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The Role of Sensorimotor Difficulties in Autism Spectrum Conditions

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The Role of Sensorimotor Difficulties in Autism Spectrum Conditions

P. Hannant

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Abstract

The Role of Sensorimotor Difficulties in Autism Spectrum Conditions

Autism Spectrum Conditions (ASC) refers to neurological differences in how individuals communicate and interact socially, in addition to repetitive, restricted interests and behaviours. Since the first pioneering reports of ASC, research has attempted to uncover the cause of the diverse range of related difficulties, generating a number of theories behind the roots of the condition. Such theories do not incorporate all of the symptoms associated with ASC, and progress may have been hampered by the lack of consideration for the non-social difficulties that are apparent, such as repetitive behaviours. The research presented in this thesis aims to address the restricted integration of these non-social difficulties, in the form of sensorimotor adversities, by first analysing the evidence for sensorimotor difficulties in ASC from both a psychological and biological perspective. The thesis then goes on to demonstrate the presence of sensorimotor difficulties in ASC using both small scale objective and large scale subjective studies. Following this, the thesis compares and contrasts ASC to a similar neurodevelopmental disorder, Developmental Coordination Disorder (DCD), in order to identify any similarities and differences between the two conditions. Thereafter, presenting a new theoretical framework to explore and understand how ASC may develop: 'The Sensorimotor Theory'. Finally, by adopting this new theory as a foundation for ASC, current sensorimotor interventions are explored and a pilot study for a new intervention is trialled, measuring both the psychological and biological impact of GABA Oolong tea.

1. Introduction

Autism Spectrum Conditions (ASC) refers to neurological differences in how individuals communicate and interact socially, in addition to repetitive, restricted interests and behaviours, as well as atypical motor mannerisms (unusual movements) and sensory responsivity (APA, 2013). ASC usually becomes apparent within the first three years of life and symptoms are life long (Grinker, 2016).

ASC varies greatly between individuals and is an umbrella term for a spectrum that encapsulates the aforementioned behaviours to differing degrees of severity (Figure 1; Diagnostic and Statistical Manual 5th Edition (DSM-5: APA, 2013). However, the most widely used Autism Spectrum considers the level of functional language to be the most prominent factor that assigns severity. This is apparent in the DSM-4 (APA, 2000) criteria where ASCs were apportioned into separate profiles, such as Asperger's disorder and Autistic disorder where the level of language acquisition differs between them. The international classification of diseases, tenth edition (ICD-10), more commonly used in the UK as a diagnostic tool, echoes the DSM-4 differentiated criteria. However, the anticipated ICD eleventh edition is likely to be significantly influenced by the overarching terminology of the DSM-5 (National Autistic Society).

Generally, people on the autism spectrum have great difficulty making and / or maintaining friendships and understanding social rules. They typically have one or very few interests or behaviours that they engage in repeatedly and find changes in routine difficult. Individuals on the autism spectrum may or may not have difficulties with language acquisition, learning and lowered intelligence.



"If you've met one person with autism, you've met one person with autism," -- Dr. Stephen Shore.

1.1 **Theories of Autism Spectrum Conditions**

Since the first pioneering reports of ASC (Kanner, 1943; Asperger, 1944) half a century of research has attempted to uncover the cause of the diverse range of difficulties in ASC, and so a large number of theories behind the fundamental roots of the condition have been proposed. One infamous example is the 'Refrigerator mother' (Bettleheim, 1967), coined very early on by noting the 'lack of warmth' among parents of children with ASC. This theory is now understandably completely discredited. However, theories of ASC have mirrored the zeitgeist of their time and it was not until the mid 1980s that theories began to relate some of the core elements of autism to cognitive impairment. Amongst others, six main cognitive theories of ASC have since been reported, 'Theory of Mind', 'Central Coherence', 'Executive Functioning', 'Amygdala', 'Extreme Male Brain' and 'Multi-deficit'. However, none of these theories appear to have completely fulfilled the criteria given by Cramer (2013) used to judge the merit of theories: how comprehensive the theory is, i.e. does it explain all symptoms; the precision and testability, i.e. can the theory be used to predict if an individual has ASC; parsimony, does it explain ASC in relatively few terms; falsifiability, can it be disproved easily; heuristic value, can it generate thoughts and perspectives; applied value, have effective solutions/interventions been generated from the theory; and universal, does it apply to all individuals with ASC. Comparing past theories of ASC to these criteria enables future theories to be evaluated fairly and thoroughly. Thus, in order to propose a framework for a new theory in this thesis, each of these are considered below.

The Theory of Mind Hypothesis – 1985

'Theory of Mind' (ToM) is based on the understanding that individuals with ASC have difficulty "imputing mental states to themselves and others" (Premack & Woodruff, 1978, p. 515), such as understanding that others have beliefs, desires and intentions that are different to their own. See Figure 2.





First order reasoning



Second order reasoning

Figure 2 Theory of Mind demonstrating the first and second order reasoning scenarios

This theory explains the difficulties in social integration in ASC and was first evidenced by Baron-Cohen et al., (1985) where he demonstrated that 80% of children with ASC failed an unexpected transfer task based on an 'I thinks he thinks' first order scenario. However, initially the 'Theory of Mind' was quite vague and general, and overtime the theory has been repeatedly challenged, advanced, progressed, refined and simplified. For example Happé, (1994) stated that 20% of the children in Baron-Cohen's study were able to complete the task and suggested that the Theory of Mind explained some cognitive impairment but not all facets of the disorder. In order to support ToM, Baron-Cohen went on to change the theory to a delay as opposed to a deficit in ASC (Baron-Cohen, 1989) by using a second order false-belief task based on a 'I think, he thinks, she thinks' scenario (see figure 2). However, this too was challenged when Bowler (1992) showed that 73 per cent of adolescents with ASC also passed a second-order false belief task. Over time the ToM tests moved into the area of face and language processing and it was in this area that Jolliffe and Baron-Cohen (1999) were able to show that even at the very highest functioning end of ASC individuals had difficulty comprehending nonliteral language. Theory of Mind has been research widely as it can be tested relatively easily, such as in the 'Sally-Anne Scenario' (Baron-Cohen et al., 1985), the 'Strange Stories Test' (Happé, 1994), the 'Eyes Task' (Baron-Cohen et al., 1997) and the 'Frozen Eyes' task (Back et al., 2007).

As Theory of Mind research has grown and become more refined, it is widely accepted that this is a symptom of ASC and generally underpins current DSM-5 Criteria: "Deficits in nonverbal communicative behaviours used for social interaction, ranging, for example, from poorly integrated verbal and nonverbal communication; to abnormalities in eye contact and body language or deficits in understanding and use of gestures; to a total lack of facial expressions and nonverbal communication". This understanding has led to interventions being devised. Examples include a 'Mindreading DVD' that teaches emotion-recognition to people with ASC (Baron-Cohen et al., 2002), which is currently being evaluated in a treatment group; and a randomised controlled trial of Theory of Mind training that included skills such as listening to others, understanding the difference between fantasy and reality and recognising emotions in others, which demonstrated improvements in the ability to reason about beliefs and false beliefs, and on the understanding of mixed and complex emotions (Begeer et al., 2011).

When comparing the ToM to Cramer's (2013) criteria above it fulfils five of the seven criteria: testability, parsimony, falsifiability, heuristic value and applied value. However, it does not appear to be universal as only 80% of the ASC individuals could not successfully complete the 'Sally-Anne Scenario' described above (Baron-Cohen et al., 1985). It is also not comprehensive, in that it does not explain the atypical sensory responsivity and some of the more unusual motor behaviours that are also present in ASC, such as stimming.

The Central Coherence Theory – 1989

This theory considers an information processing style rather than а deficit, and specifically focuses on the ability to process information in context (Frith, 1989; Hill & Frith, 2003; Frith & Happé, 1994; Happé, 1996). The theory of Central Coherence was first evidenced in ASC by Frith (1989) and suggests that individuals with ASC have weak global or central coherence, thereby instead of processing information in a global whole, information is processed in a detailed focused fragmented way. For example, due to superior local processing, when compared to typically developing individuals, participants with ASC are thought to do comparably well in the embedded figures test (Jolliffe & Baron-Cohen, 1997; Witkin, 1971) where the presence of a target figure must be located within a complex background (See Figure 3). Such ability is comparable to the story of the five blind men and the elephant, where an individual lacking central coherence would build a picture based only on the separate parts of the elephant rather than seeing the elephant as a whole, such as believing the elephant is a tree having only locally processed the leg.

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Figure 3 Central Coherence Theory: The Embedded Figure Test (Witkin, 1971)

The Theory of Central coherence has been tested frequently, such as in embedded figure and block design tasks (Shah & Frith, 1983 and 1993). Individuals with ASC were shown to score above average on the Children's Embedded Figures Test and were faster at producing 40 different block designs than typically developing counterparts. It was argued that children with ASC do better on such tasks as they included smaller segmented components and therefore lacked a more global form. Other tests of Central coherence include less susceptibility to visual illusions, such as that shown by Happé (1996) in the Titchener illusion, where the presence of two circles around two smaller, identically sized circles, gave the impression that these circles were different sizes. It is thought that visual illusions require the global processing of the whole form.

Having developed over time, the 'Weak Central Coherence Theory' first originated as a very general hypothesis of difficulties in global processing. Since then many studies have challenged the theory and in doing so have helped to clarify the concepts within. Such as: studies in

boundary extension by Chapman et al., (2005) demonstrating that boys with ASC had a similar boundary extension to typically developing controls, therefore challenging the limited visual attention spread hypothesis by Mann and Walker (2003); research by Rodgers (2000), where a hierarchisation deficit model (specifically failing to place special status on elements of perceptual stimuli) appeared to be a more satisfactory explanation of visual functioning than weak central coherence; studies that have failed to demonstrate superior visual performance (Brian & Bryson, 1996); and failed contextual suppression, where deficits in suppression were not seen across all participants with ASC (the ability to reject irrelevant ambiguous information) (Norbury, 2005). Norbury also noted that only those with decreased functional language demonstrated weak central coherence, thus suggesting a link to language ability. Moreover, Roper & Mitchell (2001) were unable to replicate the susceptibility to illusions reported by Happé (1996), and using size illusions (Muller-Lyer, Ponzo, Hat and Titchener's circles, Roper & Mitchell (2001) went on to demonstrate that individuals with ASC were just as susceptible to illusions as TD. The theory has now evolved into a theory of more superior local processing, less of a deficit than a different processing style and part of multi-deficit theory that no longer tries to explain the differences of ASC in one overarching theme (Rajendran & Mitchell, 2007).

Weak central coherence is widely reported and prevalent in ASC (Pellicano et al., 2006), has been found to have moderate correlations to the 'Theory of Mind' (Burnette et al., 2005), and has been positively correlated with better planning, inhibition and shifting abilities in executive functioning (Pellicano et al., 2006). Furthermore, support and interventions often include remediation in this area, such as books based on seeing the 'big picture' in social awareness training (Collucci, 2011), or comic strip conversations to help individuals see other parts of a situation (Gray, 1994).

When comparing the Central Coherence Theory to Cramer's (2013) criteria above it fulfils five of the seven criteria: testability, falsifiability, heuristic value, applied value and parsimony. However, it does not appear to be universal or comprehensive, as it only explains the detailed focus cognitive style in ASC, and does not explain other symptoms such as the atypical sensory and motor behaviours that are also present.

The Executive Functioning Theory (or Executive Dysfunction Theory) – 1991

In contrast to other theories of ASC, the Executive Functioning Theory was not born from neurotypical research, but rather specific brain injury of the frontal lobe. Patients who presented with such brain damage also exhibited symptoms similar to those lacking in 'Theory of Mind', such as a need for sameness and difficulties in switching attention (Ozonoff, et al., 1991). Executive functioning can be described as the ability to maintain an appropriate problem-solving set for the attainment of a future goal (Rajendran & Mitchell, 2007), and is a reasonably

parsimonious umbrella term that covers other sub areas of executive functioning, such as initiation, shift, inhibition, sustaining attention, memory and emotional regulation. See Figure 4.



Figure 4 Executive Functioning

The Executive Dysfunction Theory in ASC is reported in detail by Hill (2004) and can help to describe some of the symptomology of ASC, such as rigidity and regulating emotion. Research into Executive Dysfunction in ASC has shown difficulties in: planning ability, assessed by Tower Tasks (Ozonoff & Jensen, 1999); mental flexibility, tested by the Wisconsin Card Sorting Test (WCST) (Hill, 2004; Shu et al., 2001; Verte et al., 2006); and inhibition, assessed using the Stroop effect (Ozonoff & Jensen, 1999). Executive functioning assessments such as the Behaviour Rating Inventory of Executive Function (Gioia et al., 2000) profile have also suggested global executive dysfunction in ASC. Moreover interventions have been trialled to support executive functioning in the classroom, such as flexibility and problem-solving in ASC (Kenworthy et al., 2014).

However, research has disproved the 'Executive Functioning Theory' as many studies have shown that executive functioning difficulties do not affect all individuals with ASC (Liss et al., 2001; Pellicano et al., 2006). For example, using the Stocking of Cambridge evaluation (a computerised version of the Tower Task) no planning deficit has been identified (Goldberg et al., 2005).

When comparing this theory to Cramer's (2013) criteria above it appears to fulfil five of the seven: precision and testability, parsimony, falsifiability, heuristic value and applied value. Nonetheless, this theory is not universal and other ASC communication difficulties, such as the pragmatics of

language, with only social inference and not inference being impaired (Martin & MacDonald, 2003) and sensorimotor difficulties, such as gait, low tone and motion processing, are not accounted for in this theory.

The Amygdala Theory – 2000

The amygdala is considered to be the part of the brain partially responsible for social function, the 'social brain'. The 'Amygdala Theory' is based on abnormal functioning of the amygdala and its role in social intelligence (Baron-Cohen et al., 2000). See Figure 5 for amygdala location within the brain.



Figure 5: The location of the Amygdala

The Amygdala theory is based on the increased attentiveness and decreased aggression that occurs upon amygdala stimulation. This theory helps explain the emotion recognition deficits affiliated with the ToM within ASC, as there is evidence to suggest that the amygdala plays a large part in primitive social and emotional awareness (Brown & Shafer, 1998). A review by Phelps & LeDoux (2005) has illustrated the central role the amygdala plays in implicit emotional learning and memory, emotional modulation of memory, emotional influences on attention and perception, emotion and social behaviour, and emotion inhibition and regulation. Moreover, microscopic pathological differences have been found in the amygdala in the ASC brain, where there is an increase in cell density in the presence of normal amygdala volume (Bauman & Kemper, 1994; Rapin & Katzman, 1998) and a significant decrease in bilateral amygdala. Studies have also shown differing amygdala activation to facial perception in ASC (Schultz, 2005), and a positive correlation between greater amygdala activation and social anxiety in ASC. Further evidence can be seen in functional MRI data, where individuals with ASC did not activate the amygdala at all when shown visual representations of two tasks involved in deriving social relevant information from facial stimuli (Baron-Cohen et al., 2000). Deficits in the amygdala have also been applied to a deficient reward system, with suggestions that interventions that increase motivation may be a way forward (Kohls et al., 2013).

Accordingly, such unequivocal, universal, biopsychological evidence could also be used to disprove the Amygdala theory, i.e. where individuals with ASC do access and use the amygdala during emotional, attentive and social tasks; this has not been shown.

When comparing this theory to Cramer's (2013) criteria above it appears to fulfil five of the seven: precision and testability, falsifiability, applied and heuristic value and universality. Yet the 'Amygdala Theory' is not comprehensive, as it does not explain the atypical sensorimotor behaviours that are also present in ASC. The theory is also not parsimonious, as in addition to the differences in the amygdala in ASC, other areas of the brain, such as the cerebellum (Fatemi et al., 2012) and the basal ganglia (Estes et al., 2011), have also shown pathological disparities in ASC, therefore, this theory alone appears too simple.

The Extreme Male Brain (EMB) Theory

(The Empathising-Systemising Theory, E-S Theory) – 2002

This theory, proposed by Baron-Cohen in (2002), simply classifies individuals predominantly into two categories: empathisers and systemisers. When charted on a grid, with axes of empathising versus systemising, those with ASC fit neatly into the extreme systematic area, where the other four areas are made up of extreme empathising, female brain, balanced brain, and male brain. See figure 6: Baron-Cohen's depiction of the incrementum.



Figure 6: A depiction of the two types of brain where type B is the "balanced brain" (equal systemizing and empathizing ability); type E is the "empathizing" brain, coded female; type S is the systemizing brain, coded male; and the "extreme type S" brain is coded as autistic

When looking at functional aspects of the brain, females are purported to have more of an empathic brain, and males a more systematic brain (Lawson et al., 2004). When comparing the brain of individuals with ASC, in this theory they appear to have impaired empathising and increased systemising with differences in mind reading, face reading, empathy, language development and the pragmatics of friendship as empathic characteristics, and islets of ability,

attention to detail, toy preferences, preferences for rules, obsessions with closed systems, and collecting as systematic characteristics (Baron-Cohen, 2002). Thus, appearing to fit the male profile in an extreme form. Findings have been reached by using a number of different tests such as: the Empathy Quotient, where females scored more highly than males, and individuals with ASC scored even lower than males (Baron-Cohen and Wheelwright, cited in Baron-Cohen, 2002); the 'Reading the Mind in the Eyes test', where females scored higher than males, and males scored higher than ASC (Baron-Cohen, 2002); tests of intuitive physics, where males score higher than females, and individuals with ASC score higher than females, and individuals with ASC score higher than females, and individuals with ASC score higher than males (Baron-Cohen et al., 2001); and also on a neurological level, areas of the brain such as the prefrontal cortex and superior temporal gyrus, that are on average smaller in males than in females are smaller in individuals with ASC than typical males; whilst regions of the brain that are on average larger in males than in females, such as the amygdala, cerebellum and overall brain size/weight, are larger in individuals with ASC than typical males.

In order to disprove the EMB Theory individuals with ASC would need to be able to deal with unpredictable and non-routine events, and / or have empathy. It would be doubtful these individuals would have ASC in the first instance, particularly as such symptoms form part of the DSM-5 Criteria for ASC (APA, 2013), such as reduced emotions and unusual use of objects. However, females with ASC have been identified as having significant differences in brain anatomy from controls in brain regions reported as also *abnormal* in males (Craig et al., 2007), and those individuals with ASC who experience echoemotica (described as taking on another person's emotions without realising it is not their own, Shore, 2003: p200), also might not fit into the EMB theory as they very much feel emotions of others'. Additionally studies have perhaps falsified the EMB theory, by testing associations between pre-natal testosterone and visual-spatial tasks, and pragmatic language. Both studies found no relationship (Falter et al., 2008; Whitehouse et al., 2010).

In addition to ASC, the EMB theory has been applied to other areas of study, such as exploring differences in male and female characteristics when regarding conduct problems and depression (Zahn-Waxler et al., 2008), and gaining a better understanding of emotional intelligence and the impact on job performance (Joseph & Newman, 2010).

When comparing this theory to Cramer's (2013) criteria above it appears to fulfil six of the seven: precision and testability, parsimony, falsifiability, applied and heuristic value and universality. The EMB Theory also relates well to the Weak Central Coherence Theory as it posits superior attention to detail. Yet, it is not comprehensive, as the theory does not explain the atypical sensorimotor behaviours that are also present in ASC. Moreover, this theory perhaps does not explain those individuals with ASC who have obsessions with unpredictable animals such as horses, those without ASC who are highly empathic physicists or mathematicians and a shrinking ratio of males to females being diagnosed with ASC.

The Multi-Deficit Theory

When considering each of the above theories they all appear to be testable and falsifiable with both high heuristic and applied value. Furthermore, with the origination of each theory, understanding of the difficulties within ASC has evolved and progressed. The theories have been parsimonious with the exception of the amygdala that appears to be too simple. However, few have been universal, such as the Executive Dysfunctioning Theory where generic difficulties with elements of executive functioning have been disproved, such as the ability to plan. However, all of the theories to date do not appear to be comprehensive with most appearing to only recognise the social awareness difficulties within ASC without successfully considering all of the non-social symptomology, such as repetitive behaviours, unusual motor mannerisms and atypical sensory responsivity. Consequently, some researchers have suggested that it is time to give up on finding a single unifying theory, as it may rather consist of a number of co-occurring, genetically distinct clusters of symptoms, thereby proposing a multi-deficit account (Happé et al., 2006; Happé and Ronald, 2008; Goodman, 1989). Given the heterogeneity within ASC, multi-impairment and pervasiveness of the condition, in addition to often conflicting results between studies, a multideficit theory would universally incorporate such diversity. A multi-deficit theory is at the opposite end of the theory continuum to a specific deficit, such as executive functioning or theory of mind. It would seem that the first hard evidence for such as theory came from Pellicano, et al., (2006) who found that when age, verbal ability and non-verbal ability were taken into account, few associations were found between central coherence, theory of mind and executive functioning. Furthermore, in itself the criteria for ASC covers more than one domain: deficits in socialemotional reciprocity, non-verbal communication, maintaining relationships, stereotyped and repetitive motor movements, insistence on sameness, highly restricted interests and atypical sensory reactivity. Therefore, in order to be diagnosed with the condition there should be evidence of multi-deficits.

When comparing this theory to Cramer's (2013) criteria above it appears to fulfil two of the seven: Comprehensive and universal. However, there is no simplicity to a multi-deficit theory and without specific hypotheses to explore, the ability to test this theory would be virtually impossible, with very few studies investigating a multi-deficit account. Furthermore, in order to falsify a multideficit account, a specific deficit would need to be isolated, as only specific impairments within the multi-impairment would be heuristic. A later study by Pellicano et al. (2010) would be an example of this, where a link was demonstrated between the multiple cognitive domains in ASC, suggesting that rather than ToM, executive functioning and central coherence cooccurring, early domain general skills play an important role in shaping a child's ToM. Furthermore, multi-deficit accounts have implications for treatment as therapy that may be effective in one domain, or for some individuals, may be ineffective for others.

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Additional Theories

In addition to the above theories, other ASC theories have been proposed such as the social motivation theory (Chevallier et al., 2012) and the alexithymia theory (Bird et al., 2010). With regards to the social motivation theory, Chevallier et al., (2012) proposed that it is a deficit in social motivation leading to weaker social orienteering that ultimately induces social isolation in ASC. Whereas the alexithymia theory suggests that it is alexithymia (a difficulty in recognising or distinguishing and describing feelings), and not autism, that predicts poor recognition of emotional facial expressions. However, both of these theories are associated with the amygdala and so have not been reported discretely here. For example, the aforementioned deficient reward system in the amygdala theory forms the basis of social motivation (Kohls et al., 2012) and alexithymics have reduced amygdala activity when viewing a sad face, or one that evokes disgust in comparison to controls (Kano & Fukudo, 2013).

1.2 The role of sensorimotor difficulties in autism spectrum conditions

None of the above theories, when judged on merit using Cramer's (2013) criteria, appear to be a sound theory for ASC as they do not incorporate all of the symptoms associated with ASC, with the exception of the multi-deficit theory that still lacks many of the criterion. Moreover, progress in defining an effectual theory may have indeed been hampered by the lack of consideration for the non-social difficulties seen in ASC; repetitive behaviours, narrow circumscribed interests and sensory difficulties. Yet often it is difficulties in these areas that are first likely to be present. For example, head lag in infants (defined as the head lagging behind the trunk in a pull-to-sit position) (Flanagan et al., 2012), persistent asymmetry when lying and early impairments in rolling over from back to stomach (Teitelbaum et al., 1998) and motor delays being significantly more likely to be reported first by parents (at a mean age of 14.7 months), as opposed to language development and social relatedness (Chawarska et al., 2007). Subtle symptoms of atypical sensory modulation are also present between 9-12 months of age, such as poor visual orientation, excessive mouthing of objects, delayed response to name and social touch aversions (Baranek, 1999). Early development theories such as Jean Piaget's developmental stage theory, that proposed sensorimotor integration was central to neurotypical development, also supports such findings (Piaget, 1976). Hence, although previously not considered as central to the aforementioned theories of ASC, it appears that sensorimotor difficulties could be a promising explanation for the development of ASC.

The research presented in this thesis aims to address the restricted integration of these nonsocial sensorimotor adversities into theories of ASC, and in doing so presents a new theoretical framework with which to explore and understand how ASC develops: 'The Sensorimotor Theory' of ASC. This theory and main hypotheses explored within this thesis are set out in a detailed literature review in **Output 1**.

- 1.3 Within this context the overall objectives of the thesis are to:
- Analyse evidence of motor coordination and sensory difficulties in autism spectrum conditions (ASC), from both a psychological and biological perspective, with particular reference to sensorimotor differences
- II. Demonstrate the presence of sensorimotor difficulties in ASC and their association with the severity of ASC symptoms
- III. Show associations between Developmental Coordination disorder (DCD) and symptoms of ASC to highlight that good motor coordination skills are important for effective social skills and empathy
- IV. Compare and contrast ASC and DCD to help define more specifically the differences between the two conditions with regards to their sensorimotor foundations
- V. Critically evaluate the Sensorimotor Theory against Cramer's Criteria (2013)
- VI. Consider current intervention for sensorimotor difficulties in ASC and trial a possible intervention to help lessen the symptoms and make key recommendations for the future
- 1.4 In bringing together a coherent body of research on sensorimotor skills in Autism Spectrum Conditions it is felt that the work:
 - Provides evidence of training in a number of research methods appropriate to the discipline and area of study
 - Provides evidence of knowledge in the general field of autism spectrum conditions and other neurodiversities impacted by sensorimotor impairment
 - Demonstrates evidence of originality (as evidenced through publication in peer reviewed journals and through the impact of the research on practice)
 - Demonstrates the ability for critical independent judgement in comparing and contrasting Autism Spectrum Conditions to other sensorimotor disorders, in providing a theoretical framework and a reflective evaluation of researcher development
 - Demonstrates a common theme with a link throughout the work
- 1.5 The submission is subsequently organised into five main sections:
 - An autobiographical context for the research
 - A chronological description presenting the development of the portfolio
 - An analysis of the portfolio of evidence
 - An analysis of impact of the research in terms of its contribution to knowledge and practice and how others have appraised it
 - A review of the research methodology that links the outputs together
 - A critical reflection of development as Research Practitioner using Gibb's reflective cycle
 - A statement on the contribution of other authors to the outputs
 - Conclusions and suggestions for further research

2. Autobiographical context

- 2.1 Work by the author in this area of research dates back to 2012. Since then a substantial body of experience and expertise has been developed. This initially included working with a number of children and adolescents with ASC in the role of a Specialist teacher, training parents and educational professionals in strategies to support the difficulties faced by children with ASC. This not only helped to develop an understanding of the different types of support available to the education profession and parents, but also generated curiosity and interest in some of the consistent patterns in autistic childhood development. A deeper analysis into the biological and psychological constructs of these patterns observed in ASC then followed. Examples, referenced in Appendix 1, include:
 - a training certificate for the Early Bird Plus programme (National Autistic Society)
 - a training certificate for the Talk Boost speech and language programme
 - a training certificate for clinical and research reliability for the Autism Diagnostic Observation Schedule
 - a training certificate for clinical and research reliability for the Autism Diagnostic Interview-Revised
 - two peer reviewed conference posters
 - two training PowerPoints, for training education professionals in Autism Spectrum Conditions
- 2.2 Research on sensorimotor skills in ASC dates from 2013 and has focused on: confirming and advancing current research in identifying sensorimotor difficulties in ASC; using both psychological and biological knowledge to explore underlying commonalities; compare and contrast similar conditions; proposing a new theory framework; and finally possible intervention. A total of **7 outputs** in the form of published papers, excerpts in Educational Publications and papers in draft / review have been produced. Outcomes have been used to inform further research, clinical practice and intervention.

3 A Table to Show a Chronological description of ASC and sensorimotor skills research

Project	Funding	Contribution / Role
(a) 2013 The Role of Sensorimotor skills in Autism Spectrum Conditions		Devised the topic and was the lead author and researcher.
(b) 2014 Sensorimotor difficulties are associated with the severity of Autism Spectrum Conditions		Principal Investigator, responsible for drawing out key issues such as the somatosensory relationship between sensorimotor skills and autism severity.
 (c) 2015-2016 Sensorimotor difficulties are associated with receptive language in autism spectrum conditions. 		Principal Investigator, responsible for drawing out and writing up key issues
(d) 2015-2016 Dyspraxia and autistic traits in adults with and without autism spectrum conditions		Joint author in which the developed concept, and the evidence upon which it is based, draws extensively upon previous Outputs, experience and teaching
(e) 2016-2017 Sensory and Motor Differences in Autism Spectrum Conditions and Developmental Coordination Disorder in Children: A Cross- Syndrome Study	Free consultant advice through partnerships with Occupational Therapist and Paediatrician	Principle Investigator responsible for the main literature review and drafting the paper for publication.
 (f) 2016-2017 What works for Improving Outcomes for Children on SEND Support: A Rapid Evidence Assessment 	Funding from DfE tendered by Prof. Julia Carroll	Author for the specialised area of Physical and Sensory Needs, in addition to joint author of teacher survey
(g) 2016-2017 A Double-Blind, Placebo- Controlled, Crossover- Designed GABA Tea Study In Children Diagnosed With Autistic Spectrum Conditions	Coventry University £10,000 AcuMedic Ltd. £3,000	Principle Investigator responsible for the main literature review and drafting the paper for publication.

4. Analysis of the portfolio of evidence

Introduction

- 4.1 The portfolio of evidence consists of published outputs, including published peer reviewed journal articles (4) and peer reviewed journal articles currently under review / or final draft form for submission (3). Some of these outputs are also based on peer reviewed published conference proceedings (2: See Appendix 1) and have been chosen specifically to reflect the way in which the research has identified, highlighted, evidenced and cascaded understanding of the association of sensorimotor skills and autism severity.
- 4.2 The following analysis of the research portfolio is organised to reflect this logic pathway. It is presented in five parts:

Chapter 1: Analyse evidence of motor coordination and sensory difficulties in autism spectrum conditions (ASC), with particular reference to sensorimotor differences

Chapter 2: Demonstrate the presence of and association of sensorimotor difficulties with effective social skills and with the severity of symptoms in ASC

Chapter 3: Compare and Contrast ASC and DCD to help define more specifically the differences between the two conditions with regards to their sensorimotor foundations

Chapter 4: Critically evaluate the Sensorimotor Theory against Cramer's Criteria (2013)

Chapter 5: Consider current intervention for sensorimotor difficulties in ASC and trial a possible intervention to help lessen the symptoms

4.3 Portfolio of Evidence – Logic Pathway



Chapter 1: Analyse evidence of motor coordination and sensory difficulties in autism spectrum conditions (ASC), with particular reference to sensorimotor differences

- 4.3.1 **Output 1**, **'The Role of Sensorimotor difficulties in Autism Spectrum Conditions'**, is a peer reviewed published journal article (Frontiers in Neurology IF 3.184) that presents the new theoretical perspective and hypotheses for the research presented in the current thesis. Specifically, it looks in depth at the role of sensorimotor difficulties in ASC from both a psychological and biological perspective. The reason for writing this paper was to develop a better understanding of the observed motor difficulties that children with ASC present with. It was apparent during the literature search that there is a plethora of research evidence that identifies difficulties in both motor coordination and sensory responsivity in ASC as single entities, yet despite the two domains being intrinsically linked, there was far less research on sensorimotor difficulties as a single functioning unit. Where there was research, this appeared to be in its infancy.
- 4.3.2 This paper discusses the psychological association of sensory reactivity and motor coordination difficulties with ASC separately. However, the paper then goes on to explore the relationship between the sensory and motor domains with a considered analysis of the feedback and feedforward programs involved in this process. Typically developing (TD) individuals appear to be able to utilise sensory feedback to coordinate movement effectively, however as movements are repeated they instil a greater degree of automaticity which develops a feedforward program, which then only requires sensory feedback to modify and correct the already coordinated movement. It would appear that children with ASC have greater difficulty coordinating the initial movement effectively, but they do acquire a feedforward program in a similar fashion to TD children. However, once a feedforward program has developed it would seem that the sensory cues and feedback needed to modify and adapt movement are not incorporated as well into the feedforward program as TD individuals. Thus indicating difficulties with sensory feedback.
- 4.3.3 Biologically, the paper examines in greater detail the role of the cerebellum and basal ganglia in sensorimotor coordination, looking specifically at the large inhibitory neurones present in both of these areas of the brain, called Purkinje cells. These neurones are considered to be the sole output of all motor coordination in the cerebellar cortex and also form a large part of the basal ganglia; associated with movement automaticity and habit. Research has found that a decreased volume of Purkinje cells is a common finding in individuals with ASC. The inhibitory neurotransmitter GABA (gamma-aminobutyric acid) and the main excitatory neurotransmitter glutamate released by these neurones also play an important role in sensory discrimination and inhibitory neurotransmitter (GABA) levels appear to be largely reduced in ASC. Moreover, a GABA receptor gene, *GABRB3*, is considered one of the key candidate genes for ASC.

4.3.4 In conclusion, this paper increases recognition of the role of sensorimotor skills in ASC, and suggests further research is needed in sensorimotor feedback and inhibition in ASC, in addition to the impact of sensorimotor difficulties on autism symptomology. Thereby, beginning the research journey within this thesis.

OUTPUT 1 (Published)

'The Role of Sensorimotor difficulties in Autism Spectrum Conditions'

Hannant, P., Tavassoli, T., & Cassidy, S. (2016). The role of sensorimotor difficulties in autism spectrum conditions. *Frontiers in Neurology*, 7.

Author Contribution:

In this publication Hannant devised the topic and was the lead author. However, Hannant, Cassidy and Tavassoli worked jointly on conducting a literature search, compiling evidence from across different disciplines, and writing the manuscript. All three authors jointly revised the paper prior to publication.





The Role of Sensorimotor Difficulties in Autism Spectrum Conditions

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In addition to difficulties in social communication, current diagnostic criteria for autism spectrum conditions (ASC) also incorporate sensorimotor difficulties, repetitive motor movements, and atypical reactivity to sensory input (1). This paper explores whether sensorimotor difficulties are associated with the development and maintenance of symptoms in ASC. First, studies have shown difficulties coordinating sensory input into planning and executing movement effectively in ASC. Second, studies have shown associations between sensory reactivity and motor coordination with core ASC symptoms, suggesting these areas each strongly influence the development of social and communication skills. Third, studies have begun to demonstrate that sensorimotor difficulties in ASC could account for reduced social attention early in development, with a cascading effect on later social, communicative and emotional development. These results suggest that sensorimotor difficulties not only contribute to non-social difficulties such as narrow circumscribed interests, but also to the development of social behaviors such as effectively coordinating eye contact with speech and gesture, interpreting others' behavior, and responding appropriately. Further research is needed to explore the link between sensory and motor difficulties in ASC and their contribution to the development and maintenance of ASC.

Keywords: autism spectrum conditions, sensory, motor, sensorimotor, repetitive behavior, cerebellum, gamma-aminobutyric acid, social cognition

INTRODUCTION

Successful social functioning requires multiple skills, such as quickly seeking out and integrating information from pertinent social cues in order to plan and carry out an appropriate response. This involves effectively coordinating non-verbal and verbal language including posture, vocal-tone, facial gesture, and eye contact with speech during a social interchange. Hence, social reciprocity requires integrating a variety of sensory information from the environment to plan and execute movement effectively. If this ability was impaired, we could predict a whole host of difficulties, from performing simple actions (such as reaching for a cup) to having difficulty seeking out pertinent social cues and even difficulties interpreting others' behavior and responding appropriately. Furthermore, novel and challenging actions may be avoided and known perfected routines preferred.

Abbreviations: ASC, autistic spectrum conditions; GABA, gamma-aminobutyric acid.

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1

Current DSM-5 criteria (1) refers to Autism as a "disorder," however in this paper, we use the less stigmatizing term "condition," recognizing that Autism includes both strengths and weaknesses, while still being a medical condition for which individuals need support. Individuals with Autism Spectrum Conditions (ASC) have difficulties with social interaction and communication, repetitive behaviors, narrow circumscribed interests, and atypical sensitivity to sensory information (1). Three decades of research have attempted to uncover the cause of this diverse range of difficulties in ASC with little success. Thus some researchers have suggested that it is time to give up on finding a single unifying theory of ASC, as it may rather consist of a number of co-occurring, genetically distinct clusters of symptoms (2-4). Progress may have been hampered by lack of research into the non-social difficulties seen in ASC; repetitive behaviors, narrow circumscribed interests, and sensory difficulties. However more recent research, such as that by Gowen and Hamilton (5), has started to explore the contribution of sensorimotor difficulties (defined as an impairment in the pathway involving motor activity triggered by sensory stimuli) to the development and maintenance of ASC. This could provide a more parsimonious explanation (compared to multi-deficit accounts) of the social and non-social difficulties that come to develop in ASC. By examining both psychological and biological evidence, this paper therefore explores and hypothesises that motor coordination and sensory difficulties in ASC may be associated with the development and maintenance of ASC symptoms.

MOTOR COORDINATION IN AUTISM SPECTRUM CONDITIONS

Unusual motor processing is associated with ASC. Initial clinical reports of ASC reported general "clumsiness" in these individuals (6-9), and this has been corroborated in more recent research (10-15). Furthermore, Fournier et al. completed a robust meta-analysis including 51 comparisons of motor ability and deduced that individuals with ASC display a pronounced motor impairment compared to neuro-typical controls, with their motor skills often fall 1.5 SDs below the typical mean (16). Green et al. (12) and Miyahara et al. (17) were able to quantify the prevalence of motor impairment by administering assessments of coordination to children with ASC, concluding that approximately 80% had definite motor impairment with 10% being borderline. The prevalence of impaired motor processing in coordination, praxis, balance, and muscle tone in ASC is also echoed in a range of other studies (5, 18-21). More narrowly, Ming, Brimacombe, and Wagner went on to identify the prevalence of specific deficits in motor skills, with hypotonia (low muscle tone) and apraxia (impaired ability to execute planned movement) being the most common deficit (51 and 34%, respectively). Additionally, a review written by Gowen and Hamilton (5) demonstrated how a number of fine and gross motor movements had been identified and reported, such as: slower repetitive hand and foot movement, slower and less accurate manual dexterity, poorer ball skills (e.g., aiming and catching), unstable balance, impaired gait (e.g., tandem gait, heel, or toe walking), reduced coordination of locomotor skills (e.g., running and jumping), and hypotonia.

Motor abnormalities in ASC are present from early infancy (22–24), such as head lag in infants (defined as the head lagging behind the trunk in a pull-to-sit position) (25). Motor delays are significantly more likely to be reported by parents as the first area of concern at a mean age of 14.7 months (26). However, Teitelbaum et al. (22) also described, in detail, coordination differences between babies with and without ASC from as young as 6 months old, such as persistent asymmetry when lying and early impairments in rolling over from back to stomach.

SENSORY REACTIVITY IN AUTISM SPECTRUM CONDITIONS

Atypical sensory reactivity, as defined here as the psychological reaction that occurs when a person is exposed to sensory stimuli, is also associated with ASC. Pioneering reports of ASC described sensory "intrusions" (6, 7). Studies corroborate these initial clinical descriptions, showing that the prevalence of sensory reactivity problems in ASC is high. Caminha and Lampreia (27) reported a 69-80% occurrence of sensory dysfunction symptoms in ASC, while (28) noted that 95% of their sample had some degree of sensory processing difficulties. Findings from sensory symptoms in ASC, such as hypersensitivity and hyposensitivity, vary depending on modality tested, level of analysis, and method used. Research has, for example, shown the existence of basic sensory reactivity symptoms such as: hypersensitivity across visual (29), tactile (30-32), auditory (33-36) and olfactory (37) domains as well as hyposensitivity to olfactory and gustatory stimuli (38, 39); differences in perceptual function (40); and proprioceptive impairment (ability to determine where the body is in space) (41, 42).

More narrowly with respect to visual processing, although visual acuity seems to be typical in individuals with ASC (43, 44, 45), children with ASC statistically; (a) score higher on the Embedded Figures test, thereby demonstrating greater field independence, or the ability to see objects as discrete and distinct from their background as opposed to central coherence (46, 47); (b) exhibit faster visual search times (47, 48), have enhanced discrimination ability (48); and (c) detect more unattended changes in natural stimuli when compared to typical developing children (49). Such findings of an apparent superior performance in basic sensory perception led Mottron et al. (50) to suggest that individuals with ASC have "enhanced perceptual functioning" (51). There is however evidence to demonstrate diminished performance on higher order visual processing tasks such as visual spatial and visual motion processing (52-54). Some studies have indicated that adults with ASC are impaired at motion processing but retain intact form processing (perhaps reflecting selective dysfunction of the magnocellular pathway - the motion processing pathway) (52, 54-56). Other studies have found atypical visual global processing within the dorsal visual pathway (57), or have found that attentional and cognitive demands of the tasks might contribute to differing results (58). For a comprehensive review of vision in ASC, see Ref. (53). Taken together, basic early stage visual processing seems to be similar to or superior in individuals with ASC; however, there appears to be compromised higher order visual processing, which could be linked to difficulties with temporal sensorimotor skills, such as motion processing.

Regarding auditory processing, findings are similar in nature with intact or enhanced basic auditory perception and potential difficulties with processing higher order complex sounds. For example, individuals with ASC show superior pitch processing (55, 59, 60). O'Riordan and Passetti (61) also report greater auditory discrimination ability in children with ASC, and Järvinen-Pasley et al. (62) show superior perceptual processing of speech in children with ASC. However individuals with ASC also show difficulties in filtering complex auditory sound such as speech from background noise and therefore can often have difficulty attending to or registering auditory information (63, 64).

In tactile processing results have also been mixed, showing hypersensitivity (31, 65, 66) to basic stimuli, as well as no differences (61). Another line of research shows differences in adaptation toward touch; Tommerdahl et al. (66) showed that participants with ASC outperformed controls in tactile acuity after short adaptation to a vibrotactile stimulus period of 0.5 s. In addition, they demonstrated that individuals with ASC do not show enhanced spatial localization after being adapted to tactile stimulus, which occurs in typical controls. A more recent study also showed differences in tactile processing in children and adults with ASC (67). Differences in auditory and tactile processing might be associated with sensorimotor difficulties in ASC as well as visual differences.

The chemical senses have been investigated less than other senses, showing mostly impaired or intact processing in ASC depending on measure used (38, 39). Suzuki et al. (39) for example, reported impaired odor identification using the University of Pennsylvania Smell Identification Test in adults with ASC. A second study by Bennetto et al. (38) reported that adolescents (10-18 years) with ASC were less accurate in olfactory identification. However olfactory detection thresholds seem to be intact in individuals with ASC (39, 68). Regarding taste processing, identification seems to be impaired in individuals with ASC. Bennetto et al. (38) found that adolescence with ASC are less accurate in identifying sour and bitter tastes but showed similar identification for sweet and salty tastes. In line with this, Tavassoli and Baron-Cohen (68) found that adults with ASC had difficulties in identifying bitter, sour, and sweet tastes. Moreover, on sensory questionnaires, individuals with ASC are reported to show more olfaction and taste sensitivity compared to individuals without ASC (28, 69-72). Individuals with ASC (55%) for example present with more clinical symptoms in smell/taste sensitivity on the Short Sensory Profile compared to children with Sensory Processing Disorder (32%) (73).

Kern et al. (74) reported that abnormal responsivity in each of the main sensory modalities (auditory, visual, touch, and oral) was not independent, showing significant correlations between them; suggesting that sensory responsivity dysfunction in ASC is global in nature. This is further supported by the recognition of increased rates of synesthesia in ASC (a condition in which a sensation in one sensory modality triggers a perception in another) (75). Additionally, a study by Stevenson et al. (76) recently demonstrated reduced multisensory integration [described as the process whereby information from all the different sensory modalities are combined to influence perception, decision and behavior (77)] in ASC, by using a sound-induced flash illusion as a measure. However, sensory abnormalities in ASC appear to have the potential to reduce with age: a cross-sectional, linear regression analysis with 104 participants, aged 3–56 years, suggested that sensory difficulties become similar to typical controls by the age of 33 years (78). A meta-analysis also showed that sensory symptoms seem to be most prevalent between the ages of 6–9 years of age (79).

IMPACT OF MOTOR COORDINATION AND SENSORY REACTIVITY IN ASC

Difficulties in motor coordination and sensory reactivity have both separately been associated with the severity of symptoms in ASC. Research and primary observations indicate that the range of motor difficulties experienced by children with ASC appear to affect the development and maintenance of their social and communicative difficulties. Examples of this include the significant impairments shown by children with ASC in skilled motor gestures, including imitation (80) and development of speech sound production (20). Children with motor coordination difficulties are less competent at recognizing emotions in others (81) and are more likely to have increased anxiety on the playground due reduced social interaction (24). Furthermore, research has shown a correlation between motor and praxis performance and social communication skills in ASC (82-87). Piek and Dyck expands on the correlation and long recognized comorbidity of Developmental Coordination Disorders (DCD) and ASC, suggesting that as the disorders typically occur together, they either have overlapping causes or that one disorder is a direct cause of the other (88). However, research also suggests that impairments in movement could be a fundamental cause of the social and communicative difficulties seen in ASC, as opposed to a peripheral feature of the condition [see Ref. (89)]. This theory is consistent with recent evidence that suggests children who display fine motor difficulties early in childhood (from 7 months old) are significantly more at risk of developing an ASC by 36 months (90). Cross syndrome studies have also shown that children with ASC have more difficulties in basic (91) and gestural motor skills (92) compared to children with Attention-Deficit Hyperactivity Disorder (ADHD) and/or DCD. These results suggest that early motor difficulties are associated with the development of social and communicative difficulties later in life. Accordingly, as autism severity is based on social communication impairments in current DSM-5 criteria (1), this may be of paramount importance in the development of ASC.

In addition to the challenging sensory hypersensitivity/overload and hyposensitivity experienced by individuals with ASC, research has also linked sensory reactivity disorders to social communication difficulties (85, 93, 94). Fitzgibbon et al. (95) proposed that both physical pain and social pain are processed atypically in individuals with ASC and insensitivity to pain, for example, could in turn limit empathy and understanding of pain in others. Recent research has also identified associations between sensory reactivity and the severity of autism such as Tavassoli et al. (33) who reported that autistic traits defined by the Autism Spectrum Quotient, correlated positively with sensory over-responsivity, and Boyd et al. (96) specifically noted high levels of hyper responsive behavior predicted high levels of repetitive behavior. Other such studies include: Ashwin et al. (37), Kern et al. (74), Hilton et al. (85), Ben-Sasson et al. (79), Lane et al. (97), Siaperas et al. (98), and Tavassoli et al. (99).

SENSORIMOTOR INTEGRATION IN ASC

The evidence reviewed so far show that sensory and motor difficulties are prevalent in ASC and impact on social functioning (86, 93). Evidence also suggests that these difficulties are present from birth, e.g., pre-social skill deficits (29, 100), and increase the risk of developing ASC by aged 3 years (90). This indicates that there is a possible impairment in the process of sensorimotor integration [a brain process that allows, by complex neural operations, the connection of the sensory and motor domains (101), p. 427] that plays a fundamental role in the development of ASC. Although further studies are needed to explore whether sensorimotor integration difficulties are unique to ASC, Gowen and Hamilton (5) also proposed that altered sensory input and variability in motor execution "together" may play a pivotal role in ASC. Researchers have linked weaker praxis and motor performance to sensory reactivity in ASC (98, 102-104). Additionally, Siaperas et al. (98) found that children with ASC demonstrated significant impairment in both motor performance and proprioceptive and vestibular processing and thus suggested that sensory difficulties are not a peripheral, but a core feature of ASC.

Sensory feedback and movement are intrinsically connected (105), as the ability to plan and execute a simple movement effectively (such as reaching for a cup), requires sensory feedback (such as your position in relation to the cup as you reach for it) in order to effectively coordinate movement while performing the action (5, 105). Any error signal (such as missing the cup) at the final stage of movement is then processed and corrected. As movements are repeated in this fashion, they become automatic, and the delay caused by continuous sensory feedback is reduced, as the motor command (feedforward program) rapidly generates a prediction of the sensory consequences of the action (106, 107). For this reason, when sensory guidance is unreliable, slow, or associated with negative effect, both the ability to first acquire a motor command, in addition to regulating a stored motor command, would be impaired, leading to limited accuracy and flexibility (105). Therefore, deficiencies in sensorimotor integration would present as difficulties in effectively utilizing sensory feedback to correct movements, resulting in coordination difficulties and sensory reactivity abnormalities comparable to those seen in ASC (100, 105-112).

A number of studies have shown difficulties in sensorimotor integration in ASC. For example, Ronconi et al. (113) demonstrated that visual attention was impaired in children due to an imbalance of sensorimotor feedforward and feedback programs, by demonstrating a slower zoom-in and zoom-out mechanism in the eye. Schmitt et al. (114) and Mosconi et al. (115) demonstrated that those with ASC were significantly less accurate when moving their eyes from a central fixation to a peripheral target, showing increased saccade variability and difficulties in decelerating saccades. Wilkes et al. (116) also showed that children with ASC were delayed in initiating a saccade when following a moving light with their eyes compared to controls. Price et al. (117) demonstrated compromised visual sensitivity to human motion, and Glazebrook et al. (118) showed that adults with ASC had difficulty coordinating both hand and eye movements, taking significantly longer to complete integrated tasks than typical controls. These low level difficulties in initiating and adjusting saccades, and coordinating hand and eye movements, could explain a range of social and communication difficulties seen in ASC. For example, delay in looking to pertinent social cues (119-121), particularly for fast paced dynamic stimuli (122-125), with resulting difficulties in early social engagement and later ability to interpret others emotions and behavior (122, 123, 126, 127).

Studies have also shown that those with ASC have difficulty integrating sensory information in motor learning. For example, when children with ASC performed a motor learning task on a touch screen, the presence of a visual distractor did not impact their performance like with typical controls (109). Gepner suggested a correlation between visuo-postural detuning and ASC severity, whereby individuals with ASC had weaker postural stability and reactivity to environmental motion (128). Similarly, children with ASC are significantly less able to correct movements from visual compared to proprioceptive feedback (128-131). Studies have also shown difficulties specifically with motor movements, which require integrating visual cues or other sensory signals (18, 103, 132), and children with ASC have difficulty specifically when tracing shapes using feedback from a mirror image, and imitating others actions (133). These results all suggest that those with ASC do not tend to incorporate other sensory inputs, particularly visual feedback, into motor learning and have difficulty coordinating visual and motor movements. These difficulties could particularly impact social learning from imitation and integration of eye movements with gesture during social communication in ASC. However, further research is needed to explore this possibility.

Despite the possible interpretations of perceptual feedback being incorrect, Vandenbroucke et al. (134), by using a forcedchoice texture segregation task, went on to suggest that with considerable practice individuals with ASC were able to compensate for the imbalance in feedback and build on a feedforward program. Larson et al. (135) also noted that the mechanisms of acquisition and adaptation of feedforward programs are indistinguishable between children with ASC and typically developing children. Furthermore, Gowen et al. (132) demonstrated that in comparison to neurotypical controls, individuals with ASC rely to a greater extent on a goal directed pathway, established in part by a feedforward program. Similarly, Rinehart et al. (136) demonstrated an intact ability to execute programed movement but atypical movement preparation. Nazarali et al. (137) demonstrated that individuals with ASC have difficulty reprograming already planned movements when given additional sensory information, and Glazebrook et al. (118) showed that individuals with ASC can use sensory input such as vision and proprioception, although the greater visual-proprioceptive integration required the more time

was taken by the ASC group to perform the movements. Thus, with age, through continual practice, coping strategies and natural development, known, repetitive movements and feedforward motor programs appear to improve in ASC.

BIOLOGICAL BASIS OF IMPAIRED SENSORIMOTOR INTEGRATION IN ASC

In addition to psychological research given above linking sensorimotor integration to ASC, there is a substantial amount of biological evidence that collaborates this relationship. For example, difficulties in sensorimotor integration in ASC have been linked to the cerebellum, such as saccadic accuracy being connected to impairment of the error-reducing function of the cerebellum (114). Other examples include a correlation in the magnitude of cerebellar hypoplasia with decreased exploration in children with ASC (138), and an association of cerebellum volume with specific difficulties incorporating visual cues in motor learning (129). The cerebellum is reported to contain pathways that link sensory signals to motor areas in the brain (139), and these are important in controlling and coordinating movement (140). Stoodley and Schmahmann (141) specifically showed an anterior sensori-motor versus posterior cognitive/emotional dichotomy in the cerebellum. Research has also shown that the cerebellum has a fundamental role in maintaining the equilibrium between feedback and feedforward programs in sensorimotor integration, for example, Kawato et al. (142) found that the cerebellum was the most likely site for feedforward programs to be stored, by using functional magnetic resonance imaging (fMRI) to measure specific brain activity during coordinated and planned movement; Brooks (105) and Mostofsky et al. (143) noted that the cerebellum was responsible for triggering learned movement (feedforward programs); and Fuentes and Bastian (144) suggested that the cerebellum is intrinsic to predicting movement outcomes.

Abnormalities in the cerebellum of individuals with ASC are one of the most consistent neuroanatomical findings (145). McAlonan et al. (146) found structural abnormalities in the cerebellum with deficits in gray and subcortical white matter. Using fMRI, atypical patterns of both cerebral activation (indirectly detected by increased cerebral blood flow) and deactivation (signaled by decreased cerebral blood flow) have been noted in ASC: where cerebral activations during a simple motor movement were found to be mainly confined to the anterior cerebellum in TD adults but also spread to the posterior cerebella examined at autopsy showed clearly defined anatomic abnormalities; most commonly a significantly decreased number of Purkinje cells, a large inhibitory neuron thought to regulate motor function (145, 150, 151).

Moreover, the basal ganglia, which is considered to be reciprocally connected to the cerebellum (152), is also reputed to play a functional role in both motor and sensory control and integration (152, 153). More specifically, it is hypothesized that within the basal ganglia, there are two distinct striatal pathways that facilitate both movement and sensory representation. Although it is unclear whether these are distinct or seemingly intertwined (154), a direct pathway appears to be responsible for facilitating movement whereas an indirect pathway is thought to inhibit both competing motor programs and afford sensory control by filtering and gating sensory input (155). The basal ganglia have been shown to have decreased volume in ASC (156). Furthermore, the striatum, one of the largest components of the basal ganglia, is reported as having excess functional connectivity in ASC (157).

In addition to anatomical differences of the cerebellum and basal ganglia being associated with sensorimotor impairment in ASC, both brain regions contain large GABAergic inhibitory neurones. More specifically, Purkinje cells, considered the sole output of all motor coordination in the cerebellar cortex (158) and "medium spiny neurons," thought to form 95% of the striatum in the basal ganglia (159). The inhibitory neurotransmitter GABA (gamma-aminobutyric acid) and the main excitatory neurotransmitter glutamate released by these neurones also play an important role in sensory discrimination in ASC (160). GABA is known to decrease the firing of neurons (161), thereby reducing and inhibiting sensory feedback. Alterations in GABAergic transmission have been associated with sleep disorders (melatonin production) (162), mood disorders, anxiety and other hyperexcitable states such as epilepsy (163, 164). GABA levels have also been shown to be lower in the auditory and motor cortices of children with ASC with a mean deficiency of GABA equating to 22 and 11%, respectively, in comparison to TD peers (165) GABAergic functioning has been implicated in tactile reactivity (166, 167). Moreover, reductions in GABAergic system have been discovered in ASC brain tissue: with significant reductions in GABA_A receptors, 63% reduction in comparison to controls (168), and a reduction by 61% of the glutamic acid decarboxylase protein (the enzyme responsible for converting glutamate into GABA) (169). Similarly, increased glutamate levels (excitatory neurotransmitter) in blood and platelets have been found in ASC subjects, suggesting impaired conversion of glutamate to GABA, consequently increasing the excitatory state of the brain (169, 170). A GABA receptor gene, GABRB3, is one of the key candidate genes for ASC as found in humans as well as in animal models (171, 172). A study by Green et al. (173) demonstrated that participants with ASC also showed stronger activation of the amygdala toward sensory stimuli, which is thought to perform a pivotal role in emotion processing and decision-making; GABAergic neurons are also present in the amygdala.

Further evidence of an imbalance in these vital neurotransmitters in ASC arise from treatments for hyperactive disorders of the auditory system, such as tinnitus and hyperacusis (a lowered threshold for discomfort from sounds that typical individuals do not find unpleasant) (174), where the administration of benzodiazepines such as Clonazepam (an allosteric modulator of the GABA_A receptor) have been used to restore the balance between inhibition and excitation in the brain (174). Banji et al. (175) also demonstrated that induced cerebellar damage in mice instigated motor clumsiness, similar to that seen in ASC, the motor clumsiness was then reduced by treating the mice with green tea extract (*Camellia sinensis*). L-Theanine is a major amino acid component found almost exclusively in green tea (176–178) and blocks the binding of L-glutamic acid to glutamate receptors in the brain (176, 178, 179), thereby perhaps aiding the improvement in motor activity by increasing inhibition of movement.

IMPACT OF SENSORIMOTOR DIFFICULTIES IN ASC

Further evidence to support our hypothesis can be found in studies that have demonstrated sensorimotor difficulties and associated biological markers specific to ASC. In particular, difficulties with accuracy, speed, and initiation of eye movements; coordination of eye and body movements; and the ability to integrate visual information into motor learning could all have a profound impact on social learning opportunities during development and maintenance of social and communication difficulties in ASC. For example, difficulty quickly moving and correcting saccades could explain the well-established lack of attention to social cues in young children who go onto develop ASC (180, 181), with a cascading effect on later social development and learning (124). Difficulties integrating eye movements with body movements could account for social communication and interaction difficulties in ASC such as integrating eye contact with gesture and speech (1). Difficulties integrating other cues, particularly visual information in motor learning, could explain the challenges faced in social imitation in ASC [e.g., Ref. (182)]. Social imitation is key for social learning and could also contribute to the development and maintenance of social difficulties in ASC. Sensorimotor impairment could also explain other autistic traits such as echolalia and repetitive behaviors. A major study in brain anatomy using MRI by McAlonan et al. put forward that the impaired inhibition of sensory feedback through defective sensory gating (the brain's selective processing of sensory stimuli) found in ASC, could lead to difficulties where the individual is unable to inhibit repetitive thoughts, actions, or speech (146). Indeed, non-ASC related research has already demonstrated links between sensorimotor control and social behavior. Skewes et al. (183), by noting how the size and precision of a visual illusion may influence visual motor behavior, suggest that potentially, the way in which sensorimotor control adapts to the opinions of others may help facilitate smoother social interaction. Hoke et al. (184) identified that the integration of sensory and motor processing underlies social behavior in tungara frogs.

A number of theories have attempted to explain the development of ASC, including ability to understand mental states to predict others behavior (Theory of Mind) (185); impaired eye gaze detection (186) or lack of early social attention in favor of objects (124). These theories have failed to explain the wide range of difficulties seen in ASC from social communication to sensory reactivity and repetitive motor movements. Sensory and motor difficulties have also been considered largely peripheral to ASC, with atypical sensory reactivity only recently being added to DSM-5 diagnostic criteria (1). However, the wide range of difficulties in ASC could be explained by using a wider perspective: a central theory of sensorimotor integration impairment and the ensuing "chain" of likely misalignments and misjudgments that follow. Early development theories such as Jean Piaget's developmental stage theory proposed that sensorimotor integration was central to neurotypical development and where a child struggles to coordinate their initial sensory experiences, further stages of development will be impaired (187).

FUTURE DIRECTIONS

To the authors' knowledge, there are no studies that have explored the impact of sensorimotor difficulties (such as saccades or impaired motor learning) on the development or maintenance of core ASC symptoms. Sensory feedback and feedforward programs are seemingly pivotal to successful sensorimotor integration. A greater understanding of these systems and the impact of sensorimotor integration in ASC may be a crucial way forward to understanding the development and maintenance of this condition. For example, research that identifies a significant correlation between motor coordination, sensory reactivity, and the severity of ASC could demonstrate that these are fundamental and pervasive difficulties associated with and reflective of the scale of the condition. Similarly, further research into the varying degrees of sensorimotor difficulties and more specifically at the level the difficulties occur, such as the more complex higher order level including anticipation and timing, may also be crucial in identifying if and which area the sensorimotor chain is affected in ASC. There also appears to be a critical window for the impact of such sensorimotor deficits on cognitive and social development: below the age of two. Consequently, interventions incorporating both structured physical tasks and sensory environments below this age should have particular focus. Such intervention is also recommended in Barenek's review of the efficacy of sensory and motor interventions in ASC (2002), where it is noted converging evidence would suggest beginning sensorimotor inventions at an early age may be beneficial. Similarly, sensorimotor integration difficulties in comorbid conditions, such as dyslexia and dyscalculia should also be explored, as finding a recurrent thread to specific learning difficulties in ASC could alter the type and time of intervention.

Additionally, the apparent deficiency of the inhibitory neurotransmitter GABA in the cerebellum of ASC individuals should also be an area for consideration, as this could have a global impact on sensorimotor planning, cognitive and social development. The introduction of a non-evasive GABA substitute, such as oolong tea, could therefore lead to a decrease in sensory feedback, supporting an equilibrium with feedforward programing and ultimately moderate planned movement.

CONCLUSION

To conclude, we hypothesize that social communication, interaction difficulties, and repetitive behaviors in ASC appear to be associated with motor coordination and sensory reactivity, specifically attaining and coordinating the delicate balance between the feedforward programs and feedback systems of sensorimotor integration. However, once reached, research indicates that in comparison to controls, the feedforward program can be maintained and utilized just as efficiently in ASC provided environmental cues stay the same. Such conclusions are reinforced when listening to the lived experiences of ASC individuals; "I can do buttons up fine, unless I concentrate too hard."

All studies included in this research have been summarized in Table S1 in Supplementary Material.

AUTHOR CONTRIBUTIONS

PH: developed topic, literature search, compiled evidence, wrote the manuscript, and completed the table. SC: literature search, compiled evidence, critical feedback, wrote the manuscript, and completed the table. TT: literature search, compiled evidence, critical feedback, and wrote the manuscript. All authors read and approved the final manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at http://journal.frontiersin.org/article/10.3389/fneur.2016.00124

Table S1 \mid Summary of all studies included in the role of sensorimotor difficulties in the development of autistic spectrum conditions.

Summary of demographics, methods and outcomes of all studies included in research.

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Conflict of Interest Statement: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Chapter 2: Demonstrate the presence of and association of sensorimotor difficulties with effective social skills and with the severity of symptoms in ASC

4.4.1 Introduction

The aim of Chapter 2 is to demonstrate that sensorimotor skills are impaired in ASC and explore some of the difficulties associated with this. In doing so it draws on three outputs: Sensorimotor difficulties are associated with the severity of autism spectrum conditions (**Output 2**), Sensorimotor difficulties are associated with receptive language in autism spectrum conditions (**Output 3**) and Dyspraxia and autistic traits in adults with and without autism spectrum conditions (**Output 4**)

4.4.2 Output 2, 'Sensorimotor difficulties are associated with the severity of autism spectrum conditions', is the empirical study that tests the hypotheses presented in Output 1 and is a peer reviewed published journal article (Frontiers in Neuroscience IF 3.398). This Output followed the presentation of data within a poster at the IMFAR conference Baltimore (International Meeting of Autism Research, Appendix 1). This paper adds to the literature by using a battery of questionnaires and observational assessments, both subjective and objective measures, to demonstrate that sensorimotor skills are impaired in ASC when compared to typically developing children, and sensorimotor skills are associated with autism severity, predicting the severity of ASC symptoms in children with ASC over and above verbal and non-verbal performance. Moreover, the paper accentuates distinct sensory elements that are strongly associated with ASC severity and anxiety. Causation of sensorimotor difficulties is not included in this study and therefore future research into sensorimotor causality is suggested by exploring the nature of the sensorimotor difficulties in ASC when compared to other related conditions (e.g. ADHD and Dyspraxia) and whether integrated sensory activities and temporal spatial coordination activities increase reciprocal social interaction capability (the focus of **Output 5**). This study also suggests that if sensorimotor difficulties are present at a child's three-year developmental review, the child's developmental progression should be monitored carefully for possible ASC symptomology.
OUTPUT 2 (Published)

'Sensorimotor Difficulties are Associated With the Severity of Autism Spectrum Conditions'

Hannant, P., Cassidy, S., Tavassoli, T., & Mann, F. (2016). Sensorimotor difficulties are associated with the severity of autism spectrum conditions. *Frontiers in Integrative Neuroscience*, *10*.

Author Contribution:

In this publication Hannant was the lead author responsible for drawing out key issues, such as the somatosensory relationship between sensorimotor skills and autism severity. Cassidy supported the assessments and jointly wrote the manuscript. Tavassoli reviewed the manuscript and advised on assessments and procedures. Mann supported in the participant assessment batteries.





Sensorimotor Difficulties Are Associated with the Severity of Autism Spectrum Conditions

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Present diagnostic criteria for autism spectrum conditions (ASC) include social communication and interaction difficulties, repetitive behavior and movement, and atypical sensory responsivity. Few studies have explored the influence of motor coordination and sensory responsivity on severity of ASC symptoms. In the current study, we explore whether sensory responsivity and motor coordination differences can account for the severity of autistic behaviors in children with ASC. Thirty-six children participated: 18 (13 male, 5 female) with ASC (ages 7-16: mean age = 9.93 years) and 18 (7 male, 11 female) typically developing (TD) children (ages 6-12; mean age = 9.16 years). Both groups completed a battery of assessments that included motor coordination, sensory responsivity, receptive language, non-verbal reasoning and social communication measures. Children with ASC also completed the Autism Diagnostic Observation Schedule (ADOS) and Autism Diagnostic Interview-Revised (ADI-R). Results showed that children with ASC scored significantly lower on receptive language, coordination, sensory responsivity and a sensorimotor subscale, Modulation of Activity (MoA) compared to the TD group. In the ASC group, MoA significantly predicted ASC severity across all ASC measures; receptive language and sensory responsivity significantly predicted parental reported autism measures; and coordination significantly predicted examiner observed reported scores. Additionally, specific associations were found between the somatosensory perceptive modalities and ASC severity. The results show that sensorimotor skills are associated with severity of ASC symptoms; furthering the need to research sensorimotor integration in ASC and also implying that diagnosis of ASC should also include the assessment of both coordination deficit and atypical sensory responsivity.

Keywords: autism, sensory, motor, sensorimotor, receptive language, social communication

INTRODUCTION

Social communication is pivotal to both a child's development and integration into society. Yet, those with an Autism Spectrum Condition (ASC) do not find it easy to communicate, interact and socialize with others: often finding it difficult to infer, intonate and interpret others' behaviors. For this reason, current diagnostic criteria for ASC (DSM-5, American Psychiatric Association, 2013) incorporate difficulties in social communication and interaction,

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as well as sensory responsivity and motor movements. Current DSM-5 criteria (American Psychiatric Association, 2013) refers to Autism as a "disorder", however in the current study, we use the less stigmatizing term "condition", acknowledging that Autism includes strengths as well as weaknesses, while still being a medical condition for which individuals need support. Much research has attempted to uncover the reasons behind the social and communicative difficulties in ASC, and has identified correlations between severity of ASC and either motor coordination or sensory responsivity separately (Ming et al., 2007; Tomchek and Dunn, 2007; Green et al., 2009; Caminha and Lampreia, 2012). More recently, research has begun to look in greater detail at the role sensory responsivity and motor coordination difficulties play together in ASC through sensorimotor integration (Dowd et al., 2012; Siaperas et al., 2012; Gowen and Hamilton, 2013). Sensorimotor integration can be defined as "a brain process that allows, by complex neural operations, the execution of a certain voluntary motor behavior in response to specific demands of the environment" (Machado et al., 2010). This study therefore simultaneously investigates whether sensory and motor differences predict the social communication and interaction difficulties seen in autism.

Unusual movements and sensory difficulties are associated with ASC. In addition to stimming and rocking, general "clumsiness" has been reported in individuals with ASC since the first pioneering reports describing this condition (Kanner, 1943; Asperger, 1944; DeMyer, 1976; Damasio and Maurer, 1978). This has been further documented in more recent research. For example, motor skill scores for children with ASC often fall 1.5 standard deviations below the typical mean (Staples and Reid, 2010; MacNeil and Mostofsky, 2012), and approximately 80% have definite motor impairment with 10% being borderline (Miyahara et al., 1997; Green et al., 2009; Fournier et al., 2010; Kopp et al., 2010; Whyatt and Craig, 2012; Gowen and Hamilton, 2013). Coordination difficulties in ASC are evident from early infancy (Page and Boucher, 1998; Ming et al., 2007; Kopp et al., 2010), with parents being significantly more likely to report motor delays as the first concern at an average age of 14.7 months (Teitelbaum et al., 1998). Initial clinical reports also described sensory "disturbances" in ASC (Kanner, 1943; Asperger, 1944). Studies indicate the occurrence of atypical sensory responsivity in ASC is high, with 69-95% showing sensory responsivity dysfunction (Matson et al., 2010; Bhat et al., 2011). Research has also demonstrated the presence of a variety of sensory responsivity impairments in ASC (Chawarska et al., 2007; Caminha and Lampreia, 2012), such as; hyper and hypo responsivity across tactile (Baranek and Berkson, 1994; Blakemore et al., 2006); visual (Nyström et al., 2015); olfactory (Suzuki et al., 2003) and auditory domains (Madsen et al., 2014; Takahashi et al., 2014; Tavassoli et al., 2014b); differences in proprioceptive impairment (the sense of relative position in space) (Tavassoli et al., 2014b); and perceptual function (Paton et al., 2012).

Difficulties in coordination and/or sensory responsivity appear to be highly prevalent in people with ASC. However, are difficulties in these domains associated with severity of ASC symptoms, particularly social and communication skills? Research suggests that motor coordination difficulties impact on a number of skills, which are key for effective social participation. For example, individuals with ASC have significant impairments in skilled motor gestures such as imitation (Mostofsky et al., 2006) and the development of speech sound production (Page and Boucher, 1998). Children who have motor difficulties, without a diagnosis of ASC, tend to show weaker empathy for others (Cummins et al., 2005) and show increased anxiety on the playground (Bhat et al., 2011). There is also evidence that early motor difficulties are key to the development of ASC; children who show fine motor difficulties in early childhood (from 7 months old) have a significantly increased risk of developing an ASC by 36 months (Landa and Garrett-Mayer, 2006). This research suggests that motor difficulties are not only highly prevalent in ASC, but might impact social and communication skills in those with and without ASC.

Research has also identified links between sensory responsivity and the severity of ASC symptoms. For example, a number of studies have shown that sensory responsivity impairments impact social interaction and communication skills (Hilton et al., 2007; Reynolds et al., 2011; Matsushima and Kato, 2013). A more specific example is shown in research by Fitzgibbon et al. (2013), who proposed that as physical and social pain appear to be processed differently in individuals with ASC, their insensitivity to pain could consequently limit the understanding and empathy for pain in others. Studies have also shown correlations between self-reported sensory overresponsivity and autistic traits in adults with ASC (Tavassoli et al., 2014b), and sensory disturbance and autism severity (Kern et al., 2007; Ben-Sasson et al., 2009; Boyd et al., 2010; Lane et al., 2010; Tavassoli et al., 2014a). Hence, difficulties in sensory responsivity have also been associated with severity of ASC symptoms, particularly social and communication skills.

Difficulties in motor coordination and sensory responsivity are highly prevalent in ASC, are each associated with severity of ASC symptoms, and there is preliminary evidence that early motor coordination difficulties increase risk of developing ASC (Landa and Garrett-Mayer, 2006). However, these studies have explored each of these domains separately, despite them being innately connected (Brooks, 1983). For example, sensory feedback is fundamental to planning and executing the movement of reaching for a cup (such as being aware of your own position both visually and proprioceptively; Brooks, 1983; Gowen and Hamilton, 2013). Any miscalculations (such as missing the cup) during the movement are then processed and modified accordingly. Movements repeated in this way then create a feedforward program that predicts the sensory consequences of the action. Movements become procedural and automatic, thereby reducing the delay of continuous sensory feedback (Wolpert and Flanagan, 2001; Todorov and Jordan, 2002). For this reason deficiencies in sensory guidance are likely to affect both the ability to acquire and modify a stored motor command, leading to limited flexibility and accuracy. This strongly suggests that sensory responsivity and

motor coordination should not be seen as distinct separate entities, but as a functional unit in the form of sensorimotor integration.

The high levels of comorbidity of ASC with sensory and motor difficulties that appear to impact adaptive functioning, in addition to their presence from birth (Brisson et al., 2012; Nyström et al., 2015), suggest that sensorimotor integration could play a key role in the development and maintenance of ASC rather than being just a symptom. Work conducted by Gowen and Hamilton (2013) support this theory; suggesting that atypical sensory input and variability in motor deployment "together" may play a crucial role in ASC. Additionally, a number of studies have shown a relationship between sensory responsivity and motor coordination delays in ASC. For example, using a cluster analysis to examine data from the Short Sensory Profile (McIntosh et al., 1999) and Vineland Adaptive Behavior Scales (Sparrow et al., 1984), Lane et al. (2010) suggested that motor coordination delays in ASC could be linked to under-responsivity in the vestibular sensory system. Glazebrook et al. (2009) demonstrated that adults with ASC had difficulty coordinating both hand and eye movements and Milne et al. (2006) found a significant relationship between visual motion responsivity and fine motor control by measuring both motion and form coherence in ASC. Gowen and Miall (2005) also identified impairments in the ability to integrate motor control successfully with sensory input by testing visually guided movement, speed complex movement, muscle tone, prediction, coordination and timing in ASC. Iwanaga et al. (2000) highlighted weaker sensorimotor integration in ASC by assessing sensorimotor functioning in higher functioning autism and autism participants using the Japanese version of the Miller Assessment for Preschoolers (Miller and Schouten, 1988). Difficulties integrating visual cues and other sensory feedback from the environment with motor movements have also been demonstrated (e.g., see Gowen et al., 2008; Dowd et al., 2012; Whyatt and Craig, 2012; Stins et al., 2015). Furthermore, specific areas of the brain associated with sensorimotor integration such as the cerebellum (Paulin, 1993; Glickstein, 1998) and the basal ganglia (Nagy et al., 2006; Chukoskie et al., 2013) have also shown abnormalities in ASC. For example 95% of autistic cerebella examined at autopsy showed clearly defined anatomic irregularities; most commonly a significantly decreased number of Purkinje cells, a large inhibitory neuron thought to regulate motor function (Bauman and Kemper, 2005; Amaral et al., 2008; Fatemi et al., 2012). A decreased volume in the basal ganglia has also been reported in ASC (Estes et al., 2011) in addition to the striatum, one of the largest components of the basal ganglia, having excess functional connectivity in ASC (Di Martino et al., 2011).

These studies have demonstrated that motor coordination and sensory responsivity are both key elements in ASC: often occurring together, working together and potentially influencing behavior together. Such difficulties could affect the necessary sensorimotor skills involved in non-verbal language and communication such as gesture and imitation, consequently leading to what is perceived as idiosyncratic behaviors i.e., eye gaze aversion, limited non-verbal communication and restricted facial gesture. These atypical behaviors in infancy could subsequently impact social learning opportunities, with a cascade effect on development of effective social communication skills (see Hannant et al., 2016, for a review).

Although research has already identified associations between either motor coordination or sensory responsivity with ASC severity in isolation, this study aims to replicate and extend these findings by looking at both these elements together. This will help future studies identify a more definitive point of difficulty within the sensorimotor chain in ASC. Specifically whether motor coordination, sensory responsivity and specifically sensorimotor integration (i.e., the neurology and connections between the two single domains) are associated with the severity of ASC symptoms. This study therefore explores whether: (1) there is a significant difference in sensory responsivity and motor coordination between children with and without ASC; and if (2) the level of sensory responsivity and motor coordination difficulties are concurrently associated with and can predict autism severity.

MATERIALS AND METHODS

Participants

The ASC group was comprised of 18 children, (13 male, 5 female) aged 7–16 (mean age = 9.9 years) and were recruited from local ASC support groups in Warwickshire, UK. Sixteen children with ASC had a pre-existing diagnosis of ASC from a trained clinician according to DSM-IV criteria. The remaining two children with ASC were currently being referred for a clinical diagnosis of ASC. ASC diagnosis was confirmed by the research team using the Autism Diagnostic Observation Schedule General—2nd Edition (ADOS-2; Rutter et al., 2012) and the Autism Diagnostic Interview—Revised (ADI-R; Rutter et al., 2005), administered by a research reliable rater.

The typically developing (TD) group was comprised of 18 children (7 male, 11 female), aged 6–12 (mean age = 9.2 years), recruited during a research event held at Coventry University, UK, involving a number of different research studies. The TD group was recruited to this event by advertising in the local media in Warwickshire, UK. The TD group included children with no disabilities or diagnoses.

Participants completed: a parent report measure of autistic traits (Social Communication Questionnaire, SCQ; Rutter et al., 2003), to ensure that no children in the TD group had significantly high levels of autistic traits; a parent report measure of sensory responsivity (Sensory Profile, SP; Dunn, 1999); a measure of visual motor integration (VMI; Beery and Beery, 2010); and a measure of motor coordination (Movement Assessment Battery for Children, MABC; Henderson et al., 2007). Standard measures of intelligence such as the full scale Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler and Hsiao-pin, 2011), which require a spoken response, appear to underestimate the abilities of children with autism (Bodner et al., 2014). Therefore, in the current study we matched participants in each group by age and using only the non-verbal

TABLE 1 | Demographic descriptives and group comparisons.

Group	Gender	Age in years	Non-verbal reasoning
ASC	13 M	9.93 ± 2.71	90.94 ± 13.28
(N = 18)	5 F		(71-112)
Control $(N = 18)$	7 M 11 F	9.16 ± 1.89	99.50 ± 12.68 (70–117)
Difference	$X^2(1, 18) = 4.05,$	$t_{(34)} = -1.00,$	$t_{(34)} = -1.98,$
	$\rho = 0.044^*$	$\rho = 0.325$	p = 0.056

Note: *denotes p < 0.05 Bonferroni Correction p = 0.025.

intelligence quotient (IQ) matrices subset of the WASI (Wechsler and Hsiao-pin, 2011) and measured receptive language IQ using the British Picture Vocabulary Scale III (BPVS-III; Dunn et al., 2009). Groups differed significantly on receptive language ability, and there was a marginally insignificant between group difference in non-verbal IQ (despite the non-verbal IQ range being similar between groups; Table 1). Therefore both receptive language and non-verbal IQ are included as covariates in the correlation and supplementary regression analysis reported below. There was a significant group difference in gender ratio (Table 1), however, there was no effect of gender on sensory responsivity or motor coordination measures in either the ASC group (MABC $t_{(16)} = 0.29$, p = 0.78; SP $t_{(16)} = 0.31$, p = 0.76) or the TD group (Movement ABC $t_{(16)} = 0.18$, p = 0.86; SP $t_{(16)} = 0.10$, p = 0.92). The TD group scored significantly lower on parent reported autistic traits than the ASC group $(t_{(24)} = 7.33, p = 0.000, d = 0.831)$, and no participants in the TD group scored above cut off indicating ASC on the SCQ (15). See Table 1 for characteristics of both groups.

Materials

Participants completed a battery of assessments, four of which were standardized (MABC, BEERY, BPVS, WASI) where a standardized score of 84 or below was considered below average and 115 or above considered above average. The other four assessments (ADOS-II, ADI-R, SCQ, SP) were criterion based with a given cut-off point. In this study raw scores were used on the MABC, in order to analyze findings in greater detail from each subset. The ADOS-II and ADI-R ASC measures were utilized in order to ensure full and robust measurement of ASC symptomology within the study. The SCQ was used as a measure of ASC symptomology across both the TD and ASC groups.

The Movement Assessment Battery for Children—2 (MABC 2; Henderson et al., 2007): This is a standardized assessment of motor coordination for children aged 3–16 years and is a revision of the Test of Motor Impairment (TOMI; Stott et al., 1984). It is comprised of three components: manual dexterity, ball skills, static and dynamic balance. Examples of test content include placing pegs onto a board, throwing a beanbag onto a target and walking heel to toe along a line. The MABC 2 was normed on 1172 children aged 3–16 years with and without disabilities. Internal Reliability includes internal consistency estimates (range = 0.92-1.00) and validity with the "Draw-a-Man" test = 0.66 (Kavazi, 2006).

The Beery-Buktenica Developmental Test of Visual-Motor Integration, Sixth Edition (BEERY VMI; Beery and Beery, 2010): This is a standardized measure of an individual's ability to combine visual perception (VP) and fine motor coordination for people aged 2–100 years. It is comprised of three parts: VMI, VP and fine motor coordination. The VMI assessment requires an individual to copy a series of developmentally progressive geometric shapes; the VP aspect involves identifying matching shapes; and the motor coordination subtest contains a variety of shape outlines that the individual draws lines within. The BEERY VMI (6th Edn.) was normed on 1737 individuals aged 2–18 years with and without disabilities, showing good inter-rater reliability (range = 0.993–0.98) and validity (range = 0.8–0.95; Beery and Beery, 2010).

Sensory Profile (SP; Dunn, 1999): A standardized parent report questionnaire for children aged 3-10 years, that assesses the frequency of a child's responses to differing sensory modulation, processing and emotional events itemized in 125 questions. The SP consists of three domains: a sensory responsivity domain, which includes auditory, visual, vestibular, tactile, oral and multi-sensory processing; a modulation domain, which includes modulating sensory responsivity with relation to endurance and tone, proprioception (body position), movement affecting activity, emotional responses and visual filtering; and a behavioral emotional response domain which considers the behavioral outcomes of sensory responsivity. Higher scores on the SP indicate behaviors closer to the norm, or average. Lower scores indicate greater deviation from the norm, and thus greater difficulties in a particular area of sensory responsivity. The SP was normed on 1187 children aged 3-14 years of age with and without disabilities. Internal reliability includes internal consistency estimates (range = 0.47 - 0.91) and convergent and discriminant validity was determined by demonstrating high correlations with scores on the school function assessment (Dunn, 1999). "Modulation of Movement Affecting Activity Level" (MoA) is a sub division of the SP that specifically measures the child's demonstration of activeness, and is considered a measure of the modulation of movement in relation to sensory responsivity or sensorimotor integration.

British Picture Vocabulary Scale—Third Edition (BPVS-III; Dunn et al., 2009): A standardized non-reading assessment of receptive language. Each item within the assessment consists of identifying the correct image out of four pictures provided, to match a given word that covers a range of subjects, such as verbs, animals, emotions, toys and attributes. The BPVS-III was normed on 1480 children aged 3–16 years with and without disabilities. Internal reliability = 0.91 and validity with the Wechsler (2005) Intelligence Scale for Children = 0.76 (Dunn et al., 2009).

The Social Communication Questionnaire—Lifetime (SCQ; Rutter et al., 2003): A standardized parent-report measure of autistic traits for children from 4 years of age. The lifetime form is composed of 40 year or no questions and is used as a screening tool indicating whether referral for diagnosis of ASC is warranted. Scores of 15 or above out of 40 indicate a possible diagnosis of ASC. Scores therefore provided an index of the number of ASC symptoms an individual exhibits, with a higher score indicating higher levels of autistic traits. Items include questions based on reciprocal social interaction, such as friendships, reciprocal conversation and patterns of behavior. The SCQ was normed on 214 children aged 2–18 years with and without disabilities. Internal reliability = 0.84-0.93 and validity with the ADI-R = 0.78 (Rutter et al., 2003).

Wechsler Abbreviated Scale of Intelligence—2nd Edition (WASI-II; Wechsler and Hsiao-pin, 2011): A brief standardized measure of verbal and non-verbal intelligence. The matrices subset was used in the current study to measure non-verbal reasoning in both groups. The WASI was normed on approximately 2900 individuals aged 6–90 years with and without disabilities. Matrix Internal Reliability = 0.87-0.94 and validity with the WRIT = 0.71 (Wechsler and Hsiao-pin, 2011).

The following diagnostic measures were also completed by the ASC group to independently confirm participants ASC diagnosis and indicate severity of ASC symptoms.

Autism Diagnostic Observation Schedule—2nd Edition (ADOS-II; Rutter et al., 2012): A standardized diagnostic instrument for diagnosis of ASC, and confirmation of ASC diagnosis for research purposes. It consists of a semistructured interview that provides a number of social presses and opportunities to code quality of social and communicative behaviors. The 2nd edition of the ADOS also includes a rating indicating the severity of ASC symptoms taking into account the person's age and expressive language level. The ADOS-II was validated on 381 individuals aged between 15 months to 40 years with and without disabilities, with a further 1139 children aged between 14 months to 16 years recruited to revise the algorithms. Inter-rater reliability showed over 80% agreement on all modules with a high level of discriminative validity between autism and TD resulting in specificities of 50-84 and sensitivities of 91-98 (Rutter et al., 2012).

Autism Diagnostic Interview—Revised (ADI-R; Rutter et al., 2005): A standardized diagnostic instrument for diagnosis of ASC, and confirmation of ASC diagnosis for research purposes. It consists of a detailed semi-structured interview to gather evidence from an informant (parent, sibling or partner of an individual) on an individual's current behavior and early development indicative of an ASC diagnosis. Interviews cover social and communication, repetitive stereotyped behaviors, sensory and motor skills, talents, and challenging behaviors. The ADI-R was validated on 50 children aged between 36–59 months with and without disabilities. Internal Reliability demonstrated 26 of 35 weighted kappa values were 0.70 or higher. The ADI-R also shows a high level of discriminative validity with Clinical Diagnosis with 24 out 25 children being correctly diagnosed using the ADI-R (Rutter et al., 2005).

Procedure

Ethical approval for the study was obtained from the local research ethics committee. After parental consent to take part in the study was obtained, the parent completed the ADI-R either

over the phone or in person with a researcher who was research reliable in both ADI-R and ADOS-II. The parent and child were then invited to a single assessment session at the University. During this session, the following assessments were carried out in random order, to counterbalance and combat order affects, by trained researchers: BEERY VMI, Movement ABC, ADOS-II, BPVS and WASI non-verbal subsets. During this time the participant's parent also completed the SP and SCQ. Before the assessment procedure each task was explained carefully and depending on autism severity, a visual timetable produced to help alleviate anxiety. During the test procedure each participant was invited to have a voluntary break after each assessment.

RESULTS

Analysis Approach

Data were analyzed using SPSS (version 22), and normality tests conducted using Skewness and Kurtosis outputs. All measured variables: ADI-R, ADOS, SCQ, MABC Total, BEERY VMI, Sensory Profile Total, MoA, BPVS III and Matrices did not deviate significantly from normal (z-scores were all <1.96). Following tests for normality, TD and ASC data (BPVS-III, MABC, SP, MoA, SCQ, BEERY VMI) were compared using Bonferroni corrected independent t-tests. Post hoc tests were then completed in order to identify specific components of the MABC and SP that differed between groups significantly. Cohen's d is used as an indicator of effect size, with 0.2 indicating a small, 0.5 medium and 0.8 a large effect. Where Cohen's d was >1 the difference between the two means was considered larger than one standard deviation. Pearson correlations between all measures (ADOS, ADI-R, SCQ, MABC, BEERY VMI, SP, MoA, BPVS-III, Matrices) were calculated for the ASC, TD and combined ASC and TD groups. These correlations were then followed up by two separate supplementary analyses: stepwise linear regressions, using autism severity as outcome measures (ADOS-II, ADI-R and SCQ), and the MoA, SP, MABC, BPVS and Matrices scores as predictors; and an additional correlation of the separate components of autism, coordination and sensory measures. Post hoc power analyses on the multiple regression model were conducted using G * Power 3 (Faul et al., 2007) to compute the achieved statistical power for each model. Results showed all models, with the exception of Model 1 in the ADOS model (statistical power of 0.67), achieved statistical power >0.89 at the alpha level of p = 0.05 with a sample size of 18.

Do Children with ASC Show Significant Sensory and Motor Difficulties?

Table 2 shows results of comparisons between the ASC and TD groups on all measures. Bonferroni corrected independent samples *t*-tests showed that children with ASC had significantly lower: receptive language ability (BPVS; $t_{(34)} = -4.00$, p < 0.001, d = 0.1.33); motor coordination skills (MABC; $t_{(34)} = -4.56$, p < 0.001, d = 1.52); sensory responsivity (SP; $t_{(34)} = -7.69$, p < 0.001, d = 2.56); (MoA; a sensorimotor component of

TABLE 2 Dependen	t variable of	descriptives	and compari	son of means.
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Group	BPVS standardized score	MABC composite total	Sensory profile total	SCQ score	BEERY VMI (ASC <i>N</i> = 17)	Modulation of Activity
ASC (N = 18)	88.56 ± 14.08	51.61 ± 15.69	228.22 ± 37.44	18.94 ± 7.94	84.24 ± 21.27	18.61 ± 3.17
Control ($N = 18$)	106.00 ± 12.02	74.28 ± 14.12	312.11 ± 27.19	3.83 ± 3.68	100.17 ± 8.93	23.11 ± 3.58
Difference	$t_{(34)} = -4.00,$	$t_{(34)} = -4.56,$	$t_{(34)} = -7.69,$	$t_{(24)} = 7.33,$	$t_{(21)} = -2.86,$	$t_{(34)} = -4.00,$
	p < 0.001,	p < 0.001,	p < 0.001,	p < 0.001,	p = 0.009,	p < 0.001,
	<i>d</i> = 1.33	<i>d</i> = 1.52	d = 2.56	<i>d</i> = 2.44	<i>d</i> = 0.97	<i>d</i> = 1.33

Note: Effect size (Cohen's d) is denoted in bold: 0.2 = small, 0.5 = medium, 0.8 = large. Bonferroni corrected p-value = 0.008.

the SP; $t_{(34)} = 4.00$, p < 0.001, d = 1.33); and higher parent reported autistic traits (SCQ scores; $t_{(24)} = 7.33$, p < 0.001, d = 2.44), than the TD group, all with medium to large effect sizes. After Bonferroni correction, there was a marginally nonsignificant group difference in visual-motor integration (BEERY VMI; $t_{(21)} = 2.86$, p = 0.009, d = 0.97).

Post hoc independent bonferroni corrected *t*-tests then explored which specific components of the MABC and SP were significantly different between the TD and ASC groups. For the MABC, results showed that the manual dexterity composite was significantly different between groups with large effect ($t_{(28.96)} = 5.562$, p < 0.001, d = 1.85) and the balance composite was also significantly different between groups with large effect ($t_{(34)} = 2.531$, p = 0.016, d = 0.84). However the aiming and catching composite was not significantly different between groups ($t_{(34)} = 1.999$, p = 0.054, d = 0.67; Bonferroni corrected p = 0.017).

For the SP, results are shown in descending effect size according to Cohen's *d*: poor registration $(t_{(25,92)} = 7.273, p = <0.001, d = 2.42)$, emotional regulation $(t_{(34)} = 6.788, p < 0.001, d = 2.26)$, low endurance $(t_{(24,42)} = 5.858, p < 0.001, d = 1.95)$, attention composites $(t_{(25,91)} = 4.738, p < 0.001, d = 1.58)$, sedentary $(t_{(34)} = 3.709, p = 0.001, d = 1.24)$ and fine motor skill composites $(t_{(34)} = 3.631, p = 0.001, d = 0.85)$ respectively were significantly different between groups with large effect. However, oral sensitivity $(t_{(34)} = 2.809, p = 0.008, d = 0.94)$, sensory seeking $(t_{(27,39)} = 2.392, p = 0.024, d = 0.80)$, and sensory sensitivity $(t_{(34)} = 2.114, p = 0.042, d = 0.71)$ composites of the SP were not significantly different between groups after Bonferroni correction p = 0.006.

Are Sensory Responsivity and Motor Coordination Associated with ASC Symptom Severity?

Pearson correlations were calculated in each group separately (ASC and TD) and with both groups combined (whole sample), between all variables (ADOS, ADI-R, SCQ, SP, MoA, MABC, BEERY VMI, BPVS and Matrices; **Table 3**).

Table 3 shows results of the correlation analysis. In the ASC group (n = 18), in addition to co-linearity between the three autism measures (ADI-R, ADOS and SCQ) and the two coordination measures (MABC and BEERY VMI), the SP and BPVS significantly correlated with the ADI-R (Current; SP r = -0.657, p = 0.002; BPVS r = -0.737, p < 0.001)

and SCQ (SP r = -0.836, p < 0.001; BPVS r = -0.561, p = 0.008) with medium to large effect. The MABC significantly correlated with the ADOS-2 (r = -0.647, p = 0.002) with medium to large effect, and the Matrices and BPVS with a small to medium effect (Matrices r = -0.495, p = 0.018; BPVS r = -0.464, p = 0.026). The Matrices also demonstrated correlation with the BEERY VMI in this group (r = -0.600, p = 0.005) with medium to large effect. In the TD group (n = 18) the SP showed some correlation with the SCQ with small to medium effect) r = -0.419, p = 0.042). The Matrices and BPVS (r = -0.656, p = 0.002) significantly correlated with medium to large effect. The BEERY VMI also demonstrated some correlation with the BPVS and Matrices with small to medium effect (Matrices r = -0.407, p = 0.047; BPVS r = -0.493, p = 0.019). When the TD group were added to the ASC group (n = 36) both the SP and MABC significantly correlated with autism severity levels in the SCQ (SP r = -0.893, p < 0.001; MABC r = -0.598, p < 0.001). Furthermore, when the groups were combined the SP also correlated with the MABC and BEERY VMI (MABC r = -0.542, p < 0.001; BEERY VMI r = -0.404, p = 0.008). The BPVS showed medium effect correlations across all variables (SCQ r = -0.645, p < 0.001; MABC r = -0.603, p < 0.001; BEERY VMI r = -0.542, p < 0.001; SP r = -0.544, p < 0.001; MoA r = -0.534, p < 0.001) and the Matrices demonstrated small to medium effect correlations across all variables when both groups were combined (SCQ r = -0.328, p = 0.025; MABC r = -0.518, p = 0.001; BEERY VMI r = -0.568, p < 0.001; SP r = -0.335, p = 0.023; MoA r = -0.376, p = 0.012; BPVS r = -0.539, p < 0.001).

The MoA in the ASC group (MoA; an independent sensorimotor variable within the SP) significantly correlated with all three ASC measures (ADI-R r = 0.482, p = 0.021; ADOS r = 0.518, p = 0.014; SCQ r = 0.540, p = 0.010) and the MABC (r = 0.477, p = 0.023). In the TD the MoA significantly correlated only with the BEERY VMI (r = 0.553, p = 0.009). However, MoA demonstrated significant correlation with the SCQ (r = -0.650, p < 0.001) and BPVS (r = 0.534, p < 0.001) with medium to large effect when the TD group's data were added to the ASC group's data, and small to medium effect with the MABC Total (r = 0.442, p = 0.004) BEERY VMI (r = 0.446, p = 0.004) and Matrices (r = 0.376, p = 0.012).

In the ASC group, supplementary analyses were then performed using stepwise multiple regression with autism severity as outcome measures (ADOS-II, ADI-R Current and

	ADIR TOTAL	ADOS TOTAL	SCQ	MABC TOTAL	BEERY VMI	SP TOTAL	SP MoA	BPVS
				ASC GROUP (n =	- 18)			
ADOS TOTAL	0.566**							
SCQ	0.831***	0.479*						
MABC TOTAL	0.324	0.647**	0.241					
BEERY VMI	0.112	0.336	0.015	0.611**				
SP TOTAL	0.657**	0.256	0.836***	0.087	0.054			
SP MoA	0.482*	0.518*	0.540*	0.477*	0.165	0.524*		
BPVS	-737**	0.375	0.561**	0.464*	0.379	0.339	0.302	
WASI MATRICES	0.076	0.031	0.036	0.495*	0.600**	0.065	0.246	0.297
				TD GROUP (n =	18)			
MABC TOTAL			0.264					
BEERY VMI			0.351	0.043				
SP TOTAL			0.419*	0.144	0.189			
SP MoA			0.253	0.181	0.553**	0.461*		
BPVS			0.024	0.299	0.493*	0.057	0.335	
WASI MATRICES			0.366	0.353	0.407*	0.249	0.254	0.656**
				Whole SAMPLE (n	= 36)			
MABC TOTAL			0.598***					
BEERY VMI			0.379*	0.572**				
SP TOTAL			0.893***	0.542***	0.404**			
SP MoA			0.650***	0.442**	0.446**	0.692***		
BPVS			0.645**	0.603**	0.542**	0.544**	0.534**	
WASI MATRICES			0.328*	0.518**	0.568**	0.335*	0.376*	0.539**

TABLE 3 | Correlation analysis (r) for autism symptom measures, coordination and sensory responsivity in autism spectrum conditions (ASC), typically developing (TD) and combined ASC and TD group.

Note: *p < 0.05, **p < 0.01, ***p < 0.001. Required $r \ge 0.522$ for sample size and $\alpha 0.05$ to achieve a Statistical Power = 0.8. Correlations in bold indicate results > required effect size.

SCQ), and the SP, MABC, BPVS and Matrices scores as predictors. BEERY VMI was not included as a predictor in this analysis, due co-linearity with the MABC. Therefore if included, the BEERY VMI would have reduced the statistical power of this analysis to find a significant effect of motor coordination with autism symptom severity.

Table 4 shows results of the supplementary stepwise regressions. In the ASC group the BPVS and SP were retained as significant predictors of ADI-R Total (Current); the MABC and Matrices were retained as a significant predictors of ADOS-II total; and the SP and BPVS were retained as significant predictors of the SCQ. The SP and BPVS were also significant predictors of autism levels in the SCQ when the TD group was added to the ASC group. In summary, results showed that the SP scores significantly predicted parent reported autism symptom severity (ADI-R and SCQ), and the MABC measures significantly predicted an in-person measure of autism severity (ADOS-II). The BPVS and Matrices predicted some, but not all, of the parent reported social and communication skills in ASC. Figure 1 demonstrates visually how the predictors correlated with the autism measures.

Table 5 shows results of a separate linear regression analysis for the main effect of MoA (an independent sensorimotor variable within the SP), predicting ASC symptom severity (ADOS, ADI-R and SCQ). Results demonstrated that MoA significantly predicted ASC symptom severity both in the ASC group across all diagnostic measures (ADOS, ADI-R and SCQ), and when the TD group's SCQ data were added to the ASC group's SCQ data.

Table 6 shows results of a further analysis in the ASC group, where individual ASC measures, coordination and sensory components were correlated in order to identify any specific links. Measures were divided into motor, sensory and sensorimotor. Results demonstrate significant correlations with autism measures in the sensorimotor components alone. Furthermore, vestibular (balance), auditory, tactile and oral motor responsivity appear to be correlated with more than one autism component.

DISCUSSION

The current study investigated whether children with ASC show significant difficulties in sensory responsivity and motor coordination compared to children without ASC, and whether these difficulties were significantly associated with ASC symptom severity. Results showed the children with ASC had significant motor coordination, sensory responsivity and receptive language difficulties compared to age and non-verbal IQ matched children without ASC. Analysis of the different subcomponents of sensory responsivity and motor coordination difficulties showed that the children with ASC had specific impairments in particular domains; the more significant sensory responsivity

Step	Variable	В	SE B	В	Cum R ²
		ADI-R Total (Cu	rrent) (<i>n</i> = 18)		
1	Constant	70.31	10.16		
	BPVS	0.50	0.11	0.74***	0.74
2	Constant	87.54	9.67		
	BPVS	0.39	0.10	0.58**	0.85
	SP TOTAL	0.12	0.04	0.46**	
		ADOS-II Tot	al (n = 18)		
1^	Constant	24.00	3.39		
	MABC TOTAL	0.21	0.06	0.65**	0.65
2	Constant	11.44	5.97		
	MABC TOTAL	0.29	0.06	0.87***	0.77
	Matrices	0.18	0.08	0.47*	
		SCQ (n	= 18)		
1	Constant	59.40	6.72		
	SP TOTAL	0.18	0.03	0.84***	0.70
2	Constant	69.93	7.23		
	SP TOTAL	0.16	0.03	0.73***	0.79
	BPVS	0.18	0.07	0.31*	
		SCQ with ASC and	TD group (<i>n</i> = 36)		
1	Constant	55.67	3.89		
	SP TOTAL	0.16	0.01	0.89***	0.80
2	Constant	63.32	4.59		
	SP TOTAL	0.14	0.02	0.77***	0.83
	BPVS	0.14	0.05	0.23*	

TABLE 4 | Stepwise multiple regressions for autism symptom measures in ASC group.

Note: *p < 0.05, **p < 0.01, ***p < 0.001. All Models with the exception of 1^ achieved >0.89 Statistical Power for a sample size of 18 and α 0.05 1^ Statistical Power = 0.67.

impairments in the ASC group were in poor registration, low endurance, emotional regulation, attention, sedentation and fine motor skills. The more significant motor coordination impairments were in manual dexterity and balance. These results confirm previous reports showing significant difficulties in sensory responsivity and motor coordination in those with ASC in comparison to TD controls, such as Matson et al. (2010) and MacNeil and Mostofsky (2012), respectively. Our analysis of the key areas of difficulty within these respective domains also mirror previous research, such as that by Tomchek and Dunn (2007), where 90% of ASC participants demonstrated a significant difference when compared to TD controls in the underesponsive domains of the SP and Siaperas et al. (2012) who showed that individuals with ASC presented with difficulties in both balance and fine motor skills.

Results also showed significant correlations between sensory responsivity, motor coordination and the severity of ASC symptoms in the ASC, TD and combined ASC and TD groups. Results demonstrated possible co-linearity between the BPVS and communication measures of autism (ADI-R and SCQ) and the Matrices and BEERY VMI, which are both measures of spatial reasoning. However, the correlation between the BPVS and Matrices was only significant, with medium to large effect, in the TD group: not the ASC group, perhaps indicating an association between non-verbal and verbal performance in this group alone. We were interested in whether sensory and motor difficulties could account for ASC symptom severity in ASC, TD and combined groups and therefore followed up these significant correlations with supplementary regression analyses, controlling for group differences in receptive language and non-verbal IQ. Results showed that difficulties in sensory responsivity and motor coordination in both the ASC and combined ASC and TD groups significantly predicted severity of ASC symptoms, over and above receptive language ability and non-verbal IQ. However, the presence of a significant predictive relationship depended on the type of measures used. Specifically, sensory responsivity difficulties significantly predicted the level of ASC symptoms in both parental report measures of ASC (ADI-R and SCQ), whilst motor coordination difficulties significantly predicted the level of autism symptoms measured by the ADOS-II, an in person measure of ASC through examiner observation. One could argue that this correlation between the parental measures and the observational measures is unsurprising. However, these measures were not analogous. For example, the ADOS-2 does not specifically measure coordination and the ADI-R does not specifically measure sensory responsivity. Therefore these correlations were unlikely to have occurred



ADI-R TOTAL Current; SPTOTAL, Sensory Profile Total; MABCTOTALCOMP, MovementABC Total Composite Score.

if sensory responsivity or motor coordination skills were in no way related to severity of autism symptoms. Additionally, a medium to large effect correlation between the SCQ, SP and MABC was apparent when ASC and TD data were merged. MoA, a subset of the SP that measures activeness and exploration, giving an indication of sensorimotor integration (sensory environment governing movement), was also associated with ASC severity across all diagnostic measures (ADOS-II, ADI-R and SCQ). The fact that the presence of a predictive relationship between sensory responsivity and motor coordination and autism symptoms differed according to the diagnostic measure used (ADOS or ADI-R), could have also been due to the differing properties of these instruments. For example, the ADOS-2 is a short time window of current behavior, approximately 1 h interaction with an examiner. This gives few opportunities for the child to display sensory abnormalities (except with a spinning disk and pin art during the break), or poor attention given the

TABLE 5	Overall model fit	beta values	t-values.	and <i>p</i> -values are	e displayed for	each diagnostic n	neasure.
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ASC symptom measure		Modulation of activity			
ADIR-TOTAL	$R^{2} = 0.321$	$F_{(1,16)} = 7.550$ $F_{(1,16)} = 5.751$ $F_{(1,16)} = 6.569$ $F_{(1,34)} = 24.849$	$\beta = -0.566, t = -2.748$	p = 0.014	
ADOS-TOTAL	$R^{2} = 0.264$		$\beta = -0.514, t = -2.398$	p = 0.029	
SCQ	$R^{2} = 0.291$		$\beta = -0.540, t = -2.563$	p = 0.021	
SCQ with ASC and TD group $(n = 36)$	$R^{2} = 0.422$		$\beta = -0.650, t = -4.985$	p = 0.0001	

All values indicate that Modulation of Activity is also a significant predictor of autism severity for each diagnostic measure. Note: Using G* Power 3 (Faul et al., 2007) an effect size (β) of 0.522 was considered significant at 0.8 Power and 18 ppts).

TABLE 6 Correlation analysis (r) for specific autism cor	mponents, coordination components and sensory	components in ASC group ($n = 18$).
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Measure	ADIRC	ADIRRSI	ADIRRSB	ADOSC	ADOSRSI	ADOSRRB	SCQ
				Motor Measures			
BEERY MC	0.207	0.009	0.032	0.252	0.380	0.032	0.032
				Sensory Measures	3		
BEERY VP	0.195	0.109	0.074	0.338	0.282	0.145	0.109
SPVIS	0.280	0.338	0.390	0.280	0.032	0.134	0.338
			S	ensorimotor Measu	res		
BEERY VMI	0.093	0.106	0.301	0.422*	0.372	0.057	0.015
MABCMD	0.152	0.331	0.006	0.595**	0.535*	0.200	0.256
MABCAC	0.253	0.076	0.174	0.309	0.117	0.188	0.092
MABCB	0.322	0.448*	0.322	0.531*	0.523*	0.344	0.238
SPAUD	0.652**	0.694**	0.521*	0.307	0.178	0.251	0.861***
SPVEST	0.201	0.179	0.590**	0.173	0.053	0.084	0.381
SPTOUCH	0.334	0.522*	0.626**	0.506*	0.365	0.069	0.615**
SPORAL	0.576**	0.310	0.693**	0.217	0.197	0.464*	0.574**

Note: *p < 0.05, **p < 0.01, ***p < 0.01. Required r = >0.522 for sample size and α 0.05 to achieve a Statistical Power = 0.8. Correlations in bold indicate results > required effect size. Key: ADIRC, ADI-R Communication; ADIRRSI, ADI-R Reciprocal Social Interaction; ADIRRSB, ADI-R Restricted Stereotyped Behaviors. ADOSC, ADOS Communication; ADOSRSI, ADOS Reciprocal Social Interaction; ADOSRB, ADOS Stereotyped Repetitive Behaviors. BEERY MC, BEERY VI Motor Coordination; BEERY VP, BEERY VI Visual Perception; BEERY VMI, BEERY VI Visual Motor Integration. MABCMD, MABC Manual Dexterity; MABCAC, MABC Aiming and Catching; MABCB, MABC Balance. SPVIS, Sensory Profile Visual; SPAUD, Sensory Profile Auditory; SPVEST, Sensory Profile Vestibular; SPTOUCH, Sensory Profile Tactile; SPORAL, Sensory Profile Oral Motor.

1:1 nature of the interaction. In addition scoring opportunities of sensory responsivity is limited; specifically the ADOS-2 only has two items, which take into account sensory abnormalities such as seeking behaviors. Hence, it is unlikely that the type of behaviors seen in the ADOS-2 would correlate with the SP, which takes into account sensory responsivity across a variety of context and time points in children with ASC, particularly as this measure was significantly characterized by poor registration and attention. In contrast, the ADI-R is a parent-report measure, with current behavior ranging from yesterday to months ago, with more emphasis and opportunity for parents to report on sensory and attention difficulties compared to the ADOS-2. Furthermore, the ADI-R places less emphasis on non-verbal language and gesture based on the proportion of algorithm questions (the ADOS-II having 50% of codes based on social interaction and the ADI-R Current algorithm 38%). Given that the ADOS-2 and ADI-R give different priorities to different behaviors and thus are utilized together as diagnostic instruments, could explain why sensory and motor skills are associated with different autism measures.

The results from the *post hoc* detailed analysis of the relationships between motor, sensory and sensorimotor abilities and the specific components of ASC, demonstrated a significant sensorimotor association with ASC severity across the ASC domains. Moreover, the specific sensorimotor domains correlated are all instrumental to somatosensory perception. For example tactile, acoustic and vestibular information respond using mechanoreceptors to skin or head displacement (Yeomans et al., 2002) and somatosensory awareness also occurs orally to ensure

alertness to objects in the mouth (Haggard and de Boer, 2014).

The strong correlation between both sensory responsivity and motor coordination with the SCQ; results from the supplementary analysis showing that sensorimotor skills, MoA, sensory responsivity and motor coordination all predicted measures of ASC symptoms over and above receptive language and non-verbal IQ both in the ASC group alone, and the combined ASC and TD groups; and the specific correlation of the senses involved in somatosensory perception such as touch, vibration and pressure, with the different components of autism, would suggest that sensory and motor difficulties may together, in the form of sensorimotor integration, significantly impact the severity of ASC symptoms. These results are consistent with previous research, such as that by Gowen and Hamilton (2013), who suggested that atypical sensory input and variability in motor deployment simultaneously could potentially impact on severity of ASC symptoms such as social and communication skills. Additionally, Ozonoff et al. (2008) demonstrated atypical MoA and exploration in children with ASC from an early age by showing toddlers with ASC who spent significantly more time spinning and rotating objects when compared to their TD peers, and Kawa and Pisula (2010) showed 4-5 year olds differed in both object exploration and time spent in more visually complex zones, which was significantly decreased in comparison to other areas. Research has also shown that this reduced exploration and activity shown in ASC individuals is linked to the cerebellum, an area of the brain known to be linked to sensorimotor integration (Glickstein, 1998), and that measures of decreased activity significantly correlates with cerebellar hypoplasia (decreased cerebellum size) in ASC (Pierce and Courchesne, 2001).

Recent research has also shown that these difficulties with sensorimotor integration are particularly key in ASC. For example, Whyatt and Craig (2012) found that the main motor skill deficits in ASC are those that demand more perceptionaction coupling and anticipatory control such as with manual dexterity. This finding is consistent with the current study's results, which showed a specific difficulty in manual dexterity in the ASC compared to the TD group. Participants with ASC also had significantly more difficulty with the MABC, compared to the BEERY VMI. As most of the tasks in the MABC require ability to time movements in response to sensory feedback such as when catching a ball or timed peg turning, as opposed to VMI alone as with the BEERY VMI, this suggests more specific issues with temporal sensorimotor adjustment as opposed to VMI in ASC (Price et al., 2012; Bo et al., 2014). Again, these anticipatory and predictive adjustments of motor programs that are paramount to movement occur in the cerebellum (Koziol et al., 2012), an area of the brain where abnormalities are consistently found in individuals with ASC (Fatemi et al., 2012).

Results from the current study suggest that difficulties in sensorimotor skills are significantly associated with the social communication difficulties and behaviors characteristic of ASC. It would be beneficial to identify which aspect of the sensorimotor motor chain may be affected in ASC. Vandenbroucke et al. (2009) suggested that with significant practice, individuals with ASC could create successful feedforward motor programmes. Larson et al. (2008) reinforced this finding by showing that the mechanisms for acquisition and adaptation of feedforward programs in children with ASC were equal to TD children. Studies have also highlighted difficulties in incorporating sensory input, such as environmental cues, into motor planning (Gowen and Miall, 2005; Gowen et al., 2008; Dowd et al., 2012). Nazarali et al. (2009) also found that individuals with ASC had difficulty reprogramming pre-planned movement. This evidence suggests that a feedforward program can be established by individuals with ASC, but once established, the essential environmental information that is usually used to fine-tune any movement is not utilized effectively. Such physical difficulties in adapting to environmental prompts from an early age would likely have huge impact on an individual's ability to detect, understand and react to social information appropriately. Furthermore, exploration would likely be decreased which in turn would instigate a negative cycle: limited exploration, limited sensory feedback, limited sensorimotor planning, cerebellar hypoplasia, decreased exploration and so on.

A limitation of the current study is that it includes a reasonably small sample (18) in each group. However, the analysis demonstrated medium to large effect sizes alongside Bonferroni correction, and thus demonstrated significant effects. A power analysis also showed that 18 participants had sufficient statistical power to detect a medium to large effect in our supplementary regression analysis. Although

we would be unable to detect smaller effect sizes, results from our correlational and supplementary regression analyses nevertheless showed significant associations between sensory, motor and sensorimotor skills, and severity of ASC symptoms, over and above receptive language and non-verbal IQ, with medium-large effect sizes. We also replicated these effects found in the correlation and supplementary regression analyses in a larger combined ASC and TD sample of 36 individuals. This suggests that difficulties in these areas are significantly associated with severity of ASC symptoms. A questionnaire-based study exploring sensorimotor questions would be advantageous in order to help verify our current findings in a larger sample. Another limitation of this study is that we cannot infer causality using our correlational design. Longitudinal studies are needed to explore the causal relationship between early sensory and motor difficulties and their impact on later social and communicative functioning. Intervention studies that demonstrate that improvement of sensorimotor abilities also improve social and communicative abilities in ASC individuals are also important in order to establish causation. Additionally, the SP does not solely look at sensory perception, items also capture behavioral outcomes and emotional responses, such as "has temper tantrums". Many of these are part of, or co-morbid with ASC. Finally, the difference in matrices performance between the groups approached significance (without Bonferroni correction), though it was clearly smaller in effect size than the differences found in the MABC and SP. Our supplementary regression analysis also controlled for group differences in receptive language and non-verbal IQ, and still found significant results showing associations between sensory, motor, and sensorimotor skills and ASC symptoms. Hence, differences in verbal and non-verbal performance between groups were unlikely to have invalidated the results.

In conclusion, this study suggests that the social communication, interaction and behavioral difficulties that children with ASC experience can be predicted by difficulties in sensorimotor abilities, suggesting possible impairment of sensorimotor integration. Current diagnostic criteria for ASC (DSM-5, American Psychiatric Association, 2013) incorporate difficulties in sensory responsivity and motor movements, although it is not clear whether these are symptomatic of, or instrumental to autism. This study has not included causation, but perhaps it will prompt future research into sensorimotor causality by exploring: the nature of the sensorimotor difficulties in ASC compared to other related conditions (e.g., ADHD and Dyspraxia) to ascertain how sensorimotor integration differs across syndromes; micro-movement methodology (Torres et al., 2013) to further investigate disruptions in sensorimotor integration; and whether integrated sensory activities and temporal spatial coordination activities increase reciprocal social interaction capability. Furthermore, if a child's 3-year developmental review presents weaknesses in sensorimotor skills, alongside intervention this information should be noted and employed as an important assessment and possible ongoing

observation tool for ASC diagnosis purposes and measures of severity.

AUTHOR CONTRIBUTIONS

PH: developed study topic and designed the study, literature search, recruited participants, collated and analyzed data, wrote the manuscript and completed the tables. SC: supported design of the study, collated and analyzed data, gave critical

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Conflict of Interest Statement: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Copyright © 2016 Hannant, Cassidy, Tavassoli and Mann. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution and reproduction in other forums is permitted, provided the original author(s) or licensor are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms. 4.4.3 The research thus far in this thesis has explored the effect of sensorimotor skills on non-verbal social integration and communication skills, such as gesture, imitation and proximities. However, sensorimotor skills may also impact on the acquisition of verbal language in ASC.

Output 3, 'Sensorimotor difficulties are associated with receptive language in autism spectrum conditions', is a journal article (currently under review for a Special Edition in Early Education and Development Journal IF 1.183) that shows that children with ASC scored significantly lower on receptive language, motor coordination and visual-motor integration than typically developing (TD) children. The results of this study also show that the TD group's receptive language was significantly associated with visual perception, whereas the ASC group's receptive language was significantly associated with balance. Additionally, balance significantly correlated with visual perception in the ASC group alone. This finding built on previous research by Glezerman (2012) that suggested individuals with ASC map word meaning on to a situational 'sensorimotor' component of the brain, as opposed to the categorical and empirical component. Furthermore, these results also support a possible 'embodied cognition theory' where it is thought conceptual information is represented within the sensorimotor systems (Mahon and Caramazza, 2008). Thus suggesting that sensorimotor skills and experiences may have a greater influence on the understanding of language in ASC than visual representation alone.

4.4.4 Accordingly, the findings from this study could impact on present interventions that rely heavily on visual imagery to develop language in ASC, such as picture exchange systems and pictorial representations of emotion and therefore warrants further investigation. If language is associated with sensorimotor abilities and experiences rather than visual representation in ASC, language programmes and assessments may benefit from including such pedagogy in the form of objects, sensory experiences, life experiences and photographs in addition to sensorimotor integration programmes. A possible area for future investigation would be to explore whether the substitution of pictorially represented images with real-life images in language assessments alter outcomes of receptive language assessments favourably.

OUTPUT 3 (Under Review)

'Sensorimotor Difficulties are Associated With Receptive Language in Autism Spectrum Conditions'

Hannant, P. (2016) Sensorimotor difficulties are associated with receptive language in autism spectrum conditions. *Special Issue Early Education and Development Journal*

Author Contribution:

Hannant was the sole author and lead investigator for Output 3. Hannant's knowledge of language development in typically developing children led to a literature search on semantic language development in ASC, generating a study that found correlations between sensorimotor skills and language in ASC. Hannant led the study and write up with the support of helpful discussions and written feedback from Cassidy and Carroll.

Early Education and Development Sensorimotor Skills are Associated with Receptive Language in Autism Spectrum Conditions

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Abstract:	A number of studies have evidenced marked difficulties in language in autism spectrum conditions (ASC). Studies have also shown that language and word knowledge are associated with the same area of brain that is also responsible for visual perception in typically developing (TD) individuals. However in ASC, research suggests word meaning is mapped differently, on to situational sensorimotor components within the brain. Furthermore, motor coordination is associated with communication skills. The current study explores whether motor coordination and visual perception are impaired in children with ASC, and whether difficulties in coordination and visual perception correlate with receptive language levels. 36 children took part: 18 with ASC and 18 TD children, matched on age and non-verbal reasoning. Both groups completed the Movement ABC, Beery-Buktenica Developmental Test of Visual-Motor Integration, British Picture Vocabulary Scale and Matrices (WASI). Results showed that ASC children scored significantly lower on receptive language, coordination and visual motor integration than the TD group. In the TD group receptive language significantly correlated with visual perception; in the ASC group receptive language significantly correlated with balance. These results imply that sensorimotor skills are associated with the understanding of language in ASC and thus the relationship between sensorimotor experiences and language warrants further investigation.

Sensorimotor Skills are Associated with Receptive Language in Autism Spectrum Conditions.

Abstract

A number of studies have evidenced marked difficulties in language in autism spectrum conditions (ASC). Studies have also shown that language and word knowledge are associated with the same area of brain that is also responsible for visual perception in typically developing (TD) individuals. However in ASC, research suggests word meaning is mapped differently, on to situational sensorimotor components within the brain. Furthermore, motor coordination is associated with communication skills. The current study explores whether motor coordination and visual perception are impaired in children with ASC, and whether difficulties in coordination and visual perception correlate with receptive language levels. 36 children took part: 18 with ASC and 18 TD children, matched on age and non-verbal reasoning. Both groups completed the Movement ABC, Beery-Buktenica Developmental Test of Visual-Motor Integration, British Picture Vocabulary Scale and Matrices (WASI). Results showed that ASC children scored significantly lower on receptive language, coordination and visual motor integration than the TD group. In the TD group receptive language significantly correlated with visual perception; in the ASC group receptive language significantly correlated with balance. These results imply that sensorimotor skills are associated with the understanding of language in ASC and thus the relationship between sensorimotor experiences and language warrants further investigation.

Keywords: Autism, sensorimotor, receptive language, visual perception, balance, embodied cognition, BA 37

Introduction

The ability to communicate effectively is a fundamental milestone in development and is critical to learning, socialising, behaviour and emotional well-being (Lindsay & Dockrell, 2010). However, children with an Autism Spectrum Condition (ASC) find it difficult to communicate and interact with others from infancy (Dawson et al., 2000). Consequently, current diagnostic criteria for ASC (DSM-5, APA, 2013) include social communication and interaction difficulties, in addition to unusual sensory responsivity and motor movements. (Current DSM-5 criteria (APA, 2013) refers to Autism as a "disorder", the current study uses the less stigmatising term "condition"; acknowledging both strengths and weaknesses in Autism, while still being a medical condition for which individuals need support).

A number of studies have demonstrated language difficulties in ASC. Weaknesses in all areas of language have been shown using a large battery of standardised assessments to measure language in ASC children (Jarrold et al. (1997). Measuring language ability in children with ASC and Speech and Language Impairment (SLI) using the British Picture Vocabulary Scale (BPVS; Dunn & Dunn, 2009) and the Clinical Evaluation of Language Fundamentals (CELF-3, Semel et I., 1995) also showed a flat profile in language in ASC (Loucas et al. 2008). More recently a meta-analysis in receptive and expressive language skills in ASCs has shown that both areas of language are equally decreased in ASC children in comparison to typically developing peers (Kwok et al., 2015), although comprehension and semantics have also been highlighted as being most affected (Boucher, 2012). A longitudinal study showed that children with a receptive language disorder are often difficult to distinguish from those with an ASC in terms of their language outcomes (Howlin et al., 2000). Other studies that demonstrate differences in language include Dunn et al. (2008), Dunn & Bates (2005), Bishop & Norbury (2002), Norbury (2005), Lloyd et al. (2006), Ungerer & Sigman (1987) and Luyster et al. (2008). Moreover, language difficulties are a requisite to a diagnosis of ASC according to current DSM-5 criteria (APA, 2013).

Identifying reasons behind such language and communication difficulties in ASC would be an important advancement in understanding the symptoms. Both visual perception and sensorimotor skills have been associated with communication and social abilities. More specifically, visual perception (Ardila, 2011) and the emergence of sitting skills (Libertus

and Violi, 2016), being associated directly with receptive language in typically developing (TD) individuals, and impairments in motor abilities with SLI (Iverson & Braddock, 2011) and the development and severity of social skills in ASC (Landa & Garrett-Mayer, 2006; Green, et al., 2009; Ming, et al., 2007). The 'embodied cognition hypothesis' should also be considered here, where it is thought conceptual information is represented within the sensorimotor systems (Mahon and Caramazza, 2008), for example: the 'action-sentence compatibility effect' (Borreggine and Kaschak, 2006), where sentence meaning interacts with movements made during oral sentence presentation; or olfactory anchors that stimulate memory (Engen, 1991). Moreover, a systematic lag between the age of the earliest memory and the age of acquisition of the associated word has been observed (Morrison and Conway, 2010), which is thought to reflect the formation of the conceptual knowledge required from details in the episodic memories and situational contexts. This raises the question as to if and why there are different cognitive relationships in language between TD controls and the ASC group; this study therefore investigates the possible effects of visual perception and sensorimotor abilities on receptive language in ASC.

Difficulties in language have been shown to effect social communication skills in individuals with and without ASC. For example: when measuring social confidence, preschool children preferred playmates with similar linguistic skills (Brighi et al., 2015); using a longitudinal study on children from 2.5 years to 5.5 years of age, ASC children who had deficits in receptive and expressive language growth had persistently high trajectories using calibrated severity scores (Venker et al. 2014); and the social functioning in two groups of individuals, one with ASC and one with SLI, from initial studies at age 7-8 to a follow-up study at 23-24 years of age, had similar and significant difficulties in behavioural patterns, social functioning, jobs and independence (Howlin et al., 2000). It is therefore important to identify cognitive processes that impact development of language in ASC.

In ASC motor coordination difficulties are understood to have a negative impact on social interaction skills and participation. This can be seen in infants with ASC or at risk of developing ASC, where atypical fine motor skills have been observed, such as reduced grasping activity and less mature object manipulation (Libertus, et al., 2014). Moreover, a relationship between early motor delay and communication delay in infants with ASC has also been documented (Bhat, et al., 2012). These findings are further strengthened by

correlations between the severity of ASC symptoms and motor coordination (MacDonald et al., 2013; Green, et al., 2009; Ming, et al., 2007). More specific observations demonstrate that significant impairments in motor skills appear to result in limited gesture in ASC (Mostofsky et al., 2006): in turn this restricted gesture has been identified as a significant predictor of receptive language in pre-schoolers with ASC (Luyster et al., 2008). Difficulties with speech pronunciation due to oral motor difficulties could also impact on social acceptance and interaction in ASC (Page and Boucher, 1998). Moreover, children who have fine motor difficulties in early childhood (from 7 months old) are considered to be more at risk of developing an ASC by 3 years old (Landa & Garrett-Mayer, 2006). As social communication is a key symptom of ASC and the severity appears to be associated with motor impairment (Hannant et al., 2016), the possible association with difficulties in language should be explored further.

Motor coordination is thought to be associated with social skills: however visual feedback is essential to the planning and performing of motor movements (Brooks, 1983; Wolpert and Flanagan, 2001). For example an individual needs to be responsive to where items are visually in order to grasp, reach or avoid them. Consequently any cognitive difficulties in visual guidance are likely to affect the ability to acquire and modify a motor command for effective motor coordination. Adults with ASC have difficulty coordinating hand / eye movements (Glazebrook et al., 2009). Additionally, by measuring both form and motion coherence in ASC, a link between visual motion responsivity and fine motor control has been observed (Milne et al., 2006). Difficulties integrating visual cues from the environment with motor movements have also been demonstrated (Dowd et al., 2012). Furthermore, motor coordination deficits in children with a Developmental Coordination Disorder (DCD) were significantly related to their visual perceptual deficits (Cheng et al., 2014).

There are two distinct areas of the brain involved in language: Broca's area, which appears to be involved in the sequencing processes (grammar) of language; and the temporal Wernicke's area, which appears to be linked to the semantic/lexical system (Ardila ,2011). Interestingly, the area BA 37 within the Wernicke's area, is associated with both receptive language and the visual representation of auditory linguistic information, determining that this particular area of the brain not only has language but also visual perception functions (Ardila et al., 2015; Milner and Goodale, 2008; Pammer et al., 2004; and Stewart et al. 2001). However, this association seems to differ in individuals with

developmental disorders such as ASC and SLI. By analysing a number of imaging studies of BA 37 activation during sentence comprehension, Glezerman (2013) found that instead of mapping words on to the categorical and empirical components of the left hemisphere (LH), individuals with ASC and SLI would primarily match word meaning to the situationalexperiential contexts of the right hemisphere (RH). Moreover, Glezeman suggests that only in ASC is word meaning mapped onto the situational sensorimotor level in the RH BA 37. Therefore, the Broca's area may well be important in linking difficulties in visual perception and/or sensorimotor skills to receptive language difficulties.

Research has found correlations between receptive language and visual perception in typically developing individuals, and between coordination and social communication severity in ASC. Furthermore, studies have also indicated disparity in word mapping within the BA 37 area of the brain: where TD individuals map word meaning on to the left hemisphere categorical visual components and ASC appear to map on to right hemisphere sensorimotor components. However presently there is limited research into the causes of receptive language difficulties in ASC. Exploring how and why receptive language in ASC differs to typically developing children may help guide intervention and assessment processes. Therefore this study explores whether: 1) there is a significant difference in receptive language ability, visual perception and motor coordination between children with and without ASC; and if 2) visual perception and motor coordination correlate with receptive language ability in children with and without ASC.

Method

Participants

Two groups of children took part in a larger study exploring the impact of sensorimotor skills on social and communication skills: an ASC group and a TD group. The ASC group were recruited from local ASC support groups in Warwickshire, UK and was comprised of 18 children, (13 male, 5 female) aged 7-16 (mean age – 9.9 years). All children with ASC had a pre-existing diagnosis of ASC from a trained clinician according to DSM-IV criteria. ASC diagnosis was also confirmed using the Autism Diagnostic Observation Schedule General – 2nd Edition (ADOS-2) (Rutter, et al., 2012) and the Autism Diagnostic Interview Revised (Rutter, et al., 2005), administered by a research reliable rater. The TD group were recruited by advertising in the local media in Warwickshire, UK and was comprised of 18 children (7 male, 11 female), aged 6-12 (mean age = 9.2 years). The TD group had no

known disabilities or diagnoses. All participants completed: a measure of receptive language IQ (BPVS-III; Dunn & Dunn, 2009); performance IQ (WASI matrices subset; Wechsler, 2011); a measure of visual motor integration (Beery & Beery, 2010); a measure of motor coordination (Movement ABC; Henderson et al. 2007); and a parent report measure of autistic traits (Social Communication Questionnaire; SCQ; Rutter, et al., 2003). Participants were matched on age and performance IQ, but not gender (table 1). However, there was no effect of gender on receptive language, visual perception or motor coordination measures in either the ASC group (BPVS t(16)=1.25, p=.23; BEERY VP t(16)=1.17, p=.26; Movement ABC t(16)=.29, p=.78) or the TD group (BPVS t(16)=.00, p=1.00; BEERY VP t(16)=.47, p=.65; Movement ABC t(16)=.18, p=.86. No participants in the TD group scored above cut off indicating ASC on the SCQ (15). See Table 1 for characteristics of both groups.

Table 1: Demographic descriptives and group comparisons.

Crown	Condor	Ago in Vooro	Non-Verbal			
Group	Gender	Age in rears	Reasoning			
ASC	13 M	9.93±2.71	90.94±13.28			
(N=18)	5 F		(71-112)			
Control	7 M	9.16±1.89	99.50±12.68			
(N=18)	11 F		(70-117)			
Difference	X ² (1,18)=4.05,	<i>t</i> (34)=-1.00,	<i>t</i> (34)=-1.98,			
	<i>p</i> =.044*	<i>p</i> =.325	<i>p</i> =.056			
Note: * denotes p<0.05 Bonferoni Correction p=.025						

Materials

Participants completed a battery of four assessments that were standardised (MABC, BEERY, BPVS, WASI) where a standardised score of 115 or above was considered above average and 84 or below was considered below average. A further assessment (SCQ) was criterion based with a given cut-off point. Raw scores were used on the MABC, in order to measure any correlation more accurately on each subset.

The Movement Assessment Battery for Children – 2 (Movement-ABC 2; Henderson, Sugden and Barnett 2007): A standardised assessment of motor coordination for children aged 3 - 16 years which is comprised of three components: manual dexterity, ball skills, static and dynamic balance. Examples of test content include placing pegs onto a board, throwing a beanbag onto a target and walking heel to toe along a line. The Movement-ABC 2 was normed on 1172 children aged 3-16 years with and without disabilities. Internal Reliability includes internal consistency estimates (range = .92-1.00) and validity with the 'Draw-a-Man' test = 0.66 (Kavazi, 2006). Test Duration: 20-40 minutes

The Beery-Buktenica Developmental Test of Visual-Motor Integration, Sixth Edition (*BEERY VMI; Beery, Beery and Buktenica, 2010*): A standardised measure of an individual's ability to combine visual perception and fine motor coordination for people aged 2-100 years which is comprised of three parts: visual motor integration, visual perception and fine motor coordination. The visual motor integration (VMI) assessment requires an individual to copy a series of developmentally progressive geometric shapes; the visual perception (VP) aspect involves identifying matching shapes; and the motor coordination subtest contains a variety of shape outlines that the individual draws lines within. The BEERY VMI (6th Ed) was normed on 1737 individuals aged 2-18 years with and without disabilities. Inter-rater reliability (range = .93-.98) and validity (range = .80-.95) (Beery & Beery, 2010). Test Duration: 10-15 minutes

The British Picture Vocabulary Scale III (BPVS-III; Dunn, Dunn, Styles and Sewell, 2009): A standardised non-reading assessment of receptive language. Each item within the assessment consists of identifying the correct image out of four pictures provided, to match a given word that covers a range of subjects, such as verbs, animals, emotions, toys and attributes. The BPVS-III was normed on 1480 children aged 3-16 years with and without disabilities. Internal reliability = .91 and validity with the Wechsler Intelligence Scale for Children (2005) = 0.76 (Dunn et al., 2009). Test Duration: 10-15 minutes

The Social Communication Questionnaire - Lifetime (SCQ; Rutter, Bailey, & Lord 2003): A standardised parent-report measure of autistic traits for children from 4 years of age. The lifetime form is composed of 40 yes or no questions and is used as a screening tool indicating whether referral for diagnosis of ASC is warranted. Scores of 15 or above out of 40 indicate a possible diagnosis of ASC. The SCQ was normed on 214 children aged 2-18 years with and without disabilities. Internal reliability = .84-.93 and validity with the ADI-R = 0.78 (Rutter et al., 2003) Test Duration: <10 minutes

Wechsler Abbreviated Scale of Intelligence – 2nd Edition (WASI-II; Wechsler & Hsiao-pin, 2011). A brief standardised measure of verbal and performance intelligence. The matrices subset was used in the current study to measure non-verbal reasoning in both groups. The WASI was normed on approximately 2900 individuals aged 6-90 years with and without

disabilities. Matrix Internal Reliability = .87-.94 and validity with the WRIT = 0.71 (Wechsler & Hsiao-pin, 2011). Test Duration: <10 minutes

The following diagnostic measures were also completed by the ASC group to independently confirm participants ASC diagnosis and indicate severity of ASC symptoms:

Autism Diagnostic Observation Schedule – 2nd Edition (ADOS-II; Rutter, et al., 2012): A standardised diagnostic instrument for diagnosis of ASC, and confirmation of ASC diagnosis for research purposes. It consists of a semi-structured interview that provides a number of social presses and opportunities to code quality of social and communicative behaviours. The 2nd Edition of the ADOS also includes a rating indicating the severity of ASC symptoms taking into account the person's age and expressive language level. The ADOS-II was validated on 381 individuals aged between 15 months to 40 years with and without disabilities, with a further 1139 children aged between 14 months to 16 years recruited to revise the algorithms. Inter-rater reliability showed over 80% agreement on all modules with a high level of discriminative validity between autism and TD resulting in specificities of 50 to 84 and sensitivities of 91 to 98 (Rutter et al., 2012). Test Duration: ≈60 minutes

Autism Diagnostic Interview – Revised (ADI-R; Rutter, et al., 2005): A standardised diagnostic instrument for diagnosis of ASC, and confirmation of ASC diagnosis for research purposes. It consists of a detailed semi-structured interview to gather evidence from an informant (parent, sibling or partner of an individual) on an individual's current behaviour and early development indicative of an ASC diagnosis. Interviews cover social and communication, repetitive stereotyped behaviours, sensory and motor skills, talents, and challenging behaviours. The ADI-R was validated on 50 children aged between 36 to 59 months with and without disabilities. Internal Reliability demonstrated 26 of 35 weighted kappa values were .70 or higher. The ADI-R also shows a high level of discriminative validity with Clinical Diagnosis with 24 out 25 children being correctly diagnosed using the ADI-R (Rutter et al., 2005) Test Duration: ≈180 minutes

Procedure

The local research ethics committee gave ethical approval for the study. Following parental consent to take part in the study, the parent completed the ADI-R either in person or over the phone with a researcher who was research reliable in both ADI-R and ADOS-II (ASC group only). Both child and parent then attended a single assessment session at the

University. During this session, the following assessments were carried out in random order to counterbalance and combat order affects: BEERY VMI, Movement ABC, ADOS-II (on ASC group only), BPVS and WASI performance subsets; the participant's parent also completed the SCQ. Before the assessment procedure each task was explained carefully and depending on autism severity, a visual timetable produced to help alleviate anxiety. During the test procedure each participant was invited to have a voluntary break after each assessment. Additionally, each task was both demonstrated and practiced according to the manual instructions to ensure understanding.

Results

Analysis approach

Data were analysed using SPSS (version 22), and normality tests conducted using Skewness and Kurtosis outputs. All measured variables: Movement ABC (composite scores); BPVS III, BEERY VMI, Visual Perception and Motor Coordination did not deviate significantly from normal (*z* scores were all <1.96). Following tests for normality, TD and ASC data (BPVS-III, MABC, SCQ, BEERY) were compared using Bonferroni corrected independent t-tests. A correlation analysis was then performed between the BPVS-III, MABC subset scores and BEERY VMI subset scores. Cohen's d is used as an indicator of effect size, with 0.2 indicating a small, 0.5 medium and 0.8 a large effect. These correlations were then followed up by a stepwise linear regression, using receptive language as the outcome measure (BPVS), and age, Matrices, visual perception and MABC balances scores as predictors. Post Hoc power analyses on the multiple regression model were conducted using G*Power 3 (Faul et al., 2007) to compute the achieved statistical power for each model. Results showed for Model 2 in the TD group a statistical power of 0.6 was achieved, and in the ASC group a statistical power >0.8 was achieved, with a sample size of 18.

Do children with ASC show significant receptive language, visual perception and motor difficulties?

Table 2 shows results of comparisons between the ASC and TD groups on all measures. Bonferroni corrected independent samples *t*-tests showed that children with ASC had significantly lower receptive language ability (BPVS), motor coordination skills, visual motor integration (VMI), visual perception (VP) and higher parent reported autistic traits (SCQ scores) than the TD group, all with large effect sizes according to Cohen's *d*. With Bonferroni correction the difference in balance and BEERY VMI was not considered significant (p=.016; p=.009 respectively); however the effect size of the difference between groups in these subsets is still noteworthy.

Group	BPVS Standardised Score	MABC Composite Total	MABC Balance	MABC Manual Dexterity	SCQ score	BEERY VMI (ASC N=17)	BEERY VP
ASC	88.56± 14.08	51.61±15.69	22.67±8.17	16.56±8.60	18.94±7.94	84.24±21.27	92.94±14.85
(N=18)	70-119	25-71	8-33	5-32	8-35	48-120	69-130
Control	106.00±12.02	74.28±14.12	28.89±6.49	29.94±5.51	3.83±3.68	100.17±8.93	110.06±7.60
(N=18)	77-127	46-92	17-36	14-38	0-13	82-116	95-125
	<i>t</i> (34)=-4.00,	<i>t</i> (34)=-4.56,	<i>t</i> (34)=-2.53,	<i>t</i> (29)=-5.56,	<i>t</i> (24)=7.33,	<i>t</i> (21)=-2.86,	<i>t</i> (25)=-4.35,
Difference	<i>p</i> =.000*,	<i>p</i> =.000*,	<i>p</i> =.016,	<i>p</i> =.000*,	<i>p</i> =.000*,	<i>p</i> =.009,	<i>p</i> =.000*,
	<i>d</i> =-1.33	<i>d</i> =-1.52	<i>d</i> =84	<i>d</i> =-1.85	<i>d</i> =2.44	<i>d</i> =98	<i>d</i> =-1.45

Table 2: Dependent variable descriptives and comparison of means.

Note: Effect size Cohen's d: 0.2 = small, 0.5 = medium, 0.8 = large.

* Significant results after bonferroni corrected p value = .007

Key: BPVS: British Picture Vocabulary Scale III; MABC: Movement Assessment Battery for Children;

SCQ: Social Communication Questionnaire; BEERY: The Beery-Buktenica Test of Visual Motor Integration

Does visual perception and motor coordination correlate with receptive language ability? In both the ASC and TD group, separate and joint correlation analyses were conducted between receptive language (BPVS-III) and the MABC total, balance and manual dexterity composite and BEERY visual perception and visual motor integration composites. The WASI matrices was also included in order to determine any similarities in correlations with the BPVS III, as they both rely on visual processing to access the test.

Table 3 shows results of the correlations. In the ASC group the MABC balance subset significantly correlated with receptive language (BPVS III) (r = .57, p = .007). In the TD group the BEERY VP significantly correlated with receptive language (BPVS-III) (r = .57, p = .007). The matrices significantly correlated with the BPVS III in the TD alone (r = .66, p = .002), with no significant relationship in the ASC group (r = .30, p = .116). When the ASC and TD groups were combined all variables, BEERY (VMI and VP), MABC (Total, Balance and Manual Dexterity) and Matrices significantly correlated with the BPVS-III respectively (VMI r = .54, p < 001; VP r = .56, p < 001; MABC total r = .60, p < 001; MABC Balance r = .59, p < 001; MABC Manual Dexterity r = .60, p < 001; and Matrices r = .54, p < 001). Figure 1 demonstrates visually how visual perception and balance correlated with receptive language between groups.

				MABC			
				Manual	MABC	MABC	WASI
		BEERY VMI	BEERY VP	Dexterity	Balance	TOTAL	MATRICES
BEERY VP	ASC (18)	.797***					
	TD (18)	.134					
	TOTAL (36)	.745***					
MABC Manual	ASC (18)	.536 [*]	.410 [*]				
Dexterity	TD (18)	223	150				
	TOTAL (36)	.541***	.570***				
MABC Balance	ASC (18)	.554 [*]	.556**	.663**			
	TD (18)	.101	.093	.580**			
	TOTAL (36)	.513**	.542***	.694***			
MABC TOTAL	ASC (18)	.611**	.497*	.924***	.840***		
	TD (18)	.043	084	.781***	.908***		
	TOTAL (36)	.572***	.560***	.915***	.872***		
WASI MATRICES	ASC (18)	.600**	.517 [*]	.465*	.446*	.495*	
	TD (18)	.407*	.307	.316	.444*	.353	
	TOTAL (36)	.568***	.518**	.496**	.513**	.518**	
BPVS	ASC (18)	.379	.257	.399	.573**	.464*	.297
	TD (18)	.493*	.567**	.272	.345	.299	.656**
	TOTAL (36)	.542***	.564***	.600***	.588***	.603***	.539***

Table 3: Correlation analyses (*R*) for all measures across ASC, TD and Total group.

Note: * *p* <0.05, ** *p* < 0.01, *** *p* < 0.001. Required Effect Size = .522 (Power = .80, α = 0.05, 18 Sample Size) Correlations in **bold** indicate results > required effect size Key: BPVS: British Picture Vocabulary Scale III; MABC: Movement Assessment Battery for Children; BEERY: The Beery-Buktenica Test of Visual Motor Integration

Figure 1: Scatterplots to show correlations with receptive language in the ASC and TD groups



Key: BPVS: British Picture Vocabulary Scale III; MABCBCOMP: Movement Assessment Battery for Children Balance; BEERYVP: Visual Perception

Does visual perception correlate with balance?

The correlational analysis demonstrated that visual perception (BEERY VP) significantly correlated with balance (MABC Balance) in the ASC group alone (r = .56, p = .008). There was no correlation between balance and VP in the TD group (r = .09, p = .356).

Table 4 shows results of the supplementary stepwise regressions. In the TD both Matrices and Visual Perception were retained as significant predictors of receptive language (BPVS). In the ASC group the MABC Balance composite alone was retained as a significant predictor of receptive language (BPVS). These predictors were retained when both age and performance IQ were included. In summary, results showed that the visual perception scores significantly predicted TD receptive language and the MABC balance measures significantly predicted receptive language in ASC when accounting for age and performance IQ as the Matrices predicted some, but not all, of the receptive language in TD individuals.

Step	Variable	В	SE B	В	Cum R ²			
Typically Developing Group (n=18)								
1	Constant	44.11	17.93					
	MATRICES	.62	.18	.66**	.66			
2	Constant	-14.54	30.15					
	MATRICES	.50	.17	.53**	.76			
	BEERYVP	.64	.28	.40*				
ASC Group (n=18)								
1	Constant	66.18	8.49					
	MABC BALANCE	.99	.35	.57**	.57			

Table 4: Stepwise multiple regressions for receptive language in the TD and ASC group.

Note: * p < 0.05, ** p < 0.01, *** p < 0.001.

Typically Developing Model 2 achieved 0.6 Statistical Power for a sample size of 18 Autism Spectrum Conditions Model 1 achieved >0.8 Statistical Power for a sample size of 18

Discussion

The current study investigated whether children with ASC show significant difficulties in receptive language, visual perception and motor coordination when compared to TD children and whether visual perception and motor coordination are associated with receptive language. Results demonstrated that children with ASC had significant receptive language, visual perception and motor coordination difficulties when compared to age and performance IQ matched children without ASC. These results confirm findings with regards to impaired language by Dunn et al. (2008) and coordination by MacDonald et al. (2013) and add to findings with regards to visual perception in ASC, by suggesting visual perception is a comparable area of weakness in ASC. When considering whether visual perception and motor coordination are associated with receptive language ability, the results showed distinct differences between children with and without ASC. In TD children, visual perception was shown to significantly correlate with receptive language with medium to large effect. However, in the ASC group visual perception was not associated with receptive language: instead balance significantly correlated with receptive language with medium to large effect. Furthermore, data analysis showed a correlation between visual perception and balance in the ASC group alone with medium effect. Studies have shown a similar link between visual perception and balance in both SLI (Nicola et al., 2015) and DCD (Cheng et al., 2014), with the same discrepancy echoed: no link between visual perception and motor skills in the TD children.

Differences in the processing of receptive language in ASC could be associated with visual processing difficulties and a consequential over-reliance on the other senses, thus processes that rely heavily on visual representation, such as language, may also be affected. Evidence for this can be seen in research conveying that children with ASC rely more heavily on proprioceptive feedback than visual for the correction of movement (Marko et al., 2015; Izawa et al., 2012; Schmitz et al., 2003; Gepner and Mestre, 2002). Additionally, studies have identified deficits in ASC when compared to controls in areas of visual processing such as visual attention (Ronconi et al., 2013), accuracy when moving eyes from a central fixation point to a peripheral target (Schmitt et al., 2010; Mosconi et al., 2013) and delay in initiating a saccade when following a moving light (Wilkes et al., 2015). The visual processing difficulties and over-reliance on body position could also accentuate the aforementioned 'embodied cognition hypothesis' (the representation of conceptual information within the sensorimotor system, Mahon and Caramazza, 2008), thus impacting on the empirical and visual acquisition of language. This could also account for the relationship between coordination and language in infants, such as the link between sitting and language (Libertus and Violi, 2016).

However, the differences in how semantic information is processed in ASC is also noteworthy. For example children with ASC differed from TD children by not utilising semantic priming when asked to name near-semantically related words, such as truck-car (Kamio et al., 2007). Moreover, the aforementioned review by Glezerman (2012) suggests that due to 'reversed lateralization' as described previously in the BA 37, individuals with ASC map word meaning onto the situational 'sensorimotor' components of the RH BA 37, as opposed to the categorical and empirical components of the LH.

The mapping of language and associations of language with sensorimotor experiences would help to explain some learning difficulties in ASC, such as hyperlexia (superior word-reading skills in comparison to comprehension, Newman et al., 2007) and real-life difficulties faced by individuals with ASC on a daily basis, such as non-verbal communication issues, as both rely heavily on the visual representation of meaning rather than experience.

It was difficult to decide on background variables to match the ASC and control group. Matching controls with language and verbal IQ was not appropriate as we were examining the unusual language profile in ASC. Similarly, we did not want to use a non-verbal IQ test that relied heavily on visuo-spatial skills. For this reason, the matrices subset of the performance IQ was chosen as a non-verbal IQ match. However it is important to note that the matrices assessment is similar to the BPVS III in that the participant has to select one of a set of visual representations as the answer. Additionally, in the TD children the BPVS III correlated with the matrices score (r = .66, p = .002), consequently this measurement was also not entirely suitable. Thus, participants were matched firstly on age and then on matrices performance. This finding would also suggest that the BPVS III is not a suitable assessment to measure performance in children with ASC.

A limitation of the current study is that it includes a reasonably small sample (18) in each group. However, Bonferroni correction was utilised and analysis was shown to have medium to large effect sizes, hence there was sufficient statistical power for the analysis. Furthermore, the study gathered a rich dataset consisting of a number of high quality standardised measures and independent confirmation of ASC diagnosis, which may not have been possible for a larger sample size. Another limitation is the cross-sectional nature of the data collection, which does not allow for causal interpretations. Finally, the difference in nonverbal IQ between the groups approached significance (without Bonferoni correction), though it was clearly smaller in effect size than the differences found in language and visual perception.

In conclusion, this study indicates that receptive language in ASC is correlated with sensorimotor skills as opposed to visual perception, as seen in TD. This finding, in addition to previous research with regards to 'embodied cognition' and differences in semantic mapping, suggests that the receptive language difficulties that children with ASC experience could be related to their sensorimotor experiences. Accordingly, the findings from this study could impact on present interventions that rely heavily on visual imagery to develop language in ASC, such as picture exchange systems and pictorial representations of emotion and therefore warrants further investigation. If language is associated with sensorimotor abilities and experiences rather than visual representation in ASC, language programmes and assessments may benefit from including such pedagogy in the form of objects, sensory experiences, life experiences and photographs in addition to sensorimotor integration programmes. A possible area for future investigation would be to explore whether the substitution of pictorially represented images with real-life images in language assessments alter outcomes of receptive language assessments favourably.

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4.4.5 **Output 4, 'Dyspraxia and autistic traits in adults with and without autism spectrum conditions'** is a journal article published in Molecular Autism (IF 4.961) and is a large cross-sectional study of over 5,000 individuals that supports the findings within **Outputs 1-3**. This Output followed the presentation of data within a poster at the IMFAR conference Salt Lake City (International Meeting of Autism Research, Appendix 1). The paper presents the first evidence that the rate of self-reported dyspraxia is significantly higher in those with ASC (6.9%) compared to the general population without a diagnosis of ASC (0.8%). Furthermore, the paper shows that in the general population, dyspraxia was associated with significantly higher autistic traits and lower empathy.

This suggests that motor coordination skills are important for effective social skills and empathy whilst highlighting similarities between ASC and Developmental Coordination Disorders (DCD). Thus, it is important that clinicians are aware of the impact and importance of motor coordination skills for wider social functioning and empathy so that they are able to offer timely and appropriate support for these individuals.

OUTPUT 4 (Published)

'Dyspraxia and Autistic Traits in Adults With and Without Autism Spectrum Conditions'

Cassidy, S., Hannant, P., Tavassoli, T., Allison, C., Smith, P., & Baron-Cohen, S. (2016). Dyspraxia and autistic traits in adults with and without autism spectrum conditions. *Molecular Autism*, 7(1), 48.

Author Contribution:

Whilst Cassidy was the lead author for Output 4, and responsible for the main literature review and drafting the paper; the concept, and the evidence upon which it is based, draws extensively upon the previous Outputs and the experience and teaching of children with ASC by Hannant, and crucially their insight into the coordination and sensory difficulties that individuals with ASC encounter on a daily basis. Hannant assisted in conceptualisation, the literature review and writing of the manuscript. The paper was jointly revised by all six authors prior to submission for publication.

RESEARCH

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Dyspraxia and autistic traits in adults with and without autism spectrum conditions

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Abstract

Background: Autism spectrum conditions (ASC) are frequently associated with motor coordination difficulties. However, no studies have explored the prevalence of dyspraxia in a large sample of individuals with and without ASC or associations between dyspraxia and autistic traits in these individuals.

Methods: Two thousand eight hundred seventy-one adults (with ASC) and 10,706 controls (without ASC) self-reported whether they have been diagnosed with dyspraxia. A subsample of participants then completed the Autism Spectrum Quotient (AQ; 1237 ASC and 6765 controls) and the Empathy Quotient (EQ; 1147 ASC and 6129 controls) online through the Autism Research Centre website. The prevalence of dyspraxia was compared between those with and without ASC. AQ and EQ scores were compared across the four groups: (1) adults with ASC with dyspraxia, (2) adults with ASC without dyspraxia, (3) controls with dyspraxia, and (4) controls without dyspraxia.

Results: Adults with ASC were significantly more likely to report a diagnosis of dyspraxia (6.9%) than those without ASC (0.8%). In the ASC group, those with co-morbid diagnosis of dyspraxia did not have significantly different AQ or EQ scores than those without co-morbid dyspraxia. However, in the control group (without ASC), those with dyspraxia had significantly higher AQ and lower EQ scores than those without dyspraxia.

Conclusions: Dyspraxia is significantly more prevalent in adults with ASC compared to controls, confirming reports that motor coordination difficulties are significantly more common in this group. Interestingly, in the general population, dyspraxia was associated with significantly higher autistic traits and lower empathy. These results suggest that motor coordination skills are important for effective social skills and empathy.

Keywords: Autism spectrum conditions, Dyspraxia, Co-morbidity, Autistic traits, Social skills

Background

Is ability to effectively coordinate, plan and carry out movements associated with successful social functioning? Dyspraxia is characterized by pronounced difficulties in the selection, timing and spatial organization of purposeful movement and coordination [1] and is thought to arise from atypical neural connections in the cerebral cortex [2]. Individuals with autism spectrum conditions (ASC), who have pronounced difficulties with social interaction, also exhibit atypical motor movements [3]. In fact, original clinical reports of ASC reported general 'clumsiness' in these individuals [4, 5], which have

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²Autism Research Centre, Department of Psychiatry, University of Cambridge, Douglas House, 18B Trumpington Road, Cambridge CB2 8AH, UK Full list of author information is available at the end of the article been recently confirmed in a number of research studies [6, 7]. Children (without ASC), who have dyspraxia, also exhibit social and emotional difficulties [8]. However, little research in ASC or the general population has explored the association between dyspraxia and social or emotional skills in adulthood [9]. This is the purpose of the current study.

Studies of children with ASC have demonstrated significant motor difficulties in these individuals [10, 11]. Motor skill scores for children with ASC often fall 1.5 standard deviations below the typical mean [12, 13], and approximately 80% have definite pronounced motor difficulties with 10% being borderline [6, 14–18]. Atypical motor skills in ASC are present from early infancy [19–21] and reported by parents as one of their first areas of concern (average age 14.7 months) prior to seeking an ASC diagnosis [22]. Motor difficulties in



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children with ASC may be associated with their social and communicative difficulties. For example, children with ASC show significant difficulties in skilled motor gestures, such as imitation [23]. Empathic ability is also reduced in children (without ASC) who exhibit motor difficulties [8], and research has shown correlations between motor coordination and social communication skills in children with ASC [24–26]. Approximately 2–6% of children (without ASC) from the general population have dyspraxia [27]. These children exhibit difficulties in social skills, social phobia, empathy [8, 28], maintaining peer relationships and increased anxiety [29–31]. This suggests that children without ASC, but with dyspraxia, exhibit traits associated with ASC, particularly in social interaction, empathy and social anxiety.

There are however very few studies on dyspraxia in adults with and without ASC—all of the aforementioned studies involved children. However, the limited number of available studies suggests that in those with and without ASC, motor difficulties continue into adulthood [32, 33]. There is also preliminary evidence from a small number of studies that general population young adults (aged 16–25 years) with dyspraxia may have many of the same difficulties as in childhood [34, 35]. There is also high risk of these individuals experiencing mental health problems, low self-esteem and emotional difficulties, exacerbated by low occupational attainment [9]. It is also currently unclear how many adults with and without ASC have dyspraxia or the impact of this on their social skills.

To address this gap in research, the current study aimed to explore the prevalence of dyspraxia in a large population sample of adults with and without ASC and associations between dyspraxia and autistic traits in these individuals. We utilize online self-reported diagnosis of dyspraxia, alongside validated measures of autistic traits (the Autism Spectrum Quotient (AQ) [36]) and empathy (the Empathy Quotient (EQ) [37]). These measures have been validated for use in those with ASC and the general population, to reliably quantify individual differences in autistic traits and empathy in those with and without ASC. This allows us to assess whether the presence of dyspraxia is associated with significantly increased autistic traits in a large population sample, consisting of over 2500 adults with ASC and over 10,000 adults without ASC. Recruitment of high-functioning adults over 18 years old also allows us to explore associations between dyspraxia and autistic traits independent of intellectual disability or age-related effects through development.

If movement difficulties are significantly associated with social, communication skills and empathy, then individuals with ASC and co-morbid dyspraxia may have a significantly higher number of autistic traits than individuals with ASC without co-morbid dyspraxia. Additionally, individuals (without ASC) who have dyspraxia may also have significantly higher autistic traits than those without dyspraxia or ASC. If this were the case, then this would suggest that movement difficulties are associated with autistic traits in those with and without ASC and could be a prime target for intervention to improve social skills in these groups.

Methods

Participants

Participants completed questionnaires online through one of two websites (www.autismresearchcentre.com or www.cambridgepsychology.com). Controls (without ASC) were only included if they did not report having a child or other family members with autism, to avoid including those with the 'broader autism phenotype' [38]. Individuals with a diagnosis of bipolar disorder, epilepsy, schizophrenia, attention deficit/hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD), learning disability (LD), intersex/transsexual condition or psychosis were excluded from the control group.

After exclusions, 2871 participants reported having a formal clinical diagnosis of ASC (70% male). A majority (n = 2056) had Asperger syndrome; the remaining participants reported having high-functioning autism (n = 287), autism (n = 302), atypical autism (n=43), pervasive developmental disorder (n = 124) and autism spectrum condition (i.e. participants who did not specify a subtype) (n = 59). The control group (without ASC) was comprised of n = 10,706 individuals (41% male), who reported they had no diagnosis of ASC. Participants were aged between 18 and 75 years old (Table 1).

A majority of the individuals in the ASC group provided information on type of education (mainstream, home, special) (n = 2473, 86%), and of these individuals, a majority reported having attended mainstream school (n = 1949, 78.8%). In total, 1284 (44.7%) of the ASC group also provided information on current occupation, and of these, a majority (n = 846, 65.9%) were employed, n = 233 (18.1%) individuals were in full-time study and n = 202 (15.7%) individuals were unemployed. In the control group, n = 5490 (51.3%) individuals provided information on their education type, and of these, a majority (n = 5358, 97.6%) reported having attended mainstream education. In total, 6011 (56.1%) of the control group provided information on their occupation, and of

 Table 1
 Self-reported dyspraxia in adults with ASC vs. adult controls without ASC

	Dyspraxia	No dyspraxia
ASC (n = 2871)	199 (6.9%)	2672 (93.1%)
Control (<i>n</i> = 10,706)	91 (0.8%)	10,615 (99.2%)
Total (<i>n</i> = 13,577)	290 (2.1%)	13,287 (97.9%)

these, n = 4931 (82%) were currently employed, n = 967 (16.1%) were in full-time study and n = 113 (1.9%) were unemployed.

Measures

When registering in the CARD, participants provided demographic data, including age, biological birth sex and educational and occupational attainment, and any diagnoses from a trained clinician, including ASC and dyspraxia. Participants then complete questionnaires designed to quantify autistic traits. We extracted data from two of these self-report questionnaires. The AQ [36] quantifies individual differences in autistic traits, in adults with average or above average intelligence quotient (IQ). The EQ quantifies individual differences in empathizing ability [37].

Statistical analysis

Chi-square analyses were used to compare the prevalence of dyspraxia in the ASC and control groups, with odds ratios used as a measure of effect size. Large samples increase the robustness of ANOVA to violation of normality and homogeneity of variance. Separate twoway ANCOVAs, including age as a covariate, were conducted on AQ and EQ data, with two between-subjects factors of 'diagnosis' (ASC vs. control) and 'dyspraxia' (dyspraxia vs. no dyspraxia). The presence of significant diagnosis-by-dyspraxia interaction effects indicates that the effect of dyspraxia on autistic traits is dependent on ASC diagnosis. Significant interaction effects were followed up by simple main effects analysis, to establish whether the effect of dyspraxia on autistic traits was present in each diagnostic group. Effect sizes were calculated using partial eta squared (η^2) for main effects, interactions and simple main effects. For partial eta squared (η^2), 0.01 represents a small, 0.06 a medium and 0.14 a large effect.

Results

Table 1 shows the frequency of self-reported dyspraxia in the ASC and control groups. Table 2 shows the means for age, AQ and EQ for (1) adults with ASC with dyspraxia, (2) adults with ASC without dyspraxia, (3) controls with dyspraxia, and (4) controls without dyspraxia.

Table 2	Mean	age,	AQ	and	EQ	by	diagnostic	group
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	Dyspraxia?	Age (SD)	AQ (SD)	EQ (SD)
ASC	Yes	29 (12.4)	37.6 (7.5)	20 (12)
	No	35.7 (13.2)	38.5 (6.8)	17.6 (9.9)
Control	Yes	25.3 (10.1)	23.4 (8.2)	39.8 (14.4)
	No	31.4 (13.5)	17.7 (7.5)	45 (14.2)
Total	Yes	27.9 (11.9)	33.4 (10)	25.2 (15.3)
	No	32.1 (13.5)	20.7 (10)	41 (16.8)

Dyspraxia

Participants with ASC were significantly more likely to report a clinical diagnosis of dyspraxia (6.9%) than controls (0.8%) ($X^2(1) = 400.5$, p < 0.001; OR 8.69).

AQ

A between-subjects ANCOVA showed a significant main effect of age (F(1, 7997) = 131, p < 0.001, $\eta^2 = 0.02$). After controlling for the effect of age, there was a significant main effect of dyspraxia, where participants with a diagnosis of dyspraxia self-reported significantly higher levels of autistic traits (mean = 33.4) than those without a diagnosis of dyspraxia (mean = 20.7) (*F*(1, 7997) = 16.58, p < 0.001, $\eta^2 = 0.002$). Results also showed a significant main effect of ASC diagnosis, where participants with a diagnosis of ASC self-reported significantly higher levels of autistic traits (mean = 38.4) than those without ASC (mean = 17.8) (F(1, 7997) = 624, p < 0.001, $\eta^2 = 0.07$). Lastly, there was a significant interaction between the presence of dyspraxia and ASC diagnosis (F(1, 7997) = 22, p < 0.001, $\eta^2 = 0.003$). Simple main effects analysis showed a significant effect of dyspraxia in the control group $(F(1, 7997) = 28, p < 0.001, \eta^2 = 0.003)$; controls with dyspraxia self-reported significantly higher levels of autistic traits (mean = 23.4) than controls without dyspraxia (mean = 17.7). There was no significant effect of dyspraxia in the ASC group (F(1, 7997) = 0.3, p = 0.5, $\eta^2 = 0.001$).

EQ

A between-subjects ANCOVA showed a significant main effect of age (*F*(1, 7271) = 18, p < 0.001, $\eta^2 = 0.002$). After controlling for the effect of age, there was no significant main effect of dyspraxia; participants with a diagnosis of dyspraxia did not self-report significantly different levels of empathy (mean = 25.2) than those without a diagnosis of dyspraxia (mean = 41) (F(1, 7271) = 0.6, p = .4, $\eta^2 = 0.001$). However, results did show a significant main effect of ASC diagnosis, where participants with a diagnosis of ASC self-reported significantly lower empathy (mean = 17.8) than those without ASC (mean = 45) ($F(1, 7271) = 289, p < 0.001, \eta^2 = 0.04$). Lastly, there was a significant interaction between the presence of dyspraxia and ASC diagnosis (F(1, 7271) = 8, p < 0.01, $\eta^2 = 0.001$). Simple main effects analysis showed a significant effect of dyspraxia in the control group $(F(1, 7271) = 4, p = 0.04, \eta^2 = 0.01);$ controls with dyspraxia self-reported significantly lower empathy (mean = 39.8) than controls without dyspraxia (mean = 45). There was no significant effect of dyspraxia in the ASC group ($F(1, 7271) = 3.6, p = 0.06, \eta^2 = 0.001$).

Discussion

This study aimed to explore for the first time whether dyspraxia was significantly more prevalent in adults with ASC compared to controls without ASC and associations between dyspraxia and autistic traits in adults with and without ASC. Results showed that adults with ASC self-reported a significantly higher rate of dyspraxia (6.9%) than adults without ASC (0.8%); the relative odds of having a diagnosis of dyspraxia were 8 times higher in adults with ASC compared to controls without ASC. These results show for the first time that the prevalence of dyspraxia is significantly higher in adults with ASC compared to controls without ASC. These findings reflect previous research, showing that motor coordination difficulties are highly prevalent in ASC [6, 7]. Furthermore, these findings add to the small body of currently available evidence showing that the difficulties associated with dyspraxia in childhood persist into adulthood [33, 35].

Results also showed that the association between dyspraxia and levels of autistic traits and empathy differed according to the presence of co-morbid ASC. Specifically, diagnosis of dyspraxia was only significantly associated with self-reported autistic traits and empathy if participants did not have co-morbid ASC. Controls without ASC, with a diagnosis of dyspraxia, self-reported a significantly higher number of autistic traits and significantly lower levels of empathy than controls without ASC or dyspraxia, whereas those with ASC and co-morbid dyspraxia did not self-report significantly different levels of autistic traits or empathy compared to those with ASC without co-morbid dyspraxia.

These results suggest that motor coordination difficulties are significantly associated with social skills and empathy in adults without ASC, whereas co-morbid dyspraxia in adults with ASC is not significantly associated with increased difficulties in social communication skills and empathy. One possible explanation for this finding is that dyspraxia and ASC symptoms may overlap, particularly as both conditions are seemingly associated with atypical development of neurons within the cerebral cortex [2, 7, 39]. For example, previous research has shown that the difficulties individuals with dyspraxia experience are somewhat similar to the difficulties people with ASC experience. Empathy, for example, is significantly reduced in children (without ASC) who exhibit motor difficulties [8]. Hence, the association between dyspraxia with social skills and empathy in those without diagnosis of ASC would be greater than in those with co-morbid ASC.

Another possibility is that dyspraxia may be underdiagnosed in those with ASC. Motor difficulties are highly prevalent in people with ASC [6, 7], and these difficulties may be viewed as part of their ASC, as opposed to requiring another co-morbid diagnosis. However, given the small but growing body of evidence showing the importance of motor coordination in social skills in both the general population (without ASC) and those with ASC, recognition and diagnosis of these difficulties is key to access appropriate support and treatment. A small body of evidence suggests high risk of adults with dyspraxia, without co-morbid ASC, experiencing mental health problems, low self-esteem and emotional difficulties, exacerbated by low occupational attainment [9]. Results from the current study add to this literature, suggesting that these individuals also experience difficulties in social skills and empathy, characteristic of ASC. Future research will need to explore whether improving motor coordination early in childhood, or in adulthood, improves these poor outcomes.

The current study has a number of strengths as well as limitations. It contributes to an under-explored area of research-dyspraxia in adulthood and the prevalence and impact of dyspraxia on autistic traits in adults with and without ASC. It also utilized measures of selfreported autistic traits (AQ) and empathy (EQ), which both have undergone substantial reliability tests and have excellent psychometric properties [40, 41]. The current study also analysed data from a very large population sample-over 2800 adults with ASC and 10,000 control adults without ASC. These large numbers were necessary in order to explore differences in autistic traits between the groups, considering that only 0.8% of control adults had a diagnosis of dyspraxia. One limitation is that in order to achieve this large sample, self-report measures of dyspraxia and ASC diagnoses were necessary. However, previous research has shown significantly high concordance rates between self-reported and clinically confirmed diagnoses [42, 43]. Additionally, participants provide details on when and where they received their ASC diagnosis when they register in the research database, to ensure that these self-reported diagnoses are valid. Hence, it is unlikely that the self-report methods utilized in the study significantly invalidated the results.

Another potential limitation is the use of the term 'dyspraxia' in the current study. This could have led to an under-reporting of this diagnosis in the control group, due to lack of familiarity with this term. It may also be the case that the likelihood of receiving a dyspraxia diagnosis maybe unevenly distributed across subsets of those with ASC. As discussed above, it is possible that dyspraxia is under-diagnosed in those with ASC, and this may differ by subtype. This could have meant that the rate of dyspraxia in both the control and ASC groups could have been under-estimated in the current study. However, if anything, this means that the rate of dyspraxia diagnosis is a conservative estimate in the current study. If an alternative label, or in person measures, were used, the rates could potentially have been higher in both groups. Future research studies will need to explore whether these rates of dyspraxia are replicable

in a large representative sample of those with and without ASC using in person assessments, across the autism spectrum. However, taken together, this is the only and the largest study to date that has explored the prevalence of dyspraxia and associations between dyspraxia and autistic traits in those with and without ASC.

Conclusions

In conclusion, the current study reports the first evidence that dyspraxia is significantly more prevalent in adults with ASC compared to controls without ASC, confirming previous reports that motor coordination difficulties are highly prevalent in these individuals. Interestingly, the presence of dyspraxia was significantly associated with difficulties in social skills and empathy, particularly in those without co-morbid ASC. These results suggest that adults with dyspraxia demonstrate a significantly increased number of autistic traits compared to the general population (without ASC or dyspraxia) and thus experience similar difficulties to adults with ASC. This is the first evidence of the significant association between motor coordination difficulties with social skills and empathy in adults in the general population and adds to the limited available literature showing a host of poor outcomes in adults with dyspraxia (without ASC). Clinicians must be aware of the impact and importance of motor coordination skills for wider social functioning and empathy and offer appropriate support and treatment for these individuals.

Abbreviations

AQ: Autism Spectrum Quotient; ASC: Autism spectrum condition; EQ: Empathy Quotient

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Availability of data and materials

The datasets analysed during the current study are not publicly available due to the terms and conditions participants agree to when they register in CARD, but are available from the corresponding author on reasonable request.

Authors' contributions

SAC study conception and design, data analysis, manuscript writing and final approval of the manuscript. PH contributed to the conception and design, critical revision and final approval of the manuscript. TT contributed to the design, critical revision and final approval of the manuscript. CA helped in the data extraction and preparation, critical revision and final approval of the manuscript. PS was responsible for the data extraction and preparation,

critical revision and final approval of the manuscript. SBC helped design the study, critical revision and final approval of the manuscript. All authors read and approved the final manuscript.

Authors' information

Not applicable.

Competing interests

The authors declare that they have no competing interests.

Consent for publication

Not applicable.

Ethics approval and consent to participate

A favourable ethical opinion was obtained from the Psychology Research Ethics Committee, Coventry University, UK, to conduct secondary analysis of anonymized data from the Cambridge Autism Research Database (CARD), from the Autism Research Centre, University of Cambridge, UK. Participants register to join the CARD and provide their consent online, where they also have opportunity to read the Terms and Conditions, which includes details of how their data will be stored and used in a variety of research projects in an anonymized form. Participants with and without ASC then provide questionnaire and performance data and indicate their willingness to be re-contacted with invitations to participate in new research studies. This consent procedure for participants to register in the CARD was approved by the Psychological Research Ethics Committee (PREC), University of Cambridge, UK.

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Chapter 3: Compare and Contrast Autism Spectrum Conditions (ASC) and Developmental Coordination Disorder (DCD) to help define more specifically the differences between the two conditions with regards to their sensorimotor foundations

4.5.1 Introduction

The aim of Chapter 3 was to compare the sensorimotor skills and underlying mechanisms in ASC to those in DCD in order to: build on **Output 4** in demonstrating that adults with DCD also display autism symptomology; increase knowledge of the researched differences in sensorimotor coordination between the two sensorimotor conditions; and to critically evaluate sensorimotor difficulties in DCD without comorbid ASC.

- 4.5.2 **Output 5, 'Sensory and Motor Differences in Autism Spectrum Conditions and Developmental Coordination Disorder in Children: A Cross-Syndrome Study'**, is currently under review in the Journal of Autism and Developmental Disorders (IF: 3.321) and explores the similarities and differences in coordination, sensory responsivity and social behaviours between 77 children with ASC or DCD, aged 7-21 years of age, using a detailed online questionnaire that incorporates the Developmental Coordination Disorder Questionnaire (DCDQ; Wilson et al., 2007), the robust 312 question Sensory Profile Checklist-Revised (Bogdashina, 2016) and the Autism Quotient (Auyeung et al., 2008).
- 4.5.3 Results support and underpin the work in **Outputs 1, 2 and 4** demonstrating that the presentation of coordination difficulties in children with ASC is very similar to those with DCD and should be treated as such.
- 4.5.4 Results also show that the sensory profiles between ASC and DCD are shown to differ, in that children with ASC have greater auditory and visual sensitivity, whilst children with DCD have greater proprioceptive sensitivity. Furthermore, these differences show 'good' validity in this study as a means of discriminating between the two conditions.
- 4.5.5 These differences also support previous research in the apparent underlying causes of motor coordination difficulties in both conditions, such that ASC coordination difficulties are reportedly linked to visual processing impairments including visual feedback and motion coherence, whilst DCD coordination difficulties appear to be linked to spatial processing, the formation of a feedforward program and form coherence. However, auditory processing sensitivity was also identified in the ASC sensory profile as a difference between the two movement conditions. This may also be related to the visual modality, as the regulation of visual movement has been associated with the auditory spatial channel and locus of sound (Butterworth & Castillo, 1976; Perrott, 1990). Therefore the relationship between the two modalities warrants further investigation.

- 4.5.6 In addition to a different sensory profile, the AQ also appears to discriminate between the two conditions with 'excellent' validity. However results from this study propose that a higher cut-off point should be taken into consideration for children with DCD in order to allow for co-occurring presentations in the two conditions, as also noted in **Output 4**. The best cut-off score on the AQ to help distinguish between DCD and ASC, that maximises (sensitivity + specificity) is 89.5. At this score, the sensitivity is .85 and the specificity is .84 (1 specificity = .16).
- 4.5.7 It is hoped that this study may not only help to discriminate between two often phenotypically similar conditions, but in doing so has furthered knowledge into the root of the conditions and ultimately their therapy.

OUTPUT 5 (Under Review)

'Sensory and Motor Differences in Autism Spectrum Condition and Developmental Coordination Disorder in Children: A Cross-Syndrome Study'

Hannant, P., Van de Weyer, R., & Cassidy, S. (2017). Sensory and Motor Differences in Autism Spectrum Conditions and Developmental Coordination Disorder in Children: A Cross-Syndrome Study. *Journal of Autism and Developmental Disorders*

Author Contribution:

For Output 5, Hannant and Cassidy devised the topic and Hannant was the lead investigator and main author. Hannant created the online questionnaire, analysed the data and completed the literature search. Van de Weyer and Mooncey supported recruitment and Hannant, Cassidy, Van de Weyer and Mooncey worked jointly on compiling evidence from across different disciplines, and writing the manuscript. All four authors jointly revised this paper prior to publication.

Journal of Autism and Developmental Disorders

Sensory and Motor Differences in Autism Spectrum Conditions and Developmental Coordination Disorder in Children: A Cross-Syndrome Study. --Manuscript Draft--

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Abstract:	Research has shown that Developmental coordination disorder (DCD) can present with similar symptomology as Autism Spectrum Conditions (ASC). This paper explored similarities and differences in coordination and sensory responsivity between DCD and ASC. 77 children took part: 42 with ASC, 26 with DCD and 9 with ASC and DCD. All groups completed parent report measures online that included motor coordination, sensory responsivity and social communication. Results showed no significant differences in coordination, and significant differences in sensory responsivity between ASC and DCD (increased visual and auditory responsivity and decreased proprioception). Exploratory analysis showed good validity in these differences. These results suggest that ASC coordination difficulties are linked to visual processing impairments, whilst DCD coordination difficulties are linked to spatial processing.

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Chapter 4: Critically evaluate the Sensorimotor Theory against Cramer's Criteria (2015)

4.6.1 Combined with the mounting evidence for sensorimotor difficulties in ASC, the research presented in this thesis suggests that impaired sensorimotor function in ASC is prevalent, present from an early age, is associated with symptom severity and is able to distinguish between 'like' conditions: with particular reference to using visual information to both guide and follow movement. Accordingly, a sensorimotor theory for ASC appears to be a conceivable possibility. Thus, 'The Sensorimotor Theory' is evaluated against Cramer's (2013) criteria below in order to judge its merits as a theory.

4.6.2 The Sensorimotor Theory

The 'Sensorimotor Theory' is based on the delicate balance between the feedforward and feedback systems of sensorimotor integration and is discussed in detail in **Output 1**. The intricate arrangement can be seen in Figure 7 below (Wolpert & Flanagan, 2001) and shows how motor coordination is dependent on sensory feedback for both accurate and adaptive movement. For example the ability to plan and execute a simple movement requires sensory feedback (the FEEDBACK Model). However, even when movements are repeated and become automatic forming a feedforward program (the FEEDFORWARD Model), and the need for sensory feedback is reduced as the program generates a prediction of the sensory consequences of the action, any error signal at the final stage of movement still needs to be processed, and corrected by sensory feedback. Accordingly, when sensory guidance is impaired both the ability to first acquire a motor command, in addition to regulating and adapting a stored motor command, would be impaired, leading to limited accuracy and flexibility. It would appear that individuals with ASC have impaired neural inhibition within the sensorimotor neurones of the brain. Thus, 'The Sensorimotor Theory' is an imbalance in the excitation and inhibition of sensorimotor integration impacting considerably on sensory feedback.

Figure 7: The sensory feedback and feed forward input to motor coordination (Wolpert & Flanagan, 2001)

When comparing this theory to Cramer's criteria (2013) above it appears to fulfil six of the seven:Comprehensive - Yes: This theory explains all of the difficulties associated with ASC:

- a. Non-verbal language Outputs 2 and 4, suggest that sensorimotor difficulties impact on a number of skills, which are key for effective social participation. For example, Output 2 shows that sensorimotor skills are associated with autism severity, where incorporating sensory and environmental cues into motor planning impacts on social interaction and communication. Output 4 demonstrates that adults with dyspraxia are more likely to display higher levels of autism symptomology than controls and are also more likely to have difficulty with empathy. Our research is corroborated in further studies, such as individuals with ASC having significant impairments in skilled motor gestures such as imitation (Mostofsky, et al., 2006) and the development of speech sound production (Page & Boucher, 1998). Children who have motor difficulties, without a diagnosis of ASC, tend to show weaker empathy for others (Cummins, et al., 2005) and show increased anxiety on the playground (Bhat, et al., 2011). Anxiety in individuals with ASC is also associated with sensorimotor behaviours and higher levels of insistence on sameness and routine (Rodgers et al., (2012), which can also interfere with social relationships.
- b. Language Output 3 suggests that receptive language in ASC is correlated with sensorimotor skills as opposed to visual perception, as seen in TD. This finding, in addition to previous research with regards to the 'embodied cognition hypothesis' (where

it is thought conceptual information is represented within the sensorimotor systems; Mahon and Caramazza, 2008), and differences in semantic mapping (Glezeman, 2013), suggests that the receptive language difficulties that children with ASC experience could be related to their sensorimotor experiences; also indicating that visual perception is a comparable area of weakness in ASC.

- c. Sensory Research and **Output 2** has demonstrated the presence of a variety of sensory responsivity impairments in ASC (Chawarska, et al., 2007; Caminha & Lampreia, 2012), such as; hyper and hypo responsivity across tactile (Baranek and Berkson, 1994; Blakemore et al., 2006); visual (Nyström et al., 2015), olfactory (Suzuki et al., 2014) and auditory domains (Tavassoli et al., 2012; Jüris, 2013; Takahashi et al., 2014; Madsen et al., 2014); differences in proprioceptive impairment (the sense of relative position in space) (Tavassoli, et al., 2013); and perceptual function (Paton, et al., 2012). Research has also identified links between sensory responsivity and the severity of ASC symptoms. For example, a number of studies have shown that sensory responsivity impairments impact social interaction and communication skills (Matsushima & Kato, 2013; Hilton, et al., 2007; Reynolds, et al., 2011).
- d. Motor –Outputs 2 and 5 show that motor difficulties are prevalent in ASC and are broadly similar to other developmental coordination disorders. Research adds that motor skill scores for children with ASC often fall 1.5 standard deviations below the typical mean (MacNeil & Mostofsky, 2012; Staples & Reid, 2010), and coordination difficulties in ASC are evident from early infancy (Ming, et al., 2007; Page & Boucher, 1998; Kopp, et al., 2010), with parents being significantly more likely to report motor delays as the first concern at an average age of 14.7 months (Teitelbaum, et al., 1998). Additionally, the basal ganglia, an area of the brain decreased in volume in ASC and associated with sensorimotor integration (Estes et al., 2011), is thought to be involved in automaticity and motor habit (Nagy et al., 2006; Chukoskie et al., 2013; Ashby et al., 2010; Yin & Knowlton, 2006).
- e. Empathy Fitzgibbon, Segrave, Fitzgerald and Enticott (2013) proposed that both physical pain and social pain are processed atypically in individuals with ASC and insensitivity to pain, for example, could in turn limit empathy and understanding of pain in others. Also the aforementioned 'Amygdala Theory' discusses the important role the amygdala plays in emotional learning and memory, emotional modulation of memory, emotional influences on attention and perception, emotion and social behaviour, and emotion inhibition and regulation (Baron-Cohen et al., 2000. The amygdala receives a great deal of sensory input and has the highest density of GABA_A receptors in the brain. GABA is the inhibitory neurotransmitter that plays an important role in sensory discrimination in ASC (Puts et al., 2011; **Outputs 1 and 7**). Therefore, although not tested explicitly, sensorimotor difficulties may also lead to underlying difficulties in empathy. Additionally, alexithymia (difficulties in identifying and describing feelings) is also associated with atypical scan paths of the eyes and mouth, further suggesting

sensorimotor involvement (Bird et al., 2010)

- 2) Precision and testability Yes: The sensorimotor theory can be tested subjectively through questionnaires such as the DCDQ '07 (Wilson et al., 2007) and the Sensory Profile-2 (Dunn, 2014), or objectively through assessments such as the Movement ABC (Henderson, et al., 2007), or by measuring areas of sensorimotor activation in the brain. For example difficulties in sensorimotor integration in ASC have been linked to the cerebellum, such as saccadic accuracy being connected to impairment of the error-reducing function of the cerebellum (Schmitt et al., 2014). Research has also shown that the cerebellum has a fundamental role in maintaining the equilibrium between feedback and feedforward programs in sensorimotor integration (Kawato et al., 2003).
- 3) Parsimony Yes: The 'Sensorimotor Theory' appears to be a simple or parsimonious theory that encompasses all of the aforementioned areas, such as social communication, motor mannerisms, hypo or hyper sensory responsivity and atypical empathy.
- 4) Falsifiability Yes: The 'Sensorimotor Theory' can be challenged and unwittingly has been, such that Baron-Cohen, in his reply to Haydn Ellis (Baron-Cohen, 2005) suggested that motor clumsiness in ASC did not tally with his own personal experience in running a clinic. However, the prevalence of motor difficulties in ASC is very high with approximately 80% reportedly having definite motor impairment with 10% being borderline (Green, et al., 2009, Fournier, et al., 2010; Miyahara, et al., 1997; Gowen & Hamilton, 2013; Whyatt & Craig, 2012, Kopp et al., 2010), whilst studies indicate the occurrence of atypical sensory responsivity in ASC at approximately 95% (Matson, et al., 2010; Bhat, et al., 2011). Furthermore, the differences in sensorimotor skill may be due to the underlying variation in ability to temporally control movement (Whyatt & Craig, 2013). **Output 5** also set out to challenge 'The Sensorimotor Theory' in order to better understand why similar movement conditions such as DCD do not result in ASC. However, this paper then went on to find that despite similar traits in DCD (as in **Output 4**) the causation of movement difficulties in both conditions is likely to be disparate.
- 5) Heuristic value Yes: The initial analysis of the role of sensorimotor skills in ASC has already been read over 2000 times and has been cited in published material on four occasions since its publication. Furthermore, the recognition of movement as being an important underlying factor for ASC has also been recognised in a research topic in the Frontiers Integrated Neuroscience Journal (Torres & Donnellan, 2015).
- Applied value Yes: With an understanding of the fundamental sensorimotor difficulties in ASC and how they impact development, early intervention can be delivered, such as support with tracking movement, sensory integration (**Output 6**) and GABA substitution (**Output 7**).
- 7) Universality No: However, with up to 90% of ASC individuals demonstrating motor difficulties, and up to 95% atypical sensory responsivity (see above), this theory is close to meeting criteria, in addition to the vast amount of research to substantiate this point having found both motor (Kanner, 1943; Asperger, 1944; DeMyer, 1976; Damasio, 1978; Berkeley et al., 2001; Mari et al., 2003; Green et al., 2009; Hughes, 1996; MacNeil and Mostofsky, 2012; Staples and Reid, 2010), and sensory (Kanner, 1943; Asperger, 1944; Caminha and Lampreia, 2012; Tomchek & Dunn,

2007; Nyström et al., 2015; Baranek and Berkson, 1994; Blakemore et al., 2006; Paton et al., 2012; Tavassoli et al., 2013; Jüris, 2013; Takahashi et al., 2014; Madsen et al., 2014; Bennetto et al., 2007; Suzuki et al., 2014) difficulties in ASC.

4.6.3 'The Sensorimotor Theory' can also account for other theories of ASC. For example, the weak coherence theory could be linked to diminished performance, or compromised higher order visual processing tasks such as visual spatial and visual motion processing (Koldewyn et al., 2009; Simmons et al., 2009; Spencer et al., 2000;). Some studies have indicated that adults with ASC are impaired at motion processing but retain intact form processing (perhaps reflecting selective dysfunction of the magnocellular pathway - the motion processing pathway) (Koldewyn et al., 2009; Koldewyn et al., 2011; Spencer et al., 2000; McCleery et al., 2007). Other studies have found atypical visual global processing within the dorsal visual pathway (Pellicano et al., 2005). Additionally, the extreme male brain theory, or E-S Theory, could be partially linked to atypical eye fixation perhaps inducing alexithymia. For example, higher order visual motion processing difficulties could affect the scan paths that ultimately lead to alexithymia (Bird et al., 2010) perhaps leading to decreased empathy. Empathy in the EMB Theory could also be affected by differing sensory signals received by the amygdala (Puts et al., 2011) perhaps due to the decreased GABA levels (Fatemi et al., 2009).

Chapter 5: Consider current intervention for sensorimotor difficulties in ASC and trial a possible intervention to help lessen the symptoms

4.7.1 Introduction

The aim of Chapter 5 is to apply the Sensorimotor Theory to possible intervention. In doing so it draws on two outputs: What Works for Improving Outcomes for Children on SEND Support: A Rapid Evidence Assessment (**Output 6**) and A Double-Blind, Placebo-Controlled, Crossover-Designed GABA Tea Study In Children Diagnosed With Autistic Spectrum Conditions (**Output 7**)

- 4.7.2 **Output 6, 'What Works for Improving Outcomes for Children on SEND Support: A Rapid Evidence Assessment'**, is a Department for Education Publication (In Press) that examines current support and intervention in the classroom for children with sensorimotor difficulties, and as sensory and motor impairments are intrinsically linked (Brooks, 1983), goals that encompass sensorimotor skills need to be built into the child's education, as without intervention no natural process will rectify such difficulties (Green et al., 2008).
- 4.7.3 This paper first considers adaptations that can be made for these children using the M.A.T.C.H acronym: Modifying the task; Altering expectations; Teaching specific strategies; Changing the environment; and Help by understanding (Missiuna, 2004). In addition to physical adaptations, this paper also advises on assistive technology.

- 4.7.1 The second part of this paper looks in greater detail at support with handwriting and fine motor control and asserts that at school age a top-down approach has been deemed more successful than remediation of motor difficulties (Cramm and Egan, 2015). Examples of top-down strategies include direct and task-driven instruction of handwriting having significantly greater results than motor instruction (Santagelo & Graham 2016; Hoy et al., 2011; Feder & Majnemer, 2007; Denton et al., 2006; Weintraub et al., 2008), with greater than 10 hours of direct instruction being considered effective (Santagelo & Graham 2016). Some physical adaptations aimed at bottom up approaches that have proven beneficial are also discussed (Srivastava, 2016; Zwicker and Hadwin, 2007).
- 4.7.2 The third part of this paper considers support for children with gross motor difficulties in a similar fashion. After indicating that early intervention is best practice (Baranek, 2002; Missuna et al., 2003; Blauw-Hospers et al., 2007; Goodway & Branta, 2003), by school age current research also favours approaches that focus more on top down acquisition of specific skills based on child chosen functional goals (Mandich et al., 2001; Caçola, et al., 2016). Example remediation includes approaches such as CO-OP (Cognitive Orientation to daily Occupational Performance (Rodger & Bradenburg, 2009). However, successful bottom-up approaches are also discussed, such as a 10 week table tennis training programme, (Tsai, 2009) or Physical Education lessons that focus heavily on balance training, such as controlled jumping, rotations and trampolining (Giagazoglou et al., 2015).
- 4.7.3 Finally, this paper considers Sensory interventions which fall into two broad categories: Ayres Sensory Integration (ASI) and Sensory-Based interventions (SBI), where although literature to support these as evidence-based programmes is still in its infancy, moderate evidence has been found to support intensive and individualised ASI programmes for improving functional outcomes (Watling & Hauer, 2015). Moreover, as with coordination, if the individualised programmes are tailored around a specific goal, the impact is greater still (Schaaf et al., 2014, Case-Smith et al., 2015)

OUTPUT 6 (Published)

'What Works for Improving Outcomes for Children on SEND Support: A Rapid Evidence Assessment'

Carroll, J., Johnson, H., Bradley, L., Crawford, H., Hannant, P. & Thompson, A., (2017). What Works for Improving Outcomes for Children on SEND Support: A Rapid Evidence Assessment. *DfE Publication*

Author Contribution:

For Output 6, Hannant was the lead author on the physical and sensory section of the publication. Specifically: **Page 14**, Teacher Awareness; **Pages 34-37**, Handwriting and Fine motor control; and **Pages 61-67**, Physical and Sensory Support. Carroll worked jointly on conducting a literature search and revising the manuscript.



SEN support: A rapid evidence assessment

Research report

July 2017

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Executive Summary

Background

Children and young people on SEN support are learners who have been identified with special educational needs (SEN) and require different or additional support to meet these needs, but do not have a statement of SEN or an Education, Health and Care plan (EHCP). It can be difficult for education professionals to know how best to support these individuals. However, in recent years there has been a dramatic change in the quality, quantity and availability of research evidence to support teaching, and this report aims to summarise recent research that addresses the issue of what practice is effective for learners on SEN support in mainstream schools and colleges.

Approach

The evidence was collected by means of a rapid evidence assessment, a search of the literature aimed at drawing out key findings in an area. The literature search focused on studies of approaches, strategies or interventions supporting children and young people with SEND in mainstream schools and colleges. Over a thousand papers were reviewed, with over 500 meeting criteria for inclusion.

Key Findings

The report is structured in terms of the four broad areas of need highlighted in the SEND Code of Practice 2015, with an additional introductory section on overarching issues that draw across more than one type of need.

- There is good quality research evidence about effective interventions in the areas of cognition and learning, social, emotional and mental health, and communication and interaction. However, the evidence about high quality teaching and adaptations that can support these needs is significantly less extensive.
- A key finding was the important role of training for all education professionals. Teaching assistants can provide good quality intervention if they are well trained, while even highly qualified professionals have less impact if they do not understand the principles and motivation behind the approach they are using.
- A second overarching finding related to the role of each stage of the graduated approach advocated in the SEND Code of Practice. While this review focused on interventions and support strategies, it was clear that detailed assessment of individual children is necessary to select the most appropriate approach, and

progress should be monitored when using any intervention to assess whether it is effective for that particular child.

A third broad finding relates to transfer. It can be tempting to assume that training to remediate a particular weakness will automatically improve the target academic skill (be it motor skills to improve handwriting, phonological skills to improve reading or memory skills to improve learning) – known as transfer. However, such transfer should not be assumed. In most cases, the evidence suggests that training needs to explicitly link the tasks being practised to an academic skill. Phonological training is most effective when explicitly linked to spelling and reading, and motor practise is most effective when explicitly linked to writing. Educators should be wary of programmes that do not make this link explicit.

Gaps in the evidence base

- The research evidence for supporting physical and sensory needs is much less extensive than for the other three areas of need and is often based on small scale case studies.
- Most existing high quality research is based on work in primary schools. There is much less research carried out in secondary schools and colleges.
- Most research studies provide a comparison between a particular approach and a 'no-treatment' control. This makes it very difficult to know which of two different approaches is likely to be more effective.
- Very little research examines individual differences in responsiveness to interventions. While all teachers know that certain approaches work better for some children than others, there is very little evidence as to why this occurs.

There is good quality research evidence to back up a range of approaches in supporting literacy difficulties, socio-emotional development and language and communication. This allows us to draw some conclusions about effective provision in these areas. More research is needed to understand which approach is better than another, and why some approaches work better for some students than others.

Introduction

The population of children and young people on SEN support

Around 12% of pupils in primary school and 10% of pupils in secondary school are on SEN support (DfE, 2016). In mainstream Further Education (FE) colleges, around 19% of 16-19 year olds and 16% of 19-24 years olds have a self-declared learning difficulty or disability (LDD), the majority of whom will be on SEN support (DfE, 2014). SEN support is the term given to children and young people who have been identified with special educational needs or disabilities (SEND) and require support, but who do not have an EHC plan or statement of SEN. Almost all children on SEN support are educated in mainstream schools rather than special schools or units.

Figures 1.1 and 1.2 show the proportion of children on SEN support in maintained mainstream schools with each primary type of need. Within primary schools, the most common primary types of need are speech, language and communication needs (28.4%); moderate learning difficulties (26.7%); social, emotional and mental health difficulties (16.1%) and specific learning difficulties (10.8%).



Figure 1.1 Number of pupils on SEN support in mainstream primary schools by type of need

In secondary schools, moderate learning difficulties are the most common type of need (27.0%), followed by specific learning difficulties (23.0%) and social, emotional and mental health difficulties (19%). The incidence of speech, language and communication

as a primary type of need is significantly reduced in secondary school (9.0%). Other primary types of need in children on SEN support include autistic spectrum disorder (4.7%), physical disability (2.2%), hearing impairment (1.6%) and visual impairment (0.9%).



Figure 1.2: Number of pupils on SEN support in mainstream secondary schools by type of need

It is important to remember, however, that these statistics reflect only the primary type of need for each individual child, and many are likely to have multiple types of need. Furthermore, some of the descriptors used do not indicate the types of support an individual needs. For example, one child with an autistic spectrum disorder may need support with language and with planning, while another might need support with social interaction and communication. In addition, these needs will differ according to the age of the individual and the demands set for them. A student with specific learning difficulties in further education may have greater issues with planning and self-organisation than with word reading and spelling. For these reasons, we have chosen to structure the review of interventions in terms of the aims of each intervention or adaptation, rather than in terms of the group of individuals for whom it is aimed. Nonetheless, where interventions are particularly aimed at specific groups, we will highlight this.

The research context

Over the past fifteen years, there has been a greatly increased emphasis on using research evidence to guide teaching practice. This has been exemplified in recent years

by the work of the Educational Endowment Foundation, which has funded a large number of research studies in different areas of education. Their focus has been on supporting children experiencing economic disadvantage, but much of their work is highly relevant for this review.

With this change has come an understanding that existing education and psychology research is not always well designed to provide guidelines for practice. Historically, education research has tended to be difficult to generalise to other contexts, as it is often small scale and specific to a particular situation. Conversely, developmental psychology research has focused on the child and taken relatively little notice of the context in which that child is learning.

In the past fifteen years, researchers have started to carry out larger scale studies with higher experimental rigour, combining psychology and education approaches. However, it can be difficult to draw conclusions from these studies for a different reason – the careful experimental control, examinations of intervention fidelity and randomisation mean that they create a very different context from that of a typical classroom. Ideally, for each intervention, we would have three types of research:

- Efficacy studies to demonstrate that the intervention can change outcomes;
- Effectiveness trials to demonstrate it can work in a normal school or college context, and
- **Process evaluations** to examine which elements of the intervention contributed to its success.

In reality, it is rare that we have all of these types of evidence.

There is also an issue with the comparisons used for the particular approach or intervention. It is rare that studies compare two different interventions: normally a particular intervention is examined in comparison to 'business as usual' or no-treatment control groups. This makes it difficult to know, in many circumstances, which of two interventions is likely to be most effective. This is a significant issue for future research.

Our task is to consider the existing research with respect to children with SEN in the classroom, and to highlight findings about what approaches may be most successful for children with different types of needs. However, we acknowledge that it can be difficult to draw those conclusions for some of the reasons outlined above, and therefore we point the reader to original sources and to relevant further resources where possible.

Method

The basis of this report is a Rapid Evidence Assessment (REA). The overarching question for the REA was as follows:

What strategies and approaches have been shown to be effective for supporting children and young people with special educational needs to achieve well in mainstream schools and colleges?

REA methodology guides recommend focusing on a single database (Thomas, Newman & Oliver, 2013). We used ERIC (Education Research Information Complete), the most comprehensive education database in the world. The initial search was carried out by an experienced research assistant.

The following search terms were used:

Special educational needs/difficulties/disorder/impairment

AND support/intervention/teaching/pedagogy/approach

AND learning /autism /attention/ dyslexia /speech /language / reading /hearing /visual /sensory /physical/social /emotional /behavioural/ conduct

The following exclusionary criteria were used:

- Studies were limited to those published between 2000 and 2016
- Studies must be carried out with children or young people aged between 4 and 19.
- Studies must report on an empirical investigation of an intervention, adaptation or approach: general good practice guides were not eligible for inclusion unless they reported on data.
- Single case studies were excluded, but multiple case studies were included in areas where there was little other research (autism, sensory difficulties and physical difficulties).
- Studies must be published in English in a peer-reviewed journal, but could be carried out internationally.
- Approaches were limited to those feasible in UK mainstream schools or colleges (e.g. approaches including medication were excluded).

The initial searches produced a list of 722 papers, which was saved to a shared Paperpile database. They were given preliminary labels according to the broad topic covered (i.e. language and communication, social, emotional and mental health, behaviour management, general cognitive skills, physical and sensory impairment). The papers were allocated to specialists with different areas of expertise for assessment (Professor Carroll: specific learning difficulties, speech and language difficulties and hearing difficulties; Dr Crawford: autism and learning difficulties; Dr Johnson: emotional and mental health; Dr Bradley: attention and behavioural difficulties; Ms Hannant: physical and sensory difficulties; Ms Thompson: executive function and learning difficulties).

Each of these experts then supplemented the database with further relevant papers as necessary when they found a search area had been under-represented. The advisory board also provided advice on further sources of evidence. This resulted in a final total of 1046 papers.

Following this, all studies were given three separate labels: a 'topic' label indicating which area or areas this paper was relevant to; a 'design' label indicating what research design was used, and an 'ecological validity' label indicating whether the research had been carried out in mainstream schools or colleges in the UK, in other settings, or by trained specialists (e.g. speech and language therapists or counsellors). Studies showing high experimental rigour and high ecological validity (e.g. randomised controlled trials carried out in UK schools) were given most weight in the report.

During this stage, papers were carefully examined to ensure they fit the criteria above, resulting in 505 papers being excluded. The majority of these papers did not report on empirical data but provided general reports of good practice.

The narrative report was supplemented where appropriate from findings from recent high quality reports carried out by the Department for Education, the Educational Endowment Foundation or relevant UK charities such as the National Deaf Children's Society.

Overarching issues

In reading the papers selected in the rapid evidence assessment, there were some issues that cut across the different areas of need. We have chosen to highlight some of these issues below.

Working with parents and families

The SEND Code of Practice (2015) highlights the vital role that parents play in supporting children and young people with SEND. This modified Code of Practice follows the Lamb report (2009), which suggested that parents were sometimes excluded or marginalised in decisions about their child, whereas they should be considered partners in their child's education, and a process of communication and engagement with parents is key.

Parents are the best placed individuals to provide details on the health and early development of their child and the support they have received outside the school system, particularly when a school is first assessing a child's needs. They can also provide information on whether any difficulties have been noted at home or elsewhere, to help to understand whether they are limited to or exacerbated by the school environment. All of this information is crucial for a thorough assessment of a child's strengths and weaknesses.

Parents of students with SEND may require additional support from educational settings in order for them to feel comfortable to be open and honest about their child's needs. Parents are often required to take on an 'advocate' role for their child and this can be a time consuming and emotionally laden responsibility. They may also have different concerns to other parents, for example, they may be more worried about whether their child is safe in school or college, whether they are making friends or what other parents may think of their child, rather than focusing on academic performance.

A key issue to consider is the goal of any support or intervention. In recent years there have been a number of debates raised about different interventions which have been put in place to support children and young people, particularly with regard to individuals with ASC. While these arguments are beyond the scope of this rapid evidence assessment it is important to acknowledge them and to remember that therapy or support should be based around the needs of the individual learner and their family's views rather than a desire to make a child 'fit in' to a classroom situation.

Parents can also provide support and consistency for any approaches or interventions used at school. The <u>EEF-Sutton Trust Toolkit</u> includes a review of 14 meta-analyses on the topic indicates that parental involvement in children's learning has a small positive effect, though most of the studies cited focus on young children and many on parents reading with their child. However, parents sometimes need support to know how best to
help their child: Maloney et al (2015) find that parents who are anxious about maths and help with maths homework can have a negative, rather than a positive, impact.

Assessment of learners' strengths and weaknesses

Our rapid evidence assessment focused on support, rather than identification or assessment. However, a recurrent theme across the literature is that the most effective support relies on a full and recent assessment of a child or young person's individual strengths and weaknesses. A wide range of underlying difficulties can cause certain symptoms or behaviours. For example, difficulty in following classroom instructions could indicate hearing difficulties, language difficulties, attention difficulties, short-term memory difficulties or frustration at other, seemingly unrelated situations such as friendships or home life. A child often seeming worried or anxious could have learning difficulties, sensory processing issues, worries from outside of school and so on. Until these different options have been explored and a full picture of a child or young person's strengths and weaknesses, in terms of cognitive skills, relevant medical issues such as hearing and vision, and family support, motivation and engagement is considered, support is likely to be sub-optimal.

This assessment does not need to be lengthy in most cases. Discussion with the parents is likely to provide much useful background information. Observation of how the child behaves in the classroom, small groups and individually can also provide a guide on what aspects of the learning environment a child finds most difficult. Hearing and sight tests can be carried out at the GP or the opticians at the request of the parents. This can usefully supplement information from academic tasks completed in school time.

Supporting learners with SEND in a classroom

Teacher awareness and effective training

Typically, teachers, especially secondary school teachers, receive minimal information on SEND as part of their initial teacher training. This knowledge is often something that comes informally, piecemeal and from experience. There are some good reasons for that: children and young people with SEND show a huge variety of individual needs, and it is not normally useful to assume that 'all' those with a particular need will require the same type of support. Mitchell (2014) suggests rather that:

"my strong advice to you is that you should develop a repertoire of such strategies nested within your own philosophy...professional wisdom, and above all knowledge of the characteristics and needs of your students". We intend that this report, and the associated reports, will help teachers to develop this repertoire.

Classroom based versus individualised support

The requirement for children and young people with SEND to have their needs met in inclusive educational settings has become established over the past two decades. The SEND Code of Practice advises that schools should provide a culture that has high expectations for those with SEND and must facilitate participation and achievement.

An issue that is often raised is the role of mixed ability classes or groups versus setting by ability. The <u>EEF-Sutton Trust Toolkit</u> indicates that setting is disadvantageous to lower achieving learners in most cases. Being in lower sets is associated with lower self-belief on the part of the learners, as well as lower expectations on the part of the teachers. Both of these factors combine to mean that that lower achieving students do less well on average in classrooms where they are placed in ability groupings.

Creating a truly inclusive classroom is challenging, and as Mitchell (2014) highlights, most of the research studies examining classroom climate do not focus on learners with special educational needs, and are therefore beyond the scope of this review. However, it is clear that an environment in which learners feel emotionally safe, in which there are clear rules, predictable consequences and positive goals is good for those with and without SEND.

In recent years, schools have made more use of techniques such as collaborative learning or peer tutoring in order to support mixed ability teaching. These approaches can be very helpful for learners with SEND, as well as for typical learners, as described in the <u>EEF-Sutton Trust Toolkit</u>. However, peer tutoring needs to be carefully planned and managed, with appropriate training for the tutors and a task set at the correct level, where it is challenging but achievable for the tutee. Mitchell (2014) notes that learners with SEND can also be effective tutors, particularly tutoring younger children, and that this can help to build confidence and consolidate knowledge. For more information, see the section below on Peer-mediated social skills training.

Effective use of Teaching Assistants

In UK schools, teaching assistants form a significant part of the workforce, providing much of the support for children and young people with additional needs. The Educational Endowment Foundation has recently published a <u>review</u> of the effective use of teaching assistants, and our review of the existing evidence on SEN support is in line with the conclusions from this report. Teaching assistants can certainly provide benefits to learners with special educational needs, but they need to be appropriately supported to do so.

For example, teaching assistants can deliver structured, evidence-based interventions effectively to individuals or small groups, but they should be given proper training, a thorough knowledge of a child's strengths and weaknesses, and adequate time to prepare and record their teaching.

Teaching assistants should be working as part of a team with the classroom teacher, SENCO and relevant professionals to ensure that the additional support is linked to the curriculum and that the teaching assistant is following best practice. In many cases the teaching assistant is able to spend more time than the class teacher working closely with and getting to know an individual child, and their knowledge of that child should form part of the overall planning process.

Motivation and engagement

There is a growing body of evidence that a child's motivation and engagement is a key factor in how they respond to support or intervention. Motivation can be divided into two types: intrinsic motivation (doing an activity because you want to) or extrinsic motivation (doing a task for external reward or acknowledgement). Intrinsic motivation is associated with good progress in school, while extrinsic motivation is associated with poorer progress. This may be partly because it can encourage a 'surface' approach to task completion, where a child aims to complete a task as quickly and easily as possible, rather than trying to understand the task and produce their best work. Unfortunately, children with SEND often have low intrinsic motivation to complete academic tasks for multiple reasons, including because of prior experience of failure, and educators are often tempted to provide extrinsic motivators as a substitute.

Ideally, it is better to foster intrinsic motivation for a task rather than to use extrinsic motivation (tangible rewards, merit points or golden time). Extrinsic motivators can have a role in some situations (for example, to encourage novel behaviours to become habitual), but should not generally be relied upon long-term. Positive verbal feedback does not act as an extrinsic motivator in this way (Deci, Koestner & Ryan, 1999), and is positively associated with intrinsic motivation.

Intrinsic motivation can be increased in the following ways:

- By setting tasks that are challenging but achievable, to allow experience of success in academic tasks. This allows the development of self-efficacy (belief that one can achieve a particular task, Bandura, 1990)
- By highlighting the intrinsic value of the task (how it will be useful in the future) rather than focusing on exam results or external rewards.
- By allowing learners some degree of autonomy in setting their goals or selecting the materials to use.

• By encouraging a 'growth mind-set' that all learners make mistakes and that mistakes can be learning opportunities rather than failures (Dweck, 2000).

Deci & Ryan (2000) argue that the distinction between extrinsic and intrinsic motivation is too crude. Learners may do a task for a fully extrinsic reason (e.g. to directly receive a reward), for a mainly extrinsic reason (e.g. in the expectation of receiving social recognition), for a mainly intrinsic reason (e.g. because doing school work is what they are supposed to do) or for a fully intrinsic reason (e.g. they get inherent satisfaction from the task). Partially intrinsic motivations are also associated with good academic progress, and so it is useful to encourage partially intrinsic motivation as well as fully intrinsic motivation.

Supporting students post-16

Learners between the ages of 16 and 18 are expected to either be in education or undertaking an apprenticeship or traineeship. This therefore means that many more students with SEND require support with their education after the age of 16. The government has produced a guidance document for further education establishments implementing the SEND Code of Practice. This sets out that colleges have a duty to use their best endeavours to provide support for individuals with SEND who are registered with their institution. In some cases, this will be up to the age of 25.

There is, unfortunately, a much weaker evidence base on what works to support students with SEND in adolescence and young adulthood. There are so many differences in students of this age group in comparison to younger ages that it is very difficult to draw conclusions on what works from studies on school children. We highlight some of the findings here, and return to specific findings with post-16 students in later sections where possible.

Some research has examined the characteristics of individuals who have successfully overcome learning difficulties. While these are often qualitative, retrospective reports, they can provide helpful information. Reis et al (2000) argue that schools focus on remediation of basic skills rather than developing compensatory strategies for weaknesses. It is likely that compensatory strategies are particularly useful at the college level, when students start to manage their own learning to a greater extent. Compensation strategies highlighted by successful students include study skills, time management, organisation, memorisation strategies and the use of assistive technologies such as Dictaphones and speech to text programmes (Ries et al., 2000). Many of these are discussed in more detail in the section on executive function below.

Transitions to employment

Hakkarainen et al (2016) examined the education and employment outcomes at age 20 of learners who had had literacy, numeracy, behavioural or social difficulties at age 15. They found that having literacy difficulties or behavioural difficulties was associated with not graduating from high school, while numeracy difficulties and social difficulties were associated with individuals being not in education, employment or training (NEET), raising the possibility that different sets of skills are associated with educational success and career success. However, it is difficult to generalise from this work, as the sample size is relatively small (590 students across the range of ability) and the study is carried out in Finland, in a different educational context.

Goldman-Mellor et al (2016) examine the characteristics of young British people who are NEET. They found that individuals with a history of behavioural difficulties, attention deficit hyperactivity disorder (ADHD) or depression in childhood are much more likely to be NEET at age 18, even after accounting for socio-economic background, cognitive abilities and reading skill. Adolescents with mental health difficulties are particularly vulnerable to becoming NEET in adulthood, and therefore should be carefully supported and prepared for the transition to employment.

Literacy support in key stage 4 and post-16

- Less strong support for specific interventions for older learners, though these can be useful if they are supplemental to mainstream provision rather than replacing it
- Provision needs to be catered around the wishes of the learner
- Assistive technology can be very useful in supporting older learners

Supporting writing skills

Writing is a common area of difficulty for children and young people with SEND. Effective writing is key to allowing a child to communicate and demonstrate their knowledge. The effectiveness of writing is a mediating factor on academic outcomes and associated life chances and therefore of strategic importance to those with SEND. Learners with literacy difficulties usually also demonstrate writing difficulties, and these can be more difficult to resolve than reading difficulties.

Berninger (1999) argues that successful writing depends on a triad of skills: transcription skills (handwriting, spelling and punctuation knowledge); language skills (vocabulary and knowledge of argument structure) and working memory (required to hold information in mind while writing). When pupils are asked to do a task involving writing-by-hand/writing there are several concurrent demands being made of them. The outcome of their work and barriers for access to education encountered in this activity are mediated by any one or more areas of difficulty described below, which may not be immediately obvious. It is therefore important to assess a child's skills in each area. Although the outcomes of almost all pupils' work in mainstream education have a common presentation, most often work completed in school books or on test papers, they are in fact very specific and individual in their formulation. Consequently, deficits need to be explicitly addressed at an individual level. Recognising that there can be multiple levels of developmental need and difference for a child with 'writing difficulties' regardless of whether they are numerical, symbol or alphabetic in form is an essential first step, and while addressing these areas of deficit ensuring that barriers to educational access are mitigated or removed. Information in the Speech and language and 'Thinking Skills' interventions sections. Support for language difficulties and executive function difficulties are dealt with elsewhere in this review.

The sections below outline some evidence demonstrating the complex nature of writingby-hand difficulties, the impact of technology on this activity, and the broad area of writing. Harris & Graham (2013), studied the transition from novice to competent writer and observed for this process there were many levels of changes and transitions pupils undergo around knowledge about writing, motivation to engage and what was termed writing behaviours.

Support for learners with fine motor and handwriting difficulties

Clear, legible and fluent handwriting is necessary to embed spelling patterns (Cunningham & Stanovich, 1990) and also to ensure that a child can adequately demonstrate their abilities in other areas of the curriculum. Handwriting is associated with other tasks that require detailed finger movements, such as using cutlery or scissors accurately, (Smits-Engelsman, 2001), finger gnosis (recognition) (Feder & Majnemer, 2007) and with motor learning, described by Zwicker and Harris (2009) as "a set of processes associated with practice or experience leading to relatively permanent changes in the capability for movement". As a consequence, studies have shown that both *motor instruction* and / or the *direct teaching of handwriting* greatly increases legibility.

However, problems in 'bottom up' visual motor integration and fine motor control do not appear to preclude the development of functional handwriting (Klein et al, 2011), as proprioceptive information and motor learning appear to be the fundamental elements of handwriting (Vinter & Chartel, 2010). Consequently, by school age, current best practice favours a top down approach, focusing on handwriting practice and the development of essential motor programs (defined as communications in the central nervous system that are based on past experience and can generate planned postural adjustments and movements, Brooks 1983), as opposed to remediation of motor difficulties (Cramm and Egan, 2015). This direct instruction of handwriting has been shown to result in significantly greater legibility and fluency than non-instruction and motor instruction (Santangelo & Graham 2016; Hoy et al, 2011; Feder & Majnemer, 2007; Denton, 2006). More specifically, hand manipulation and kinaesthetic awareness training have been shown to not be as effective, and sometimes deemed ineffective, without handwriting practice (Datchuk, 2015); with greater than 10 hours of direct instruction being considered necessary (Santangelo & Graham 2016). This can be observed where kinaesthetic training has been compared to direct task driven instruction. A study by Weintraub et al, (2008) targeted at a sample of 55 children between the second and fourth grades, demonstrated that task driven *direct handwriting instruction*, such as using different writing tools in different activities to consistently practice handwriting, made and maintained significantly more gains in handwriting when compared to kinaesthetic training based on a multisensory program that included making letters from play dough, writing in rice and oral letter stating, with some handwriting instruction. Direct teaching of handwriting can include the following: individualised instruction, using technology, copying models from memory, using self-evaluation, facilitated peer modelling and evaluation, and teaching letters with motor models (Santangelo & Graham, 2016; Case-Smith, 2014).

The top down approach is favoured as best practice. However, visual perception difficulties appear to be associated with the speed of handwriting (Brown & Link, 2016; Poon, et al 2010) and therefore bottom up approaches can be successful in this area. For

example, computerised *visual perception training* increased the speed of handwriting in Chinese first grade children perceived to have handwriting difficulties by their teachers (Poon et al 2010). Additionally, physical intervention on postural stability and vestibular awareness has also been shown to predict manual skill (Flatters et al., 2016), although the outcome measure in this instance was tracing and not handwriting.

There are also some physical adaptations aimed at bottom up approaches which have proven beneficial (Srivastava, 2016; Zwicker & Hadwin, 2007). Examples here include *Thera bands* to strengthen finger muscles, using paper towels under pages to increase awareness of pressure, using textures to finger trace letter shapes and a slanting desk to support vision and posture. Thus, difficulties in handwriting practice would benefit from consultation between health and educational professionals as this leads to tailored co-teaching and frequent concise feedback (Case-Smith et al, 2014). Interestingly, adapting pencil size and width do not appear to impact writing, despite children in preschool appearing to prefer short skinny pencils and children in kindergarten picking long oversized pencils to write with (Sinclair & Szabo, 2015).

While some handwriting programmes follow goal driven handwriting practice and others have sensory considerations, some programmes include cognitive interventions and are based on learning theories that involve self-instruction and verbal mediation, such as including the ability to name and identify letters, modelling, imitation, discussion and practice (Zwicker and Hadwin, 2007; Graham et al, 2000). An example of such an intervention is the randomised control trial by Zwicker and Hadwin (2007), which assigned 72 first and second grade Canadian children to cognitive intervention, multisensory intervention or a control group. The first-grade children's handwriting improved with or without intervention, however the second-grade children showed a sizeable improvement with cognitive intervention compared to multisensory.

Additionally, it is important that self-esteem is considered in handwriting practice and child directed and self-determined goals used as important motivators (Hoy et al 2011; Datchuk, 2015). Examples of such incentives have been identified in technology 'treatment media' (Poon, et al 2010) where children have been eager to participate in visual perception and *visual motor integration* training, although this intervention was found to increase speed of handwriting; not legibility. This finding was mirrored in Santangelo & Graham's meta-analytic review (2016) and in a small-scale U.S. study by Wells et al., (2016) demonstrating that traditional methods of handwriting practice are deemed more successful in terms of letter recognition and legibility, whereas letter instruction using tablets increased productivity.

Sumner, Connelly and Barnett (2014) and Prunty (2016) have examined the difficulties shown in poor writers using tracking technology. They found that these children could form letters as quickly as typical writers, but were more likely to pause within and

between words. This suggests that writing difficulties are often not explained by difficulties in letter formation but by broader planning difficulties.

Handwriting Support

- Difficulties in handwriting are often associated with difficulties in planning rather than movements
- Evidence suggests that 'top down' or 'cognitive' approaches are most effective that is, children practicing writing letters rather than working on general motor movements
- This is particularly suitable for younger learners.

Developing the content of writing

Torgerson and Torgerson's (2014) examined the effectiveness of a writing strategy approach (instruction that involves explicitly and systematically teaching steps necessary for planning, revising, and/or editing text). This robustly designed study examined one approach, *Self-Regulated Strategy Development* (SRSD). They found a large positive impact on the writing skills of pupils who were not expected to meet the minimum threshold for literacy at the end of KS2. However, this intervention had no significant impact on spelling, grammar or reading, so evidence of transfer was limited.

Andrews et al (2004) found the teaching of grammar was better accomplished by using the strategy of *sentence combining* than formal instruction. Sentence combining strategy is where sentences of increasing complexity are created and grammar and syntactical rules are applied within the activity to achieve this. This is consistent with Graham and Perin's (2004) meta-analysis which found a small statistically significant negative effect on writing for the explicit and systematic teaching of part of speech and structure of sentences. One consideration therefore for children with SEND is to make use of sentence combining strategies to both secure grammar and increase the efficiency and effectiveness of higher order expression. This can aid ability to work within specified time frames by creating less volume of writing to express ideas and thoughts.

Self-regulated strategy development

- Explicit teaching of the steps needed for planning, writing and editing text is effective in improving the writing of children in key stage 2
- Does not generalise to related skills such as reading and spelling

Type of need 4: Sensory and/or physical disabilities

In the SEND Code of Practice sensory and / or physical needs are categorised as one of the four broad areas of need and support. This is a wide ranging category including children and young people with physical disabilities, vision (VI), hearing (HI) and multisensory impairments (MSI). We have chosen to also include in this area the following needs: co-ordination difficulties (including both fine motor difficulties such as handwriting and gross motor difficulties), and sensory processing difficulties (often, though not always, associated with ASC). Approximately 2.2% of children on SEN support have physical disabilities as their primary type of need, while 1.9.% have a hearing impairment, 0.9% have a visual impairment and 0.2% have a multi-sensory impairment. Because some of the needs covered are relatively rare, schools may have little experience on how to support children with these specific needs.

The impact of sensory processing, coordination, sensorimotor difficulties or impairment of the senses not only hinders learning and cognition but can have a pervasive and serious effect on the emotional wellbeing of children and young people, and further impacts life chances in adulthood (Gagnon-Roy 2016). In the following sections, we first address motor difficulties (both in terms of gross motor difficulties and fine motor difficulties), visual impairment, hearing impairment and sensory processing difficulties

High Quality Teaching and adaptations for motor difficulties

For many individuals with physical needs, physical adaptations in the environment are very effective ways to ensure learning is accessible. In many cases these do not require research evidence as their effectiveness is immediately obvious. These could include: ramps, writing slopes, stand / sit stations, spring-loaded scissors and pencil grips. Considering purposeful adaptations using the M.A.T.C.H acronym can have considerable benefits: Modifying the task; Altering expectations; Teaching specific strategies; Changing the environment; and Help by understanding (Missiuna, 2004). It is often assumed that such adaptations are universally employed as aids in physical conditions. However, without a firm understanding of the difficulties children and young people with physical needs face in the classroom, these aids will be less effective. Providing adaptations with support from a specialist professional is likely to be the most effective approach.

The <u>Dyspraxia Foundation</u> also provides guidance on supporting children with coordination difficulties within the classroom, which is useful regardless of whether or not a child has a diagnosis of dyspraxia. This guidance is separated out by type of difficulty experienced by the child, for example difficulties with balancing, self organisation, bumping into people or objects, handwriting or getting dressed.

In addition to physical adaptations, assistive technology is also useful within the classroom. When taking the child's age into account the following technologies can be beneficial: touch typing (which is shown to improve legibility and motivation; Klein et al, 2014); ergonomic keyboards; predictive text; speech to text and mind mapping software; and assistive APPs such as those that allow information to be typed directly on to worksheets (such as *SNAP TYPE*) and those that help line up columns in mathematics (such as *MOD MATH*). However, it should be observed that the use of assistive technology does require extensive training and supportive environments.

As children grow older, it may be most effective to focus support around specific functional goals selected by the child. For example, considering from the child's perspective: a specific social interaction problem, a particular loop in handwriting that is impeding letter formation or a particular sporting activity, and then identifying and developing the sensory and motor behaviours and procedures that support the activity (Schaaf et al, 2014).

Support for learners with gross motor difficulties

Children who have difficulties with coordination struggle to engage in activities that form an essential part of child development. Such activities include not only educational tasks, but also the taking part in and understanding social activities, and adapting functionally to home life (Wang et al, 2009). Research has indicated that the participation in childhood and educational activities impacts cognitive, affective and physical development (Mandich et al, 2001). Moreover, many children with atypical coordination also display challenging behaviour and attention difficulties (Tsai, 2009), where motor problems can go unnoticed (Rivard et al, 2007). Yet, children who do not have their coordination needs supported often have on-going academic and social frustration and go on to have mental health challenges (Gagnon-Roy, 2016), and are more likely to live with their parents in adult life when compared to children with other learning difficulties (Kirby et al, 2008).

The prevalence of gross motor coordination difficulties should be considered. In Europe it is thought that the current prevalence of Developmental Coordination Disorder or Dyspraxia is between 5-6% of children, although other quoted percentages fall between 5-20% (European Academy of Childhood Disabilities Guidelines, 2011). In addition to this, motor coordination deficits are often found to be co-occurring in other neurodevelopmental conditions such as ADHD, language impairment, ASC, sensory processing disorders and specific learning difficulties such as dyslexia. Furthermore, conditions such as prematurity, cerebral palsy, traumatic brain injuries and malignancies also present with severe motor difficulties.

In a similar pattern to fine motor skills, early intervention that helps support the underlying processing deficits and facilitates neuromaturation are recommended as minimum 'best practice' (Baranek, 2002; Blauw-Hospers et al, 2007; Goodway & Branta, 2003), and

historically this pathway to intervention has continued through school age. However, at this stage, current research favours approaches that focus more on the acquisition of specific skills based on child chosen functional goals (Mandich, et al, 2001; Cacola et al, 2016; Sugden and Chambers, 2007). Such remediation suggests that intervention needs to be functional, goal-based, structured, errorless and non-generalised (meaning that learning needs to be practised in different environments and conditions; Jackson, 1999).

One such approach is CO-OP (Rodger & Bradenburg, 2009). CO-OP is a task-orientated problem-solving approach that has three main objectives: skill acquisition in a child chosen task; development of cognitive strategies to acquire and apply the task; and the ability to generalise and transfer the learned skills to various scenarios and applications (Missiuna et al, 2001), where skill acquisition is achieved in stages, first with verbal guidance from a therapist, then self-dialogue and eventually on to independent application (Polatajko & Mandich, 2004). In a Canadian pilot study, targeting twenty 7-12 year olds, CO-OP intervention was compared to more traditional methods focusing on the 'bottom up' motor aspects of skill acquisition, such as the multi-sensory and biomechanical approaches. The results demonstrated the child task directed CO-OP approach had greater benefits and more long-term gains (Miller et al, 2001). This method was also found to be beneficial in a UK based longitudinal cross-over intervention study by Green et al (2008), where one hundred children aged between 5 and 10 years old received CO-OP. This finding was reiterated in a critical review by Armstrong (2012) where CO-OP was found to be the most effective method of improving occupational performance. Such child centred approaches may also be underpinned by the Self Determination theory where the interpersonal behaviour of significant others in the child's life (such as parents and teachers) have the ability to influence the child's motivational behaviour, both intrinsically and extrinsically (Katartzi & Vlachopoulos, 2011).

CO-OP

- Effective in improving motor skills
- Flexible approach suitable for a wide range of tasks
- Should be delivered by a trained therapist

However, as for fine motor skills and handwriting, despite top down approaches showing more promise at school age, bottom up approaches should not be discarded, as a longitudinal study over a period of three years based in Sweden demonstrated improvements in motor skills and attention with extended physical activity and motor skill training (Ericcson 2008). Furthermore, a 10 week table tennis training programme, based in a Taiwan school setting, showed significant improvements in motor and cognitive function in children aged 9-10 years of age with coordination difficulties (Tsai, 2009). Physical Education lessons that focus heavily on balance training, such as controlled jumping, rotations and trampolining, have also proven to be significantly effective in

improving motor function by reducing movement flaw, side asymmetry and redundant movement while improving body posture, contraction, force, rhythm, balance and body stability (Giagazoglou et al, 2015).

Exercise training programmes

- Suitable for a wide range of ages and abilities
- Engaging and fun for the pupil
- Evidence suggests it is beneficial, though not as beneficial as other approaches

Attention to balance within *interactive video games*, now a pervasive part of modern society, has also been successfully explored. For example, in a study based in the Netherlands recruiting forty-eight children aged 6-12 years of age, balance was the specific focus, such as video games that require the use of a balance board. This study showed significant improvements in balance, bilateral coordination, speed and agility (Jelsma et al, 2014). However, technological intervention has also been used as a potential task driven intervention, that increases opportunities to practice motor skills and thereby influence overall movement outcomes as opposed to centring specifically on balance (Straker et al, 2015). In these trials participants were encouraged to play a number of motion-based games that involved both upper and lower limb movements, in addition to both fine and gross motor skills. Results demonstrated that despite perceived differences in motor skills from the participants, no significant gains were achieved. A similar study demonstrated that motion based video games did not improve objectively measured physical activity and sedentary time in these children (Howie et al, 2016).

As parents and educators often have a limited understanding of identifying and supporting a child's specific coordination needs, many interventions involve input from professionals who can help children achieve a purposeful activity or functional outcome (Watemberg, 2007). A scoping review by Camden et al (2015) discusses the need to organise these services efficiently to increase awareness, define the graduated approach and work collaboratively with parents and educators to offer evidence based interventions, as best practice. Further studies have demonstrated that the teaching principles associated with physical and occupational therapy underpin motor skill acquisition, such as being able to give clues to adjusting body position in order to perform a task or sharing knowledge about executing movement in a certain way. Such knowledge transfer further supports the aforementioned cognitive approaches, such as CO-OP, where therapist knowledge is considered a pre-requisite for the use of the cognitive strategies involved (Niemeijer et al, 2006). However, time constraints and funding can make such collaboration ambitious and so exploring knowledge transfer systems and partnerships that are both effective and economical is vital when considering value for money. Online training has been trialled in Canada, where a selfhelp tool was designed to give parents information on coordination difficulties in different

environments, in addition to strategies to support their children to improve skills, and then transfer these skills to those involved in their care. Understanding, changing attitudes and trialling strategies were the main outcomes of the self-help tool (Camden et al, 2016). However, such systems to disseminate and impart knowledge are known to have high attrition rates, in addition to perhaps generating resentment between educators and parents (who are sometimes not recognised or regarded as the professionals). Three way partnerships, between parent, educator and therapist have been identified in a Partnering for Change (P4C) Model (Missiuna et al, 2012). In this model, the child remains the centre of focus, however as opposed to the therapist becoming directly involved, the emphasis is on coaching teachers to increase their awareness of and ability to support children with coordination impairment. Thus, the model is based around the imparting of knowledge and research about the needs of the child.

Support for learners with sensory processing difficulties

Atypical sensory reactivity has a widespread impact on daily life skills and has a high prevalence rate in a number of childhood conditions, including ASC, ADHD, developmental coordination disorder and sensory processing disorder. While atypical sensory processing can be hard to identify for an inexpert, a number of books are rich in examples of how to recognise symptoms and make reasonably straightforward adaptations to help reduce sensory reactivity in the classroom, such as chewellery, diffuser necklaces, textured materials and ear plugs. Furthermore, a number of National Health Service <u>Occupational Therapy guidelines</u> are also available and are often divided into the type of sensory processing difficulty present.

Sensory interventions to address such difficulties fall into two broad categories: *Ayres Sensory Integration* (ASI) and *Sensory-Based interventions* (SBI), where ASI targets neuropsychological mechanisms that process sensation and SBI uses a number of strategies that use sensory input to effect behaviour change (Watling & Hauer, 2015). Literature to support these as evidence-based programmes is still in its infancy and more classroom based studies are needed. However, a recent review by Watling & Hauer (2015) found moderate evidence that intensive and individualised *Ayres Sensory Integration* improves functional outcomes. Moreover, as with coordination, if the individualised programmes are tailored around a specific goal, the impact is greater still (Schaaf et al, 2014; Case-Smith et al, 2015).

A pre-and post-test study in Iran, targeted at thirty-four children aged 4-8 years of age, demonstrated that sound therapy could reduce symptoms for children who display atypical auditory sensitivity such as poor registration, hyperacusis or tinnitus, (AbediKoupaei et al, 2013). *Sound therapy* has also been shown to enhance spatial-temporal performance (Jenkins, 2001), visual-motor integration (Hall & Case-Smith, 2007) and reduce postural sway (Ross & Balasubramaniam, 2015), although the latter

study was carried out on adults without sensory processing difficulties. Sound therapy involves listening to some filtered sounds, such as sounds of the mother of the child, Mozart's music or white noise (Ross & Balasubramaniam, 2015). A study based in Hong Kong, has also demonstrated that visually, ambient prism lenses, which have a pair of wedge prisms as opposed to refractive lenses, can support posture and have a positive impact on behaviour in children (Carmody et al, 2001).

As mentioned previously, a number of books and guidelines from professionals are available that can help teachers support children with sensory processing needs. However, a child's sensory profile is specific to the individual, consequently, a 'one size fits all' approach is not possible. Recognising how specific aspects of a child's sensory reactivity impacts on their education, social awareness and daily life can channel precise support where it is necessary. Therefore, further evidence based studies on interventions focussing on some of the common overriding everyday themes of children with sensory processing disorders, are essential in order to help integrate these children into classroom life and alleviate the many demands on the child as they sit in a lesson, even before they begin to learn.

Supporting learners with visual impairment

Teaching a child or young person who has impaired vision can bring with it new challenges for teachers within a mainstream school, however with training and an understanding of the difficulties these learners face, it is possible for inclusive practice to occur and for these children to progress through the education system alongside their peers. Adaptations, such as Braille, have an immediately obvious impact on how these children and young people can access the curriculum. However, as with all types of SEND, it is also important to consider the child's social and emotional wellbeing as part of their package of support.

The classroom environment

The Royal National Institute for the Blind (RNIB) provides <u>useful guidance</u> on supporting children and young people with visual difficulties within the classroom. This guidance is divided by age (including learners accessing Further Education) and national curriculum subject.

For example, the lighting, windows and wall displays in a classroom can have a significant effect on how well a pupil with visual impairment can move around the space. When communicating with pupils, using their name first allows the visually impaired child to be aware when you are talking to them. It is important to recognise that a visually impaired child might not have access to non-verbal communication such as eye contact, which can make it more difficult to communicate effectively.

Unfortunately, while high quality 'best practice' guides exist, as described above, there is relatively little direct research evidence about high quality classroom teaching to support pupils with visual impairments. Nonetheless, evidence based best practice includes teaching pupils with VI within the mainstream classroom and not as a parallel lesson alongside and adapting the curriculum to encourage social participation of the student, helping to develop their social skills (Davis & Hopwood, 2002). An example of such teaching could be to include trained peer tutors who are able to provide instruction, feedback, support and monitor their tutees' behaviour. Such peer mentoring has been shown to increase the scores and confidence of children with vision difficulties (Wiskochil et al, 2007). However, perhaps the most important pre-requisite of teaching a child with vision impairment is for the teacher to work with professionals with specialist knowledge, which can be filtered down to those who work with the child (Douglas et al, 2011; Davis & Hopwood, 2002).

Adaptations

Often assistive technology is utilised to help children and young people with vision impairment. Examples include screen-reading or text-to-speech software, scanners with OCR (Optical Character Recognition) and refreshable braille displays, large font size and filtered backgrounds. However, a number of studies have demonstrated that the impact of such software is often associated with the knowledge level of those who use it, with teachers highlighting the importance of adequate training (Wong & Cohen, 2016). Also, updating information and communication technologies regularly is essential as technology changes rapidly (Fichten et al, 2009).

Although adjustments within the classroom are crucial (such as adjusting light, considering furniture placement, having large print) / Braille and talking books) to enable access to the curriculum, Douglas et al (2011) noted that teachers educating children with visual impairment should also ensure that the children themselves are suitably equipped to independently access resources.

Supporting learners with hearing impairment

In recent years, recognition and support for children with hearing impairment has improved considerably, with newborn hearing screening picking up many children with hearing loss at birth, and hearing aids and cochlear implants becoming much more widespread. Hence, many children who might previously have been unable to access spoken language can now perceive and produce speech well. These children might have been educated in specialist schools in the past, but now typically access a mainstream curriculum. However, that is not to say these children do not face difficulties in the classroom. Children with all types of hearing loss (whether mild, moderate, severe, profound or transient) are at increased risk of having speech, language and communication needs (Dalton, 2011), and these needs should be supported as necessary.

The National Deaf Children's Society (NCDS) provides <u>useful information</u> on supporting children with hearing loss within the classroom, including a series of commissioned research reports.

High Quality Teaching

It is important to remember that while children and young people with hearing loss may be able to perceive speech well in quiet, one to one settings, hearing and understanding in a classroom setting can be much more demanding. This is true for learners with mild and moderate hearing impairment, or transient hearing impairment due to glue ear, as well as for learners with severe or profound hearing impairment (Archbold et al, 2015). As with visual impairments, schools should work closely with specialist teachers of the deaf to ensure the support provided is of the best quality.

The classroom environment

There is evidence that creating good listening conditions in the classroom will have wide benefits, improving the learning of children and young people with all types of hearing loss, including mild and moderate hearing loss and transient hearing loss due to glue ear. It is also likely to help children who have difficulty focusing their attention (Dockrell & Shield, 2006). A <u>National Deaf Children's Society</u> report described measures that are most likely to be effective. These include reducing reverberation in classrooms by using carpeting and fabric wall displays, using soft pads on chair and table legs. There is also evidence that using a sound field classroom amplification system can improve academic performance across the class (Taub et al, 2003).

Students with hearing impairment in Further Education

The majority of deaf school leavers go on to further education in the UK, but statistics suggest that they often do not progress well in this environment, with many students dropping out or gaining no qualifications. A recent research report (Young et al, 2015) suggests that it is important that students with hearing impairments should be given *student-centred support*: in other words, the student is given whatever tools are needed to communicate effectively and is aware of all the possible options and support available to them, and is given structured support for both educational and emotional development.

Adaptations

There are some straightforward things that teachers can do to support children and young people with hearing loss. For example, ensure that the teacher is well lit and facing the student to maximise the chances of successful lip-reading, and ensure that

they can use any hearing loops or similar equipment easily. It is also useful to ensure children with hearing difficulties are seated at the front of the class and away from environmental noise sources (e.g. ventilation systems, traffic noise).

Cued speech

Cued speech is a system of hand gestures used to disambiguate speech sounds for individuals who are speech reading (lip-reading). There is a body of good quality research in France and elsewhere (Bouton et al, 2011; Leybaert, 2003) suggesting that cued speech is a useful way to improve spoken language and phonological awareness, though at present this is not widely used in the UK. LeBlanc (2004) describes the successful use of cued speech in an American school.

Interventions

Literacy interventions

Children and young people with hearing loss typically have difficulties with a range of oral and written language skills, even if they are using hearing aids or cochlear implants (Vermeulen et al, 2012). Nonetheless, they are able to learn spoken language, and this is beneficial for learning to read (Bergeron et al, 2009). There is evidence that young children with moderate to severe hearing loss, but some speech perception skills, can benefit from an intervention that combines phonics and vocabulary tuition with dialogic storybook reading (Lederberg et al, 2014). Children with hearing loss can also benefit from input from specialist speech and language therapists (Herman et al, 2015).

However, it remains the case that many deaf adolescents show low literacy attainments (Harris & Terlektsi, 2011), even those who have had cochlear implants in the first few years of life. It seems that even children who have made good early progress start to fall further behind their peers in secondary school (Geers et al, 2008). There is an evidence gap in terms of understanding how to best support adolescents with hearing impairment.

Language interventions

It is established that the levels of English vocabulary a child with hearing impairment shows is a key predictor in their academic progress (Maybery et al, 2011). Messier & Wood (2015) show that e-books with embedded word definitions can be an effective tool in increasing vocabulary in children with cochlear implants.

The Nuffield Foundation (2009) describe an approach to reading and language for deaf children that focuses on increasing morphological knowledge. It is well established that teaching morphology is a useful approach to teaching literacy for hearing children, and as learning morphological spelling patterns does not depend on phonology to the extent that learning phonics does, one might expect this would be an effective approach, but

unfortunately research on this programme has not yet been published in a peer-reviewed journal.



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- 4.7.7 **Output 7, A Double-Blind, Placebo-Controlled, Crossover-Designed GABA Tea Study In Children Diagnosed With Autistic Spectrum Conditions,** will be a journal article intended to be submitted to Molecular Autism, that builds on early intervention by considering sensorimotor pathology, neurology and endocrinology differences in children with ASC, and in doing so by explores the effect of introducing a non-evasive GABA substitute, in the form of GABA Oolong tea, on the sensory, motor, emotional, anxieties and sleep of children with ASC. The detailed background and theory under-pinning this study is also presented in **Output 1**.
- 4.7.8 The results of this study show that GABA Oolong tea significantly improved manual dexterity in children with ASC. Results also demonstrated that sensory responsivity improved in two-thirds of the participants and autism symptomology decreased in over half, with four of these individuals being re-classified on the DSM-5 scale, using % improvement in proportion to placebo. Cortisol Awakening levels also decreased in over two thirds of participants. GABA Oolong tea did not appear to impact actual sleep time
- 4.7.9 The results of this study also indicated that children with ASC also benefited from Theanine tea, although these benefits appeared more sporadic with only 2 showing improvements in both sensory and motor control. (L-Theanine is an amino acid analogue of L-glutamate and L-glutamine, readily crosses the BBB, and is thought to increase by proxy the GABA levels within the brain (Nathan et al., 2006; Lardner, et al., 2014) by exerting an indirect action on GABA_A receptors (Egashira, et al., 2007).
- 4.7.10 This study concluded that the sensorimotor abilities and DSM-5 symptomology of children with ASC can benefit from the administration of GABA in the form of Oolong tea. Therefore, with the sensorimotor phase of development being a crucial step in successful social and cognitive exploration, further investigation into this neurotransmitter appears to be central to our understanding of ASC.

OUTPUT 7 (Current Draft)

'A Double-Blind, Placebo-Controlled, Crossover-Designed GABA Tea Study in Children Diagnosed with Autistic Spectrum Conditions'

Hannant, P., Joyce, A., Cassidy, S & Renshaw, D. (2017). A double-blind, placebo-controlled, crossover-designed GABA tea study in children diagnosed with autistic spectrum conditions

To be submitted to Molecular Autism

Author Contribution:

Hannant devised the topic and was the lead investigator and author for Output 7. However, this particular study was separated into two very distinct disciplines, sensorimotor and sleep. For this reason Joyce also played a large role in the implementation of this study, supporting recruitment, participant assessment, actigraph data analysis and hormone assaying. Renshaw also supported the endocrinology aspect of this study. Hannant was responsible for the sensorimotor aspect of this study. Joyce and Cassidy worked jointly with Hannant on writing the both the application for funding and manuscript. All three authors will jointly revise this paper prior to publication.
Summary of Portfolio of Evidence

4.8.1 The review of the portfolio of evidence has analysed theories of ASC to date, using a set criterion to judge theories on merit (Cramer, 2013). The portfolio then went on to explore the differences in sensorimotor coordination between ASC and TD on both a psychological and biological level (Output 1), demonstrating both the presence of these difficulties in ASC and their association with autism verbal and non-verbal symptomology (Outputs 2, 3, 4 and 5). It has analysed the similarities and differences between sensory responsivity, motor coordination and autism symptomology in like conditions (ASC and DCD) (Output 5) and has identified possible underlying variations between them. Following the knowledge and information acquired on this journey, the portfolio went on to critically evaluate 'The Sensorimotor Theory' using the given criterion (Cramer, 2013), demonstrating that as a theory of ASC, 'The Sensorimotor Theory' met six out of seven points, with the seventh point, universality, being very closely met with a plethora of evidence to substantiate the point. The portfolio then concluded with an overview of evidence based current sensorimotor interventions (Output 6) and finally proposed a possible intervention to help manage ASC symptoms in children (Output 7). Moreover the review has demonstrated a clear connection between the research outputs as highlighted through the logic pathway diagram (Section 4.3). Impact is explored further in the following section.

5. Impact of the research in terms of its contribution to Autism Spectrum Disorders

- 5.1.1 This portfolio makes a significant contribution to academic research as it furthers the understanding of motor coordination and sensory responsivity difficulties in ASC. Current diagnostic criteria for ASC incorporate social communication, repetitive motor movements and atypical sensory reactivity as symptoms. However, this collection of papers begins to explore and shift current thinking; that ASC is indeed a sensorimotor disorder with its foundations rooted within the delicate balance of feedback and feedforward programming of sensory perception. The impact of this research is evident in both the quality of journals, and the amount of views / downloads and citations (Appendix 2) within the first nine months of publishing.
- 5.1.2 This research applies a range of investigations to both demonstrate the presence of and association of sensorimotor difficulties with autism symptomology in addition to identifying similarities and differences between ASC and like conditions. It also offers a unique insight into the possible development and maintenance of autism symptoms.
- 5.1.3 Significant contributions are also made to applied research in that clinicians and educators should also be aware of the sensorimotor difficulties in ASC and how these may present both physically and within the classroom (PowerPoint : Appendix1). With the understanding that ASC is sensorimotor based, interventions should focus on movement and experience. This

understanding is acutely important in supporting children with ASC as presently individuals with ASC are largely sedentary, yet it is seemingly movement, sensory input and environmental experience that helps alleviate the symptoms and should be a critical component in ASC therapy. Moreover, this research recommends additional observations during the diagnostic process in order to more accurately define phenotype.

6. Research methodology that link the outputs together

6.1 The aim of this section of the thesis is to demonstrate the logic that links the outputs together. This is evidenced conceptually, methodologically, and theoretically.

Conceptualisation

- 6.2 Outputs are linked in a logical sequence that builds a picture of knowledge growth, demonstration, evaluation, interpretation and intervention for sensorimotor difficulties in ASC. Through a study of sensorimotor patterns in ASC the portfolio of evidence presented a series of outputs derived from research managed by the author since 2013. As highlighted in section 3.2, there is a clear logic pathway that links these outputs together:
 - Firstly through **Output 1** evidence of motor coordination and sensory difficulties in ASC is analysed, with particular reference to sensorimotor difficulties. (Hannant et al., 2016)
 - Secondly, drawing on Outputs 2&3 the research then demonstrates the presence of and association of sensorimotor difficulties in ASC with severity of symptoms (Hannant et al., 2016; Hannant, et al., 2017 (Currently under review)
 - Thirdly, drawing on Output 4&5, coordination difficulties in ASC are compared to a like condition DCD (Developmental Coordination Disorder) and similarities and differences in sensorimotor presentation and their foundations are considered and identified (Cassidy, et al., 2016; Hannant et al., 2017 (Currently under review)
 - Fourthly, the research then evaluates current evidence based intervention and proposes a 'bottom up' intervention to help manage some symptomology of ASC. This is done through **Outputs 6&7** (Carroll, J et al., 2017; Hannant et al., 2017 (Current Draft).
 - Finally, and drawing upon the research of the previous stages, 'Sensorimotor Theory' is proposed within this thesis.

Methodologies

6.3 Research objectives have been met through applying robust methodologies across the studies, based on high quality synthesis of the available evidence and construction of a novel and robust theoretical framework. Reviews of academic literature and published reports formed key components of the research presented in all outputs.

- 6.3.1 Quantitative analysis has been utilised based on both parent criterion based questionnaires, participant observation, technology (Qualtrics online questionnaire and database and SPSS), hormonal assays and participant standardised assessment; all conducted by the author. Research design has been robust, for example including double blinded research, counterbalancing, subjective and objective data and gold standard diagnostic materials. In order to achieve this additional training has been completed in Gold Standard Autism Diagnostic instruments to ensure that the research is reliable and accurate. A number of field experts have also been collaborated with, such as Cassidy, Tavassoli, Van de Weyer, Mooncey and Baron-Cohen to ensure up to date and current thinking is utilised, and appropriate training and support received.
- 6.3.2 In addition to psychological research, biological knowledge has also been applied in order to bridge two distinct domains, thereby increasing the novelty and importance. Hannant's background in both Biochemical sciences and Psychology has enabled the research to be explored from a different angle enveloping the pervasive nature of sensorimotor difficulties within ASC. This includes a deeper understanding of neurochemistry and endocrinology.
- 6.3.3 Logically the research concludes with intervention recommendations, thus secondary research, in the form of reviews of published reports and academic literature, is triangulated by quantitative data analysis (both primary and secondary) and individual case analysis in order to inform and underpin intervention formulation.

7. A critical reflection of development as Research Practitioner using Gibb's reflective cycle

Introduction

7.1 The purpose of this section is to reflect critically on the journey of development throughout this portfolio using a chosen model, in this instance 'Gibb's Reflective Cycle (1988). This cycle considers a description of the journey, an evaluation about the experience, an analysis, conclusion and an action plan for future development.

7.2 Description

The journey to build this portfolio has involved developing a variety of skills. These included a number of primary and secondary literature searches based on furthering knowledge, and also turning around a rapid literature review for the Department of Education. In addition to this it was important to train to both clinical and research reliable standard for the delivery of Autism Diagnostic assessments such as the Autism Diagnostic Observation Schedule (Lord et al., 2012) and the Autism Diagnostic Interview – Revised (Le Couteur, 2003). Quality assurance through moderation exercises of these assessments furthered these skills. An in depth understanding, training and employment of ethical procedures has also been paramount throughout this process. Successful recruitment of participants has been crucial and this has been achieved in a number of ways, ranging from visiting and contacting support groups, writing to schools, accessing large

databases, such as the Cambridge Autism Research Database and through local advertisement. Networking and widening access to the field of disciplines within the sensorimotor area of research has also been essential, not only being able to access a wider scope of knowledge in various specialisms but also in being able to discuss methodology and analyse findings with experienced colleagues. Training in writing and reviewing papers has also been helpful and to this end the understanding of the difference between 1*, 2*, 3* and 4* REFable papers has helped structure the pathway of this portfolio. Successful writing has been demonstrated in the publication of outputs in addition to the application for funding for the final study. Finally, the presentation of findings in both lectures, training and poster presentations concludes each small section of the portfolio, where the awareness of differing levels of knowledge in a room, presenting information in an interesting and accessible style and being able to cascade knowledge, whilst also continually learning, has been an important part of the journey.

7.3 An Evaluation

During the development of this portfolio there have been a number of successes and a number of difficulties. The successes have included: the broadening and expansion of knowledge within the area of sensorimotor difficulties, both in ASC and other similar conditions; the building of quantitative analysis abilities particularly statistical data analysis; the pleasure of meeting both parents and children who are impacted by neurodiversity and also colleagues from various disciplines who support these people in many different ways, ranging from occupational therapists to sleep specialists; developing hypotheses that can be refuted; and perhaps in a small way, helping children with ASC in the future. Difficulties have mainly involved recruitment and differences in subjective analysis, although the process from final draft to publication has also been challenging at times.

7.4 Analysis

The three main difficulties experienced during the development of this portfolio have been recruitment, subjective analysis and publication. When analysing recruitment it would appear that finding children with ASC has not been the obstacle, it is more finding the parents who can commit to the studies. Each of the studies within this portfolio have been very in depth and robust. For example, the first study included both an ADOS-II (Lord et al., 2012) and ADI-R (Le Couteur, 2003) to ensure gold standard diagnostic procedures were in place; these assessments can take up to 4 hours to complete. The cross-syndrome study included a very robust sensory profile checklist of 302 questions and the final tea study required four university visits and precise tea making. All time consuming processes. Also, most of the studies within the portfolio have relied heavily on detailed subjective analysis has also been included, personal opinions can vary, such as in the GABA tea study, where qualitative views of the tea did not correspond to the questionnaires. When considering the publication aspect, the challenges that arose here was patience.

7.5 Conclusion

When considering the three main difficulties throughout the process, the recruitment aspect could have been supported by perhaps shortening the commitment required, whilst keeping the robustness of the studies intact. For example the sensory profile checklist used in the cross syndrome study could have been adapted and shortened. However, where it is important to complete time consuming activities, perhaps greater funding would support local visits to families, or increase incentives to take part, such as reimbursements for time. With respect to subjective analysis, despite the inclusion of objective analysis and ensuring test-retest reliability within the assessments, a more qualitative design may have been helpful, for example developing a thematic analysis based on structured conversations with parents following each tea trialled. Finally, draft to publication is a process that develops with experience, such as being able to name possible reviewers as networks widen and identifying areas of possible critique before submission.

7.6 Action Plan

For future studies, in addition to building a bank of active participants, the commitment required by participants needs to be taken into account in order to help increase recruitment. Parents and children are more likely to participate in activities that do not consume large amounts of time and that do not require large amounts of effort. With subjective analysis, these could in the future be supported by more qualitative discourse where the examiner might identify differences not considered by the participants when filling out questionnaires. Finally, when submitting papers identifying suitable journals, recognising possible areas of critique and knowing more specialised reviewers through networking will develop over time. More specifically, networking is an area that would be of enormous benefit and should be broadened over time, as both the discourse and knowledge of those who have been in the field for longer would be of great value.

8. Contribution of other people to the outputs and research

The research draws upon data gathered over three years from a variety of sources including local autism groups, typically developing children and data from the ARC (Autism Research Centre) in Cambridge.

- For Output 1, Hannant devised the topic and was the lead author. However, Hannant, Cassidy and Tavassoli worked jointly on conducting a literature search, compiling evidence from across different disciplines, and writing the manuscript. All three authors jointly revised the paper prior to publication.
- Hannant worked jointly with Cassidy and Tavassoli on Output 2 producing the paper that explored correlations between sensorimotor skills and autism symptom severity. In this publication Hannant was the lead author responsible for drawing out key issues, such as the somatosensory relationship between sensorimotor skills and autism severity. Cassidy supported the assessments and jointly wrote the manuscript. Tavassoli reviewed the manuscript and advised on assessments and procedures. Mann supported in the participant assessment batteries.
- Hannant was the sole author and lead investigator for Output 3. Hannant's knowledge of language development in typically developing children led to a literature search on semantic language development in ASC, generating a study that found correlations between sensorimotor skills and language in ASC. Hannant led the study and write up with the support of helpful discussions and written feedback from Cassidy and Carroll.
- Whilst Cassidy was the lead author for **Output 4**, and responsible for the main literature review and drafting the paper; the concept, and the evidence upon which it is based, draws extensively upon the previous Outputs and the experience and teaching of children with ASC by Hannant, and crucially their insight into the coordination and sensory difficulties that individuals with ASC encounter on a daily basis. Additionally, Hannant also assisted in the literature review and writing of the manuscript. The paper was jointly revised by all six authors prior to submission for publication.
- For Output 5, Hannant and Cassidy devised the topic and Hannant was the lead investigator and main author. Hannant created the online questionnaire, analysed the data and completed the literature search. Van de Weyer and Mooncey supported recruitment and Hannant, Cassidy, Van de Weyer and Mooncey worked jointly on compiling evidence from across different disciplines, and writing the manuscript. All four authors jointly revised this paper prior to publication.

- For Output 6, Hannant was the lead author on the physical and sensory section of the publication. Specifically: Page 14, Teacher Awareness; Pages 34-37, Handwriting and Fine motor control; and Pages 61-67, Physical and Sensory Support. Carroll worked jointly on conducting a literature search and revising the manuscript.
- Hannant devised the topic and was the lead investigator and author for Output 7. However, this particular study was separated into two very distinct disciplines, sensorimotor and sleep. For this reason Joyce also played a large role in the implementation of this study, supporting recruitment, participant assessment, actigraph data analysis and hormone assaying. Renshaw also supported the endocrinology aspect of this study. Hannant was responsible for the sensorimotor aspect of this study. Joyce and Cassidy worked jointly with Hannant on writing the manuscript. All three authors will jointly revise this paper prior to publication.

9. Conclusions and suggestions for further research

- 9.1 In the introduction to the thesis six objectives were presented. These have to a large extent been achieved. The portfolio of evidence analysed motor coordination and sensory difficulties in autism spectrum conditions (ASC), from both a psychological and biological perspective, with particular reference to sensorimotor differences, and went on to highlight possible areas of significant interest involved in these difficulties. At the same time the research has demonstrated the presence of these sensorimotor differences and similarities between ASC and a similar movement condition accentuated the importance of good motor coordination skills in social awareness, whilst also noting distinct presentations between the conditions. The research was then culminated together in order to test the merits of 'The Sensorimotor theory'. Finally, interventions were considered and trialled.
- 9.2 In bringing together a substantial body of research it is felt that PhD equivalence is achieved. Firstly, the portfolio provides evidence of training in a number of research methods appropriate to the area of study. These include: literature reviews, training to use assessments for research purposes, moderating assessments, recruitment, networking with wider disciplines, quantitative data analysis, online questionnaire creation and cascading results. Secondly, the research provides significant evidence of knowledge in the field of ASC, with a specific understanding in the area sensorimotor skills. Thirdly, it presents evidence of originality, evidenced through publication in peer-reviewed journals and citations (Appendix 2). Fourthly, the research demonstrates the ability for critical independent judgement in suggesting adapting current intervention strategies and trialling another. Fifthly, the portfolio of evidence presents a clear and logical pathway linking the research together (See section 4.3). Also, the final study secured both internal and external funding, thus indicating the impact of the research presented in this thesis.
- 9.3 Critical analysis has been demonstrated within this thesis by:
 - i Using Cramer's (2013) criteria to judge the merit of different theories of ASC, including the proposed theory within this thesis
 - ii In literature searches and rapid evidence assessment in order to rate and select studies that demonstrated high experimental rigour and high ecological validity
 - iii In noting limitations to included studies
 - iv In comparing like conditions in order to falsify theory
 - v In noting advantages and disadvantages in current intervention and proposed intervention
- 9.4 Future research within 'The Sensorimotor Theory of Autism' should be in three areas:
 - 1) Widening the GABA tea trial to include more children, with longitudinal benefits
 - 2) Looking in greater depth at the impact of the auditory spatial canal on vision, or contrariwise

3) A large study, which would hopefully include educational colleagues, in improving school readiness as children enter school by assessing not only language skills, but also sensory responsivity, and fine and gross motor coordination, thereby picking up difficulties in these areas before they impact on emotional well-being and learning.

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Appendix 1:



EarlyBird Plus Programme

Licensing Agreement

THIS AGREEMENT is dated 13TH day of MARCH 201.4

BETWEEN

 The National Autistic Society an incorporated charity registered in England (company registered number 1205298) registered charity number 269425 of 393 City Road London EC1V 1NG ("the Charity")

PENNY HANNANT OF LEICESTER City (2)

("the Licensed Trainer")

WHEREAS

The Charity is the beneficial owner of the intellectual property rights in materials relating to an autism specific management programme known as the NAS EarlyBird Plus Programme ("the Materials")

The Charity is the beneficial owner of the registered trademark "NAS EarlyBird Plus" ("the Mark") and wishes to maintain the integrity of the Mark

For the same reason the Charity wishes to ensure that only trainers accredited by the Charity offer training in the Materials

The Charity has agreed to permit the Licensed Trainer to use the Materials and the Mark on the terms set out below

IT IS AGREED as follows:

- 1. The Charity hereby confirms the grant to the Licensed Trainer with immediate effect from the date of this Agreement for a period of 3 years of a non-exclusive licence to use the NAS EarlyBird Plus Programme Materials and the NAS EarlyBird Plus Mark (subject to the obligations imposed by way of the licence terms detailed below) for the purpose of training persons in the use of the NAS EarlyBird Plus Programme and for no other purpose; for the avoidance of doubt the restriction on using the Materials or the Mark or both includes their use jointly or severally in connection with any other programme of training or management
- 2. The Materials (in whole or part) are not to be translated by the Licensed Trainer nor is the Licensed Trainer to enable assist or procure any other person or body to translate the Materials or any part thereof without the express prior written approval of the Charity
- Customer Satisfaction Data from the use of the Materials are to be sent to the Charity as follows:
 - a) Attendance sheet
 - b) Copies of evaluation sheets from 8 Group Sessions and 2 Home Visits
 - c) Copies of Post-Programme and Follow-up Questionnaires
- 4. The full framework of the Materials is to be maintained, including "Group Sessions 1 to 8" and "Home Visits A & B" as indicated in the Materials

1

- 5. No photocopying of the Materials is permitted other than the "Appendix" shown as "Training Manual" in the Materials and then only as indicated therein
- 6. The Licensed Trainer will not do anything which conflicts with the objects or policies of the Charity or which might prejudice its reputation or goodwill
- 7. The Licensed Trainer will notify the Charity promptly on becoming aware of any unauthorised use by any third party of the Materials or the Mark or of materials or a logo similar to them and will at the expense of the Charity take such action as the Charity may reasonably require to restrain such use
- 8. The Licensed Trainer will arrange and maintain the following insurance cover or ensure that it is arranged and maintained on the Licensed Trainer's behalf by the Licensed Trainer's employer providing proof thereof to the Charity upon request:

Employer's Liability	:	£10 million
Public Liability	2	£ 5 million
Professional Indemnity	:	£ 1 million

- 9. Subject to clause 10 this Agreement shall continue until terminated by effluxion of time
- This Agreement shall be terminable at any time by the Charity if the Licensed Trainer fails to remedy any material breach on its part of this Agreement within thirty days of written notice by the Charity
- On and after termination of this Agreement by whatever means and whatever time the Licensed Trainer will cease to use the Materials and the Mark in any way or manner whatsoever
- 12. Any notice or other document to be given under this Agreement shall be in writing and shall be deemed duly given if sent by first class recorded delivery post to the last known address of the party to be served any such notice being deemed to be served on the second business day following the day of posting
- 13. This Agreement shall be governed and construed according to the laws of England

Signed by the Licensed Trainer:

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Whose signature is witnessed by:

(Address)

NAS EarlyBird Centre Barnsley Road Dodworth Barnsley S75 3JT

EarlyBird Centre Trainer

Signed on behalf of the Charity by a deputed signatory:

laa

Talk Boost Licensed Tutor

This is to certify that

Penny Hannant

is a Licensed Talk Boost Tutor certified to deliver Talk Boost Training.

Effective date: 18th November 2013

Licensee Registration Number: 5935697

This certificate is valid for 3 years from the date shown above (subject to payment of annual support fee).

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Virginia Beardshaw Chief Executive, I CAN



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Clinical Reliability

from 23rd to 27th September 2013

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20th May 2016

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Sensorimotor Difficulties Are Associated With the Severity of Autism Spectrum Conditions.

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(2) Autism Research Centre, University of Cambridge, Cambridge, UK.
(3) Seaver Autism Centre, Icahn School of Medicine, Mount Sinai, US.

INTRODUCTION

- Recent research has found significant sensory processing and motor coordination difficulties in Autism Spectrum Conditions (ASC)¹⁻⁵.
- However, few studies have examined the relationship between both motor coordination and sensory processing on severity of ASC.
- This study explores whether: 1) Children with ASC have significant sensory and motor difficulties compared to matched controls; and 2) Sensory and motor difficulties can predict the severity of ASC symptoms.

METHODS

Participants

- 36 children, 18 (13 male, 5 female) with ASC and 18 (7 male, 11 female) from a sample population, matched in age and nonverbal reasonina.
- ASC children were recruited from a local support group and diagnosis was confirmed and severity measured using both the ADOS-II and the ADI-R.

Measures

- Movement Assessment Battery for Children (Movement-ABC 2) An observation and outcome based assessment that comprises: manual dexterity, ball skills and static and dynamic balance.
- BEERY VMI 6th Ed. A measure of an individual's ability to combine visual perception and fine motor coordination.
- The Sensory Profile A parent report questionnaire that assesses the frequency of a child's responses to differing sensory processing, modulation and emotional events.
- Modulation of Movement a section of the Sensory Profile that measures the level of activity based on sensory outcomes.
- The British Picture Vocabulary Scale III a non-reading assessment of receptive language, dependent on identifying the correct image to match a given word.
- WASI a measure of expressive language and performance intelligence.
- The Social Communication Questionnaire (SCQ) A broad parent report measure of social communication difficulties in ASC

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RESULTS

Table 1. Means were compared between groups (ASC and controls), across all variables using independent Bonferroni corrected t-tests. Significant differences were found with medium to large effect; with ASC children showing significant difficulties across domains.

Measure	ASC Mean	Control Mean	Significance	Effect Size
Movement-ABC Total	51.6±15.7	74.3±14.1	p<0.001	d=1.52
Sensory Profile Total	228.2±37.4	312.1±27.2	p<0.001	d=2.56
BPVS-III	88.6±14.1	106.0±12.0	p<0.001	d=1.33
Modulation of Movement	18.6±3.2	23.1±3.6	p<0.001	d=1.33
BEERY VMI	84.2±21.3	100.2±8.9	p<0.01	d=0.97
SCQ	18.9±7.9	3.8±3.68	p<0.001	d=2.44

The manual dexterity composite and balance composite of the Movement ABC were significantly lower in ASC compared to the control group with large effect (Manual Dexterity: $t_{(28.96)}$ =5.562, p=<.001, d=1.85; Balance $t_{(34)}$ =2.531, p=.016, d=.84)

In the Sensory Profile, significantly lower results, when compared to the control group, are shown in descending order of size: poor registration (*t*_(25.92)=7.273, p<.001, d=2.42); emotional regulation ($t_{(34)}$ =6.788, p<.001, d=2.26); low endurance ($t_{(24,42)}$ =5.858, p<. 0001, d=1.95); attention composites (t_(25.91)=4.738, p<.001, d=1.58); sedentary $(t_{(34)}=3.709, p<.001, d=1.24)$ and fine motor skills $(t_{(34)}=3.631, p<.001, d=0.85)$. Oral sensitivity, sensory seeking and sensory sensitivity were not shown to be significantly different between groups after Bonferroni correction.

Table 2. Stepwise multiple regression analyses were conducted with autism severity as the outcome measure (ADOS-II, ADI-R and SCQ), and Sensory Profile Total, Movement ABC, WASI Matrices and BPVS as predictors.

ASC group final models (n=18)			
Predictors	ASC Measure	Cum R ²	B, SE, eta
BP∨S Sensory Profile	ADI-R Total	.85	BPVS B=.39, SE B=.10, β =.58** Sensory Profile B=.12, SE B=.04, β =.46**
Movement ABC Matrices	ADOS-II Total	.77	MABC B=.29, SE B=.06, β =.87*** Matrices B=.18, SE B=.08, β =.47*
Sensory Profile BPVS	SCQ	.79	Sensory Profile B=.16, SE B=.03, β =.73*** BPVS B=.18, SE B=.07, β =.31*
9	ample Populatio	on and AS	C group final models (n=36)
Sensory Profile BPVS	SCQ	.83	Sensory Profile B=.14, SE B=.02, β =.77*** BPVS B=.14, SE B=.05, β =.23*

p <0.05, ** p < 0.01, *** p < 0.001.

Modulation of Movement significantly predicted autism symptom severity both in the ASC group across all diagnositic measures (ADOS, ADI-R and SCQ) and when the TD group's SCQ data were added(all p < 0.05).

Table 3. A Correlation Analysis (r) for specific autism, coordination and sensory components in ASC group (18).							
Measures	ADIR- C	ADIR-RSI	ADRI-RSB	ADOS-C	ADOS-RSI	ADOS-RRB	SCQ
MABC Dexterity	0.152	0.331	0.006	0.595**	0.535*	0.200	0.256
MABC Balance	0.322	0.448*	0.322	0.531*	0.523*	0.344	0.238
SP Auditory	0.652**	0.694**	0.521*	0.307	0.178	0.251	0.861***
SP Vestibular	0.201	0.179	0.590**	0.173	0.053	0.084	0.381
SP Touch	0.334	0.522*	0.626**	0.506*	0.365	0.069	0.615**
SP Oral	0.576**	0.310	0.693**	0.217	0.197	0.464*	0.574**

CONCLUSION

- Children with ASC showed significant difficulties in sensory and motor skills compared to controls, across a number of measures.
- After controlling for group differences in verbal and non-verbal ability, sensory processing (Sensory Profile) significantly predicted the level of autism symptoms measured by the ADI-R and SCQ, and motor coordination skills (Movement ABC) significantly predicted the level of autism symptoms measured by the ADOS-II.
- Specific sensorimotor correlations shown are all instrumental to somatosensory perception. These results show that sensorimotor difficulties significantly account for autism symptom
- severity across a number of gold standard diagnostic measures. Future research should explore whether improving sensorimotor skills in children with ASC could improve their social and communication skills.

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Dyspraxia and autistic traits in adults with and without Autism Spectrum Conditions.

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 (5) CLASS Clinic, Cambridgeshire and Peterborough Foundation NHS trust, Cambridge, UK.

INTRODUCTION

- The ability to effectively coordinate, plan and carry out movements effectively is key for successful social functioning.
- Autism Spectrum Conditions (ASC) are frequently associated with motor and coordination difficulties.¹²
- Recent studies have shown that motor difficulties impact social and communication skills in ASC.3 4
- Dyspraxia is characterized by pronounced difficulties in motor coordination.5
- To date, there have been no studies that have: 1) explored the prevalence of dyspraxia in a large sample of individuals with and without ASC; or 2) explored the relationship between dyspraxia and autistic traits in the general population.

METHODS Participants

- 2,871 adults with ASC (70.3% male) and 10,706 controls without ASC (42.3% male) were recruited through the Cambridge Autism Research Database (CARD).
- Participants self-reported whether they had been diagnosed with dyspraxia.
- A majority of participants completed self-report measures of autistic traits (1,237 ASC and 6,765 controls) and empathy (1,148 ASC and 6,676 controls) online through the Autism Research Centre and Cambridge Psychology websites.

Measures

Empathy Quotient (EQ) A self-report questionnaire that quantifies individual differences in empathy. 80% of patients with ASC score less than 30 on the EQ, compared to 12% of the general population.

Autism Spectrum Quotient (AQ) A self-report questionnaire to quantify autistic traits in adults with at least average IQ. 80% of patients with ASC score equal to or greater than 32 on the AQ, compared to only 2% of the general population.

RESULTS

Table 1: Self-Reported Dyspraxia in adults with ASC vs. adult controls without ASC.

	Dyspraxia	No Dyspraxia
ASC (n=2,871)	199 (6.9%)	2,672 (93.1%)
Control (n=10,706)	91 (0.8%)	10,615 (99.2%)
Total (n=13,577)	290 (2.1%)	13287 (97.9%)

 Individuals with ASC were significantly more likely to self-report dyspraxia than controls without ASC (OR 9, 95% Cl 6.7 - 11.2, p<0.001)

Table 2: Mean age, AQ and EQ by diagnostic group

	Dyspraxia?	Age (SD)	AQ (SD)	EQ (SD)
150	Yes	30 (12.4)	37.6 (7.5)	20 (12)
ASC	No	35.7 (13.2)	38.5 (6.8)	17.6 (9.9)
Control	Yes	25.3 (10.1)	23.4 (8.2)	39.3 (14.5)
Control	No	31.4 (13.5)	17.7 (7.5)	45 (14.2)
Total	Yes	27.9 (11.9)	33.4 (10)	25.2 (15.3)
Total	No	32.1 (13.5)	20.7 (10)	41.2 (16.6)

- There was a significant interaction between ASC diagnosis and dyspraxia for AQ (F(1,7998)=22.4, p<0.001) and EQ (F(1,7820)=8.4, p=0.004) scores.
- ASC group: there was no significant effect of co-morbid dyspraxia on AQ (p=.22) or EQ (p=.1) scores.
- Control group: those with dyspraxia had significantly higher AQ (p<.01) and EQ scores (p<.05) than those without dyspraxia.

CONCLUSION

- Dyspraxia is significantly more prevalent in adults with ASC compared to individuals without ASC, confirming reports that coordination difficulties are significantly more common in this clinical group.
- In the general population, dyspraxia was associated with significantly higher self-reported autistic traits, and with significantly lower empathy.
- These results suggest that the addition of motor coordination difficulties in ASC does not further impact their high levels of autistic traits. However dyspraxia is associated with more autistic traits in individuals without ASC.

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Session Outline

- Myths and Facts
- Triad of Impairment
- Sensory
- Supporting Behaviour

History

Behaviours associated with Autism have been noted throughout history

- Leo Kanner 1940s (Classic Autism, USA)
- Hans Asperger 1940s (Asperger Syndrome: Austria)
- Lorna Wing 1970s (Autism Spectrum UK)

However, recognition of the disorder is relatively new and we still have much to learn





Myth & Facts

- Myths and facts activity
- In small groups discuss each statement, decide on whether the statement is true or false
- Feedback

Myths

- Simply another term for learning difficulties
- Once speech begins, other difficulties disappear
- Always occurs with an area of genius ability
- Results from bad behaviour and naughtiness
- Due to bad parenting

Г

• A middle class disorder

Facts

- More likely in boys than girls
- A developmental disorder involving the brain
- People with ASD experience the world differently
- May be accompanied by differing degrees of learning difficulties
- A lifelong disability
- Genetically linked
- Diagnosed by identifying differences in 'triad'

Au	tism	Spectro	um
The Autistic Spe	ctrum?		
Asperger's	HFA	Kanner Autism	PDD-NOS
Autism H	FA PDD	NOS Traits	No Autism

The Tip of the Iceberg



















Triad of Impairments

Activity

Think of a pupil you know with ASD

Highlight on the Triad sheet where they present

Feedback

Individual Sensory Profile

- Please listen carefully to each statement
- Put your hand up if you agree
- Keep an eye on the responses of others





Additional Factors

- Anxiety inwards and outwards
- Compulsions
- Everyone with Autism is an individual first. They are influenced by their own experiences, environment and personality. No two people with Autism are the same

Understanding Autisms

- Visual thinkers and processors
- Single channel (only one concept at a time)
- Concrete and literal
- Weak organisational skills including concepts of time
- Often associated with coordination difficulties
- Poor self-awareness
- Weak inference skills
- Like routine and structure

Golden Rules for Supporting Behaviour

- Understanding the autism
- Analyse the behaviour
- Use structure
- Change yourself rather than the child
- Reduce anxiety
- Be consistent
- Plan transitions

Understanding Behaviour

- Iceberg detective
- STAR
- Sensory Profile
- Low arousal





Summary

- Autism is a developmental disorder involving the brain
- It is a social disorder effecting your ability to predict and interpret social content
- Deficits in:
 - Communication
 - Theory of Mind (Simon Baron-Cohen)
 - Executive function
 - Central coherence (Uta Frith)
 - Sensory Integration (Olga Bogdashina)







1

PSYCHOLOGY, BEHAVIO

Coventry





2





<u>Cellence</u> <u>With Impact</u>

Coventry W











4













Appendix 2:

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