with water and a short break before returning to desk-based lessons.

Sessions were conducted in the mornings to ensure enough time to complete testing before the end of the school day on the necessary days. Measures of change were taken on the last day of each week, and were tightly time-controlled. That is, if a child was tested at 11am during the first week, it was ensured they were tested at the same time on the second week to eliminate effects of fluctuating mental fatigue levels (van der Linden, Frese and Meijman, 2003). Stress measures were collected prior to the computer-based EF tasks, to ensure that participation in EF tasks did not cause any additional stress, particularly in those with performance anxiety.

Choice of Activities

It is well established that individuals with autism demonstrate decreased flexibility and increased aversion to changes in routine (DSM-IV:299.00; American Psychiatric Association, 2000). Due to the fact that this research was conducted in a secondary school, it was acknowledged that this intervention may present the adolescents with disruption to their expected daytime routines. In order to increase the probability of compliance and to decrease feelings of anxiety or distress due to such disruption, this study was designed to provide participants with a choice of desirable activities. Research by Orsmond, Krauss and Seltzer (2004) highlighted that adolescents on the autistic spectrum prefer activities without social demands. Pan and Frey (2006) extended these findings to physical exercise, noting that individuals with ASD may be more likely to participate in such activities when given choices that are not based on opportunities to socialise. Thus, participants were provided with a choice of activities which were both appropriate and effective in increasing participation and maintaining exercise intensity. The physical activity session did not require any more from the child than a typical physical education class at school. At no point was a participant asked to engage in a dangerous activity, nor were they expected to continue should they not feel able to.

Length of Exercise Sessions

The decision to aim for 20 minutes of high-intensity exercise during the physical activity weeks was based on the guidelines supplied by the UK Department of Health (UKDH, 2011). That is, at least 60 minutes' engagement in appropriate physical activities on most or all days of the week, including 20 minutes of continuous moderate- to vigorous sessions at least three times per week (UKDH, 2011). Thus, such length of engagement was deemed suitable to produce the desired outcomes whilst reducing the likelihood of negative outcomes, including fatigue and exercise-related physical risks.

<u>Education as Usual (EDU)</u>

During control weeks, education remained as usual for the pupils. However, it was arranged with staff that during these weeks that should their regular weekly physical activity class fall on a day of testing, the content of that class was either primarily theory-based, or involved lower intensity activities such as light ball games, which have not previously been found to impact experimental results (Kern, Koegel and Dunlap, 1984). Again, stress and EF assessments were conducted on the last day of the week following their final session. As in studies by Seigel and Manfredi (1984) and Tuckman and Hinkle (1986), physical activity outside of sessions was not restricted, modified or monitored for either groups.

4.1.3. Results

The statistical package SPSS 17.0 for Windows (Statistics, SPSS, 2008) was used for all analyses. Since analysis method varied for each measure, further details are outlined in each respective section. Descriptive statistics for the stress and enjoyment measures are presented in Tables 8 to 11.

4.1.3.1. Session Order

As in the analysis of Hillman et al. (2009), preliminary analyses were conducted to explore any influence of session order. A repeated measures ANOVA was conducted separately for each dependent variable (education as usual and intervention scores on the SSS and pre-post exercise survey), with session order as a between-subjects variable (2 levels: *Exercise first, EDU second,* versus *EDU first, Exercise second*). No significant main effects or interactions of session order were found for any variable ($p > 0.05$).

4.1.3.2. Heart Rate

Descriptive statistics were performed on recorded HR of participants. Analysis revealed an average of 150 beats per minute (BPM) across all participants (M= 150.75, SD= 21.84, Range= 96-177) at the peak of exercise engagement, equivalent to around 73% Max HR. An independent samples t-test was conducted to explore any differences in HR between diagnostic groups. Findings revealed no significant differences, with ASD HR (M= 154.26, SD= 18.43) statistically similar to that of the SEN participants HR (M= 148.07, SD= 24.09) across both intervention groups, $t(51) = -1.024$, $p = 0.311$.

4.1.3.3. Stress

Groden's Stress Survey Schedule

Overall scores

Table 8: Scores on the SSS at education as usual and post-intervention

	Diagnostic group	Mean	Std. Deviation
Education as usual (EDU)	**ASD**	107.71	24.377
	SEN	114.38	26.118
	Total	**111.36 ***	**25.326**
Intervention	**ASD**	97.04	23.876
	SEN	106.55	23.026
	Total	**102.25 ***	**23.674**

* represents statistical significance

A 2x2 mixed factorial ANOVA was conducted, with intervention phase (EDU versus exercise) as the within-subjects factor, and group (ASD versus SEN) as a between-subjects factor. A significant main effect of intervention phase was found, with both groups reporting lower levels of stress following the exercise period (ASD M= 97.04, SD= 23.88; SEN M= 106.55; SD= 23.67) when compared to education as usual (ASD M= 107.71, SD= 24.38; SEN M= 114.38, SD= 26.12), $F(1,52) = 19.920$; $p < 0.001$, $\eta_p^2 = .281$. There were no significant effects or interactions that included diagnostic group. Overall, the SEN group reported moderately higher stress levels than the ASD group at both EDU and exercise, but these differences were non-significant ($p > 0.05$).

The Pre-Post Exercise Questionnaire

Table 9: Scores on the PPEQ at education as usual and intervention

	Diagnostic group	Mean	Std. Deviation
Education as usual (EDU)	**SEN**	6.86	3.502
	ASD	6.96	3.127
	Total	**6.91 ***	**3.307**
Intervention	**SEN**	5.76	2.911
	ASD	5.79	2.859
	Total	**5.77 ***	**2.860**

A repeated measures ANOVA revealed a significant effect of intervention phase (education as usual versus exercise) at the group level, $F(1,51) = 7.535$, $p < 0.05$, $\eta_p^2 = .129$. No further main effects or interactions of diagnostic group were found. Overall, participants reported higher levels of stress and negative experiences at EDU (ASD M= 6.96, SD= 3.31; SEN M= 6.86, SD: 3.50) when compared to intervention (ASD M= 5.79, SD= 2.86; SEN M= 5.76, SD= 2.91), $t(52) = 2.777$, $p < 0.05$. Descriptive analyses highlighted that both ASD and SEN groups agreed with the statement 'I look forward to attending school' at a higher frequency during the intervention period (77.3%) than when compared to EDU (60.3%) in the Pre-Post Exercise Questionnaire.

4.1.3.4. Exercise Enjoyment Survey

Table 10: Responses to "Do you enjoy physical exercise classes in school?"

	Education as usual (EDU)		Exercise	
	ASD	**SEN**	**ASD**	**SEN**
"No"	79.2% (19)	34.5% (10)	29.2% (7)	20.7% (6)
"Yes"	20.8% (5)	55.5% (19)	70.8% (17)	79.3% (23)

Chi-Square Tests of Independence were performed for EDU and for intervention responses to explore the differences between groups at each time-point. EDU results revealed a significant difference between responses, $X^2 = 8.8557$, $p < 0.005$, with ASD more likely than SEN participants to respond with “No”. Post-intervention results revealed no significant difference between groups, $X^2 = 0.1547$, $p > 0.6$, with no difference in the probability of the SEN or ASD groups responding “Yes”.

As displayed in Table 8, the incidence of ASD participants reporting that they enjoy exercise at school increased. Increases were also observed in the SEN group, although not to the same degree. Further binomial tests revealed that the ASD group were more likely to say “No” than “Yes” at EDU ($p < 0.005$), and more likely to say “Yes” at post-intervention ($p < 0.05$). The SEN group were almost significantly more likely to say “Yes” at EDU ($p < 0.1$), and were significantly more likely to say “Yes” than “No” at post-intervention ($p < 0.005$).

<u>Response Categories</u>

Participant responses were categorised into 6 distinct themes: break-related (e.g. ‘*because we get time out from our desks*’, ‘*because it disturbs me in the classroom and I want to go back*’), health-related (e.g. ‘*I know it is good for my heart*’, ‘*it gets me too out of breath*’), peer/social-related (e.g. ‘*I can play with my friends*’, ‘*the other children don’t choose me*’), teacher-related (e.g. ‘*my teacher joins in so it’s funny*’, ‘*we see if we are faster than our teacher*’), activity-related (e.g. ‘*I love running*’, ‘*I’m not very good at the games*’) and energy-related (e.g. ‘*I prefer to relax*’, ‘*I get too tired*’).

The reasons for both enjoying (‘Yes’) and not enjoying (‘No’) school-based physical exercises classes are displayed in Table 9. As is demonstrated by the above examples, the same response category was often given from both a positive and negative perspective.

Table 11: Frequency of responses given for exercise class enjoyment or non-enjoyment segregated by response category

	Education as usual				Exercise			
	ASD		SEN		ASD		SEN	
	"No"	"Yes"	"No"	"Yes"	"No"	"Yes"	"No"	"Yes"
Break		2		4	2	6		3
Health		2	1	1				
Peers/Social	2		1	10				9
Teacher						9		9
Activity	12	1	3	5	1	2	3	5
Energy	5		5	5	4		3	3

EDU reports

The most reported category for not enjoying participation in exercise at school in the ASD group was type of available activity. The most common reason for enjoying exercise for the SEN participants was related to peer engagement. Unlike the SEN group, no ASD child reported any social benefits of school-based exercise. For those SEN participants who reported disliking exercise classes, unwanted energy exertion was most prominent.

Post-Intervention

Following the intervention period, the most common reason given for enjoyment of school-based exercise for ASD participants was the involvement of their teacher or other member of staff. Unwanted energy exertion was most prominent in the ASD group who reported disliking participation. As in the ASD group, one of the most common reasons for reporting exercise enjoyment in the SEN group was staff involvement. Unlike the ASD group, however, peer interaction was deemed equally as important.

4.1.4. Discussion

The aim of this study was to explore the short-term effects of acute aerobic exercise on the reported stress of school-aged children with ASD and other SEN. A further aim was to successfully incorporate the exercise programme into the school day using structured ABA-based intervention ingredients. Fifty-four school-aged children participated in one week of additional exercise classes implemented during the school day, and one week of education as usual in a counterbalanced order. Reports of stress were collected within 90-minutes of completing the final session of acute exercise, and on the last day of an education as usual control condition. In line with the hypothesis, results revealed a significant decrease in reported stress in both ASD and SEN groups following the intervention programme when compared to education as usual. Furthermore, using established behavioural techniques, the programme was successfully integrated within the school week, with only 3.4% of children not consistently complying throughout the intervention phase.

In line with our predictions, these results support the previous findings of Roth (1989) and Broman-Fulks et al. (2004), who discovered improvements in self-reported anxiety following 20-minute bouts of physical exercise. The present findings also compliment the previous analyses of affect-related exercise outcomes in children (Tomson et al., 2003) and adolescents (Steptoe and Butler, 1996). Although there is no current comparable research in those with ASD, results of those in other clinical populations are supported by these results. For example, Bartholomew, Morrison and Ciccolo (2005) discovered improved psychological wellbeing in patients diagnosed with major depressive disorder following just 30 minutes of moderate intensity aerobic exercise.

However, despite these consistencies, the present results are not in line with all affect-related ASD findings. Namely, whilst some researchers purport higher levels of stress in the ASD population (Groden, Cautela, Prince & Berryman, 1994), in this study, higher stress levels were reported by the SEN group at both EDU and intervention phases. A potential explanation for this effect, however, is the reduced self-report abilities of those with ASD. That is, those with ASD may be less accurate at reporting their own emotions, with previous research suggesting a higher rate of false positive reports from those with ASD and comorbid anxiety (Mazefsky, Kao & Oswald, 2011). Nevertheless, it is important to consider that whilst between-groups comparisons may be limited for this reason, participation in exercise did significantly alter self-reported stress responses for those with ASD and those with SEN, suggesting a level of sensitivity in these measures.

With regards to programme implementation, only two of the 55 children failed to participate in continuous exercise for at least 20 of the 30 minute sessions. Both displayed avoidance behaviours, such as attempting to leave the group or sitting down, that were not affected by the introduction of motivators. For the rest of the group, individual and group level praise, as well as self-motivation to reach the targets and receive reinforcements set by teachers, appeared to ensure maintained participation. As suggested by Pan and Frey (2006), the opportunity for appropriate choices may have contributed to motivation, allowing a level of control that is often not part of the daily school curriculum. As discussed in Chapter 2, this 'shared control' and following the child's lead is a frequent part of many other conventional interventions for ASD populations (Ingersoll, 2010; Hardan et al., 2014). The present results therefore provide support for incorporating structured ABA-techniques into tasks that may already exist within the school curriculum. In order to explore the importance of this aspect further, future research may seek to compare two physical exercise intervention groups; one using the current structure and ingredients, and one conducting classes as usual, without such a framework. Should group differences occur in terms of participation and enjoyment levels, this could have further significant impacts upon field-based practices and academic establishments. For now, the present results are excitingly valuable in terms of maintained engagement, especially given the report of Finkelstein et al. (2010) regarding a lack of exercise motivation in this population. Further, given that this field-based study was primarily teacher-led, we present the efficacy of transferring ABA-based techniques to field-based implementers. As reviewed in Chapter 2, the current evidence-base for ASD interventions is limited by a lack of transference to the real-world, as in our presented study.

Relevantly, enjoyment of the current programme was measured using a short survey. It was predicted that children would report higher enjoyment of exercise programmes following the intervention period than at intake. Supporting these predictions, reports of exercise class enjoyment rose from 20.8% to 70.8% in ASD participants, and from 55.5% to 79.3% in SEN participants. Interestingly, no children with ASD reported any social benefits of exercise classes, with some reporting this as a reason to dislike school-based exercise. On the other hand, children with other SEN reported the social aspect as one of the most prominent reasons for enjoying exercise at both pre and post intervention periods. This supports research by Finkelstein, Nickel, Barnes and Suma (2010), who report that the social aspect of physical exercise may indeed be a reason for avoidance of such activity in the ASD population.

In our study, a particularly interesting finding is the response rate related to teacher involvement after completing the programme. In this intervention, emphasis was made regarding praise from teaching staff, as well as active involvement to maintain interest and energy exertion levels. Classroom teachers and teaching assistants joined in with jogging, aerobics and indoor activities, perhaps leading to an increase in pupil-teacher relationship and quality of interaction. Notably, previous research by Kaale et al. (2012) has reported improvements in child-teacher interactions following a structured, teacher-led intervention period. Although teachers in the present study subjectively reported a greater rapport with their class following these sessions, it is for future research to determine the power of pupil-level teacher involvement in physical exercise activities and the outcome of school-based intervention programmes.

Despite this research being limited in that the physiological underpinnings of these changes were not measured, previous research into multiple populations provides possible explanations. Firstly, according to the distraction hypothesis, it is possible that children reported reduced stress and negative affect due to a novel change in their regular school routines. That is, children were provided with more opportunity to engage in physical exercise as a 'distraction' from classroom-based learning than usual. This explanation is particularly interesting and warrants further research, given that ASD individuals are often associated with negative feelings when experiencing a change to their established routine. A further explanation is that of increased feelings of self-mastery and accomplishment. As mentioned, the ABA-based nature of this programme ensured that each child's motivation was at optimal levels, with praise and reward given for engagement and cooperation. Such increase in immediate positive feedback may in turn positively impact affect scores, particularly if considered alongside theories of self-mastery and accomplishment (Petruzzello et al., 1991).

Importantly, many theorists and researchers do not discard underlying physiological effects of exercise, with many suggesting a combination of mechanisms in this positive affect-exercise relationship. With this in mind, there are numerous other explanations that may account for these effects within 90 minutes of high-intensity exercise that relate directly to physiological underpinnings. These include upregulation of emotion-related endorphins and monoamine neurotransmitters (serotonin, dopamine and noradrenaline; Knöchel et al., 2012; Sarris et al., 2012); a reduction in glucocorticoid stress hormones (Eyre & Baune, 2012c) and increased opioid activation in the central and peripheral nervous systems (Harber & Sutton, 1984). Future research is required to build upon the present study, utilising a similar well-controlled, double-blind design and incorporating similar physiological measures as previously cited in other clinical and non-clinical populations (Harber & Sutton, 1984; Roy & Steptoe, 1991; Eyre & Baune, 2011; Lautenschlager et al., 2012).

The above explanations raise an interesting question: when considering longer-term engagement in physical exercise, are these effects on stress sustained in the absence of a distraction or novelty effect, or when temporary neural arousal may have returned to baseline levels? Although motivation and contingent reinforcement are key ingredients of many current long-term interventions (Rogers & Lewis, 1989; Koegel, 2000; Vismara & Lyons, 2007), attrition rates and reduced motivation of certain populations in physical exercise programmes are high (Salmon, 2001). Therefore, whilst the current results provide novel findings, they are limited only to the short-term effects of exercise in the initial week of exposure.

In sum, the aim of this study was to explore the effects of an acute exercise programme on the reported stress of pupils with ASD and other SEN. A large-scale within-subjects design was used which randomised participants at the classroom level to receive a bout of high-intensity aerobic exercise, and a period of education as usual in a counterbalanced order. Measures of self-reported stress and exercise programme enjoyment were collected to assess the effects of increased school-based exercise when compared to their current school programme. Results revealed that participating in acute, high-intensity (at least 70% of Max HR) aerobic exercise led to immediate reductions in self-reported stress in both ASD and SEN participants when compared to the education as usual control condition. Potential explanations for these results are immediate and concurrent neural arousal, improved self-perception and mastery, and/or the effects of novelty or distraction. However, importantly, if distraction, novelty or mastery indeed account for these results, it is perhaps not illogical to assume a potential fall-off of this initial effect. That is, when children adapt to such changes and engagement in more frequent physical exercise is no longer novel, such mood-improving effects may no longer be as prominent. This question is important, especially when exploring physical exercise as a potential long-term intervention for this population. A further study was conducted to explore this notion, and will be presented in the next section.

CHAPTER 4B: STUDY 2 - THE EFFECTS OF A CHRONIC EXERCISE INTERVENTION ON SELF-REPORTED STRESS IN SCHOOL-AGED CHILDREN WITH ASD AND OTHER SEN

4.2.1. Introduction

In addition to the discussed acute effects, a variety of research studies have been conducted to explore the impact of chronic physical exercise interventions on reported and physiological signs of stress and anxiety. Whilst immediate and concurrent physiological arousal is of great interest, longer-term programmes that can be integrated into an individual's lifestyle and adapted to their physical abilities are perhaps most clinically relevant (Salmon, 2001). The results of Study 1 evidence the efficacy of a school-based exercise programme after acute bouts of exercise, with affect measured within 90 minutes of the final session. Children with ASD and other SEN reported decreased levels of disorder-related stress following exercise when compared to education as usual. Various explanations may account for these changes, such as the effects of novelty (Motl & Gosney, 2007), distraction (Bahrke and Morgan, 1978), and immediate physiological arousal (Lautenschlager et al., 2012). However, it is both clinically and practically relevant to explore whether these positive effects uphold following chronic engagement over a longer period, particularly when effects of novelty, distraction, and immediate physiological arousal are likely to have subsided.

4.2.1.1. Behavioural Evidence

Indeed, a wealth of literature supports the upheld impacts of physical exercise on affective states and traits. For example, a recent study by Carraro and Gobbi (2012) investigated the effects of a 12-week exercise programme for adults with mild to moderate intellectual disabilities (ID). The researchers randomised 27 individuals to either an exercise group, or a painting control group. Both groups received two sessions per week, last one hour each. The exercise sessions were modified based on the skill and fitness of participants, and involved group activities to ensure a social aspect of each session. Self-reported anxiety scores were collected at EDU, midway through the programme (6 weeks), and upon completion of the programme (12 weeks). It was discovered that those in the exercise treatment group demonstrated significant improvements in their state anxiety scores over time when compared to the control group, although both groups showed overall reductions in negative scores. The heightened improvements in the exercise group not only evidence physical activity as a viable intervention for individuals with ID, but that the effects also remain prominent over time, with each session retaining efficacy. This certainly appears to contradict the idea that exercise sessions may be effective only due to an initial novelty effect (Motl & Gosney, 2007).

In a similar study by Carmeli, Barak, Morad and Kodesh (2009), 16 individuals with a dual diagnosis of both mild ID and anxiety were investigated. Acknowledging that these individuals are at risk of having a sedentary and unhealthy lifestyle due to their anxiety, the researchers aimed to explore an intervention option that may improve quality of life. Participants were randomly assigned to receive either aerobic exercise or leisure activities, or were placed in a non-exercise control group. The exercise group participated in three sessions per week for 6 months. Levels of anxiety and quality of life were assessed using the Hamilton Anxiety Scale (HAM-A; Hamilton, 1969). Results revealed significant and clinical improvements in HAM-A scores for both aerobic exercise and structured leisure activities. No significant improvements were found in the control group. Carmeli and colleagues concluded that either physical exercise or leisure activity programmes are efficient at improving the emotional wellbeing of those with ID. Despite the small sample size and self-report measures, these results support the findings of Carraro and Gobbi (2012), building on the growing literature which evidences this link.

Although most research to date has explored the effects of physical exercise and stress in the adult population, there is a limited amount of research that confirms such effects in youths with autism. In one such study, Hiller, Murphy and Ferrara (2011) ran an 8-week exercise intervention for adolescents and young adults with ASD, measuring self-reported anxiety and cortisol levels throughout. Findings revealed significant reductions in cortisol following each 75-minute exercise and relaxation session. These results were mirrored by the participants' self-reported anxiety scores. However, although the reported anxiety levels did not decrease overall upon completion of the programme, the authors suggest that this may be due to the limited number of sessions across the intervention (once per week). Thus, a consistent exercise programme may be effective for both short- and long-term feelings of stress, and a larger number of measures could highlight multiple benefits of an exercise intervention programme for individuals with autism.

4.2.1.2. Exercise Intensity: Does it matter?

Sensory Integration Therapy

Interestingly, a number of studies have demonstrated that only certain types of high-intensity physical activities positively impact performance (e.g. Kern, Koegel and Dunlap, 1984). However, theories of Sensory Integration (SI) attribute the effects to participating in physical exercises that are specifically loaded onto three basic senses: tactile, vestibular and proprioceptive (Ayres, 1972). These senses, as well as visual, auditory, olfactory and gustatory, are organised in the brain to provide information on one's body and environment, allowing appropriate responding. Whilst no individual has perfect integration, this innate process is believed to form the foundations of learning and behaviour (Ayres and Robbins, 2005). Specifically, the tactile system is responsible for sensations of touch, pain, pressure and temperature (Foss-Feig, Heacock and Cascio, 2012). Individuals with ASD are often reported to exhibit tactile defensiveness, resulting from an over-stimulation of the brain in the processing of these senses (Tomchek and Dunn, 2007). Symptoms may include hypersensitivity to touch and pain in some individuals, whilst others experience hyposensitivity to these same senses. Such misrepresentations often cause negative emotional responses, distractibility, and irritability in hypersensitive children, whilst hyposensitive children may seek stimulation and become more prone to injuries. Indeed, both ends of this sense spectrum may be experienced by the same child (Baranek, 2002; Rogers and Ozonoff, 2005). Meanwhile, the vestibular system is responsible for the detection of movement and positioning of the head, whilst the proprioception system is responsible for the awareness of body position, motor planning, and praxis (Ayres and Robbins, 2005). Dysfunctions in either of these systems often lead to extremely high or low activity levels, impulsivity, anxiety, and under-achievement in school (Piek and Dyck, 2004).

Sensory Integration therapy therefore provides physical exercises that target deficits within these systems, aiming to build upon these senses and associated developmental and behavioural skills. For example, targeting proprioception deficits, such exercise often includes actions that require 'crossing the midline' (moving a hand or foot into the space of the other) – a bilateral skill that may not have developed typically in children of this population, but is needed for academic skills such as reading and writing (Ayres, 1972). SI therapists maintain that in affected populations, each side of the brain is not successfully communicating, resulting in decreased coordination, decreased motor control, and difficulties achieving higher level skills (Cermak, Quintero and Cohen, 1980; Ayres and Robbins, 2005). Thus, when compared to aerobic exercise, emphasis in SI therapy is placed on the movement aspect of the intervention, as opposed to the level of intensity. Most notably, as outlined in Chapter 3, children with ASD are widely reported to possess such deficits and impairments in motor skills (Lang et al., 2010), generating great interest in SI therapy as a potential motor-based intervention for this population.

4.2.1.3. Study aims and hypothesis

The primary goal of this study is to assess the effects of a 6 week (30 session) motor-based intervention programme, and whether these effects on the same measures hold up beyond any potential novelty or distraction effects of Study 1. Whilst one group of participants will undergo a longer-term version of the high-intensity aerobic exercise programme in Study 1, another group will engage in a low-moderate intensity SI-based intervention for the same duration. Although intensity will be considered as a factor, intervention effects will primarily be compared to baseline (education as usual) conditions in a within-subjects design.

Since the previous study uncovered positive effects in both children with ASD and those with other developmental conditions, it is hypothesised that a chronic intervention program will also have similar effects across all participants. It is predicted that participating in 6 weeks of motor-based intervention will reduce self-reported stress in both ASD participants and those with other developmental conditions. Specifically, the stress effects found in Study 1 will uphold - and perhaps be strengthened - as a result of a longer programme. Since at least 24 hours will have passed between the final exercise sessions and testing, any effects found are unlikely to be attributed to immediate physiological changes. Furthermore, effects of the sensory integration training and aerobic exercise programme are not predicted to differ, since the previous literature is not vast enough to make directional predictions.

4.2.2. Methods

4.2.2.1. Participants

Table 12: Participant demographic information for Study 2

Diagnoses	Group	*n*	Chron. age	Gender	Order	BAS verbal	BAS Non-Verbal
ASD	**CK:** 10 **AE:** 7	17	**M:** 12.04 **SD:** 0.70	14 males 3 females	**EDU first:** 8 **Exercise first:** 9	**M:** 75.35 **SD:** 15.22	**M:** 37.12 **SD:** 8.02
SEN	**CK:** 13 **AE:** 10	23	**M:** 11.98 **SD:** 0.61	12 males 11 females	**EDU first:** 12 **Exercise first:** 11	**M:** 76.65 **SD:** 14.41	**M:** 34.57 **SD:** 12.82

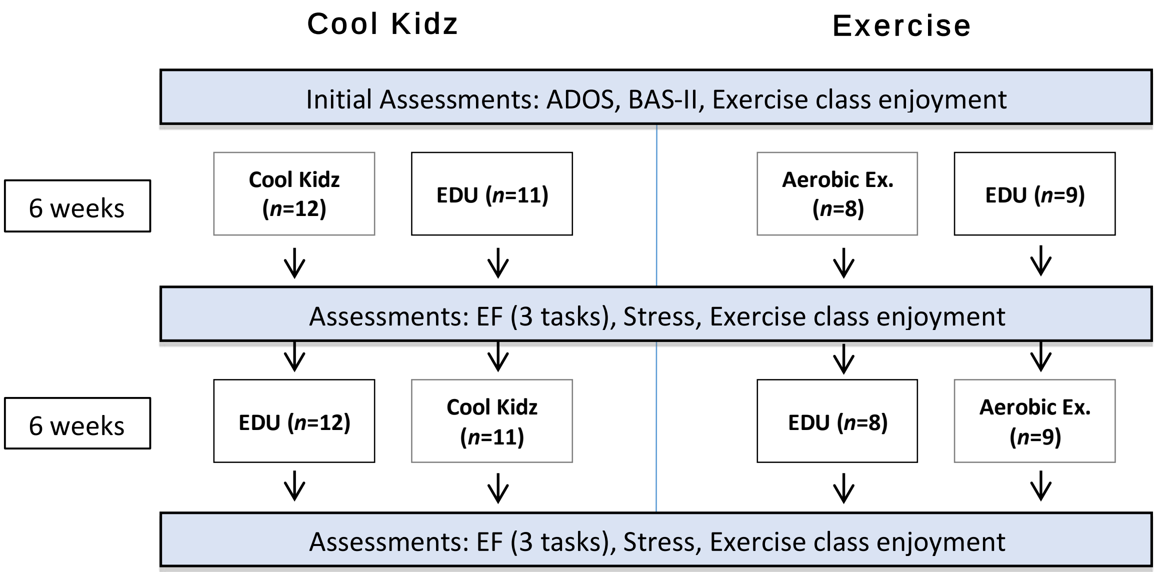
Forty participants who were not part of Study 1 were recruited from a secondary school in the United Kingdom through opt-out letters sent home to parents and carers. Opportunity sampling was used to collect participants, resulting in a limit to number and age range. In total, 17 participants with ASD symptoms (3 females, mean age= 12.04, SD= 0.70), and 23 with SEN (11 females, mean age= 11.98, SD= 0.61) were recruited. Pupils were attending the Special Educational Needs school due to possessing educational statements with Moderate Learning Difficulties (MLD), Autism Spectrum Disorders (ASD) or Behavioural, Emotional and Social Disorder (BESD) as their primary needs. Seven of those with ASD obtained their diagnosis through an external Educational Psychologist, with additional verification obtained via administration of the Autism Diagnostic Observation Schedule – Generic (ADOS-G; Lord et al., 2000) by a research-trained examiner. Due to the nature of the education system, many pupils were still in the process of obtaining an ASD process, based on GP or school concerns. The presence of ASD traits was confirmed by administration of the ADOS. Pupils below the ASD cut-off of the ADOS remained in the SEN group, whilst those who scored above it were placed in the ASD group. The BAS-II was administered to obtain standardised verbal (*word definition* and *verbal similarities*; ASD M = 78.35, SD = 13.26; SEN M = 76.74, SD = 14.32) and non-verbal (*matrices*; ASD M = 37.12, SD = 8.02; SEN M = 34.57, SD = 12.82) abilities. Between the ASD and SEN groups, participants were matched for chronological age ($p = 0.78$), as well as on verbal ($p = 0.72$) and non-verbal abilities ($p = 0.4$).

Two types of physical exercise interventions were used in this study. When considering these groups, 23 of the pupils mentioned above participated in the Cool Kidz (CK) programme (7 females, mean age= 11.58, SD = 0.3), and 17 in the aerobic physical exercise (AE) programme (7 females, mean age= 12.68, SD = 0.3). BAS-II scores for verbal ability (CK M = 78.22, SD= 12.73; AE M = 76.35, SD = 15.31) and non-verbal ability (CK M = 39.30, SD = 11.19; AE M = 30.71, SD = 8.77), as well as chronological age were not matched between groups ($p < 0.05$). Unlike the acute exercise programme (Study 1), participation level was more lenient to reflect real-world events, accounting for illness-related school absences, as well as the fact that pupils are often required for other therapy or time-out. All participants had normal or corrected-to-normal vision. No individual qualified for participation if they had any cardiovascular or related health problems, details of which were obtained from both the school and parents. The research was reviewed and approved by the Science, Technology, Engineering, and Mathematics Ethical Review Committee at the University of Birmingham.

4.2.2.2. Design

A large scale within-subjects experimental design was adopted for these projects (see figure 6). Each participant completed six weeks of daily exercise within school hours, and six weeks of education as usual. The study used a counterbalance design to eliminate practice effects. The same assessments as in Study 1 were administered. As an exploratory factor, one half of participants completed a physical exercise programme based on the theories of Sensory Integration (the 'Cool Kidz' programme), and the other completed a high-intensity exercise programme as in Study 1. Importantly, no child took part in the cognitive testing within 90 minutes of their final session.

Figure 6: Experimental design



Cool Kidz Programme

The Cool Kidz programme was founded on the basis of sensory integration (SI) therapy, and designed as an opportunity for children with poor motor ability to begin building their vital motor skills (Ayres and Robbins, 2005). The programme focuses largely on the SI principles of coordination and activities that involve the crossing of neural networks (Ayres and Robbins, 2005). Acknowledging the link between exercise and school-related performance, it was specifically developed as a teacher-friendly manual, providing schools with comprehensive lesson plans to meet each child's needs. The programme operates on the principles that engaging in targeted gross motor exercise improves fine motor and concentration skills. The programme developers highlight the need for children to engage in 20-30 minutes of physical fitness activities each day. Specifically, the Cool Kidz programme consists of 26 lesson plans, with repetition of a lesson encouraged should children not master the activities during first exposure. Whilst some activities are likely to raise the heart rate (skipping, jumping), each session consists of low-moderate intensity exercises.

4.2.2.3. Measures

Refer to Study 1 for full descriptions of diagnostic and behavioural measures. Each participant was administered the ADOS-G, BAS-II, the SSS, and the pre-post exercise survey by University research assistants blinded to the child's experimental status. Exercise class enjoyment was measured pre- and post- each intervention phase, although reasons for enjoyment were not measured in this version.

Heart Rate

Heart rate was measured using the same ANP100 Finger Pulse Oximeter as in Study 1. Again, children in both groups clipped the monitor to their finger themselves to avoid any anxieties or sensory barriers. Heart rate was measured at the peak of the exercise session (around 15 minutes into the activities) on the day of testing. For the high-intensity PE group, the target heart rate was above 70% of participants' Max HR, with mechanisms for achieving this identical to Study 1. However, for the SI group, no target was given.

4.2.2.4. Procedure

The procedures for the physical exercise group remained the same as in Study 1. For both indoor physical exercise and Cool Kidz activities, the school sports hall or other large rooms were used. However, a key difference between Study 1 and the current study is the time between testing and the final session. It was ensured that at least 24 hours had passed between sessions and assessments, compared to the 90 minutes of Study 1. It was agreed that this time lapse was great enough for immediate physiological effects to return to baseline, whilst remaining within the timeframe of any direct effects of a chronic exercise programme.

4.2.3. Results

The statistical package SPSS 17.0 for Windows was used for all stress analyses. Since analysis method varied for each measure, further details are outlined in each respective section. Descriptive statistics for this section are presented in Tables 13-18.

4.2.3.1. Session Order

As in the analysis of Hillman et al. (2009), preliminary analyses were conducted to explore any influence of session order. A repeated measures ANOVA was conducted separately for each dependent variable (EDU and intervention SSS and pre-post survey scores), with session order as a between-subjects variable (2 levels: *Exercise first, EAU second* versus *EAU first, Exercise second*). No significant main effects or interactions of session order were found for any variable ($p > 0.05$).

4.2.3.2. Heart Rate

An independent samples t-test was used to explore differences in HR between intervention groups. Findings revealed significant differences between groups. Participants in the AE group demonstrated significantly higher HR's (M= 159.82, SD= 14.70; ~77% Max HR) when compared to the CK group (M= 102.09, SD= 13.01; ~50% Max HR); $t(38) = -13.132$, $p = 0.000$. An additional independent samples t-test was conducted to explore any differences in HR between diagnostic groups. Findings revealed no significant differences, with ASD HR (M= 119.65, SD= 30.60) statistically similar to that of the SEN participants HR (M= 131.78, SD= 32.58) across both intervention groups; $t(38)= 1.195$, $p = 0.24$.

4.2.3.3. Stress

Groden's Stress Survey Schedule

Overall scores

Table 13: Scores on the SSS at EDU and intervention

	Diagnostic group	Mean	Std. Deviation
Education as usual	**ASD**	112.47	18.104
	SEN	127.00	23.653
	Total	**120.83 ***	**22.427**
Exercise	**ASD**	107.41	19.650
	SEN	111.57	19.040
	Total	**109.80 ***	**19.164**

* represents statistical significance

A 2x2 mixed factorial ANOVA was conducted, with intervention phase (EDU versus exercise) as the within subjects factor, and diagnostic group (ASD versus SEN) as a between-subjects factor. A significant main effect of intervention phase was found, with both groups reporting lower levels of stress following the exercise period (ASD M= 107.41, SD= 19.65; SEN M= 111.57, SD= 19.040) when compared to EDU (ASD M= 112.47, SD= 18.104; SEN M= 127.00, SD= 23.653), $F(1,38) = 10.045$; $p < 0.05$, $\eta_p^2 = .209$. There were no significant effects or interactions that included diagnostic group. Overall, the SEN group reported moderately higher stress levels than the ASD group at both baseline and exercise, but these differences were non-significant ($p > 0.05$).

Intervention type

To explore any differential effects of intervention type (Cool Kidz and aerobic exercise), a repeated measures ANOVA was conducted, with intervention phase as the within subjects factor, and intervention type as the between subjects factor. A significant main effect of intervention phase was found, with both intervention groups reporting lower levels of stress following the exercise period (CK M= 111.74, SD= 19.475; AE M= 107.18, SD= 18.997) when compared to EDU (CK M= 119.35, SD= 20.435; AE M= 122.82, SD= 25.285), $F(1,38) = 12.594$; $p = 0.001$, $\eta_p^2 = .249$. There were no significant effects or interactions that included intervention type ($p > 0.05$), with both the Cool Kidz and aerobic exercise performing similarly.

Pre-Post Exercise Questionnaire

Table 14: Scores on the PPEQ at EDU and intervention

	Diagnosis	Mean	Std. Deviation
Education as usual	**ASD**	6.71	3.216
	SEN	5.61	2.589
	Total	**6.08 ***	**2.886**
Exercise	**ASD**	5.76	2.840
	SEN	5.00	1.954
	Total	**5.33 ***	**2.368**

* represents statistical significance

A repeated measures ANOVA revealed a significant effect of intervention phase (EDU versus aerobic exercise) at the group level, $F(1,38)= 4.889$; $p < 0.05$, $\eta_p^2 = .111$. Overall, participants reported higher levels of stress and negative experiences at EDU (ASD M= 6.71, SD= 3.22; SEN M= 5.61, SD: 2.59) when compared to intervention (ASD M= 5.76, SD= 2.84; SEN M= 5.00, SD= 1.95), $t(39) = 2.211$, $p < 0.05$. A further repeated measures ANOVA was conducted to explore between-group differences for ASD and SEN participants. No significant effects of diagnostic group were found at baseline or EDU ($p > 0.05$).

Intervention type

To explore any differential effects of intervention type (Cool Kidz and aerobic exercise), a repeated measures ANOVA was conducted, with intervention phase as the within subjects factor, and intervention type as the between subjects factor. There were no significant effects or interactions that included intervention type ($p > 0.05$), with both the Cool Kidz and aerobic exercise performing similarly.

4.2.3.4. Exercise Enjoyment Survey

Table 15: Responses to "Do you enjoy physical exercise classes in school?" in the AE group

	Education as usual		**Exercise**	
	ASD	**SEN**	**ASD**	**SEN**
"No"	52.9% (9)	76.9% (10)	17.6% (3)	26% (6)
"Yes"	46.1% (8)	23.1% (13)	82.4% (14)	74% (17)

Chi-Square Tests of Independence were performed for EDU and for intervention responses to explore the differences between groups at each time-point. No significant differences were found between responses at EDU ($p > 0.05$), or at post-intervention ($p > 0.05$). However, as displayed in Table 16, the incidence of ASD and SEN participants reporting that they enjoy exercise at school increased. Further binomial tests revealed no significant differences in "Yes" and "No" responses at EDU in the ASD group ($p > 0.05$), but that ASD participants were more likely to say "Yes" at post-intervention ($p < 0.02$). In the SEN group, no significant differences were found between EDU "Yes" and "No" responses, but the SEN group were significantly more likely to answer "Yes" at post-intervention ($p < 0.05$).

Table 16: Responses to "Do you enjoy physical exercise classes in school?" in the CK group

	Education as usual		**Exercise**	
	CK	**AE**	**CK**	**AE**
"No"	43.5% (4)	52.9% (9)	17.4% (4)	35.3% (6)
"Yes"	56.5% (13)	47.1% (8)	82.6% (19)	64.7% (11)

To explore any differences between the Cool Kidz and aerobic exercise groups at EDU and intervention, further Chi-Square Tests of Independence were performed. No significant differences were found between responses at EDU ($p > 0.05$), or at post-intervention ($p > 0.05$). Further binomial tests revealed no significant differences in responses at baseline in the CK group ($p > 0.05$), but that the CK group were more likely to say "Yes" at post-intervention ($p < 0.001$). In the AE group, no significant differences were found at EDU ($p > 0.05$) or intervention ($p > 0.05$).

4.2.4. Discussion

The aim of this study was to determine the effects of a chronic physical exercise programme on the reported stress of a group of school-aged pupils with ASD and other SEN. Forty participants underwent a 12-week programme consisting of 30 exercise sessions over six weeks, and education as usual for six weeks in a counterbalanced design. Participants received moderate intensity SI therapy, or high-intensity aerobic exercise. Measurements of self-reported stress and exercise programme enjoyment were taken after each phase. Importantly, to further build upon Study 1, participants were measured at least 24 hours following their final session, when temporary physical arousal as a result of exercise engagement are likely to have subsided. Results from Study 2 indicate that participating in either forms of exercise intervention leads to a significant overall reduction in reported stress. Overall reports of stress on the SSS were higher following EDU than following exercise. This effect is present in children with ASD and those with other SEN. Overall, children with SEN showed trends in reporting higher levels of stress than those with ASD. When considering types of stress, those which were more loaded onto cognition were reported as more stressful for both groups than those which were more environment-related. However, there were no differential effects of intervention on types of stress, suggesting that whilst physical exercise reduced overall stress, the level at which different types of stress affect participants remained proportional.

These results support the previous findings of Carmeli et al. (2009) and Carraro and Gobbi (2012), who both discovered superior anxiety-related improvements of physical exercise programmes above sedentary control periods. Importantly, contrary to results found for other measures (Levinson & Reid, 1992), the level of exercise intensity does not appear to be an influential factor in the success of exercise programmes. Both groups in the present study reported decreased stress and increased coping abilities, despite one group participating in a lower intensity programme. These findings support the conclusions of Lawlor and Hopker (2001), who found no differential effects of exercise type on affect outcome. This is certainly noteworthy when considering that physiological changes may primarily account for the exercise-stress relationship. That is, perhaps moderate intensity exercise is sufficient enough to promote the neural activity required for reduced feelings of stress. For example, upregulation of emotion-related endorphins and monoamine neurotransmitters that lead to increased positive emotion may be equally present in lower and higher exercise intensity (Lautenschlager et al., 2012; Sarris et al., 2012).

Relevantly, one of the primary justifications for exploring prolonged effects of exercise following Study 1 was to more accurately explore potential mechanisms for this effect. Interestingly, as discussed, some maintain that the core reasons for the stress-exercise relationship in the general population are the effects of distraction (Petruzzello et al., 1991) and novelty (Motl & Gosney, 2007). Importantly, these increases in exercise classes were implemented throughout half of an entire school term, reducing the likelihood that such classes remained novel or distracting to the regular routine. Furthermore, others maintain that an increase in body temperature is a driving factor behind improved reporting of emotion (DeVries, 1981; Craft & Perna, 2004). However, whilst exercise-induced temperature increases return to baseline within 90 minutes, in our study, participants were assessed at least 24 hours following their final session. Therefore, the thermogenic model cannot directly account for the findings of maintained effects outside of this post-exercise window. One explanation that does appear to hold up in the present results is the theory of cognitive dissonance (Festinger, 1957). In the present study, children were required to increase exercise participation as part of their school timetable – a behaviour that many (47.5%) reported as a negative experience at intake. However, this theory maintains that individuals possess a drive to avoid conflicts between attitudes, beliefs and behaviours, leading many to adapt their cognition in line with experiences. Based on this model, it is therefore possible that children adapted their attitudes towards school-based exercise, increasing their enjoyment in daily participation, and in turn improving self-reported affect.

A further psychological explanation that may account for these findings is that of the 'self' – namely, self-concept and self-mastery. Notably, across both groups, increases in reports of exercise class enjoyment occurred, with the most prominent reason being enjoyment of the activities. Since this reasoning was also present in the aerobic exercise group – who engaged in activities that were already an established aspect of their existing classes – this effect cannot be attributed to practicing new skills. Instead, as in Study 1, the structured introduction of individual and group-level praise, motivation and reward may explain improvements in self-perception, in turn increasing exercise class enjoyment, and decreasing self-reported levels of stress. Based on previous research and current practices, interventions that incorporate motivation and contingent rewards are highly successful (Rogers & Lewis, 1999; Koegel, 2000; Koegel, Koegel & McNerney, 2001; Vismara & Lyons, 2007). However, studies to date that have explored the relationship between ASD and response to praise has produced mixed results (Rigsby-Eldredge & McLaughlin, 1992; Kasari, Sigman, Baumgartner & Stipek, 1993; Smith, Greenberg, Seltzer & Hong, 2008). The possibility that the results are related to self-perception through praise and reinforcement certainly requires further investigation, and if validated, may be widely transferable to other aspects of the school day.

Importantly, as in Study 1, this study aimed to control practice effects using a counterbalanced design. This is particularly crucial given that self-reporting emotions may be somewhat positively affected by practice in this population. In order to explore this relationship, session order was analysed to uncover any potential false positives for those participating in exercise intervention second, having already experienced a testing session. This exploration proved interesting in two ways: first, there were no significant effects of session order, suggesting that outcomes were a direct result of the intervention programme, rather than practice. Second, given that there were no superior effects of participating in exercise intervention first, this data suggests a lack of longevity of effects with regards to the stress and exercise. That is, the effects in the group who underwent intervention first were back to baseline levels after six weeks of education as usual. Further research is required to follow up on the long-term outcomes, since this was not an established measure of the current research studies.

In sum, this study built upon the findings of Study 1 by demonstrating a prolonged stress effect of chronic exercise programme participation. Participants with ASD and other SEN reported reduced feelings of stress and increased coping abilities following 6 weeks of increased physical exercise classes throughout the school week, compared to a control period of education as usual. Furthermore, whilst Study 1 evidenced immediate positive effects of high-intensity physical exercise on reported feelings of stress, this study discovered positive effects of both moderate and high-intensity programmes. Previous research suggests numerous explanations for these effects, including self-perception, as well as relevant neural changes and activity. This is the first series of studies to date which explore the impact of school-based physical exercise programmes on reported stress in the ASD population. Whilst further research is required to expand on the sample size and variety of measures, these findings present physical exercise of moderate or high-intensity as a cost-effective coping mechanism for perceived stress in individuals with ASD and other SEN. However, there are certainly limitations to these study, which will be discussed in Chapter 6.

CHAPTER 5: THE EFFECT OF A STRUCTURED EXERCISE INTERVENTION ON THE EXECUTIVE FUNCTION PERFORMANCE OF SCHOOL-AGED CHILDREN WITH AUTISM SPECTRUM DISORDERS AND OTHER SPECIAL EDUCATIONAL NEEDS

5.0.1. Introduction

Along with facilitations in emotional wellbeing as explored in Chapter 4, other research has demonstrated that the benefits of exercise can extend to specific cognitive systems, with improvements to executive functioning (EF) being one such system well-documented among the literature (Brisswalter, Collardeau and Rene, 2002; Hall et al., 2001; Colcombe and Kramer, 2003; Hillman, Snook and Jerone, 2003). EF is considered as the control processes which are responsible for managing a number of cognitive operations, including planning, inhibition, working memory and attention (Salthouse et al., 2003). There is considerable evidence demonstrating the importance of EF in a child's development. Indeed, intact executive functions are frequently associated with increased self-control (Barkley, 2001), task initiation (Gilotty et al., 2002), organisation (Gioia et al., 2000), and flexibility (Kleinhans, Akshoomoff & Delis, 2005).

Given these associations, it is commonly suggested that children who are unable to plan, switch between tasks and inhibit certain behaviours may face great difficulty achieving at school (St Clair-Thompson & Gathercole, 2006). Importantly, such deficits are implicated in the development of both their psychological processes and social behaviours (Morgan & Lilienfeld, 2000). Because of these factors, there has been a plethora of literature exploring the link between EF and clinical disorders such as ADHD and ASD, which often embody deficits in many of these components, including impulse control, attention and organised search (Willcutt et al., 2005; Biederman et al., 2007; Pellicano, 2012). For example, the executive dysfunction theory of autism is one of a number of theories which have been proposed to help explain characteristics of ASD throughout the lifespan, and is particularly useful in accounting for a number of non-social features such as repetitive and obsessive behaviours, rigid interests and perseveration (Turner, 1999; Lopez, Lincoln, Ozonoff & Lai, 2005). Although the primary EF model no longer dominates current causal understandings of autism, deficits in EF remain a highly recognisable and an acknowledged characteristic of this population (Pellicano, 2012). By and large, individuals with ASD display specific impairments in the domain of flexibility, demonstrated by their inflexible responses on the Wisconsin Card Sorting Test (WCST), whereby they continue to perseverate despite experimental feedback which urges the individuals to change their thought processes (Sandson & Albert, 1984). This type of behaviour on the WCST, as well as on other tasks which gauge EF, is often indicative of more generalised behaviours including uninterruptable activities or speech, rigid routines and repetitive motor activities (Liss et al., 2001; South et al., 2007).

Such deficits in EF are often observed in school-based classrooms. For example, a key aspect of EF is inhibitory control, which enables individuals to filter out/suppress irrelevant external stimuli or internal impulses that may otherwise be distracting (Dowsett & Livesey, 2000). Within the school environment, this may include classroom disturbance, such as increased corridor noise or another teacher entering the classroom, or the urge to leave their seat or distract a peer. Deficits in this inhibitory behaviour in the classroom are widely observed in children with ASD (Jahromi, Bryce, & Swanson, 2013). Likewise, another branch of EF is attentional switching – deficits in which make it difficult for an individual to switch from one set of rules or tasks to another. Again, this is particularly evident in those with ASD in the form of rigidity and inflexibility with changes in plans and routines (Liss, Saulnier, Fein & Kinsbourne, 2006).

Given the wide application of EF skills, a number of researchers have focused on assessing the effects of programmes that aim to improve EF performance. Successful programmes in multiple populations include computerised training (Thorell, Lindqvist, Nutley, Bohlin and Klingberg, 2009), martial arts (Lakes and Hoyt, 2004), and aerobic exercise (Tomporowski, 2003; Chang, Labban, Gapin and Etnier, 2012). Indeed, physical activity in the form of aerobic exercise and sport is a frequent aspect of the school curriculum in Western society. Along with the established physical health benefits, physical exercise as an intervention for EF-regulated improvements is of high interest for both financial and relative ease-of-access reasons. In this chapter, the behavioural and physiological evidence for the beneficial effects of physical exercise on EF will be presented. Since the outcomes and mechanisms for single bouts (acute) and longer-term programmes (chronic) are believed to differ, these two camps of research will be discussed separately. Finally, two novel research studies will be presented, which both examine whether or not this relationship exists in children with ASD and SEN, from both a short-term (Study 1) and longer-term (Study 2) engagement perspective.

5.0.1.1. Behavioural Evidence: Acute Physical Exercise

The short term effects of high-intensity physical exercise are well documented. Whilst a large proportion of exercise-based research focuses on physical fitness and long-term programmes, research involving acute bouts of exercise in a variety of experimental designs is gaining momentum. Importantly, as reported in a study by Levinson and Reid (1992), the positive effects of vigorous exercise may only be evident within 90 minutes of engagement, with targeted behaviours returning to baseline after this period. The interest in this field is evidenced by the various reviews and meta-analyses relating specifically to the effects of physical exercise on cognition (Tomporowski, 2003; Brisswalker, Collardeau and René, 2002; Lambourne and Tomporowski, 2010; Chang, Labban, Gapin and Etnier, 2012). Although results vary from no effects (Bard and Fleury, 1978; Cote, Salmela and Papthanasopoloulu, 1992; Cian et al., 2000) to positive facilitations (McGlynn, Laughlin, and Rowe, 1979; Hogervorst, Riedel, Jeukendrup and Jolles, 1996), there is general consensus in the field that acute bouts of exercise do indeed affect cognitive task performance.

Notably, many studies that demonstrate positive results adopt well-controlled randomised designs. Chang and Etnier (2009) randomly assigned 41 healthy adults to receive either 45 minutes of resistance exercise training, or a 45-minute period of quiet reading. Cognition was measured both pre and post each session using the Stroop Test (Stroop, 1935) and Trail Making Test (TMT, Soukup, Ingram, Grady and Schiess, 1998). Analysis of results revealed that physical exercise significantly improved performance on the Stroop test, although not on the TMT. Interestingly, the authors maintain that this differential effect may be due to exercise affecting specific types of executive function. That is, whilst the TMT is loaded onto cognitive flexibility, the Stroop task performance is indicative of inhibition, interference, and speed of processing. The authors conclude that 45 minutes of moderate physical activity is superior above seated reading in improving both higher level processes, such as shifting of habitual responses, as well as lower level cognitive processes, such as speed of processing. However, although promising, transferability of results must be handled with caution, given the laboratory-based and volunteer-reliant nature of this research. Despite these shortfalls, this research supports a wealth of preceding acute exercise studies, including those demonstrating benefits in processing and decision speed (Hogervorst, Riedel, Jeukendrup and Jolles, 1996; Adam, Teeken, Ypelaar, Verstappen & Paas, 1997; Davranche, Burle, Audiffren & Hasbroucq, 2006) and interference (Lichtman & Poser, 1983; Sibley, Etnier & Le Masurier, 2006).

Such effects appear prominent throughout the lifespan. Hogan, Mata and Carstensen (2013) investigated age differences in the impact of acute exercise on cognitive performance. 114 participants aged between 19 and 93 years were randomly assigned to receive either 15 minutes of stationary cycling at moderate intensity, or a seated computer task. Participants were assessed on working memory and state affect before and after their session in a well-controlled pre-post design. In somewhat contrast to previous age-related literature (Whitbourne, Neupert, and Lachman, 2008; Ready, Marquez, & Akerstedt, 2009), the authors found positive effects of a single bout of exercise on affect and cognitive performance, which were independent of age. Response times in the working memory task significantly decreased across all ages, evidencing greater efficiency in working memory performance. However, again, caution must be taken when considering results from studies relying on volunteers who are not blinded to the hypothesis of the study.

As will be discussed in the following section, some research demonstrates both behavioural and physiological effects. A recent study by Drollette et al (2014) explored the impact of a single bout of moderate intensity exercise on cognitive control capacity. With particular emphasis on individual differences in cognition and related treatment responses, Drollette and colleagues measured the event related potentials (ERP) of 40 children completing a modified version of the Eriksen flanker task (Eriksen and Eriksen, 1974) – a measure of EF-loaded inhibitory control. Children had either completed 20 minutes of treadmill walking, or 20 minutes of seated rest. Interestingly, results revealed a greater response in those grouped as low-performers. Whilst high-performers maintained levels of accuracy, latency and P3 amplitude, low-performers improved in their accuracy performance and P3 amplitude as a result of exercise, but not sedentary sitting. Both groups demonstrated shorter P3 latency and smaller N2 amplitude following treadmill walking. Although this study supports previous results of acute exercise (see Hillman, Erickson and Kramer, 2008 and Lang et al., 2010 for further reviews), this finding of individual differences certainly brings a new perspective to the literature.

5.0.1.2. Behavioural Evidence: Chronic Physical Exercise

In addition to acute bouts of intense exercise, the effects of chronic aerobic exercise programmes on cognition have been studied for decades. Whilst the above studies demonstrate the immediate effect of physical exercise on cognitive performance, a key question remains: do these effects uphold after immediate physiological stimulation returns to baseline levels? Chronic exercise studies that contribute to this debate will now be explored, before proposed mechanisms for both types of engagement are outlined.

An early study by Tuckman, Hinkle and Scott (1986) randomly assigned school-aged children to a 12-week running programme or routine physical education classes. The programme was incorporated into the school curriculum and ran three times per week for 30 minutes. At the end of the programme, children in the aerobic running condition demonstrated improvements in cardiorespiratory fitness (physical performance and pulse rate) and creativity, as measured by the Alternate Uses test (Christensen, Guilford, Merrifield and Wilson, 1960). Since such flexible thinking and resulting creativity has been linked with executive functioning (Delis et al, 2007), the results certainly have implications for this field (Best, 2010). These findings were confirmed in a later study by Hinkle at al. (1993). Furthermore, a 15-week randomised controlled trial by Davis et al (2007) found that children in a high-dose aerobic exercise group (40 minutes of exercise daily) significantly increased their performance in the Cognitive Assessment System (CAS, Naglieri and Das, 1997), but only for the sub-scale mapped onto EF, Planning. Interestingly, such effects are not limited to observation and task performance. A small group of the same participants were reported to display increased dose-response prefrontal cortex activity as well as decreased posterior parietal cortex whilst performing cognitive tasks in a later fMRI study (Davis et al, 2011). The authors discuss that this relationship may be due to the direct neural stimulation of movement, or simply to the engagement in tasks requiring goal-directed mental processes. It is important to consider, however, that as participants only consisted of overweight children, the results may be limited to this population who may be more reactive to exercise.

The above findings are by no means restricted to the younger population, with studies reproducing these outcomes throughout the lifespan. Indeed, research into the cognitive abilities of older adults has provided strong support for this hypothesis. Kramer et al. (1999) investigated the effects of aerobic versus anaerobic exercise on a number of cognitive processes in a group of older adults. Participants were subject to a battery of tests prior to and during the six-month exercise program. The results revealed selective benefits in EF, but not other cognitive processes, following an aerobic exercise regime. More recently, Bixby et al. (2007) assessed a group of healthy older adults on their levels of physical activity and EF, as indexed by the Yale Physical Activity Survey (YPAS) and Stroop Color and Word Test respectively. After controlling for both intelligence and age, their findings revealed that YPAS scores were able to positively explain a significant amount of variance in scores of EF, but were unrelated to nonexecutive performance. A review by Colcombe, Kramer, McAuley, Erickson and Scalf (2004) confirmed these findings. In addition to the chronic intervention literature, Colcombe and colleagues summarised that longitudinal and cross-sectional research into the effects of general cardiovascular fitness (CVF) demonstrate positive associations between physical fitness and cognitive performance in older adults. That is, increased levels of exercise is considered a significant factor in increased cognitive task scores. Further, a meta-analyses of studies between 1966 and 2001 investigating the impact of exercise intervention and task performance found that not only was aerobic exercise producing larger improvements in cognitive tasks, but that the largest improvements were found in tasks loaded onto EF (Colcombe and Kramer, 2003).

Evidently, there is strong experimental evidence supporting both acute and chronic forms of exercise engagement across the lifespan. Research to date suggests that the benefits of physical exercise may be loaded onto specific forms of cognition, which is certainly an important factor to consider when examining studies will null results. Although sampling bias is a great concern across the current literature, with the majority of studies either targeting specific groups or relying on willing volunteers, there are certainly strong grounds to consider physical exercise as an effective intervention. Further behavioural evidence for clinical populations will be discussed in Study 1 and Study 2.

5.0.1.3. Mechanisms Underpinning EF and Physical Exercise

Importantly, the mechanisms through which executive functions are positively affected by physical exercise are thought to vary depending on the type of exercise and length of the programme. A compelling review by Best (2010) provides insight into this phenomenon, citing three potential pathways through which this effect exists; (I) physiological changes in the brain as a result of aerobic exercise, (II) cognitive demands commanded by goal-directed, engaging tasks such as group exercise, and (III) complex movements requiring cognitive engagement.

5.0.1.3.1. Physiological explanations

Physiological changes are frequently explored as a direct result of cardiovascular exercise in relation to cognitive and behavioural outcomes (Sibley and Etnier, 2003). Despite numerous behavioural studies, the physiological foundations of both acute and chronic exercise are yet to be agreed (Giles et al., 2014). Importantly, however, although it is believed that the cardiovascular system promotes neural changes that underpin EF, there appear to be clear pathway differences between acute and chronic aerobic exercise (Best, 2010).

Acute physical exercise

As reviewed by Meeusen, Piacentini, & De Meirleir (2001), acute, high-intensity aerobic exercise is purported to lead to immediate neurochemical changes that concurrently enhance skill acquisition related to the priming of the central nervous system (CNS; Best, 2010). Evidence for the involvement of the CNS in cognitive abilities can be drawn from models of those with Attention Deficit Hyperactive Disorder (ADHD), which attribute their cognitive and attentional deficits to underarousal of the CNS (Satterfield and Cantwell, 1974). Furthermore, Winter et al. (2007) reported a superior effect of high-intensity acute exercise on learning when compared to longer, more moderate exercise engagement. Interestingly, in their study, increased brain-derived neurotrophic factor (BDNF) and monoamine levels were noted for acute exercise, with such neural changes predicting material retention. Another proposal by Giles and colleagues (2014) maintains that high-intensity (70-75% $VO_{2\ max}$) acute exercise stimulates changes in the prefrontal cortex (PFC); an area associated with cognition. Specifically, their research led them to conclude that a single bout of intensive physical exercise promotes oxygenated and deoxygenated haemoglobin in the PFC. These findings are supported by the work of Ide, Horn and Secher (1999), who found enhanced total blood flow in frontal brain regions following acute exercise. However, although Ando, Kokubu, Yamada and Kimura (2011) discovered decreases in cerebral oxygenation at 80% $VO_{2\ max}$, Giles et al (2014) found continued increases at 84% $VO_{2\ max}$, evidencing the need for further investigation into this effect.

Further beneficial effects of acute exercise on underlying neural processes responsible for EF have been demonstrated by Hillman, Snook and Jerome (2003). In their study, a total of 20 undergraduates participated in 30 minutes of treadmill activity, with an intensity self-set by the participants to a level of 'somewhat hard to hard' on the perceived exertion scale (Borg, 1970). Results of P3 amplitude and latency were measured during a subsequent Erikson Flanker task (Eriksen and Eriksen, 1974), and compared with baseline performance in which the task was not preceded by exercise. As predicted, the findings indicated that acute cardiovascular exercise led to increased P3 amplitude, which has been theorised to reflect the allocation of mental resources which underlie executive control during cognitive tasks. Specifically, heightened P3 amplitude is reported to suggest improvements in executive control processes through improved working memory resources (Donchin and Coles, 1988) and general arousal (Magnié et al., 2000). Equally noteworthy, however, was the varied effects on P3 latency between task conditions, which the authors note evidences the need to investigate the effects of task difficulty on post-exercise performance. No effects of exercise were found for behavioural responses of the task, with both accuracy and reaction times demonstrating no relationship to cardiovascular exercise. This may be due to participants self-selecting intensity, which could have led to limited arousal. However, a later study by Hillman et al (2009) demonstrated improvements in the inhibitory performance and larger P3 amplitude of typically developing children (mean age: 9.5 years) following 20 minutes of treadmill activity controlled at 60% of their Max HR.

Finally, acute physical exercise has been associated with the upregulation of dopamine and noradrenaline as a result of acute motor cortex arousal. In addition to the evidenced improvements in mood a discussed in Chapter 4, Holzapfel et al. (2015) highlighted the positive effects of these increased neurotransmitters on motor function and EF (Brown, Marsden, Quinn & Wyke, 1984; McMorris & Hale, 2015). Given this overlap, it is unsurprising that physical exercise concurrently produces multiple behavioural and performance improvements. Likewise, although acute physical exercise primarily produces more temporary arousal than that of chronic engagement mechanisms, the positive effects of both types of engagement are logically sound, given the positive life-long link between motor and cognitive development in the cerebellum (Ridler et al., 2006). Most notably, concurrent activation of the cerebellum and PFC occurs whilst an individual engages in planning, sequencing and monitoring – skills required for both motor and EF task performance (Westendorp et al., 2011; Holzapfel et al., 2015).

Chronic physical exercise

Despite these neural overlaps, research into the topic of physical exercise intervention is divided as to whether the mechanisms behind the beneficial effects are limited to a person's overall physical fitness and/or chronic engagement in exercise, and do not extend to include temporary bouts of acute exercise. Indeed, using a similar methodology to Hillman, Snook and Jerome (2003), Stroth et al. (2009) found that high-fit participants yielded greater anticipatory task preparation and monitoring of appropriate responses, indexed by event-related potentials indices of CNV amplitudes and decreased N2 amplitudes, respectively. However, acute aerobic exercise was not related to either of these indices of EF. Furthermore, a recent study by Davis et al. (2011) demonstrated that daily bouts of exercise (20 or 40 minutes per day) spanning a three-month period resulted in dose-response benefits to both EF and mathematical ability in a group of overweight children. Importantly, these benefits were mirrored by brain activation changes in the prefrontal and posterior parietal cortices which the researchers credited solely to the exercise program.

Nevertheless, of the studies that investigate the neural correlates of chronic engagement in exercise, a large majority focus on non-human primates and rodents (van Praag, Christie, Sejnowski, & Gage, 1999; van Praag, Shubert, Zhao, & Gage, 2005; Cotman, Berchtold, & Christie, 2007). However, promising findings in such samples have pinpointed exercise-related hippocampal neurogenesis (van Praag et al, 1999), synaptic plasticity (Dishman et al, 2006; Schinder and Poo, 2000) and various growth factor upregulations, such as brain-derived neurotrophic factor (BDNF) – an important modulator in synaptic transmission which is particularly exercise-dependent (Churchill et al, 2002; Holmes, 2006). Pereira et al (2007) found evidence of increased cerebral blood volume in the hippocampus of both mice and humans following a controlled period of exercise, whilst a study with older adults by Colcombe et al (2006) found that six months of aerobic exercise led to increased grey and white matter volume, particularly in the frontal brain regions associated with EF. Relevantly, with reference to the ageing brain, Fabel and Kempermanns' (2008) conclusion draws upon the above theories collectively. Acknowledging the vast literature evidencing the link between chronic exercise and hippocampal neurogenesis, the authors suggest that robust neurogenesis in such brain areas may be a result of non-specific neural activation coupled with novel and engaging experiences.

In sum, strong experimental evidence suggests both behavioural and neural changes as a result of physical exercise. Chronic exercise programmes in which participants are exposed to repeat bouts of exercise over varying periods of time tend to primarily evidence hippocampal neurogenisis in both human and non-human rodents, whilst single, acute bouts of exercise appear to stimulate immediate arousal of the CNS, increase frontal blood flow, upregulation of certain neurotransmitters, and increase P3 amplitude. These changes and activations, in turn, appear to directly impact upon EF performance in tasks which involve structured thinking and appropriate responding. Evidence relating more specifically to the effects of acute and chronic exercise will be discussed in Study 1 and Study 2 respectively.

5.0.1.3.2. Psychological Explanations

Executive Function Components of Tasks

So far, it is apparent that physical exercise positively impacts the neural underpinnings of cognitive performance following both single and chronic bouts of cardiovascular activity. Interestingly, a finding from the meta-analysis of Sibley and Etnier (2003) revealed no significant effects of exercise type, suggesting that "any type of physical activity will ultimately benefit cognitive performance" (Sibley and Etnier, 2003, page 252). This assumption is particularly intriguing, given that not all physical activity promotes intense cardiovascular activity, such as structured walking and low-intensity yoga. Acknowledging the effect of low-moderate intensity activities, some researchers maintain that the key moderator in this exercise-cognition relationship is not pure physiological arousal, but the presence of cognitive demands in the exercise task (Sibley and Etnier, 2003; Best, 2010). Thus, whilst this theory does not dispute physiological changes, emphasis is placed on the demand, rather than intensity, of the exercise. This extends to both the behavioural demands, as well as complex motor input required in order to participate in exercise.

Cognitively-demanding physical activities

Studies involving exercise that requires planning, inhibition and the implementation of patterns and sequences demonstrate facilitations in EF. For example, Best (2010) speculates that certain physical activities, such as basketball and football, demand and engage effective cognitive processes to enable the creating, monitoring and modifying of plans in order to cooperate with teammates and succeed in such tasks. This, in turn, may positively facilitate EF performance thereafter (Best, 2010). Importantly, since the intensity of engagement is certainly less of a focus in this assumption, research has investigated the efficacy of low-intensity physical activities that challenge EF, such as self-control and disciplined focus.

One such example is that of traditional martial arts, which requires strong EF-mediate skills in order to succeed. Lakes and Hoyt (2004) explored the effect of a three-month school-based Tae Kwon Do programme on the self-regulation of 193 children. Groups were randomly assigned at the classroom level to receive either martial arts training, or continue as usual in their standard physical education lessons. Following three months of intervention for two- to- three periods per week, only children receiving traditional martial arts training significantly improved in their EF-mediated self-regulation. Similarly, Rangan, Nagendra and Bhat (2008) hypothesised that 11-to-13- year old children receiving yoga modules as part of their education programme would demonstrate increased planning ability when compared to education as usual controls. As predicted, performance on the Tower of London task significantly improved in most subtests in both groups, with a superior effect in the yoga-based group. In both studies, the researchers attribute the effects to the more cognition-focused nature of the target programmes. Evidence for the effects of such self-control oriented programmes is also presented by others, including Trulson (1986), Chan, Sze, Siu, Lau and Cheung (2013), and in an EF intervention review by Diamond and Lee (2011).

Although these self-control and inhibition-focused activities are somewhat motor-based, some researchers have argued that the EF effects of exercise are in fact not attributable to motor activity at all, but instead the pure engagement and practising of EF skills. This notion is backed by evidence demonstrating that other forms of complex cognitive demands, such as targeted computer games, also facilitate improvements in EF. For example, Thorell, Lindqvist, Nutley, Bohlin and Klingberg (2009) examined the effects of a computerised training programme either targeting working memory (WM) or inhibitory control (IC) in a group of pre-schoolers. Following a total of 25 sessions, it was discovered that those in the WM experimental group significantly improved on both trained and non-trained cognitive tasks, including spatial and verbal WM, as well as in their attention. However, when compared to the control group, children in the IC condition displayed no additional improvements on WM and attention tasks, although improvements were found on two trained tasks. The authors report that these differential effects may be due to variations in the psychological and physiological processes that underpin these constructs. The duration of such effects are also unclear, although research by Klingberg et al (2005) demonstrates effects at 3 months post-training in ADHD participants.

5.0.1.3.3. Complex motor movements

Nevertheless, numerous findings do suggest a superior effect of motor activity, and with the additional physical health benefits that coincide with this intervention, it remains of high value in the field. Given the supporting research for each argument, it is not unwarranted to value both theories of physiological arousal and EF-loaded exercise. Indeed, combining the theories relating to the cardiovascular and cognitive demands of targeted exercises, others maintain that the specific cognitive demands of complex motor planning may play a key role. That is, complex movements stimulate the neural underpinnings of EF. In particular, Diamond (2000) highlighted the close neural connection and simultaneous activation of the dorsolateral prefrontal cortex (a crucial area for EF) with the cerebellum (a crucial area for complex coordinated movement, including planning and prediction). Both of which, she suggests, are therefore importantly coordinated for EF and motor processes, and signify a link between these two different functions. Importantly, motor processes which are deemed complex, such as bimanual coordination tasks (using each hand to complete different tasks or instructions simultaneously) are implicated in this effect, with more simple processes such as walking dismissed as weaker in this EF relationship (Diamond, 2009). As cited by Best (2010), this specific link is evidenced in the brains of animals, whereby neural growth in selected regions such as the cerebellum and hippocampus is greater as a result of complex movements than that of simpler, repetitive movements (see Carey, Bhatt and Nagpal, 2005 for review).

5.0.1.4. Temporal Properties

Perhaps most efficiently summarised by Best (2010), one reason for the differences in findings is that there are indeed differences in mechanisms depending on the type and length of exercise engagement. As discussed, an overwhelming majority of the literature demonstrates a positive relationship between physical exercise and cognitive performance, but numerous factors may influence the extent or type of benefits. These include (A) whether the exercise is intense enough to stimulate immediate physiological changes that underpin EF, and (B) whether the exercise activity possesses cognitive and/or motor demands. However, a further variant exists which may influence observed effects: the temporal properties of the exercise session and proceeding cognitive tasks.

In recent years, a growing number of meta-analyses have provided insight into temporal effects, including time of testing, as well as time of day. For example, a meta-analysis by Lambourne and Tomporowski (2010) reviewed 40 within-subjects studies specifically assessing single bouts of exercise on cognitive task performance. Interestingly, findings revealed that whilst exercise had a negative impact on cognition during physical exercise sessions, cognitive performance proceeded to improve following completion of exercise when compared to baseline. However, using broader criteria to account for various proposed mechanisms, Chang Labban, Gapin and Etnier (2012) reviewed 79 studies and discovered a small positive effect regardless of whether cognitive testing took place during or following exercise. Interestingly, the authors did find significant effects of the time of exercise, with morning exercise contributing the largest effects. Exercise performed in the afternoon or evening led to statistically insignificant results.

Another interesting factor is that of the delay between completion of exercise and engagement in cognitive tasks. Although chronic programmes should be robust to temporal delays in testing (Klingberg et al., 2005), results of acute exercise sessions may be highly sensitive and confined to the period of immediate physiological arousal. Indeed, whilst Lambourne and Tomporowski (2010) found detrimental performance during exercise, delaying cognitive testing for too long may lead to missed effects. Of the previous studies which report testing delays, Chang et al. (2012) found the strongest positive effects between 11-20 minutes following exercise. This effect was mitigated after 20 minutes. Others propose that the most effective post-exercise window for improved performance is 90 minutes (Hori et al., 1978), whilst some report a less linear effect, with target behaviour initially statistically negative compared to zero (Chang et al., 2012), and improving after 30 minutes following exercise completion (Tate & Petruzzello, 1995; Raglin & Wilson, 1996).

5.0.1.5. Summary

To summarise, physical exercise is evidenced to directly impact the physiological underpinnings of EF, such as upregulation of growth factors, as well as immediate increased cerebral blood flow. However, conflict still exists within the literature regarding whether exercise type is a factor in the success of physical activity facilitating EF. Whilst some emphasise the importance of moderate- to- high-intensity aerobic activity, others place more weight on the cognitive demands of physical exercise, particularly those that stimulate cognition through strategic and adaptive engagement. Finally, complex motor movements are reported to activate key cognitive-mediating areas of the brain such as the prefrontal cortex, as well as promote hippocampal and cerebellum adaptations. Importantly, previous results may not be as conflicting as it appears- rather, the temporal and physical properties of the exercise programmes vary too greatly to warrant comparison. Indeed, Tomporowski and Ellis (1986) maintained that contradictions and variations amongst the literature are due to researchers failing to use theory-based approaches in their exercise and cognition research. Reports of the time lapse between exercising and task performance is critical for future studies.

Of the research investigating the beneficial effects of exercise in young persons with ASD, surprisingly, very few studies have explored the direct impact on EF. However, as found in the meta-analysis of Sibley and Etnier (2003), physical exercise benefits the cognition of children with learning disabilities to the same degree as the effects found in the typically developing population. In fact, some maintain that those with lower EF control benefit most from such intervention (Flook et al., 2010). Although some studies find a differential effect (Bluechardt, Wiener and Shephard, 1995), the authors note the underpowered nature of many of these designs. Furthermore, they proceed to note physical exercise as an "important component of their education programme" (Sibley and Etnier, 2003, page 252). Thus, the common aim of the current studies is to investigate the effects of an ABA-based physical exercise programme on measures of cognitive function in school-aged children with ASD. Study 1 explores the short-term relationship, with participants receiving one week of intervention and tested within 90 minutes of their final exercise session. The aim of Study 2 was primarily to assess whether these effects hold up over a longer period of time (6 weeks), with cognitive tasks administered at least one full day after the final exercise session. A secondary aim was to investigate the differential effects of low and high-intensity programmes.

CHAPTER 5A: STUDY 1 - THE EFFECTS OF AN ACUTE EXERCISE INTERVENTION ON EF PERFORMANCE IN SCHOOL-AGED CHILDREN WITH ASD AND OTHER SEN

5.1.1. Introduction

Research into the effects of a single bout of exercise and EF is prominent in multiple populations. Given the relative ease of accessibility, the interest in physical exercise as an effective and immediate tool for improving target behaviours in clinical populations is rapidly expanding. Indeed, the benefits of physical exercise to EF have been demonstrated in stroke patients (Ploughman, McCarthy, Bosse, Sullivan & Corbett, 2008) as well as individuals with Down Syndrome (DS; Chen, Ringenback, Crews, Kulinna & Amazeen, 2014), ADHD (Pontifex, Saliba, Raine, Picchietti & Hillman, 2013) and ASD (Anderson-Hanley, Tureck & Schneiderman, 2011). This section will outline recent research studies that explore the EF-related effects of an acute bout of exercise in clinical populations, before presenting current shortfalls and novel empirical research to expand this literature.

A recent series of studies have demonstrated significant improvements in the cognitive performance of young adults with DS following acute exercise (Ringenbach, Albert, Chen, & Alberts, 2014; Chen, Ringenbach, & Albert, 2014). In one such study, participants were assigned to receive either a single bout of moderate intensity treadmill activity for 20 minutes, or a session of sedentary video watching (Chen et al., 2014). Performance on the Knock-Tap test - a measure of inhibition - significantly improved following treadmill activity when compared to the sedentary control condition. The authors propose that this positive effect may be due to enhanced neural transmission or increased levels of arousal. Interestingly, improvements in attention shifting and choice-response time were found to be insignificant, again highlighting the importance of task selection. Nevertheless, the promising findings give rise to future studies which can provide more insight using larger sample sizes in this population, as very few studies to date have focused on this relationship in DS individuals (Chen et al., 2014).

A further study by Pontifex and colleagues (2013) supported these results, demonstrating ERP effects in children with ADHD. The authors aimed to examine the effects of a single 20-minute bout of moderate intensity physical exercise on academic performance, behaviour and neurocognition. Pontifex and colleagues adopted a within-subjects design with 20 children aged between 8- and 10- years old. Participants were assessed on their inhibitory control through the Eriksen flanker task, their ERP responses, and their academic performance through the Wide Range Achievement Test - 3rd edition (WRAT3; Wilkinson, 1993). Results following moderate treadmill activity at 65-75% of their maximum heart rate were compared to their performance following a seated reading. As predicted, ADHD participants and their matched controls demonstrated improvements in inhibitory control, action monitoring processes, behaviour regulation, and overall processing speed. Such gains were implicated by the authors in academic performance, given the improved reading comprehension and arithmetic ability. Along with general gains, the ADHD-specific improvements in regulatory processes give rise to physical exercise as a non-pharmateutical treatment for this clinical population.

Most recently, Piepmeier et al. (2015) explored the impact of a single 30-minute bout of physical exercise on the cognitive performance of children with ADHD. A group of participants with an average age of 10 years and 7 months were tested on two occasions, in a counterbalanced order – once following aerobic exercise, and once following sedentary video-watching. Measures included perceived exertion, the Stroop Task, the Tower of London task, and the Trail Making Test. Intriguingly, findings revealed specific gains in cognition, with positive improvements to speed of processing and inhibitory control on the Stroop task, but not in planning, speed of processing and set shifting on the TMT. Although greater improvements were expected in the ADHD group given their pre-existing deficits, both the ADHD and typically-developing control groups improved equally. The authors highlight the support of this finding in the previous study of Pontifex et al. (2013).

Relevantly, demonstrating these results in the ASD population, a recent two-part pilot study by Anderson-Hanley, Tureck and Schneiderman (2011) investigated the influence of acute exergaming on cognition and repetitive behaviours in children and adolescents with autism. Hypothesising that a combination of mental and physical challenge would most effectively enhance such performance, Anderson-Hanley and colleagues provided participants with exercise-based virtual reality games. Children were assigned to one of two exergaming conditions (Dance Dance Revolution [DDR] or cyber cycling) and a control condition (video watching), with measures of EF and repetitive behaviour collected before and after each 20-minute session. In both studies, it was discovered that incidence of repetitive behaviour significantly decreased after each exergaming session when compared to the control condition. EF was found to significantly increase, but only on the Digit Span Backward measure (Anderson-Hanley, Tureck and Schneiderman, 2011). However, whilst these preliminary results are promising, further research is required to validate such findings. For example, the exertion and intensity of the exergaming sessions were not directly measured and, as acknowledged by the authors, this potential inconsistency could account for the variance in findings (Kern et al, 1984; Elliott et al., 1994). Furthermore, it is important to ascertain exactly which aspect of the experimental condition (i.e. mental or physical challenge) most strongly predicted such outcomes (see Thorell et al., 2009).

5.1.1.1. Study aims and hypothesis

As evidenced, there are a limited amount of large-scale studies that explore physical exercise as an intervention tool for ASD. The aims of this study are therefore to build upon the existing literature, addressing some of the methodological weaknesses and gaps in previous studies. This study will assess the short term effects of acute aerobic exercise on EF performance (inhibition and switching) in the young ASD and SEN populations. As highlighted in Chapter 4, it aims to successfully integrate the physical exercise programme into the regular school week using an ABA-style framework, utilising behavioural techniques to facilitate engagement of participants to whom physical exercise is a non-preferred/avoided activity. Given previous findings in multiple populations to date, it is predicted that engaging in high-intensity physical activity (70-80% Max HR) will lead to accuracy and latency improvements in EF tasks in both ASD and SEN participants.

5.1.2. Methods

5.1.2.1. Participants

These children are the same sample as reported in Chapter 4, Study 1. Refer to Chapter 4, Study 1 for participant and design information.

5.1.2.2. Measures

Executive Function

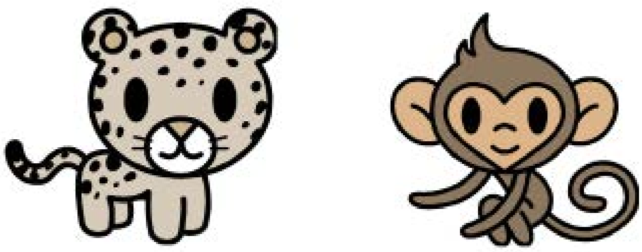
Three executive function tasks loaded onto either inhibitory control (the Pictures task), attentional shifting (the Arrows task), or both (the Eyes task), were administered on the final day of the intervention and EDU periods. These tasks were chosen due to their ability to collect rich executive function data in short, time-effective tasks whilst being particularly motivating to participants due to their computerised, game-like nature (Moore & Calvert, 2000).

Each participant completed the batch of tasks within 90 minutes of completing their exercise or academic session. The tasks chosen for this study have previously been tested on children from 4- to 7- years of age (Burns, Riggs & Beck, 2012), and were therefore deemed suitable for older children with developmental delay. In all tasks, participants were provided with two response buttons, and had to either press the button on the same side as the stimulus ('congruent' trials), or on the opposite side ('incongruent' trials), depending on the rule or direction of each trial. Inhibitory control was measured by comparing 'congruent' and 'incongruent' trial performance. Attentional shifting was measured by comparing performance in trials that used the same rule as a preceding trial, and those that had 'switched' to another rule. As described by Burns, Riggs & Beck (2012), greater performance is often found for trials that maintain consistent rules ('non-switch), as well as trials in which the stimuli are presented on the side of the correct response ('congruent').

Pictures Task

Participants are presented with two pictorial stimuli (see Figure 7) and are instructed to press the green button when the cat appears, and the blue button when the monkey appears. The green button is on the right-hand side of the participant, whilst the blue button is on the left-hand side. The stimuli appear in a random order on either the right or left side of the screen. At no point are these stimuli presented simultaneously. For half of the trials, the correct response is the button on the same side as the stimulus (congruent trials). The other half require participants to inhibit the natural instinct to press the button on the same side, and respond with the opposite button (incongruent trials). For these incongruent trials, the level of inhibitory control is evidenced by effects on accuracy and latency relative to congruent trial performance.

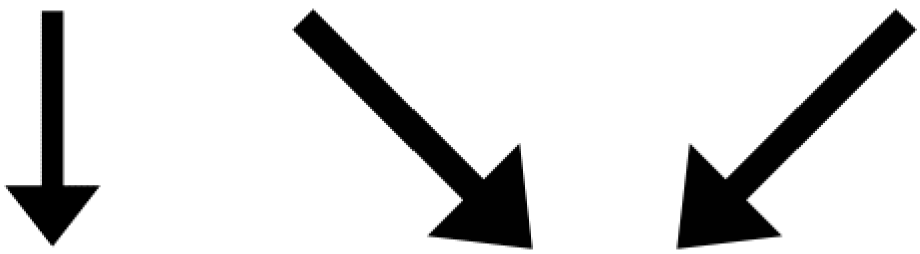
Figure 7: The Pictures Task stimuli



Arrows Task

Participants are presented with a series of directional arrows (one per trial, see Figure 8) and instructed to press the button that each arrow is pointing towards. As in the Pictures task, the response buttons are located on the right-hand and left-hand side of the participant. Two types of presentation require the participant to press the button to the right of the screen (either pointing straight downwards on the right-hand side, or pointing diagonally across at a 45-degree angle from the left-hand side). The other two types require a response with the left-hand button (either pointing straight downwards on the left-hand side, or located on the right-hand side pointing diagonally across at a 45-degree angle). Although this task has the spatial inhibitory demands of the pictures task, this use of directional arrows places additional shifting demands on participants. Importantly, participants are required to process two separate pieces of information in order to elicit the correct response: visual properties of the presented stimuli (direction of arrow), as well as converting spatial information to the rules of the task ("press the button on the opposite side" and "press the button on the same side").

Figure 8: The Arrows Task stimuli



Eyes task

Adapted from Davidson et al.'s (2006) Arrows task, the eyes task has both spatial inhibitory and shifting demands. Although this task repeats the measures of the Pictures and Arrows task, it was used as an additional measure for two main reasons: (1) given that each task was time-limited (2-3 minutes), including a third task ensured that participants were comfortable and that performance was an accurate reflection of ability, and, importantly (2) the additional social aspect of the stimuli provided an important variant that may alone affect performance in children with social conditions (Crawford et al., 2016), and may therefore be more sensitive to intervention-related change (Pan, 2010). This task involves faces as the target stimulus (see Figure 9), with directional eye gaze at either a 45-degree angle towards the opposite side of the screen, or straight downwards. As with the other tasks, the faces appear on either the left-hand or right-hand side of the screen, and participants are instructed to press the button that the eyes are gazing towards. Congruent trials are those which require pressing the button on the same side as the target stimuli (eyes looking downwards), whereas incongruent trials require that participants press the button on the opposite side to the target stimuli (eyes looking diagonally across the screen).

Figure 9: The Eyes Task stimuli

Heart Rate

As described in Chapter 4, Study 1.

Exercise Enjoyment Survey

As described in Chapter 4, Study 1.

5.1.2.3. Procedure

As described in Chapter 4, Study 1.

Executive Function tasks

Research assistants who were blind to each child's experimental status settled each participant into a quiet testing room within the school grounds. To reduce the chance of distraction, the use of classrooms was avoided. Instead, small, distraction-free rooms that participants were familiar with were deemed most appropriate. Participants were seated 50 centimetres in front of a 15" monitor which displayed each task. All participants were reminded that they can stop at any point if they felt uncomfortable, and that nobody else could see their scores. The tasks were run in the same order for each participant (the Pictures task, followed by the Arrows task, and ending with the Eyes task). Participants were read standardised instructions (see Appendix C) before beginning the practice trials. When participants had correctly responded to at least 3 of the 4 block practice trials, the task began. At this point, it was explained to participants that they would not be given feedback for the remaining trials, and to complete the task as quickly and as accurately as possible. Trials were presented for a maximum of 2500ms, with 500ms between them.

5.1.3. Results

5.1.3.1. Statistical Analysis

Data was cleaned by removing responses with a latency below 350ms, as in a previous study using the same tasks (Burns, Riggs & Beck, 2012), and excluding responses greater than 3 standard deviations from each mean. Responses were then sorted and analysed by congruency (congruent versus incongruent trials) for the Pictures and Eyes tasks, and then by switching (non-switch versus switch trials) for the Arrows and Eyes tasks. Latency was analysed only for correct trials, with incorrect or non-responded trials removed for all RT analysis. The statistical package SPSS 17.0 for Windows was used for all analyses. Descriptive statistics were obtained and approximate normal distributions were confirmed through calculating skewness and kurtosis for each variable independently, as well as the Levene's test for homogeneity of variance. Furthermore, no significant differences in distribution were found between datasets. For all analyses, the family-wise alpha level was set at 0.05. Statistical procedures for congruence and switching effects are outlined in the relevant sections below. Results are displayed in Figures 10 to 17.

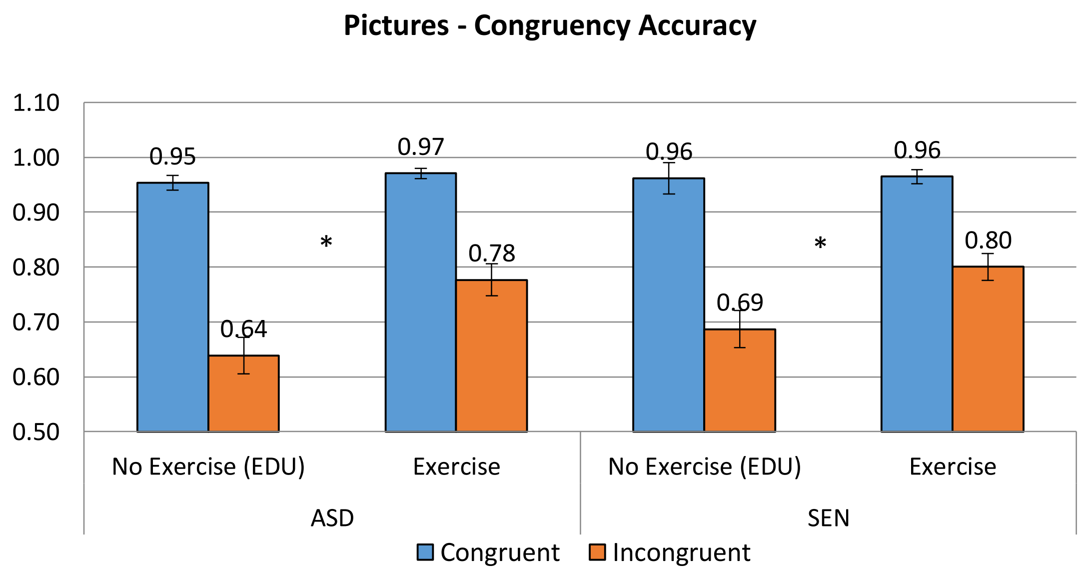
5.1.3.2. Pictures Task

Congruence

A 2 (congruency: congruent versus incongruent trials) x 2 (intervention phase: EDU versus exercise) mixed factorial ANOVA was used, with participant group (ASD and SEN) as the between-subject factor.

Accuracy

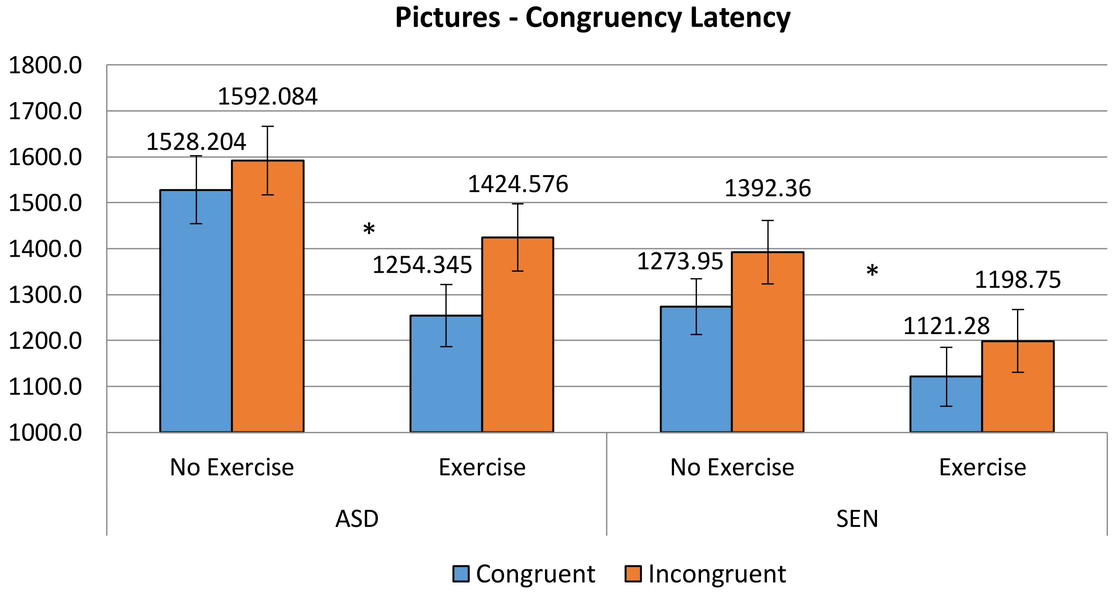
Figure 10: Accuracy of congruent and incongruent trial responses for the Pictures task



A significant main effect of congruence was found, $F(1,51) = 126.979$, $p < 0.001$, $\eta_p^2 = .713$. On average, participants performed more accurately during congruent trials than incongruent trials during both EDU and intervention phases. A main effect of intervention phase (EDU versus exercise) was also found, $F(1,51) = 25.535$, $p < 0.001$, $\eta_p^2 = .334$, as well as a significant interaction between congruence and intervention phase, $F(1,51) = 12.236$, $p < 0.001$, $\eta_p^2 = .193$. There was no significant effect of diagnostic group.

Latency

Figure 11: Latency of congruent and incongruent trial responses for the Pictures task



A significant main effect of congruence was found, $F(1,51) = 29.402$, $p < 0.001$, $\eta_p^2 = .366$, whereby congruent trials were responded to with greater speed than incongruent trials at both EDU and exercise phases. A main effect of intervention phase was also revealed, $F(1,51) = 13.517$, $p = 0.001$, $\eta_p^2 = .210$, with participants demonstrating increased latency during their EDU phase when compared to their exercise phase. Finally, a significant congruence x intervention phase x group interaction was found, $F(1, 51) = 6.632$, $p < 0.05$, $\eta_p^2 = .115$.

As a result of this complex interaction, further analysis into separate group results was conducted. A 2 (congruency: congruent versus incongruent trials) x 2 (intervention phase: EDU versus exercise) repeated measures ANOVA was performed independently for the ASD group, as well as the SEN group. *ASD:* Main effects of congruence, $F(1,23) = 9.963$, $p < 0.05$, $\eta_p^2 = .302$, and intervention phase, $F(1,23) = 7.011$, $p < 0.05$, $\eta_p^2 = .234$, were found. A congruence x intervention phase interaction rested on significance, $F(1,23) = 4.122$, $p = 0.05$, $\eta_p^2 = .152$. That is, the relative RT difference between congruent and incongruent trials were greater at post-intervention than at EDU for this group. *SEN:* Main effects of congruence, $F(1,28) = 25.062$, $p < 0.001$, $\eta_p^2 = .472$, and intervention phase, $F(1,28) = 6.278$, $p < 0.05$, $\eta_p^2 = .183$, were found. There was no significant congruence x intervention phase interaction as in the ASD group.

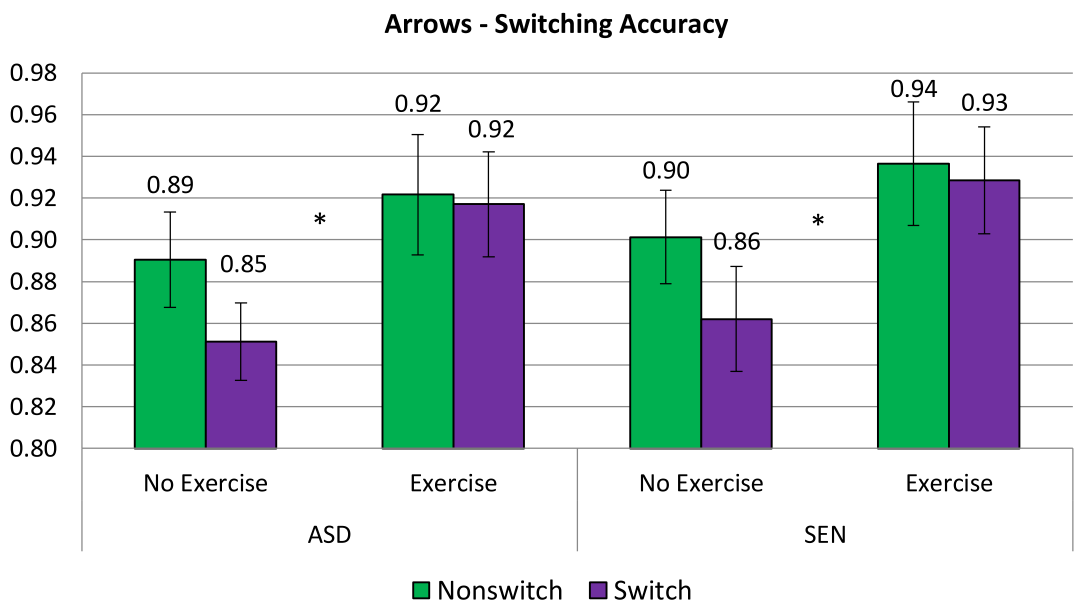
5.1.3.3. Arrows Task

Switching

A 2x2 mixed design ANOVA was used, with switching (non-switch, switch) and intervention phase (EDU, exercise) as the within-subjects factors, and participant group (ASD and SEN) as the between-subject factor.

Accuracy

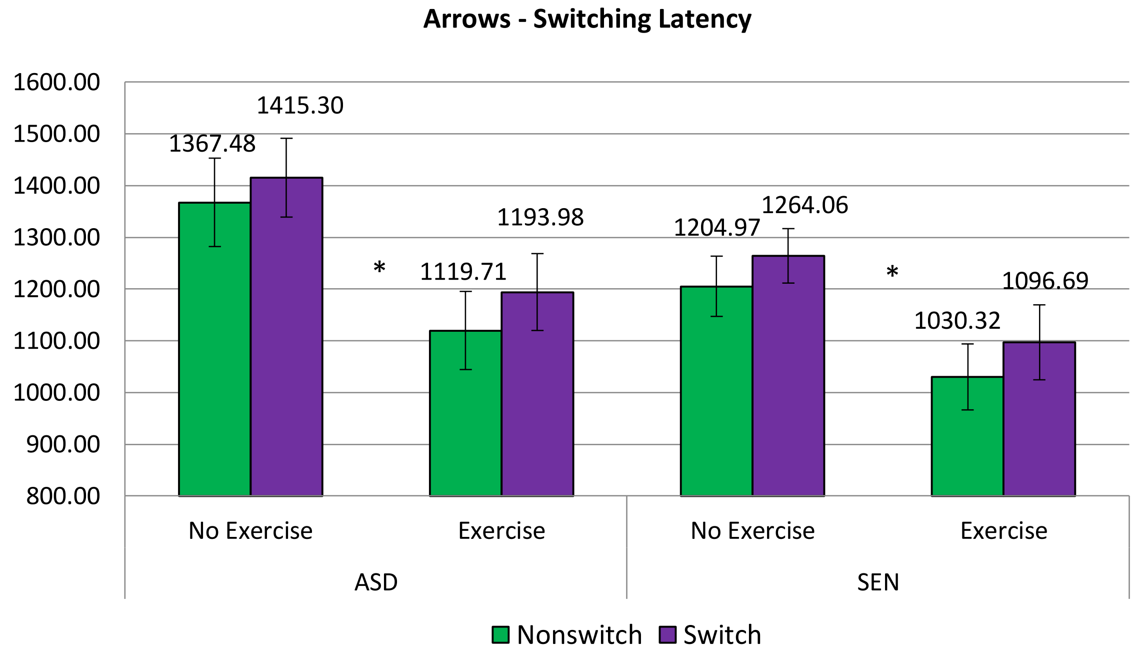
Figure 12: Accuracy of non-switch and switch trial responses for the Arrows task



Significant main effects of switching, $F(1, 50) = 6.036$, $p < 0.05$, $\eta_p^2 = .108$, and intervention phase, $F(1,50) = 4.610$, $p < 0.05$, $\eta_p^2 = .084$, were found. Participants demonstrated increased accuracy in non-switch trials when compared to switch trials in both EDU and intervention phases. Participants also demonstrated more accurate responses across all conditions in their exercise phase when compared to their EDU phase. There were no effects of group, and no significant switching x intervention phase x group interaction.

Latency

Figure 13: Latency of non-switch and switch trial responses for the Arrows task



Significant main effects of switching, $F(1,50) = 15.079$, $p < 0.001$, $\eta_p^2 = .232$, and intervention phase, $F(1,50) = 17.196$, $p < 0.001$, $\eta_p^2 = .256$ were found. Participants demonstrated decreased latency in non-switch trials when compared to switch in both EDU and intervention phases. Participants also demonstrated faster responses across all conditions in their exercise phase when compared to their EDU phase. No other main effects or interactions were found to be significant.

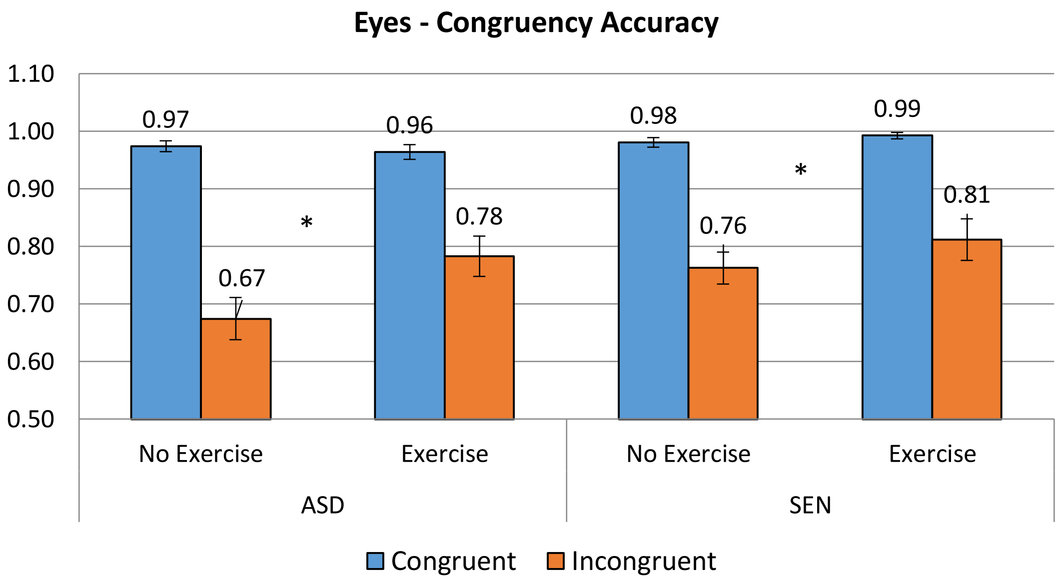
5.1.3.4. Eyes Task

Congruence

A 2x2 mixed design ANOVA was used, with congruence (congruent/incongruent) and intervention phase (EDU, exercise) as the within-subjects factors, and participant group (ASD/SEN) as the between-subject factor.

Accuracy

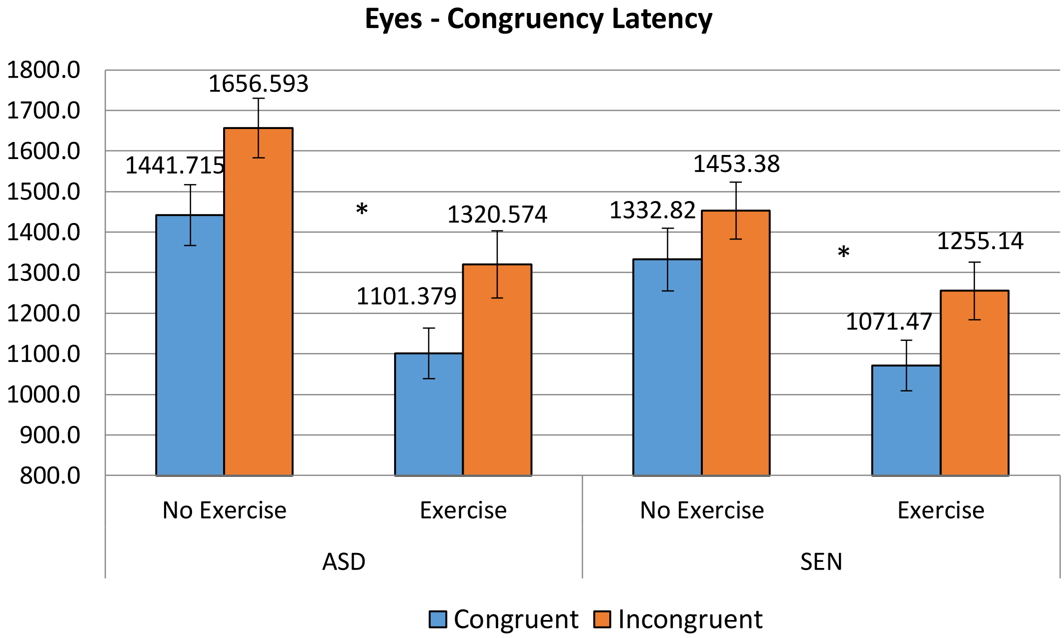
Figure 14: Accuracy of congruent and incongruent trial responses for the Eyes task



Significant main effects of congruence, $F(1, 47) = 112.664$, $p < 0.001$, $\eta_p^2 = .706$, and intervention phase, $F(1,47) = 7.402$, $p < 0.05$, $\eta_p^2 = .136$, were found. On average, participants performed more accurately during congruent trials than incongruent trials during both EDU and intervention phases. Participants also demonstrated more accurate responses across all conditions in their exercise phase when compared to their EDU phase. A significant congruence x intervention phase interaction, $F(1,47) = 6.717$, $p < 0.05$, $\eta_p^2 = .125$, demonstrated smaller congruence effects (congruence < incongruence) after exercise when compared to EDU.

Latency

Figure 15: Latency of congruent and incongruent trial responses for the Eyes task



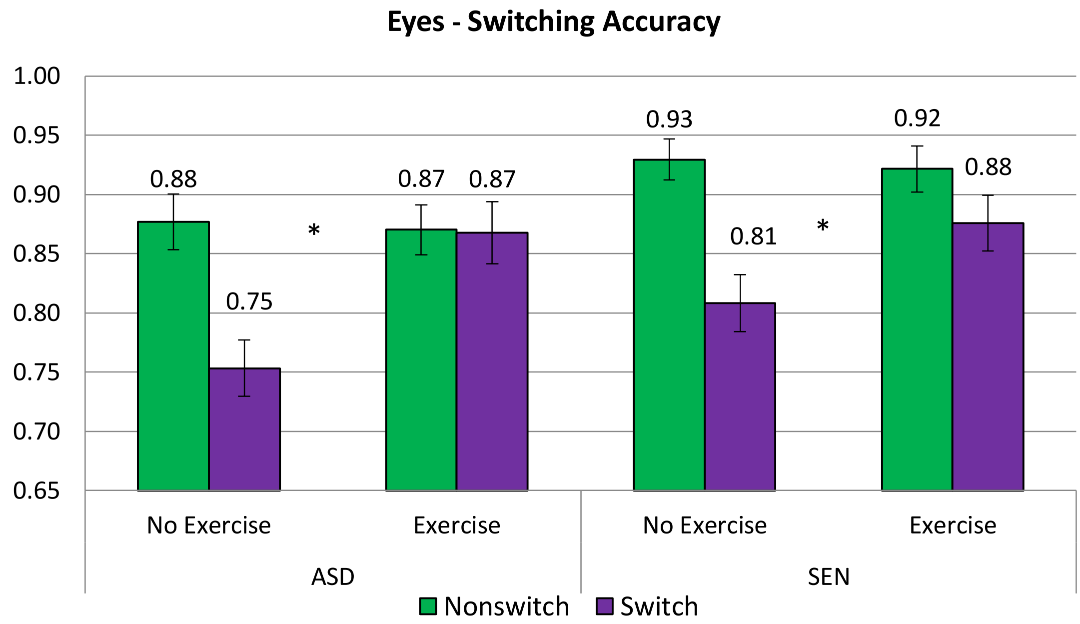
Significant main effects of congruence, $F(1, 47) = 49.599$, $p < 0.001$, $\eta_p^2 = .513$, and intervention phase, $F(1, 47) = 34.422$, $p < 0.001$, $\eta_p^2 = .423$ were found. Participant groups demonstrated decreased latency in congruent trials when compared to incongruent trials in both EDU and intervention phases. Participants also demonstrated faster responses across all conditions in their exercise phase when compared to their EDU phase. No other main effects or interactions were found.

Switching

A 2x2 mixed design ANOVA was used, with switching (non-switch, switch) and intervention phase (EDU, exercise) as the within-subjects factors, and participant group (ASD/SEN) as the between-subject factor.

Accuracy

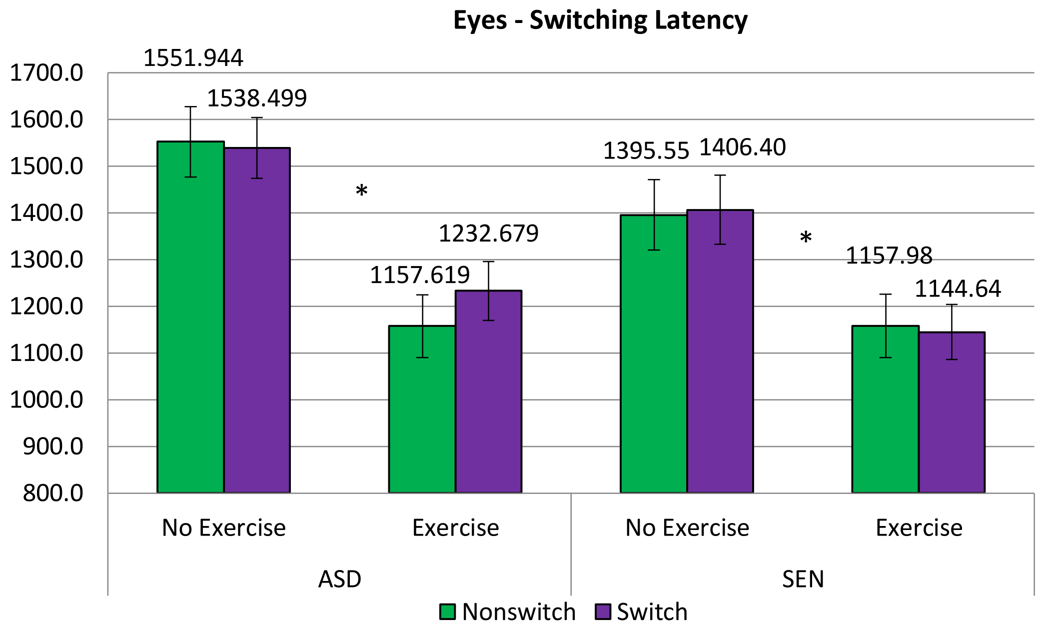
Figure 16: Accuracy of non-switch and switch trial responses for the Eyes task



Significant main effects of switching, $F(1, 50) = 28.161$, $p < 0.001$, $\eta_p^2 = .375$, and intervention phase, $F(1,50) = 7.111$, $p < 0.05$, $\eta_p^2 = .131$, were found. Participants demonstrated increased accuracy in non-switch trials when compared to switch trials in both EDU and intervention phases. A significant switching x intervention phase interaction, $F(1,50)= 15.336$, $p < 0.001$, $\eta_p^2 = .246$, demonstrated smaller switching effects (switching < non-switch) after exercise when compared to EDU.

Latency

Figure 17: Latency of non-switch and switch trial responses for the Eyes task



A significant main effect of intervention phase, $F(1,50) = 38.435$, $p < 0.001$, $\eta_p^2 = .450$, was found. Overall, participants demonstrated decreased latency in both switch and non-switch trials during the exercise phase when compared to EDU. No other main effects or interactions were found.

5.1.4. Discussion

The primary aim of this study was to explore the effects of an acute exercise programme on the cognitive performance of ASD and SEN participants. Fifty-three children (24 ASD) between the ages of 12- and 14- years old participated in one week of exercise classes implemented during the school day, and one week of education as usual in a counterbalanced order. Children were measured on their inhibitory control and attention shifting performance at the end of each phase using computer-based tasks loaded onto each skill. Inhibitory control was assessed through each participant's ability to perform as accurately and rapidly on incongruent trials compared to congruent trials. Similarly, attention shifting performance was assessed through each participant's ability to perform as accurately and rapidly on switch trials as on non-switch trials. Based on previous recommendations for acute aerobic exercise, participants were tested within 90 minutes following their final exercise session. As outlined in Chapter 4, a secondary aim was to successfully incorporate the physical exercise programme into the school week, using ABA-based methods to increase motivation and engagement. It was predicted that there would be a significant effect of physical exercise on task performance, with accuracy and latency on higher EF-loaded trials (incongruent and switch) reduced in comparison to non-EF trials (congruent and non-switch) during exercise weeks when compared to education as usual phases.

Overall, it was found that both ASD and SEN participants performed more accurately and rapidly on congruent and non-switch trials when compared to the EF-loaded incongruent and switch trials respectively. This was an expected result, evidencing the efficacy of the tasks as a measurement of EF performance. Furthermore, for every task, participants were more accurate and rapid in their responses across all trial types during exercise weeks when compared to education as usual, evidencing a general performance effect of acute exercise. Importantly, for the Pictures Eyes tasks, congruence and switching effects were found to be smaller following exercise. That is, the relative difference in accuracy between easier (congruent and non-switch) and more cognitively demanding (incongruent and switch) trials was reduced. This demonstrates a clear effect of exercise intervention on EF improvements. Importantly, due to the counterbalancing in this design, such effects are highly unlikely to be a result of practice.

These findings support the previous work of numerous researchers in multiple populations. For example, Chang and Etnier (2009) discovered comparable performance improvements in healthy adults on the Stroop test – a measure of inhibition, interference and speed of processing – following one acute session of exercise training when compared to a quiet rest control condition. Similarly, Hogan, Mata and Carstensen (2013) found cognitive improvements across age groups following just 15 minutes of exercise, whilst Drollette (2014) discovered a superior effect of acute exercise on improved accuracy on the Eriksen flanker task in participants classed as low-performers. Furthermore, previous researchers have discovered supporting results of acute exercise in similar populations to those assessed in the present study (Pontifex et al., 2013; Chen et al., 2014; Piepmeier et al., 2015). Most relevantly, Anderson-Hanley, Tureck & Schneiderman (2011) found improved EF performance on the Digit Span Backward task in children with ASD following one 20-minute exercise session when compared to a video watching control condition. The present study builds upon this previous literature, extending the findings to a school-based programme that utilises behavioural techniques to engage both ASD and other SEN pupils.

Interestingly, however, for the Pictures task, diagnosis (ASD versus SEN) mediated this relationship when considering response latency. Upon investigation, it was found that although the overall response times of children with ASD improved during exercise weeks, the difference between trial RT performances increased. Specifically, an increased difference between congruent and incongruent trials was discovered during post-intervention when compared to EDU phases, contrary to expectations. This suggests that the EF inhibition interference effect still persists in the ASD group to some extent in this task, despite improved overall EF improvements in both speed and accuracy. These results therefore appear to evidence a speed-accuracy trade off in the ASD group only. That is, whilst overall interference is reduced, it is at a cost in relative fluidity on incongruent trials. Finally, as discussed in Chapter 4, the physical exercise programme was successfully integrated into the school week, with the majority of children actively engaging and maintaining target heart rates.

Nevertheless, with the exception of the Pictures task, there were no other superior effects of diagnostic group on intervention-driven EF effects, with both ASD and SEN groups performing similarly throughout. These findings suggest that despite the reportedly lower EF abilities of ASD individuals compared to other populations (Sandson & Albert, 1984), the EF effects of exercise are not particularly heightened in ASD participants, and may apply equally to others with similar developmental delays and conditions. This interpretation supports the findings of Drollette et al. (2014), who discovered similar effects across a group categorised as 'low-performers'. Indeed, both the ASD and other SEN participants of the current study achieved lower verbal and non-verbal ability scores than is predicted for their age group, placing them within the same category as those of Drollette and colleagues' research. However, whilst the present within-subjects study aimed to highlight differences and similarities in diagnosis-driven effects, we did not design the research to directly compare these groups. Further research is required to more directly compare the effects across multiple developmental, behavioural and clinical conditions. Nevertheless, given the variety of primary and secondary needs present in many SEN classrooms, the current findings highlight the efficiency of physical exercise as an intervention for diverse groups of school-aged children. As discussed in Chapter 2, this should be the standard for future research, with emphasis placed on less strict criteria for inclusion.

Although this study did not measure physiological changes as a result of physical exercise, previous research suggests several mechanisms that may account for these EF effects. According to Meeusen, Piacentini, & De Meirleir (2001), acute, high-intensity aerobic exercise leads to immediate neurochemical changes that concurrently enhance skill acquisition related to the priming of the CNS. Another proposal by Giles and colleagues (2014) maintains that high-intensity acute exercise promotes oxygenated and deoxygenated haemoglobin in the PFC; an area associated with cognition. Similarly, Ide, Horn and Secher (1999) report enhanced total blood flow in frontal brain regions following acute exercise. Winter et al. (2007) purport increased BDNF and monoamine levels following acute exercise, with such neural changes predicting material retention. Hillman, Snook and Jerome (2003) maintain that acute cardiovascular exercise increases P3 amplitude, which has been theorised to reflect the allocation of mental resources that underlie executive control during cognitive tasks. Specifically, heightened P3 amplitude is reported to suggest improvements in executive control processes through improved working memory resources (Donchin and Coles, 1988) and general arousal (Magnié et al., 2000).

Aside from pure physiological accounts, others attribute such facilitations in EF to the engagement in cognitively-loaded physical exercise (Lakes & Hoyt, 2004) and complex motor movements (Diamond, 2000). Unlike many physiological explanations, these theories do not specifically associate the effects with the level of exercise intensity, but instead to the components of the exercise session. The participants of the present study engaged in exercise tasks that involved a level of planning and implementations of patterns and sequences, such as aerobic routines and strategic movement in team games. As reviewed by Best (2010), the results of the present study may therefore be attributable to this relevant EF engagement prior to the EF-loaded computer tasks. However, research into this field is often based on chronic programmes, making it less clear whether these effects apply to single bouts of cognitively-engaging physical exercise (Best, 2010). Further research is therefore required to explore these effects in more detail, adopting the same well-controlled design for longer-term engagement in physical exercise. Importantly, to investigate whether the effects are solely due to temporary physiological arousal in associated neural regions, EF measures should be conducted outside of the 90-minute window in which exercise-induced neural and cardiovascular arousal is most prominent. Given the success of the current study in extending the acute exercise literature to ASD and other SEN children, another experimental study will be conducted to address this further gap in the literature. Suggestions for additional future research, as well as wider implications and limitations of the present study will be discussed in Chapter 6.

In sum, this study aimed to extend the physical exercise and EF literature by conducting a school-based within-subjects study implemented within the school environment. Children with ASD and other SEN were assessed on their inhibitory and attention shifting performance following an acute period of physical exercise, and a period of education as usual. Results revealed superior EF effects of acute exercise participation within 90 minutes of engagement. Both ASD and SEN children improved in their overall speed of responding, as well as inhibitory and shifting trial accuracy and latency when compared to non-EF trials following exercise. Children were engaged in the programme using ABA-based methods to enhance motivation, including verbal and tangible contingent reinforcement. Previous research suggests that the mechanisms behind this effect may be due to temporary physiological arousal of the CNS and PFC, and/or increased P3 amplitude. Other explanations include the engagement in EF-loaded exercise tasks, facilitating improvements in proceeding EF assessments. Further research is now required to investigate whether these effects remain after exercise-induced physiological arousal returns to baseline levels.

CHAPTER 5B: STUDY 2 - THE EFFECTS OF A CHRONIC EXERCISE INTERVENTION IN SCHOOL-AGED CHILDREN WITH ASD AND OTHER SEN

5.2.1. Introduction

The benefits of chronic physical exercise are of great clinical interest. With the evidence building that assignment to exercise programmes produces multiple physical, emotional and cognitive gains, physical exercise is increasingly being considered as a cost-effective alternative for wellbeing (Salmon, 2001). Importantly, when considering clinical implications, it is crucial to take into account the variance in motor abilities across individuals with multiple conditions. Whilst some may withstand high-intensity exercise for short periods, others may more realistically achieve low-moderate activity over longer periods of time.

One such example of those with barriers to physical exercise is that of stroke patients. Kluding, Tseng and Billinger (2011) conducted a pilot study into the effects of a 12-week aerobic and strengthening exercise programme on the executive functions of stroke patients. In their pre-post study, nine chronic stroke patients completed three one-hour sessions per week. Assessments of executive functioning were conducted at baseline and following the exercise programme, which included the Digit Span Backwards (DB) task, and the Flanker task (Eriksen and Eriksen, 1974). Results revealed small improvements in working memory performance on the DB task when compared to baseline, and a significant association between improved aerobic fitness and performance on the Flanker task. However, along with the small sample size, the authors note the difficulties in recruitment and retention, given the level of ability and commitment required in order to complete participation. Although only two of eleven subjects withdrew, this discussion is certainly noteworthy when considering wider application. Indeed, Salmon (2001) reported up to a 50% attrition rate as soon as three months in the general population following initial engagement in an exercise programme.

As previously discussed, an interesting debate exists regarding the type of physical exercise that is most beneficial. An early study of three participants with ASD by Kern, Koegel and Dunlap (1984) found that mild exercise, such as ball games, had very little influence on stereotyped behaviours. Physical activities of high-intensity such as running, however, successfully reduced target behaviours. These results were later supported by Levinson and Reid (1992). More recently, Coe et al. (2006) found that academic performance was moderated by vigorous, not moderate, physical activity. Similarly, Kramer and Erickson's (2007) review of the literature led them to conclude that there is little to suggest that physical fitness correlates with cognitive change, so there must be another component that accounts for these improvements. Diamond (2015) suggests that this component may be a cognitive aspect of the exercise, such as thinking and engaging in social group behaviour.

Indeed, the core of this conflict highlighted by Diamond is demonstrated in the study of Pesce, Crova, Cereatti, Casella and Bellucci (2009), which compared two forms of physical exercise – group games versus independent circuit training - both controlled at equal intensities. Following each session, participants completed an EF-loaded word list-learning and recall task. Results revealed that whilst both groups demonstrated improvements in general memory performance, only those who participated in the group exercise condition improved on their immediate recall performance when compared to baseline. Pesce et al.

(2009) speculated that this group-specific cognitive activation may be a result of greater need for social interaction and strategic motor skills when compared to independent circuit training. Perhaps most relevant, however, is the recent study by Chan, Sze, Siu, Lau and Cheung (2013). Attempting to investigate the impact of a 4-week motor-based programme in children with autism, the authors randomly assigned participants into a traditional mind-body exercise target group, or a control group receiving Progressive Muscle Relaxation (PMR). The target exercise, Nei Yang Gong, had previously demonstrated positive behavioural and cognitive improvements in children with autism and Asperger's disorder (Chan, Sze & Shi, 2008; Chan et al., 2011), and involves structured movements that rely on self-control. As predicted, results revealed greater self-control and reductions in ASD symptomology following the target structured exercise. Children in the target condition also displayed significant increased EEG activity in the anterior cingulate cortex – an area greatly involved in inhibitory control (Botvinick, Cohen & Carter, 2004).

Despite this positive improvement found in children with autism, very few studies have attempted to explore the cognitive effects of prolonged and increased physical exercise in this group. In one somewhat similar study that investigated ASD participants, Pan (2010) explored social behaviours following a 10-week aquatic exercise programme. Sixteen children with an average age of 7 years of age participated in a 10-week water exercise swimming programme (WESP), and 10 weeks of their typical activity, in a counterbalanced order. Most relevant to cognition, children were found to more successfully inhibit impulses to behave antisocially, including managing their aggression and disruptive actions following the WESP period when compared to no-treatment control periods. Interestingly, along with motor skill mastery, the authors speculated that these results may have been attributable to participants creating a bond and receiving positive feedback from their instructor, which in turn increased feelings of competence, mastery and motivation. Furthermore, they maintained that witnessing other children behaving positively and receiving reinforcement may encourage their own behaviour. Indeed, other researchers have investigated swimming-based exercises with ASD participants and found similar results, including reductions in stereotypic movements (Yilmaz et al., 2004) and improved motor skills (Huettig & Darden-Melton, 2004).

Such success in the ASD population with lower-intensity exercise again highlights the intensity debate with reference to physical activity. As discussed, high-intensity exercise is not accessible to all individuals, especially many of those with ASD who possess characteristically heightened tactile sensitivity, often making exercise-induced temperature changes unpleasant (Kern et al., 2006). When considering prolonged engagement in physical exercise, it is perhaps more clinically applicable to investigate the effects of moderate intensity exercises that possess structure and promote complex movements. For this reason, the current study aims to explore the effects of a lower-intensity sensory integration programme that requires children to follow specific instructions and engage in physical activities loaded onto core motor skills. Furthermore, whilst research to date is promising, the majority of studies exploring chronic exercise and cognitive performance are limited to certain populations, small samples, and possess varying explanations relating to mechanisms. There is, however, general consensus that chronic exercise programmes positively impact upon physical and mental health.

5.2.1.2. Study aims and hypothesis

Given the relatively few studies in developmentally delayed populations, the aim of this study is to investigate the effects of a 12-week programme on the EF performance of children with ASD and other SEN. Each child will spend six weeks (30 sessions) participating in daily exercise classes throughout the school week, and six weeks in education as usual. Groups will be counterbalanced and randomly assigned at the classroom level. As an exploratory factor, approximately one half of participants will complete a structured physical exercise programme based on the theories of Sensory Integration. The other will complete a moderate- to high-intensity exercise programme, similar to that of Study 1. Importantly, no child will take part in any testing within 90 minutes of their final session.

The questions of particular interest in this study are whether cognitive performance continues to show improvements outside of the 90-minute post-exercise window, and whether any noteworthy differences occur between the intervention groups. It is predicted that the similar effects demonstrated between the ASD and SEN groups in Study 1 will uphold in the longer-term exercise programme. However, due to one group difference found in the Pictures task, diagnostic group will be considered in analysis. Since this study requires children to participate in a chronic exercise programme, it is hypothesised that there will be similar effects of both high and low intensity exercise.

5.2.2. Methods

5.2.2.1. Participants

These children are the same sample reported in Chapter 4, Study 2. Refer to Chapter 4, Study 2 for participant information.

5.2.2.2. Measures

Refer to Study 1 in this chapter for the experimental measures.

5.2.2.3. Procedure

The procedure for EF testing is identical to that of Study 1, although children were tested at least 24 hours following their final session. Refer to Chapter 4, Study 2 for exercise programme procedure for both groups.

5.2.3. Results

5.2.3.1. Statistical Analysis

The same procedure as described in Chapter 4, Study 2 was used for this analysis. Descriptive figures for the Pictures, Arrows and Eyes tasks are presented in Figures 18 to 25.

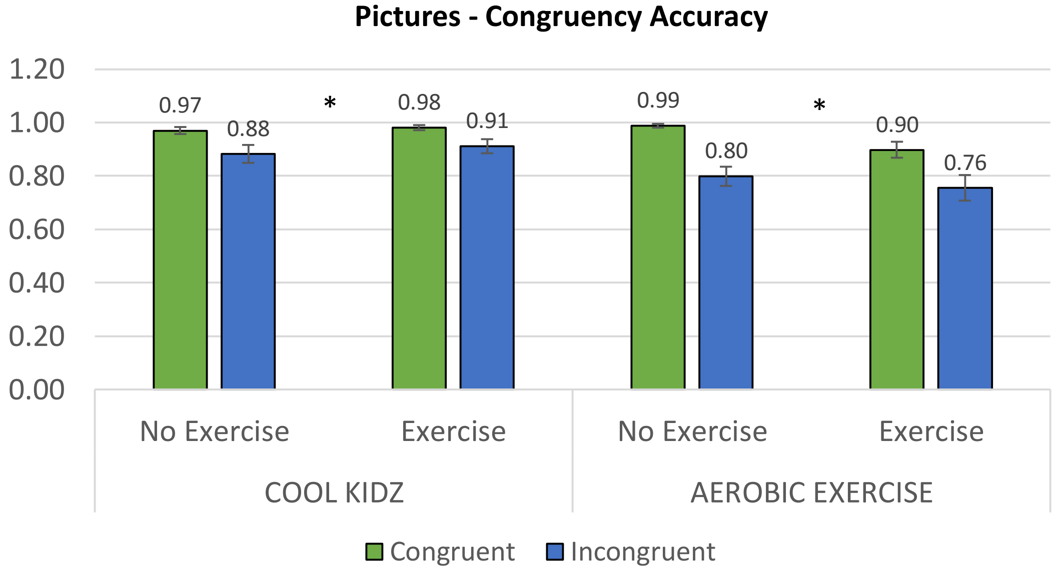
5.2.3.2. Pictures Task

Congruency

A 2 (congruency: congruent versus incongruent trials) x 2 (intervention phase: EDU versus exercise) mixed factorial ANOVA was used, with participant group (ASD and SEN) as the between-subject factor to investigate any group-level differences. No significant effects of group on congruency or intervention phase were found ($p > 0.05$), so analysis continued with both diagnostic groups combined.

Accuracy

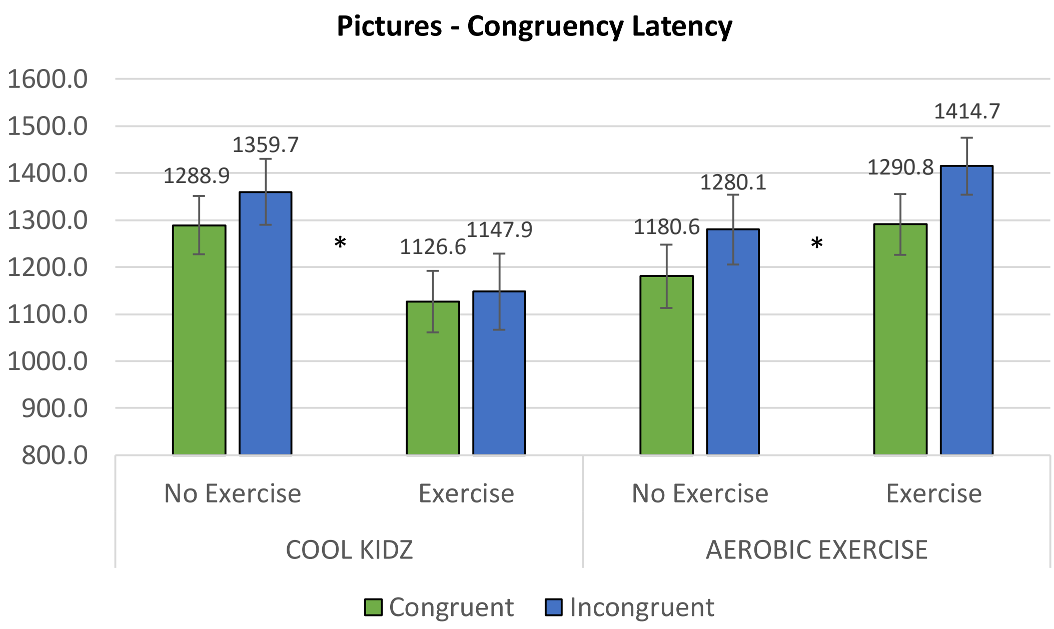
Figure 18: Accuracy of congruent and non-congruent trial responses for the Pictures task



A significant main effect of congruence was found, $F(1,37) = 35.986$, $p < 0.001$, $\eta_p^2 = .493$. On average, participants performed more accurately during congruent trials than incongruent trials during both EDU and intervention phases. A significant interaction between congruence and intervention type was found, $F(1,37) = 4.390$, $p < 0.05$, $\eta_p^2 = .106$, as well as a significant interaction between intervention phase and intervention type, $F(1,37) = 6.090$, $p < 0.05$, $\eta_p^2 = .141$. Only participants in the Cool Kidz condition performed more rapidly across all trial conditions during the post-exercise phase when compared to EDU. No other main effects or interactions were found.

Latency

Figure 19: Latency of congruent and non-congruent trial responses for the Pictures task



A significant main effect of switching was found, $F(1,37) = 13.244$, $p = 0.001$, $\eta_p^2 = .264$. On average, participants performed more rapidly during congruent trials than incongruent trials during both EDU and intervention phases. A significant interaction between intervention phase and intervention type was also found, $F(1,37) = 8.911$, $p < 0.05$, $\eta_p^2 = .828$. No other main effects or interactions were found.

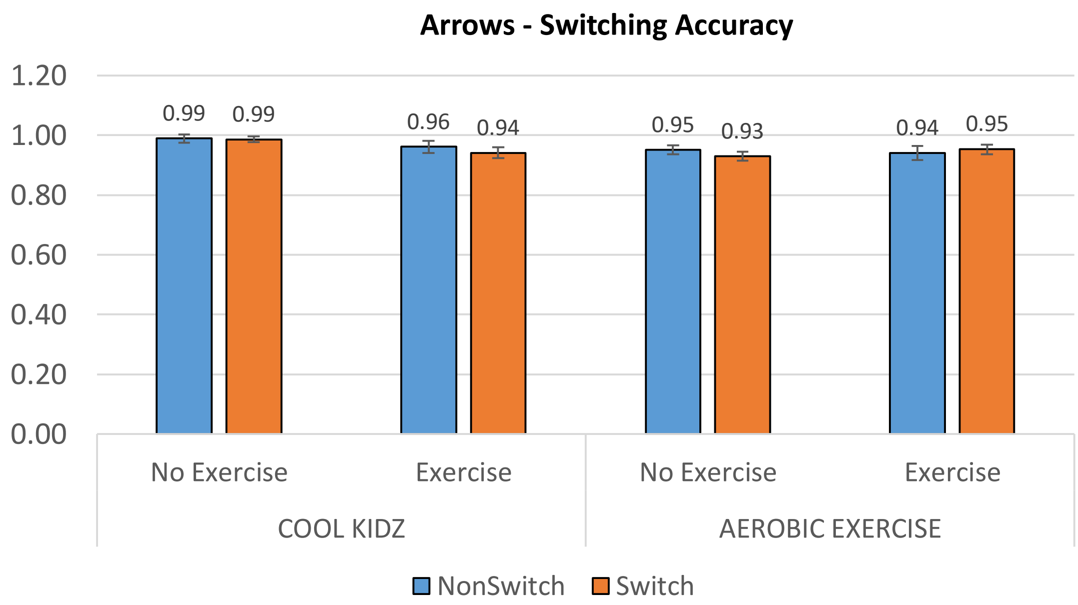
5.2.3.3. Arrows Task

Switching

A 2 (switching: non-switch versus switch trials) x 2 (intervention phase: EDU versus exercise) mixed factorial ANOVA was used, with participant group (ASD and SEN) as the between-subject factor to investigate any group-level differences. No significant effects of group on switching or intervention phase were found ($p > 0.05$), so analysis continued with both diagnostic groups combined.

Accuracy

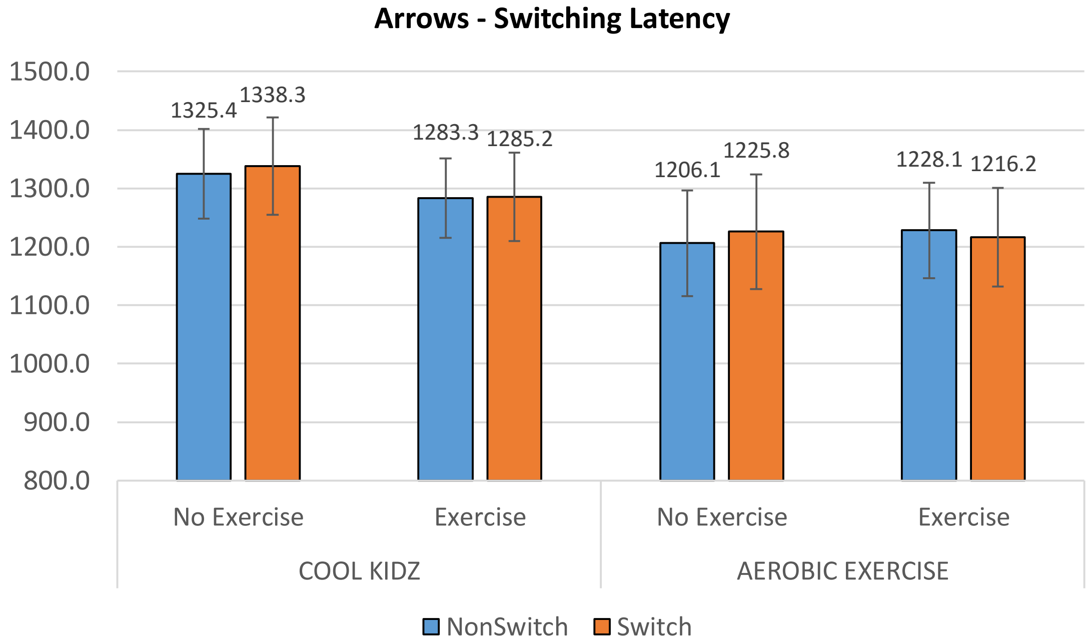
Figure 20: Accuracy of non-switch and switch trial responses for the Arrows task



No significant main effects or interactions were found ($p > 0.05$).

Latency

Figure 21: Latency of non-switch and switch trial responses for the Arrows task



No significant main effects or interactions were found ($p > 0.05$).

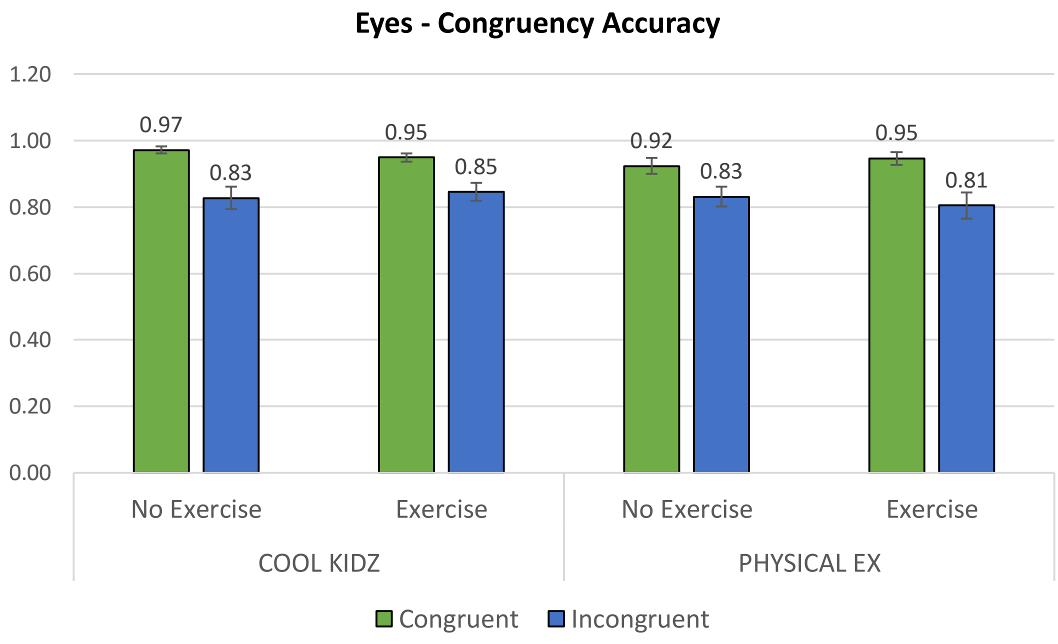
5.2.3.4. Eyes Task

Congruence

A 2 (congruency: congruent versus incongruent trials) x 2 (intervention phase: EDU versus exercise) mixed factorial ANOVA was used, with participant group (ASD and SEN) as the between-subject factor to investigate any group-level differences. No significant effects of group on congruency or intervention phase were found ($p > 0.05$), so analysis continued with both diagnostic groups combined.

Accuracy

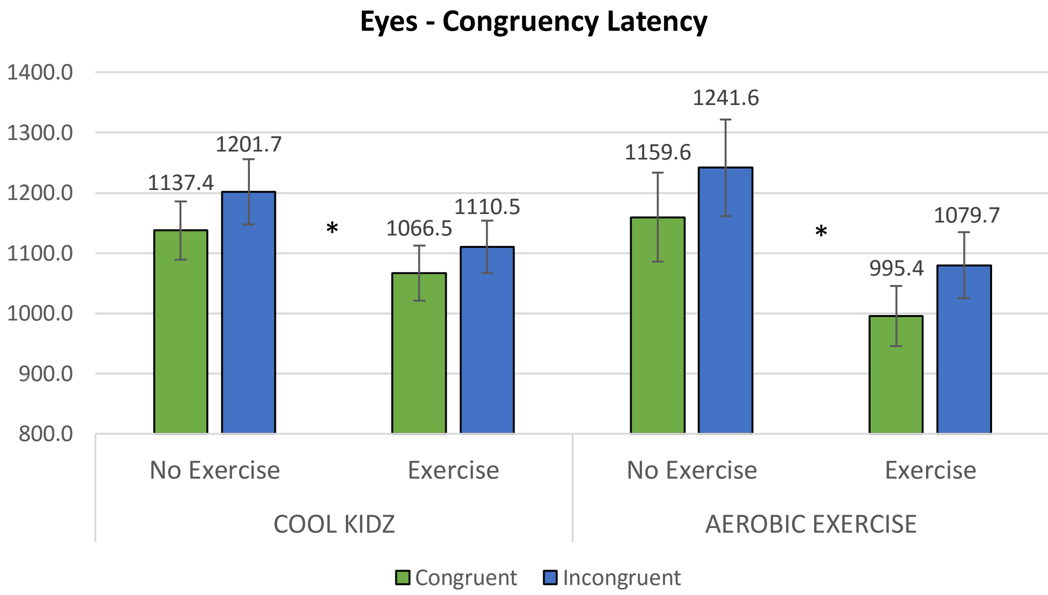
Figure 22: Accuracy of congruent and non-congruent trial responses for the Eyes task



A significant main effect of congruence was found, $F(1,35) = 50.629$, $p < 0.001$, $\eta_p^2 = .591$. Participants demonstrated increased accuracy in congruent trials when compared to incongruent trials in both EDU and intervention phases. No other main effects or interactions were found.

Latency

Figure 23: Latency of congruent and non-congruent trial responses for the Eyes task



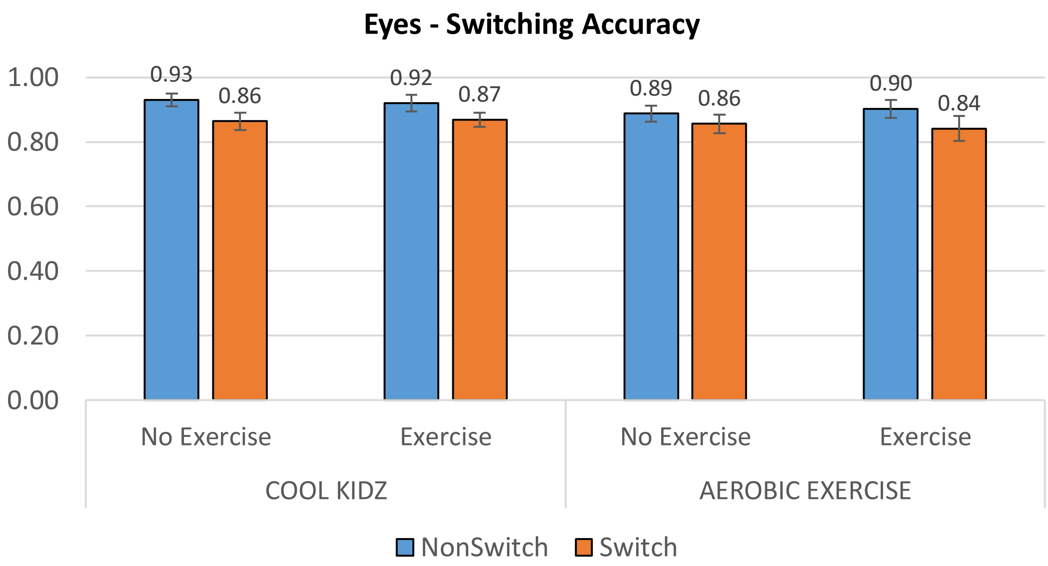
A significant main effect of congruence was found, $F(1,35) = 19.632$, $p < 0.001$, $\eta_p^2 = .359$. Participants demonstrated decreased latency in congruent trials when compared to non-congruent trials in both EDU and intervention phases. A main effect of intervention phase was also found, $F(1,35) = 6.526$, $p < 0.001$, $\eta_p^2 = .157$. Participants demonstrated faster responses across all conditions in their exercise phase when compared to their EDU phase. No other main effects or interactions were found.

<u>Switching</u>

A 2 (switching: non-switch versus switch trials) x 2 (intervention phase: EDU versus exercise) mixed factorial ANOVA was used, with participant group (ASD and SEN) as the between-subject factor to investigate any group-level differences. No significant effects of group on switching or intervention phase were found ($p > 0.05$), so analysis continued with both diagnostic groups combined.

Accuracy

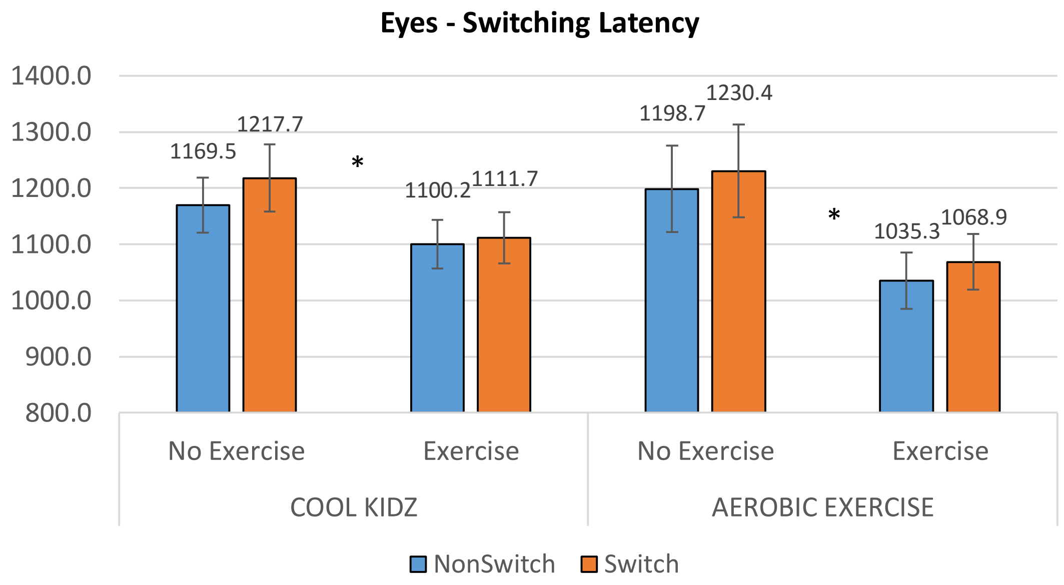
Figure 24: Accuracy of non-switch and switch trial responses for the Eyes task



A significant main effect of switching was found, $F(1,36) = 15.003$, $p < 0.001$, $\eta_p^2 = .294$. Participants demonstrated increased accuracy in non-switch trials when compared to switch trials in both EDU and intervention phases. No other main effects or interactions were found.

Latency

Figure 25: Latency of non-switch and switch trial responses for the Eyes task



A significant main effect of switching was found, $F(1,36) = 5.747$, $p < 0.05$, $\eta_p^2 = .138$. Participants demonstrated decreased latency in non-switch trials when compared to switch trials in both EDU and intervention phases. A main effect of intervention phase was also found, $F(1,36) = 1.101$, $p < 0.05$, $\eta_p^2 = .162$. Participants demonstrated faster responses across all conditions in their exercise phase when compared to their EDU phase. No other main effects or interactions were found.

5.2.4. Discussion

The aim of this study was to explore the effects of chronic school-based exercise on the cognitive performance of school-aged ASD and SEN pupils. Forty participants underwent a six-week exercise programme consisting of 30 sessions, randomised at the classroom level to receive either a low-moderate intensity SI-based treatment, or high-intensity aerobic exercise. As in our previous study, participants were tested on their inhibitory control and attentional shifting performance following the intervention programme, and following a period of education as usual. Crucially, participants were tested at least 24 hours following their final exercise session of the programme, contrasting the 90-minute testing window of Study 1.

The primary research questions were whether the effects found following acute exercise in Study 1 would uphold outside of the post-exercise arousal window, and whether there would be any superior effects of exercise intensity on EF outcomes. Although this study adopted a within-subjects design, an exploratory question was whether any outcome differences would occur between diagnostic groups (ASD and SEN), regardless of intensity. It was predicted that beneficial effects of both moderate and high-intensity programmes to EF performance would be present. Furthermore, should the results of the acute exercise of Study 1 be attributable to participating in EF-loaded exercise, similar effects were predicted to uphold in this chronic exercise study. Since a lack of previous research exists within this population, directional hypotheses could not be made regarding differential effects in ASD and other SEN participants. However, based on the results of our previous study, it was speculated that all participants would benefit equally across groups.

Contrary to predictions, no effects of intervention on EF performance (congruent versus incongruent trial responses) were found following physical exercise phases when compared to the acute exercise results of Study 1. Although the majority of results trended in the expected direction, the only significant group-level effect relevant to post-intervention changes was found in the overall mean latency of the Eyes task. Children across both diagnostic conditions and intensity groups demonstrated reduced reaction times when compared to EDU performance. Although this suggests a general fluency effect of the chronic exercise programmes, these effects were not upheld in the other tasks at the group level. However, an intervention-specific effect was found in the SI group for the latency of the Pictures task. Participants in the 'Cool Kidz' condition improved in their speed of responding across all trial conditions during the post-exercise phase when compared to EDU. This effect did not hold up in the aerobic exercise group. Additional research is required to explore these results in more detail, using a larger battery of tasks to assess both EF effects as well as overall fluency, regardless of specific inhibitory and attention shifting performance.

Importantly, however, for the Arrows task, switching effects were not present during either testing phase. That is, cognitively-demanding switch trials were responded to with similar accuracy and latency as less demanding non-switch trials. This may account for the lack of EF effects during intervention, given that participants were already performing at ceiling levels for switch and non-switch trial comparisons. Nevertheless, on the Pictures and Eyes tasks, effects of inhibition and switching were present, with more cognitively-demanding trials (incongruent and switch) responded to with less accuracy and greater reaction time than less demanding trials (congruent and non-switch).

Although the current findings contradict those of other longer-term exercise studies (Tuckman, Hinkle & Scott, 1986; Kramer et al., 1999; Davis et al., 2007; Kluding, Tseng & Billinger, 2011), when taken together with Study 1, they somewhat support multiple theories relating to temporary underlying mechanisms. For example, many researchers attribute exercise-induced EF effects to temporary cardiovascular and neural arousal

(Hillman, Snook & Jerome, 2003; Klingberg et al., 2005; Best, 2010), that return to baseline levels between 20 minutes (Chang et al., 2012) and 90 minutes (Hori et al., 1978) post-exercise. It is therefore possible that whilst both programmes temporarily enhanced EF performance in participants, the planned delay between exercise engagement and participation in the computer-based EF tasks led to missed effects (Lambourne & Tomporowski, 2010). However, since this assumption is based on the combined results of both Study 1 and 2, future research should endeavour to control and monitor any gradual reductions in post-exercise effects in this population.

The lack of EF results in the present study does not support the assumption that cognitively-loaded physical exercise leads to EF gains (Sibley & Etnier, 2003; Best, 2010). The participants in the SI-based group completed sessions that involved structured motor planning, including the implementation of patterns and sequences- skills associated with intact executive functioning. Those in the aerobic exercise group participated in games and activities that required them to create, monitor and modify plans, switch the focus of attention, cooperate with teammates, and inhibit distractions. Previous literature suggests that engaging in such physical activities improves performance on EF tasks thereafter. However, neither of the groups demonstrated improvements in EF following the 30 sessions of their programme. Again, as with direct cardiovascular and neural arousal explanations, it is possible that these effects are temporary, occurring within a limited post-exercise time window. Without direct investigation into underlying mechanisms, such assumptions are purely theory/literature based, and further research should endeavour to explore the differential effects of acute and chronic outcomes in the ASD and SEN population.

With reference to the tested population, as predicted, there were no superior effects of diagnostic group, with both ASD and SEN groups performing similarly. Diagnosis-driven effects were assessed as covariates through fitted models, but no intervention-specific effects were found. This supports the results of our previous study, which also found similar effects across all participants. Interestingly, whilst Study 1 evidenced a collective positive EF effect of acute physical exercise, this study demonstrated a comparably reduced intervention-related EF effect following a longer-term programme. These results strongly suggest a similar effect across multiple conditions, perhaps highlighting the transferability of research in other clinical populations to those with ASD. However, in relation to the null effects found compared to previous research, it is important to highlight that the current study is one of the first to measure EF as a result of a school-based programme for ASD. The positive facilitations found in typical child and adult populations are perhaps not transferable, with the effects in ASD and other SEN populations not lasting as long following exercise. Future studies should aim to investigate this interesting effect, directly comparing ASD and SEN groups with other populations.

In sum, this study aimed to extend the physical exercise and EF literature by conducting a large field-based study implemented within the school environment. Children with ASD and other SEN were assessed on their inhibitory and attention shifting performance following 6 weeks of physical exercise, and 6 weeks of education as usual. Participants were measured at least 24 hours following the final session of their programme. Results revealed a significant fluency effect across both programmes in a task loaded onto inhibitory and attention shifting performance, but no direct improvements in EF trials when compared to non-EF trials. Wider implications of this research along with current limitations and suggested future directions will be discussed in Chapter 6.

CHAPTER 6: GENERAL DISCUSSION

6.1. Introduction

For the past few decades, researchers have been investigating effective interventions that are both cost-effective and accessible for multiple disorders. As reviewed in Chapter 2, many current intervention options for this population involve behavioural strategies that target speech, language, joint attention and social play. However, an area that few clinicians target is motor skills, which have been associated with the development of a child's language and cognition. Although motor-based interventions are gaining interest, very little is known to date about their efficacy in ASD and other SEN populations. Given this gap in knowledge, the aim of this thesis was to present physical exercise as a viable intervention for this population. Incorporating the programme into the school day, this series of novel empirical research studies also aimed to demonstrate how behavioural-based methods can lead to increased motivation and participation in this population. This chapter will provide an overview of the topics and novel empirical research studies of this thesis. It will discuss combined interpretations and implications of the results, before outlining limitations and suggestions for future research directions.

This thesis first presented a comprehensive review of the current early intervention literature for individuals with autism spectrum disorders, with particular focus on behavioural, developmental, and hybrid developmental-behavioural therapies. Although the evidence-base is beginning to grow, we note a variety of limitations, including the lack of high-quality designs that use randomisation and participant blinding. This led to the conclusion that further well-controlled intervention studies are required, particularly those which attempt to explore real-world translations using key therapeutic techniques such as reinforcement and shared control. Relevantly, whilst we outlined some programmes that incorporate motor-based aspects, including gesture imitation in RIT, the vast majority directly target speech, language and social improvements through various behavioural pathways. However, a growing body of research suggests a close connection between motor development and higher order cognition, including language (Campbell, Eaton and McKeen, 2002; Bates & Dick, 2002; Robertson and Johnson, 2009).

This association led to our overview of motor development and resonance difficulties in ASD, with particular reference to implications for intervention. Here, a set of recent motor-based interventions were explored, including PROMPT and Auditory Motor Mapping. Although small-scale studies, these interventions provide promising results in those with speech and language delays and impairments. Based on these observations, we highlighted the need for new experimental research studies designed to directly examine the efficacy of motor-based interventions with this population. In line with Chapter 2, we outlined the importance of combined expertise and ingredients from traditional ABA-based and developmental intervention fields. Importantly, we maintained that the effects of these interventions appear to extend beyond simple skill learning, with benefits apparent in higher order cognitive and social processes.

Given this promising yet limited evidence outlined throughout these chapters, it was deemed important that further well-controlled intervention research be conducted in the ASD and SEN populations. Further, as concluded by Hillman et al. (2009), a recent priority in this field is the development of interventions that improve school-based performance. A particularly prominent motor-based intervention is that of physical exercise, with many studies demonstrating efficacy of moderate and high-intensity exercise in improving emotional well-being, adaptive behaviour, academic performance, and cognitive functioning in multiple populations (Kern et al., 1982; Tomporowski, 2003; Coles and Tomporowski, 2008; Pesce, et al., 2009; Ströhle, 2009; Pan et al., 2015). Importantly, physical exercise is already a frequent component of UK school curriculums, making this form of intervention both accessible and cost-effective. Therefore, combining the current knowledge with significant shortfalls in the literature, two novel large-scale experimental group design studies were developed to examine the effects of school-based physical exercise intervention programmes using key clinical components. Study 1 explored the effects of acute physical activity on the cognitive performance and reported stress of youths with ASD and other SEN. Crucially, participants were tested within 90 minutes of completing the final acute exercise session. Study 2 examined prolonged engagement in high-intensity aerobic activity and a moderate intensity SI program. Participants in this study were tested at least 24 hours after their final session.

6.2. Stress

Chapter 4 investigated the effects of short (Study 1) and longer-term (Study 2) physical exercise engagement on self-reported stress in ASD and SEN participants. When testing participants within 90 minutes of engagement in their final session of Study 1, results revealed the predicted decreases in self-reported stress across both groups. As discussed, these results support similar research in other populations, including typical adults (Roth, 1989), schizophrenics (Vancampfort et al., 2010) and individuals with depression (Bartholomew, Morrison & Ciccolo, 2005). Various psychological theories may account for these findings, such as the distraction hypothesis (Bahrke & Morgan, 1978), cognitive dissonance (Festinger, 1957; Cooper, 2011), and improved self-perceptions (Petruzzello et al., 1991). Research into physiological effects evidence further underlying mechanisms, such as upregulation of emotion-related endorphins and monoamine neurotransmitters (Knochel et al., Sarris et al., 2012), and reductions in glucocorticoid stress hormones (Eyre & Baune, 2012c) as a result of physical exercise engagement. Importantly, specific to the acute exercise of Study 1, others propose that these effects may be limited to the period at which the exercise classes are novel (Motl & Gosney, 2007), or to the duration of post-exercise body temperature increase (the thermogenic model; Craft & Perna, 2004).

In order to consider physical exercise as an effective and long-term intervention, it is important to understand whether this relationship is indeed primarily a novelty, thermogenic and/or temporary neural arousal effect, given that these underlying effects will lesson over time (Hori et al. 1978; Levinson & Reid, 1992). In an attempt to answer this questions, a second study was conducted which explored the effects of chronic physical exercise engagement in ASD and SEN participants. Children engaged in 6 weeks of education as usual, and 6 weeks (30 sessions) of either a high-intensity aerobic exercise programme similar to that of Study 1, or a moderate intensity SI-based intervention, as proposed to be effective by Diamond (2000). Despite the 24-hour delay in experimental testing, results revealed decreases in reported stress during post-intervention testing when compared to the EDU phase. There were no significant differences between scores of the children in the moderate versus high-intensity exercise programmes, suggesting that intensity may not be a mediating factor in the affect-exercise relationship. This has important implications for those with barriers to participating in high-intensity physical exercise, including motor and/or health conditions.

One of the main aims of the present studies was to incorporate the programme successfully into the school week, with particular reference to maintained child engagement. This was achieved by using a behavioural framework that utilised key components of other current behavioural and/or developmental interventions, including praise, prompting and contingent rewards. Relevantly, the discussed exercise intervention research of Throne et al. (2000) highlighted the importance of immediate feedback and its role in the positive feelings of self-mastery. The authors purport that this contingent tangible feedback may have reinforced the improved emotions assumed by the mastery hypothesis (Gauvin & Brawley, 1993) - a potential mechanism for stress-reduction as discussed in Chapter 4. Future research should therefore seek to investigate this effect, perhaps using a control condition in which participants are also reinforced for participation and improvement. This comparison would help to highlight any exercise-specific effects on stress that are superior to the components of the intervention framework. Since the present study did not measure underlying physiological changes as a result of exercise, further research of this nature would contribute greatly in this field.

The present results therefore support the use of physical exercise as both an immediate and prolonged stress/anxiety-reduction tool in the ASD and other SEN populations of this age group. Importantly, reductions in stress and anxiety levels through exercise may result in collateral gains that are commonly associated with ASD and related disorders, including stereotypic/repetitive behaviour (Joosten et al., 2009; Rodgers, Glod, Connolly & McConachie, 2012). Indeed, early studies into the effect of physical exercise and repetitive behaviours in autistic persons yield promising results, with such behaviours becoming mitigated following brief jogging sessions (Kern et al., 1982; Kern et al., 1984). When considering the learning environment, such maladaptive stereotypy is believed to negatively affect teacher-pupil interactions (Emam & Farrell, 2009), significantly reduce engagement with the environment (Koegel and Covert, 1972) and, in turn, hinder the learning process (Morrissey, Franzini, & Karen, 1992). Future research should therefore seek to investigate this relationship, paying particular attention to the collateral gains of decreased stress/anxiety, and mechanisms that underlie such effects.

6.3. Executive Functioning

Chapter 5 investigated the effects of acute (Study 1) and chronic (Study 2) physical exercise engagement on inhibitory and attentional shifting performance in ASD and SEN participants. When testing children within 90 minutes of participation in their final session, results revealed improvements in inhibitory control and attentional shifting as well as overall fluency improvements in both ASD and SEN individuals. These results supported a plethora of previous studies in other populations, including healthy adults (Chang & Etnier, 2009), and children with ADHD (Piepmeier et al., 2015) and Down's syndrome (Chen, Ringenbach, & Albert, 2014). The results also complement those of Anderson-Hanley, Tureck and Schneiderman (2011), who discovered EF improvements in children with ASD following 20 minutes of physical activity compared to those in a video-watching condition. Although there is no clear consensus within the literature, suggested mechanisms for this short term EF effect are temporary arousal in the CNS (Meeusen, Piacentini, & De Meirleir, 2001), increased frontal brain region blood flow (Ide, Horn and Secher, 1999), and heightened P3 amplitude (Hillman, Snook and Jerome, 2003) as a result of acute exercise. Alternative accounts include EF facilitations through engaging in cognitively-loaded physical exercise (Lakes & Hoyt, 2004) and complex motor movements (Diamond, 2000) that engage inhibitory, shifting and planning behaviour prior to performing experimental tasks, in turn improving performance (Best, 2010).

The wider implications of improved inhibitory and attentional shifting performance are highly important and transferable to classroom behaviour. Theoretically speaking, a child who is better able to inhibit both externally-driven stimuli (e.g. noise outside of their classroom) and internally-driven impulses (e.g. the urge to leave their seats and engage in a preferred activity) will demonstrate improved on-task behaviour in the classroom. Equally noteworthy, a child who can better cope with switching from one academic subject to another, may exhibit less frustration and, again, increased on-task behaviour.

However, as with explanations for emotional state improvements, there are certainly unanswered questions when considering acute exercise engagement and immediate EF testing. A variety of research groups have demonstrated EF effects following chronic, less intense exercise engagement, often with measures taken outside of the 90-minute arousal window. This appears to contradict mechanism explanations relating to temporary cardiovascular and neural arousal. A second study was therefore conducted to explore whether performance on these EF measures remains facilitated following a chronic programme and 24-hour delay between engagement in the final session and EF assessments. Participants were assigned at the classroom level to receive either a moderate SI-based programme, or high-intensity aerobic exercise. Findings revealed no improvements in EF following either exercise intervention when compared to education as usual, although an overall fluency effect was discovered for one task. Given that the same tasks as in Study 1 were used, these results therefore provide indirect support for theories attributing the EF effect to temporary arousal (Lambourne & Tomporowski, 2010; Best, 2010), that may subside over time (Hori et al., 1978).

Despite no direct EF improvements found in Study 2, as mentioned, we did discover an overall fluency effect in some tasks. Participants improved in their speed of responding during post-intervention assessments, with no costs to accuracy. Since these task improvements were not directly related to the intervention, this somewhat supports the research of Trudeau and Shephard (2008), who found that academic achievement is unhindered after increasing the time spent engaged in physical activity at a cost to the time spent in other subjects. These overall fluency improvements may have wider implications that extend to classroom-based activities, including on-task focus and speed of work completion. Further research is required to explore these effects, perhaps with particular attention paid to classroom observations.

Notably, when compared to other chronic exercise studies, the current programmes of 30 sessions conformed to typically reported number of sessions, which vary between eight (Chan et al., 2013) 25 (Thorell et al., 2009), 36 (Tuckman, Hinkle & Scott, 1986), and up to 130 sessions (Davis et al., 2011). Therefore, the lack of EF effects as found in these other studies are unlikely attributable to the programme’s duration, despite being limited to academic term lengths. Further, as discussed, the current programmes also conformed to activities previously found to facilitate EF effects (Pesce et al., 2009; Best, 2010; Davis et al., 2011), reducing the likelihood that the conflicting results are due to type of exercise engagement. However, aside from temporal properties of testing delay, there are indeed other explanations that may account for these differential effects, including the population tested, and type of cognitive assessments used.

Firstly, regarding the assessments, the same tasks were used in Study 2 as in the acute exercise of Study 1. Although this was intentional for comparison purposes, Chang and Etnier (2009) found differential effects of types of EF measured, with acute exercise affecting tasks loaded only onto inhibition, interference, speed of processing, but not cognitive flexibility. Similarly, in their chronic exercise study, Davis et al. (2007) found significantly improved cognitive performance only on one EF sub-scale, Planning. Thus, although the current tasks assess both inhibitory and attention shifting performance, it is certainly possible that a wider variety of EF tasks may highlight intervention effects. Equally noteworthy, in Study 1, we discovered an ASD-related effect in the Pictures task which was not present in another task (Eyes) measuring the same component of EF, inhibition. Specifically, we discovered a speed-accuracy trade off in the Pictures task, with overall EF improvements present at a cost to relative fluidity on incongruent trials. However, overall accuracy and latency improvements were found in the Eyes task with no cost to relative trial performance differences. This difference also highlights the sensitivity of EF tasks, with variations in findings both between and within types of tasks. Future research should seek to expand on these results, exploring whether these findings hold up across other measures of EF.

Secondly, since this is the first study to date to test the effects of chronic exercise on EF in the ASD population, it is possible that the effects are accurate despite the contrast with previous literature. That is, the effects found in other populations may simply not transfer to this clinical group. As mentioned in Chapter 5, children with ASD have reportedly lower baseline levels of EF, which are perhaps less susceptible to the lasting EF effects of exercise. Exploring this potential subsiding effect in the ASD population with alternative EF measures is certainly a key area for future research.

Importantly, the current tasks used to measure EF were successful at showing EDU congruency and switching effects, with the exception of the Arrows task in Study 2. The results were similar to that of previous studies that used the same or comparable tasks. For example, an exercise intervention study by Kluding, Tseng & Billinger (2012) found that in the Flanker task, congruent trials were associated with higher accuracy than incongruent trials at baseline. The stroke patients in their study scored an average of 92.4% accuracy on congruent trials, and 71.8% accuracy in incongruent trials. Following intervention, participants improved their cognitive control over incongruent stimuli by 7.7%. Furthermore, in the study of Burns, Riggs and Beck (2012) which used identical tasks, participants (4- to 7- year olds) scored an average of 94.5% across all congruent trials, and 86.8% across incongruent trials on the Pictures task, using the same amount of trials. Despite the age difference between studies, the participants of the present study possess verbal and non-verbal ages significantly lower than expected for their age group. In comparison, healthy adults have previously demonstrated around 99% and 98% congruent and incongruent trial accuracy respectively (Colcombe et al., 2004).

In addition to the benefits discussed, there are numerous wider implications when considering the impact of physical exercise on the EF performance of children, particularly those with ASD and other SEN. For example, a plethora of researchers maintain that EF is implicated in academic performance (Riggs, Blair and Greenberg, 2003; Blaire and Diamond, 2008). Relevantly, individuals with ASD have been shown to exhibit significant deficits in academic environments (Ashburner, Ziviani & Rodger, 2010), particularly with flexibility (Cannon et al., 2011). Thus, it could be proposed that improvements to academic performance in ASD populations can be achieved via developments in EF as a result of increased physical exercise (Tomporowski et al., 2008; Trudeau & Shephard, 2010). Indeed, physical exercise and academic performance have already been well-researched in healthy populations. For example, Castelli et al. (2007) observed a positive association between academic achievement and aerobic capacity, and a negative association between academic achievement and BMI. The means by which these academic gains are achieved is often thought of in relation to increased attention span through neural arousal; a key area of cognition which falls under the EF umbrella (Best, Miller & Naglieri, 2011). Indeed, the small-scale studies that have investigated these effects in the ASD population provide promising results, with physical exercise mitigating EF-regulated maladaptive behaviours (Rosenthal-Malek and Mitchell, 1997; Nicholson et al., 2011). Future research should seek to expand on current sample sizes and explore this further exercise-EF relationship in more detail.

Finally, combining the results of the present studies, it is interesting to note that outcomes related to improved stress and EF may actually be related to one another. That is, there may indeed be a link between executive functioning and levels of reported or physiological stress. A study by Leskin and White (2007) found that participants with post-traumatic stress disorder (PTSD) were more likely to demonstrate performance impairments in one of their cognitive measures, the Attentional Network Task (ANT; Fan et al., 2002). Similarly, Beers and Bellis (2002) discovered a direct link between PTSD and executive function performance in children, with those in the stress group performing poorer in attention and EF tasks than those without PTSD. It is therefore noteworthy that when intervention-related improvements of stress were found in Study 1, EF improvements were also discovered. However, whilst the literature suggests that higher stress causes more deficient EF performance, the direction of the causal relationship is unclear, and the present research does not support all research in this field. For example, Study 2 demonstrated long-term improvements in stress in the absence of long-term improvements in EF. It is therefore for future research to explore this relationship further, and to investigate whether a potential route to EF improvements is through decreasing stress in each population.

6.4. Strengths and Limitations

The present work contributes a number of novel findings in the field of ASD interventions, and builds upon the limitations and gaps in previous research. Firstly, we include a larger sample size than many studies in this field. As highlighted in a review by Petrus et al. (2008), a large amount of exercise intervention studies are underpowered. Across the seven childhood ASD exercise intervention studies that met their criteria, only 26 children were tested. These studies consisted of four single-subject designs, two group designs, and a case study. However, due to the level of resources, the limited use of school time, and the use of opportunity sampling, the present studies were limited to a maximum of 30 participants per group. Whilst this is still extremely valuable and builds upon previous literature, larger groups will benefit future research by enabling more in-depth analysis. For example, whist Study 2 included a total of 4 groups (ASD aerobic exercise, ASD Cool

Kidz, SEN aerobic exercise, SEN Cool Kidz), analysis was collapsed across diagnostic groups to maintain a reliable level of statistical power. Future research should therefore conform to, or increase on, the present standards to ensure that the quality of the ASD intervention literature continues to grow.

Equally noteworthy, previous research is often reliant on volunteers who are both somewhat motivated to exercise, and/or are not blinded to the experimental hypotheses. In the present studies, children participated as part of their school day, with participants and classroom teachers blinded to the experimental hypotheses, and research assistants blinded to each child's experimental status. Furthermore, the integration of the programme into the field was a critical aspect of the programmes. Adapting the physical exercise sessions to fit within the school day allowed the children to feel comfortable in familiar surroundings. This was particularly important, since ASD is often characterised by rigidity and inflexibility to changes, along with increased anxiety when exposed to novel or non-preferred environments (White, Oswald, Ollendick and Scahill, 2009). As discussed in Chapter 2, this field-based nature also allows for greater efficacy of application and generalisability, with effects more likely to replicate and transfer to the same environment elsewhere compared to laboratory or home-based settings.

In addition, although there has been an attempt to document a physical exercise programme in an available handbook for typically developing children (Davis et al., 2011), previous studies have not provided a full behavioural framework for engaging those with ASD and other SEN in physical exercise classes. This is important given the previous literature on the exercise avoidant behaviour of these populations (Finkelstein et al, 2010), and the key intervention ingredients that appear to contribute towards intervention success as discussed in Chapter 2. Indeed, an extremely important implication of the current findings is the increase in participation from ASD and other SEN children in school-based physical exercise. As can be interpreted from the results, adopting a behaviour framework appears to aid this population, increasing motivation to participate. Alongside this, the teacher-led nature of our framework increases the likelihood that this intervention is both transferable and accessible in other schools. This is particularly crucial, given our observations in Chapter 2 that future studies must focus on transferring such lab-based ABA techniques to field-based practitioners. Relevantly, a key aim of these studies was to contribute a cost-effect intervention, which was achieved by relying on low staff ratios and few materials. Interestingly, the involvement and interaction with their class teacher on a more personal and less formal level was most highly valued in the follow-up enjoyment questionnaires in Study 1. This should certainly be considered where possible in future school-based intervention programmes.

An important design feature of this collective group of studies is the ability to compare performance on the same measures across two different time conditions: short-term (participants tested within 90 minutes), and long-term (participants tested within 24 hours). Whilst the 90-minute timeframe has been utilised in other studies noted throughout this report, the long-term timeframe has not been as defined to date. Although Study 2 can conclude with a level of certainty that the EF effects of exercise do not last for 24 hours, the present research cannot explain whether there are indeed longer-lasting effects outside of the 90-minute physiological arousal window. It may indeed be the case that EF improvements do continue for several hours after acute or chronic exercise. Future research should seek to monitor performance at varying times after exercise, to determine precisely how long the effects last. However, chronic exercise has been shown to lead to improvements lasting longer than 24 hours, such as general academic performance throughout a school week, which was the reason for choosing this timeframe in the present study.

In addition to remaining uncertainties, there are indeed limitations to these studies, which give rise to the design of future research of this nature. Firstly, caution should be taken when considering results from the self-report stress measures. Although the participants were blinded to the research hypothesis, it is possible that positivity bias occurred, with participants not reporting their levels of stress or exercise enjoyment representatively. Equally noteworthy, as mentioned, individuals with ASD may be less accurate at reporting emotions, making individual results and between-group comparisons somewhat unreliable (Mazefsky, Kao & Oswald, 2011). Furthermore, caution must be taken when utilising and interpreting outcome measures that have not previously been demonstrated as sensitive to change. Although Groden's Stress Survey Schedule has been validated in the academic literature (Goodwin et al., 2007), the present studies are the first to use them as a measure of change. However, the current measures were deemed valuable, given that a significant proportion of items on each scale have individually been cited as improving as a result of intervention, such as flexibility (Green et al., 2007), turn-taking (Moes & Frea, 2002) and other adaptive social interactions (Cotugno, 2009) – behaviours which make up a large proportion of items on the survey. Indeed, for the purpose of the current research, these measures were sensitive to intervention-driven changes. However, to increase reliability, physiological measures of stress should be incorporated into future studies of this nature.

Secondly, as in the study by Strain and Bovey (2011), the real-world setting of this intervention led to restrictions in recruitment, randomisation, as well as information regarding diagnoses. However, whilst independent/external diagnoses of autism and other SEN were not obtained, educational diagnoses and verbal/non-verbal reasoning were confirmed by validated research-screening tools; the ADOS and the BAS-II. Since the participating school anticipated a future diagnosis for some pupils, all children were tested with the ADOS, to ensure that pupils were placed in the correct analysis group. Additionally, physical fitness measures, such as participant weight and BMI were not recorded. This was due to level of resources, as well as respecting the permissions of the participating school regarding sensitivities of pupils. Future research should certainly aim to build upon these results, observing potential increases participant fitness as a result of an extra 20-30-minute exercise class during each school day. Since previous research has predicted larger benefits in overweight children (Davis et al., 2011), it is certainly worth investigating any differential effects based on weight and existing fitness levels.

Although none of the same pupils were tested in both studies, a further limitation is the use of a single school, representing a small demographic. Again, due to the level of resources and size of research team required to carry out such a large-scale intervention investigation, the present study focused primarily on one school with a variety of pupils. However, due to the well-controlled nature of the study, findings from this intervention contribute valuable information regarding the effectiveness of increased physical activity throughout the school week in this population. Indeed, although some demographic effects may be present, it is believed that this population are less sensitive to such restrictions since the symptoms of autism vary just as greatly within demographics as they do between (Jones and Klin, 2009).

However, a particularly relevant and noteworthy limitation of the present study is the use of one age group (11- to 14- year olds). Few studies to date have accounted for age effects within children, but current literature suggests there may be developmental effects. For example, a study by Caterino and Polak (1999) found effects acute physical activity on selective attention only in fourth grade pupils, and not in those in second or third grade. Although little explanation is given for this effect, Best (2010) summarises that the relationship between EF and physical exercise may adapt throughout childhood, with varying aspects of EF sensitive to exercise depending on developmental stage. Future research should certainly aim to apply the current design and techniques to a younger cohort. This may be particularly important, given the need for early intervention as outlined in Chapters 2 and 3.

Finally, it is important to highlight the gender imbalance across groups in this study, due to the reliance on opportunity sampling. This is particularly noteworthy for studies which monitor stress-related behaviour and self-reports. Although these studies used a repeated measures design and groups were not directly compared, we do note a higher level of self-reported stress in the SEN group than the ASD group, with the SEN group including a higher number of female participants. The group-level differences in stress results can therefore not confidently be related to diagnoses, but potentially to gender imbalances, with females of this age often reporting higher levels of stress than males (Rudolph & Hammen, 1999).

Future studies should attempt to minimise this limitation, ensuring balanced male and female ratios.

6.5. Future Directions

So far, suggestions for future research have been made throughout each chapter that relate closely to the present outcomes. However, based on the successful implementation of the exercise programmes into the school day, further research should seek to build upon the design and implementation of the present study, incorporating further outcome measures.

For example, the results from small-scale repetitive behaviour and exercise research are so encouraging that physical activity has even been suggested as a prescription for such maladaptive behaviours (Lochbaum and Crews, 1995). In a review of studies examining the effectiveness of physical exercise on stereotypy reduction in children and adolescents with ASD, Petrus et al. (2008) purported that exercise intervention can indeed lead to short-term improvements in the stereotypic behaviours. However, the review highlighted the need for further research in this area with studies utilising stronger research designs, greater participant numbers and measurements of the long-term outcomes of physical exercise.

In addition, physical exercise has proven to be a useful tool in reducing aggressive behaviour in both typical and clinical populations. Gordon, Handleman and Harris (1986) found that contingent exercise lead to significant reductions in out-of-seat disruptive behaviour, noting that the use of physical exercise was much more effective and practical than other behaviour suppression techniques. A recent study by Cannella-Malone, Tullis, and Kazee (2011) investigated the effects of physical exercise on challenging behaviour in children with emotional disorders and, most relevantly, developmental disabilities. When provided with exercise opportunities eight times daily, each participant's challenging behaviour in the classroom was significantly reduced. Although this intervention schedule may be somewhat invasive during a typical school day, other studies such as those by Allen (1980) and Baumeister and MacLean (1984) have demonstrated similar reductions after just ten minutes of intensive exercise.

6.6. Summary

To conclude, the aim of this thesis was to explore the current intervention literature for ASD and related conditions, and present a motor-based intervention as a viable tool for the ASD and other SEN populations. We first systematically outlined and discussed the current evidence-base for key ASD intervention types, and provided insight into the need for more well-controlled experimental intervention research within this field. Furthermore, based on findings that this population possess key motor difficulties that are often associated with language and cognition, we then presented physical exercise as an accessible and cost-effective motor-based intervention for this population. Although promising findings have been uncovered in other groups, very few large-scale exercise intervention studies have been conducted with ASD participants.

This thesis presented the first experimental studies to investigate the effects of acute (Study 1) and chronic (Study 2) physical exercise on executive functioning and reported stress in a large-scale school-based study with this population. Study 2 also compared the effects of a moderate-intensity sensory integration programme. Results indicate that high-intensity physical exercise results in significant short-term (<90 minutes), but not long-term (24+ hours), improvements in cognitive/behavioural flexibility (executive functions) in students with ASD and students with SEN. Furthermore, both high-intensity and low-moderate intensity SI exercise resulted in significant reductions in stress, in both the short-term and long-term in students with ASD and students with SEN. These findings provide direct evidence for the effectiveness of school-based physical exercise intervention for children with developmental conditions. Future research should aim to expand on these findings, using additional EF and behavioural outcome measures, as well as assessing whether the findings transfer to other age groups.

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APPENDICES

Appendix A – Study 1 and 2 Consent Forms

Study 1 Parent Consent Form

University of Birmingham Infant and Child Laboratory Research Study
"The Effects of School-based Exercise Routines on Children and Adolescents with Autism"
Consent for Child with Autism

Why is this research study being conducted? What is its purpose?

The purpose of this study is to discover the positive effects of physical exercise in children and adolescents with Autism Spectrum Disorders (ASD).

Who is conducting this research study, and where is it being conducted?

Joseph McCleery, PhD, Natasha Elliott, BSc, and their colleagues, are conducting this study at your child's school.

How are individuals selected for this research study? How many will participate?

You are being asked to participate because your child is aged between 11-years and 15-years old and has been diagnosed with Autistic Disorder, Asperger's Syndrome, or Pervasive Developmental Disorder – Not Otherwise Specified (PDD – NOS). There will be approximately 70 participants in this study.

What do I have to do if I am in this research study?

Physical exercise sessions (20-30 minutes daily for one week): Your child will be given an extra 20-30 minute physical activity session each day for one week. Behavioural measures (such as short computer-based reaction-time based tasks and measures of repetitive behaviour) will be taken throughout the week, as well as during a week designed to maintain current physical activity frequency, but at a lower intensity. The physical activity sessions will be ran by [redacted] Physical Education teacher who your child will be familiar with, and will not require your child to take part in any activities that they are not otherwise encouraged to participate in.

Behavioural assessments (One hour): We will administer behavioural assessments of your child's developmental and language abilities as well as their communication and social skills. These will include tasks such as naming objects in pictures, using coloured blocks to create patterns, and answering simple questions. Your child will also be asked to complete a short eye-tracking task, which will monitor and record their eye movements when shown videos of people and videos of objects. You may also be asked to complete a simple, short questionnaire, which includes questions related to your child's social and communication skills.

As part of this project, two short 5-10 minute video recordings will be taken of your child to monitor relevant increases and/or reductions in certain behaviours. This is completely voluntary. In any use of these recordings, your name will not be identified. You may request to stop taping at any time during the project, and you may review any or all portions. All video recordings are kept on password protected computers and/or in a locked cabinet in the lab, and they are identified by the participants' ID numbers. A separate consent form related to the use of recorded images will be also given to you to sign.

Are there any risks associated with participating in this study?

The following considerations have been put in place to ensure that no harm will come to your child: (a) the short exercise sessions have been designed in line with current public health guidelines and will not require any more from your child than a typical physical education class at school; (b) any child with any health-related conditions or complaints, including asthma or injuries, will not qualify for participation in the study, and (c) all children will be accompanied at all times. The physical activities available to your child will be chosen by the sports coordinator on the basis that your child is capable and comfortable joining in, ensuring motivation, enjoyment and on-task behaviour. At no point will a child be asked to participate in a dangerous activity, nor will they be expected to continue should they not feel able to.

Since adolescence is a sensitive period for social and emotional development and demands, it will be ensured that all aspects of the project will neither be distressing nor upsetting for each participant. The cognitive tasks chosen for this study are designed for children from 5 years of age and your child should therefore feel no distress when undertaking such tasks. Nevertheless, they will be reminded that it is their right to decline or withdraw from all or parts of the study and that their scores on such tasks will be anonymous. Furthermore, given the nature of their autism spectrum disorders, there will be no demands placed on participants to interact with others if asked to discuss thoughts or feelings.

You are free to withdraw from the study at any time, including if your child becomes upset or unhappy.

What are the benefits of this research study?

Due to the nature and goals of this study, you may notice positive changes in your child's health, behaviour and/or cognition. Furthermore, this research project will provide important insight into the academic and non-academic needs of individuals with autism. By allowing us to work with your child, you are a significant part in the growth of our understanding of Autism Spectrum Disorders, including advancing current knowledge of appropriate programs for older children with ASD.

You should know that this is a research laboratory and that the researchers are not clinical psychologists. Therefore, we will not be able to provide you with a diagnosis in the case that your child does show signs or symptoms of autism or another disorder based on the results of the assessments. Despite this limitation, at your request, we will provide you with a brief report that includes your child's scores on the assessments and general guidelines for interpreting these scores. You are free to share with clinicians and service providers in an effort to provide them with information that may assist her or him in determining whether or not your child warrants further assessments.

If you are concerned about your child's development, other services are available. These include clinical and educational assessment and treatment services through the National Health Service (NHS). <u>Please remember that we are not a clinic; we are a research facility.</u>

Participation in this research is entirely voluntary. You may refuse to participate or withdraw at any time.

What will happen with the information obtained as part of this research study?

The records of this study will be kept private. Your child's name and the other personal details you provide will be stored. However, research data will only be identified by participant number. Computer files will be stored on password-secured computers in the School of Psychology at the University of Birmingham. Paper copies will be stored in locked filing cabinets in the Infant and Child Laboratory and/or in the office of Dr. McCleery. Only researchers and teachers directly involved in this study will have access to the information collected. In any sort of study we might publish, we will not include any information that will make it possible to identify a participant. Research data obtained from this study will be held indefinitely for use in potential follow up publications as well as in other associated studies.

Again, you may refuse to participate or withdraw at any point during the study. Following completion of the study, you may request removal of information that identifies your child by contacting Dr. McCleery or Natasha Elliott via email [redacted] or telephone [redacted] at any time.

Will I receive any payments?

Your child will receive a cinema voucher or small gift certificate for his/her participation in the study.

Agreement to Participate

I have been satisfactorily informed of the above-described procedures with its possible risks and benefits. I understand that participation in this study is voluntary. If I refuse to participate or choose to drop out of the study at any time, I understand there will be no penalty, and that this decision will not affect my relationship with the University of Birmingham. I am signing this consent form before participating in any research activities. I give permission for my/my child's participation in this study.

____________________________ ____________________________

Date | Name of Child

____________________________ ____________________________

Name of Parent or Guardian | Signature of Parent or Guardian

____________________________ ____________________________

Name of Researcher / Witness | Signature of Researcher / Witness

Study 1 Child Consent Form

University of Birmingham Infant and Child Laboratory Research Study
"The Effects of School-based Exercise Routines on Children and Adolescents" – Older Child / Teen Consent Form

Dr. McCleery, Natasha Elliott and their friends, are running this study at your school to learn more about how physical exercise can help children and teenagers. They are asking to you to be in the study because you are between 11- and 15-years old.

If you agree to help us with this study, you will be given an extra physical exercise class every day for one week. During this week, as well as during a normal week of classes, you will be asked to complete some short computer tasks and an eye-tracking task. The eye-tracker is a special video camera which only follows the movement of the eye whilst you watch short videos on a computer.

You will also be asked to complete some tasks that are similar to things you may have done at school. These will include things like naming objects in pictures, using coloured blocks to create patterns, and answering simple questions.

We will keep a short videotape of you during these weeks to remember some information, but no one other than the researchers will be allowed to watch this video or know any answers you give.

None of the things you would do in this research are dangerous. However, some people become tired or bored during parts of the experiment. If you become tired or bored, you can tell the researcher, who will then let you take a break. If you become really tired, bored, or unhappy and want to stop, then you can tell the experimenter, and they will stop the experiment.

If you participate in this study, the researchers will learn more about how exercise can help children and teenagers during the school day. You will also be able to choose a small gift, such as a cinema voucher or gift certificate, which you can then keep and take home with you.

I would like to participate in this study.

______________________________	______________________________
Date	Name / Signature of Child
______________________________	______________________________
Name of parent	Signature of Parent
______________________________	______________________________
Name of Researcher / Witness	Signature of Researcher / Witness

Video Consent Form

University of Birmingham, School of Psychology

Image and Video Release Consent Form

As part of this project, a video recording and/or photograph will be taken of your child and/or you during your participation in this research project. Please indicate below the uses of these recorded images to which you are willing to consent. This is completely voluntary and up to you. In any use of these images, your name will not be identified. You may request to stop taping at any time and review any or all portions. All video recordings are kept on password protected computers and / or in a locked cabinet in the lab, and they are identified by the participants' ID numbers.

1. The research team may record images to be used in the study. ___________ Initials

2. The images may be posted on the researcher's website. ___________ Initials

3. The images may be shown to participants in other experiments. ___________ Initials

4. The images may be used for scientific publications. ___________ Initials

5. The images may be shown at scientific meetings or conferences. ___________ Initials

6. The images may be shown in classrooms to students. ___________ Initials

7. The images may be shown in public presentations to non-scientific groups.

   ___________ Initials

8. The images/recordings may be used on television and radio. ___________ Initials

9. The images/recordings may be shown to experienced professionals from other academic / research institutes for training purposes, which may include the mailing of images/recordings through the postal service.

   ___________ Initials

You have the right to request that taping be stopped or erased at any time.

You have read the above description and give your consent for the use of recorded images as indicated above.

______________________________ ______________________________

Signature Date Witness Date

Study 2 ASD Parent Consent Form

University of Birmingham Infant and Child Laboratory Research Study
“The Effects of School-based Physical Activity Routines on Children and Adolescents with Autism” Consent for Child with Autism

Why is this research study being conducted? What is its purpose?

The purpose of this study is to discover the positive effects of physical exercise in children and adolescents with Autism Spectrum Disorders (ASD).

Who is conducting this research study, and where is it being conducted?

Joseph McCleery, PhD, Natasha Elliott, BSc, and their colleagues, are conducting this study at your child’s school.

How are individuals selected for this research study? How many will participate?

You are being asked to participate because your child is aged between 4-years and 15-years old and has been diagnosed with Autistic Disorder, Asperger’s Syndrome, or Pervasive Developmental Disorder – Not Otherwise Specified (PDD – NOS). There will be approximately 70 participants in this study.

What do I have to do if I am in this research study?

Physical exercise sessions (20-30 minutes daily for six weeks): Your child will be given an extra 20-30 minute physical activity session each day for six weeks. Behavioural measures (such as short computer-based reaction-time based tasks and measures of repetitive behaviour) will be taken throughout the final week, as well as during a week designed to maintain current physical activity frequency, but at a lower intensity. The physical activity sessions will be ran by a teacher who your child will be familiar with, and will not require your child to take part in any activities that they are not otherwise encouraged to participate in.

Behavioural assessments (One hour): We will administer behavioural assessments of your child’s developmental and language abilities as well as their communication and social skills. These will include tasks such as naming objects in pictures, using coloured blocks to create patterns, and answering simple questions. Your child may also be asked to complete a short eye-tracking task, which will monitor and record their eye movements when shown videos of people and videos of objects. You may also be asked to complete a simple, short questionnaire, which includes questions related to your child’s social and communication skills.

Are there any risks associated with participating in this study?

The following considerations have been put in place to ensure that no harm will come to your child: (a) the short exercise sessions have been designed in line with current public health guidelines and will not require any more from your child than a typical physical education class at school; (b) any child with any health-related conditions or complaints, including asthma or injuries, will not qualify for participation in the study, and (c) all children will be accompanied at all times. The physical activities available to your child will be chosen by the sports coordinator on the basis that your child is capable and comfortable joining in, ensuring motivation, enjoyment and on-task behaviour. At no point will a child be asked to participate in a dangerous activity, nor will they be expected to continue should they not feel able to.

Since adolescence is a sensitive period for social and emotional development and demands, it will be ensured that all aspects of the project will neither be distressing nor upsetting for each participant. The cognitive tasks chosen for this study are designed for children from 4 years of age and your child should therefore feel no distress when undertaking such tasks. Nevertheless, they will be reminded that it is their right to decline or withdraw from all or parts of the study and that their scores on such tasks will be anonymous. Furthermore, given the nature of their autism spectrum disorders, there will be no demands placed on participants to interact with others if asked to discuss thoughts or feelings.

You are free to withdraw from the study at any time, including if your child becomes upset or unhappy.

What are the benefits of this research study?

Due to the nature and goals of this study, you may notice positive changes in your child's health, behaviour and/or cognition. Furthermore, this research project will provide important insight into the academic and non-academic needs of individuals with autism. By allowing us to work with your child, you are a significant part in the growth of our understanding of Autism Spectrum Disorders, including advancing current knowledge of appropriate programs for older children with ASD.

You should know that this is a research laboratory and that the researchers are not clinical psychologists. Therefore, we will not be able to provide you with a diagnosis in the case that your child does show signs or symptoms of autism or another disorder based on the results of the assessments. Despite this limitation, at your request, we will provide you with a brief report that includes your child's scores on the assessments and general guidelines for interpreting these scores. You are free to share with clinicians and service providers in an effort to provide them with information that may assist her or him in determining whether or not your child warrants further assessments.

If you are concerned about your child's development, other services are available. These include clinical and educational assessment and treatment services through the National Health Service (NHS). Please remember that we are not a clinic; we are a research facility.

Participation in this research is entirely voluntary. You may refuse to participate or withdraw at any time.

What will happen with the information obtained as part of this research study?

The records of this study will be kept private. Your child's name and the other personal details you provide will be stored. However, research data will only be identified by participant number. Computer files will be stored on password-secured computers in the School of Psychology at the University of Birmingham. Paper copies will be stored in locked filing cabinets in the Infant and Child Laboratory. Only researchers and teachers directly involved in this study will have access to the information collected. In any sort of study we might publish, we will not include any information that will make it possible to identify a participant. Research data obtained from this study will be held indefinitely for use in potential follow up publications as well as in other associated studies.

Again, you may refuse to participate or withdraw at any point during the study. Following completion of the study, you may request removal of information that identifies your child by contacting Dr. McCleery or Natasha Elliott via email [redacted]) or telephone [redacted]) at any time.

If you do not wish for data to be collected from your child throughout their participation in these activities, please complete the form below and return it to your child's school.

- -

Class teacher's name____

I don't want my child ____________________________________ (name of child)

to participate in this study.

Signed___________________________ Date__________________________

Study 2 SEN Parent Consent Form

University of Birmingham Infant and Child Laboratory Research Study "The Effects of School-based Physical Activity Routines on Children and Adolescents with Autism" Consent for Child with Special Educational Needs

Why is this research study being conducted? What is its purpose?

The purpose of this study is to discover the positive effects of physical exercise in children and adolescents with Autism Spectrum Disorders (ASD). Your child will be a control participant for children diagnosed with autism.

Who is conducting this research study, and where is it being conducted?

Joseph McCleery, PhD, Natasha Elliott, BSc, and their colleagues, are conducting this study at your child's school.

How are individuals selected for this research study? How many will participate?

You are being asked to participate because your child is aged between 4-years and 15-years old and has been diagnosed with (INSERT). There will be approximately 70 participants in this study.

What do I have to do if I am in this research study?

Physical activity sessions (20-30 minutes daily for six weeks): Your child will be given an extra 20-30 minute physical activity session each day for six weeks. Behavioural measures (such as short computer-based reaction-time based tasks and measures of repetitive behaviour) will be taken throughout the final week, as well as during a week designed to maintain current physical activity frequency, but at a lower intensity. The physical activity sessions will be ran by a teacher who your child will be familiar with, and will not require your child to take part in any activities that they are not otherwise encouraged to participate in.

Behavioural assessments (One hour): We will administer behavioural assessments of your child's developmental and language abilities as well as their communication and social skills. These will include tasks such as naming objects in pictures, using coloured blocks to create patterns, and answering simple questions. Your child may also be asked to complete a short eye-tracking task, which will monitor and record their eye movements when shown videos of people and videos of objects. You may also be asked to complete a simple, short questionnaire, which includes questions related to your child's social and communication skills.

Are there any risks associated with participating in this study?

The following considerations have been put in place to ensure that no harm will come to your child: (a) the short activity sessions have been designed in line with current public health guidelines and will not require any more from your child than a typical physical education class at school; (b) any child with any health-related conditions or complaints, including asthma or injuries, will not qualify for participation in the study, and (c) all children will be accompanied at all times. The physical activities available to your child will be chosen by the sports coordinator on the basis that your child is capable and comfortable joining in, ensuring motivation, enjoyment and on-task behaviour. At no point will a child be asked to participate in a dangerous activity, nor will they be expected to continue should they not feel able to.

Since adolescence is a sensitive period for social and emotional development and demands, it will be ensured that all aspects of the project will neither be distressing nor upsetting for each participant. The cognitive tasks chosen for this study are designed for children from 4 years of age and your child should therefore feel no distress when undertaking such tasks. Nevertheless, they will be reminded that it is their right to decline or withdraw from all or parts of the study and that their scores on such tasks will be anonymous. Furthermore, there will be no demands placed on participants to interact with others if asked to discuss thoughts or feelings.

You are free to withdraw from the study at any time, including if your child becomes upset or unhappy.

What are the benefits of this research study?

Due to the nature and goals of this study, you may notice positive changes in your child's health, behaviour and/or cognition. Furthermore, this research project will provide important insight into the academic and non-academic needs of individuals with autism. By allowing us to work with your child, you are a significant part in the growth of our understanding of Autism Spectrum Disorders, including advancing current knowledge of appropriate programs for older children with ASD.

You should know that this is a research laboratory and that the researchers are not clinical psychologists. Therefore, we will not be able to provide you with a diagnosis in the case that your child does show signs or symptoms of autism or another disorder based on the results of the assessments. Despite this limitation, at your request, we will provide you with a brief report that includes your child's scores on the assessments and general guidelines for interpreting these scores. You are free to share with clinicians and service providers in an effort to provide them with information that may assist her or him in determining whether or not your child warrants further assessments.

If you are concerned about your child's development, other services are available. These include clinical and educational assessment and treatment services through the National Health Service (NHS). Please remember that we are not a clinic; we are a research facility.

Participation in this research is entirely voluntary. You may refuse to participate or withdraw at any time.

What will happen with the information obtained as part of this research study?

The records of this study will be kept private. Your child's name and the other personal details you provide will be stored. However, research data will only be identified by participant number. Computer files will be stored on password-secured computers in the School of Psychology at the University of Birmingham. Paper copies will be stored in locked filing cabinets in the Infant and Child Laboratory. Only researchers and teachers directly involved in this study will have access to the information collected. In any sort of study we might publish, we will not include any information that will make it possible to identify a participant. Research data obtained from this study will be held indefinitely for use in potential follow up publications as well as in other associated studies.

Again, you may refuse to participate or withdraw at any point during the study. Following completion of the study, you may request removal of information that identifies your child by contacting Dr. McCleery or Natasha Elliott via email [redacted] or telephone [redacted] at any time.

If you do not wish for data to be collected from your child throughout their participation in these activities, please complete the form below and return it to your child's school.

- -

Class teacher's name____

I don't want my child ____________________________________ (name of child)

to participate in this study.

Signed___________________________ Date__________________________

University of Birmingham Infant and Child Laboratory Research Study

Study 2 Child Consent Form

"The Effects of School-based Exercise Routines on Children and Adolescents" – Older Child / Teen Consent Form

Dr. McCleery, Natasha Elliott and their friends, are running this study at your school to learn more about how physical exercise can help children and teenagers. They are asking to you to be in the study because you are between 11- and 15-years old.

If you agree to help us with this study, you will be given an extra physical exercise class every day for six weeks. During one of these week, as well as during a normal week of classes, you will be asked to complete some short computer tasks, a short questionnaire and an eye-tracking task. The eye-tracker is a special video camera which only follows the movement of the eye whilst you watch short videos on a computer.

You will also be asked to complete some tasks that are similar to things you may have done at school. These will include things like naming objects in pictures, using coloured blocks to create patterns, and answering simple questions.

We will keep a short videotape of you during these weeks to remember some information, but no one other than the researchers will be allowed to watch this video or know any answers you give.

None of the things you would do in this research are dangerous. However, some people become tired or bored during parts of the experiment. If you become tired or bored, you can tell the researcher, who will then let you take a break. If you become really tired, bored, or unhappy and want to stop, then you can tell the experimenter, and they will stop the experiment.

If you participate in this study, the researchers will learn more about how exercise can help children and teenagers during the school day. You will also be able to choose a small gift, such as a cinema voucher or gift certificate, which you can then keep and take home with you.

I would like to participate in this study.

______________________________	______________________________
Date	Name / Signature of Child
______________________________	______________________________
Name of parent	Signature of Parent
______________________________	______________________________
Name of Researcher / Witness	Signature of Researcher / Witness

Appendix B - Exercise session framework

Table 6: Exercise session framework

Explanation/ Priming	Children should be given a full explanation about what will happen in the coming weeks, with opportunity to ask questions. This will ensure sensitivity to the common trait of rigidity/inflexibility with routines in this population.
Routine	Exercise breaks should delivered around the same time each morning. It important to incorporate such a change to the school timetable with its own set structure and consistency.
Familiarity	The activities available to children should be those which they are already familiar with, with no additional demands placed on children to learn new games and rules until the routine has been set.
Child choice	Where possible, classes should be given choices about which physical activities they would like to do each session. This child-directed approach gives the children a level of control, and increases the chance of enjoyment and motivation.
Teacher involvement	Teachers and classroom assistants should participate in the activities to keep the children engaged. This is also an opportunity for a different level of interaction between children and teachers.
Praise	Children should be given consistent, specific praise throughout participation. Praise should be given for flexibility and turn taking, as well as for their level of involvement. The latter is particularly effective at encouraging other children to maintain levels of energy exertion. Both group and individual level praise should be given.
Positive reinforcement	All children should obtain school-appropriate rewards, including ‘house-points’ and/or stickers on behaviour charts for maintained engagement. An explanation should be given beforehand about how they can gain these rewards, with particular emphasis on participation, not performance.
Engaging Activities	Activities that are known to teaching staff as being particularly motivating should be used. Options should include both outdoor sports as well as indoor aerobics to ensure that weather conditions do not affect the program.
Non-Social versus Social Activities	Given the social nature of some activities such as football, praise and encouragement should be given throughout with little emphasis on individual or group performance. The involvement of teachers and classroom assistants often facilitates involvement in more social activities. Children may enjoy the opportunity to ‘compete’ against these members of staff who otherwise have a much more authoritative role with them. However, the need for praise and motivational techniques may be notably reduced during less social activities, such as indoor aerobics which did not require interaction with classmates.

Getting Children Back On Task: Child-Specific Reinforcers	In the event that a child either refused or terminated participation, staff members who are highly familiar with each child should use motivator's specific to that child. For example, whereas one child may earn a star for their chart for re-joining the group, another child may more effectively be re-engaged by the proposition that completion of the activity means extra 'Chill Out' time. However, due to the established compliance expectations within school environments, most children should complete participation without the need for numerous re-engagement techniques.
Returning to Classroom	As is important with each aspect of this program, the return to classroom procedure should also be implemented with routine. Rather than transitioning from high-energy exercise straight to desk-based learning, children should first performed a 5-10 minute warm-down activity, including stretches and deep breathing. Upon returning to classrooms, cups of water may be provided to the children before they are expected to return to their desks. Children should again be praised for their participation, with specific examples of class-level behaviour or child-specific performances that particularly impressed their teachers.

Appendix C– Instructions for each EF task

<u>Pictures Task</u>

In this game, there are two animals
(SPACE)
One of them is a cat
(SPACE)
One of them is a monkey
(SPACE)
When you see the cat, press the blue button
(SPACE)
[*Cat appears* ***– prompt them to press the blue button]***
Child presses BLUE BUTTON
and when you see the monkey, press the green button
(SPACE)
[*Monkey appears* ***– prompt them to press the green button]***
Child presses GREEN BUTTON
Sometimes they appear on this side
(SPACE)
and sometimes they appear on this side
(SPACE)
Let's practice that
[4 PRACTICE TRIALS]
Let's try that again. This time the computer won't tell you when you are right. Try to press the button as quickly as possible

<u>Arrows Task</u>

This is a game with arrows. Sometimes the arrows point down
(SPACE)
And sometimes the arrows point across
(SPACE)
Sometimes they appear on this side
(SPACE)
And sometimes they appear on this side
(SPACE)
I want you to press the button the arrow is pointing towards
(SPACE)
If the arrow is on this side pointing down like this you press the blue button
(SPACE)
[*Same arrow appears* ***– prompt them to press the blue button]***
Child presses BLUE BUTTON
If the arrow is on this side pointing down like this I want you to press the green button
(SPACE)
[*Same arrow appears* ***– prompt them to press the green button]***

Child presses GREEN BUTTON

If the arrow is on this side pointing across like this I want you to press the green button

(SPACE)

*[Same arrow appears **– prompt them to press the green button]***

Child presses GREEN BUTTON

If the arrow is on this side pointing across like this I want you to press the blue button

(SPACE)

*[Same arrow appears **– prompt them to press the blue button]***

Child presses BLUE BUTTON

Let's practice that

[FOUR PRACTICE TRIALS]

Let's try that again. This time the computer won't tell you if you are right. Try and press the button the arrow is pointing towards as quickly as possible

<u>Eyes Task</u>

In this game you will see a face. Sometimes they eyes are looking down

(SPACE)

And sometimes the eyes are looking across

(SPACE)

Sometimes the eyes appear on this side

(SPACE)

and sometimes they appear on this side

(SPACE)

I want you to press the button the eyes are looking towards

(SPACE)

If the eyes are on this side, looking down like this, you press the blue button

(SPACE)

*[Same face appears **– prompt them to press the blue button]***

Child presses BLUE BUTTON

If the eyes are on this side, looking down like this, you press the green button

(SPACE)

*[Same face appears **– prompt them to press the green button]***

Child presses GREEN BUTTON

If the eyes are on this side, looking across like this, you press the green button

(SPACE)

*[Same face appears **– prompt them to press the green button]***

Child presses GREEN BUTTON

and if the eyes are this side, looking across like this, you press the blue button

(SPACE)

*[Same face appears **– prompt them to press the blue button]***

Child presses BLUE BUTTON

Let's practice that

[FOUR PRACTICE TRIALS]

Let's try that again. This time, the computer won't tell you if you are right. Press the button the eyes are looking towards as quickly as possible

Appendix D – The Stress Survey Schedule for Persons with Autism and Developmental Disabilities – rephrased statements

5. Being **around** noise or disruption by others

6. Waiting for events **that you like**

12. Being stopped from completing **something you usually do**

14. Being stopped from carrying out **something you usually do**

18. Receiving activities as rewards

20. Being **around** bright lights

21. Having unstructured, **free** time

24. Being told off

29. Being interrupted while engaging in **something you usually do**

45. Receiving prizes as rewards

49. Being told you are doing a good job

Appendix E – The Stress Survey Schedule for Persons with Autism and Developmental Disabilities - Visual Rating Scale

	SEVERE	5
	MODERATE - SEVERE	4
	MODERATE	3
	MILD - MODERATE	2
	NONE- MILD	1

Appendix F – The Pre-Post Stress Survey

Pre-Post Stress Survey
Physical Activities Can Reduce Your Stress

Place an X in front of all items that apply to you.

☐ I am excited about getting out of bed each day.

☐ I look forward to attending school.

☐ I have a good relationship with my sister(s) and brother(s).

☐ I enjoy time alone by myself.

☐ I eat as much food as I want to at every meal.

☐ I have close friend(s).

☐ I have at least one friend that I can talk things over with.

☐ I know at least one adult that I can share my problems with.

☐ I am afraid to go to after school activities at my school.

☐ I am afraid to go to social activities in my community.

☐ I have learned skills to help me adjust to stress.

☐ I have my school work ready to turn in on time.

☐ I have test anxiety.

☐ I rarely get everything ready in one day that I need to.

☐ My concentration is easily interrupted.

☐ I avoid some tasks because of the stress they cause me.

☐ I am off-task at some point every class period.

☐ I am sad a little sometimes.

☐ I worry about little and big things that I cannot control.

Appendix G – Description of the Cool Kidz Programme

Cool Kids Programme – Background

Cool Kids is an exercise programme produced jointly by the Education and Occupational Therapy Departments in Wolverhampton and supported by Semester School Sport Partnership. Its aim is to enhance children's learning by improving co-ordination, motor development, organisational skills and concentration. The programme was piloted in 2004 with whole classes and the results were very encouraging as children made more than their expected progress in handwriting, written work and reading.

Cool Kids Programme - The Theory

Children working towards being Calm, Confident and able to Concentrate

The Cool Kids programme was devised principally as an opportunity for children with poor motor ability to catch up on foundation skills. This seems to be possible through out infants and junior school and into secondary school. The programme has successfully been used for whole classes of Nursery and Reception children, who are developing their basic motor skills before the introduction of more complex movement. Adaptations of the programme are also being developed for special school application.

All children need exercise

- Getting fit and keeping fit is high on the national agenda, both for children and adults, 20 - 30 minutes per day has been suggested.
- Avoiding obesity is also a national target.
- Motivation and concentration is also linked to exercise.

Some children find fluid movement difficult

- All mainstream schools have children who under-achieve, with special schools and units also catering for such pupils. These children may have learning difficulties and/or motor difficulties, with diagnoses such as dyspraxia, autistic spectrum disorders, attention deficit disorders, difficulties with speech and language, dyslexia and other learning difficulties. Others have no "label", but just do not perform as well as expected or are somewhat lacking in concentration, motivation and application.
- These children may not achieve well in PE and games and often try to avoid these activities or misbehave in these lessons.
- These children may well have difficulty with writing, drawing, listening and staying still.
- These children may not be ready for complex exercise regimes such as aerobics or country dancing, and would view these activities as another area of failure.

The Cool Kids programme is aimed at building up the motor skills of under-achievers so that they can develop in physical activity and improve their ability to modulate their arousal levels. This would be seen in their ability to calm down and concentrate and thereby become achievers rather than under-achievers. There are indications of academic improvement including handwriting.

The programme is based on the following principles:

Body Awareness

- A picture of our body and where it is in space is essential for the development of smooth, organised gross and fine motor control.
- This picture is made up of information from joints, muscles (proprioception) and skin (tactile), letting us know which limbs are where and how much space we are occupying. The brain interprets proprioceptive information (regular repetitive muscle use through organised activity) developing a natural sense of space etc as an essential base for movement. Playing games such as "Move & Freeze", jumping or limb and trunk contact with the floor increases this awareness.
- If a child is not receiving sufficient proprioceptive information, he is likely to try to watch his limbs to ensure that they are doing what he wants them to do.
- Laterality: the understanding of "left and right" on self and others, is based on a good body awareness.

Developmental sequences– to put the foundations of movement into place

- In normal development postural control begins with head control. The child learns to lift his head in prone and begins to focus on objects of interest. Back, neck and eye muscles learn to work together. All our primary school children have been "lie-them-on-their-back" babies, which is done to prevent cot death, but may not have given them enough opportunity to strengthen their back muscles.
- The sequence is: Lying, sitting, four-point (crawling), kneeling and then standing.
- The children are encouraged to develop postural control in lying – prone (on their stomachs) and supine (on their backs) before working in sitting etc.
- They are encouraged to commando crawl and wriggle on their stomachs, to roll and crawl on all–fours in order to:
    - develop control over early reflex patterns which will help with postural control in sitting and standing
    - improve their postural control (static and dynamic balance) as a foundation for the ability to sit still
    - weight bear on their arms and thereby develop stability of shoulder, elbow and wrist in preparation for writing.

Praxis

- Planning new motor actions is dependent on sensory feedback. We rely on proprioception (joint and muscle sense), tactile and vestibular (sense of balance from the inner ear), vision and hearing to tell us if we're getting it right and exactly what adjustments we need to make.
- Children with poor motor planning (praxis) can often perform practiced actions reasonably well, but find new/ novel actions much more difficult to master. They need plenty of variety and challenge within their capability e.g. use a variety of weights and size of balls in games. Moving forward might be enough of a challenge to start with, but later moving in reverse may add to the challenge.
- These children may have great difficulty in regulating muscle tone - they throw too hard or too gently. They need lots of practice throwing a beanbag at a target or a ball to another child. To achieve smooth actions, simplify the activity, let them work lying down with

objects that are easy to handle. Also to give them a variety of weight and size in ball, balloon and beanbag to play throw and catch. In general, make it easier for them by increasing the size of the object, decreasing the speed and noting that objects moving across the floor rather than through the air are easier to track and catch.

- Remember that some children may also have difficulty with ideation (being able to see the play potential of equipment).
- And some children may have difficulty with motor execution (interference with their ability to move) for various reasons:
    - due to damage to the nervous system such as cerebral palsy
    - due to congenital orthopaedic problems such as club foot know as talipies or congenital dislocation of the hip
    - due medical problems such as painful or arthritic joints.

Bilateral and Sequencing

- Actions such as skipping, hopping, catching a ball require timing, sequencing and rhythm and the ability to use both sides of the body together symmetrically and/or reciprocally i.e. the child's movements are smooth, fluid not awkward and jerky.
- Children need to learn the motor skills of childhood e.g. ball control, skipping, if they are to have the self confidence to take part in their peers'games in the playground.
- Self confidence comes from knowing "I can" and then being motivated to try something even more difficult.

Modulation

- We all adjust our arousal levels to the demands of the environment we find ourselves in and most of us are reasonably successful most of the time. When we get it wrong we talk about being over-the-top or not being awake enough for the present activity. We use words like "paying attention" and "concentration"to describe this ability and its result has a bearing on our motivation for performing certain activities or being in certain environments.
- Underlying our ability to modulate our arousal levels is our reaction to the sensory world. Each of us has our own individual pattern of comfortable levels of sensory input and what we would find as uncomfortable. For example: each of us would be able to identify sounds that jar our nerves and others that sooth and calm us. We could all cope with some of the jarring sounds for a limited period of time, but would each have a point at which we need to escape.
- Children are no different. We need however to consider the child whose nervous system is set in such a way as to over-react or under-react to sensory input, by comparison to his/her peers. This child may well react differently to his/her classmates and be more easily distracted, anxious, wound-up or not "with–it". We then talk about them having poor concentration and often, poor motivation.
- These children need modulating sensory input. Proprioceptive input is our best modulator, so these children need controlled movement in heavy work patterns. They also need to learn to relax.

The programme works on the foundations of gross motor development which underlies and underpins fine motor development and the ability to pay attention. It offers the opportunities for the under-achievers to practice the foundation skills before they take part in exercise programmes with the rest of the school.