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A Comparison of Motor Deficits in Autism Spectrum  
Disorder and Developmental Coordination Disorder

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Doctor of Philosophy  
The University of Edinburgh  
2014

## Declaration

I hereby declare that this thesis is of my own composition, and that it contains no material previously submitted for the award of any other degree. The work reported in this thesis has been executed by myself, except where due acknowledgement is made in the text.

Louisa Miller

## Abstract

Autism Spectrum Disorder (ASD) is an umbrella term for disorders involving deficits in social interaction, stereotyped behaviours and communication difficulties. A growing area of research has recently focused on motor deficits in ASD, which have been noted in clinical observations and diagnostic criteria since autism was first described. However, motor deficits have traditionally carried little weight in the diagnostic procedure. Until recent changes to diagnostic criteria (Diagnostic and Statistical Manual 5th edition: DSM-5), a comorbid diagnosis of Developmental Coordination Disorder (DCD: a neurodevelopmental disorder affecting motor development) was not possible for those with ASD and motor deficits. This exclusion criterion prompted an investigation of the nature of motor deficits in ASD, questioning whether they are characteristically different from motor deficits in DCD. Previous literature suggested a possible double dissociation in the use of vision and proprioception to guide movement and perception in ASD and DCD, with a reliance on proprioception in ASD, and an over-reliance on vision in DCD. Motor deficits were first investigated by looking at high-level motor skills, and then more basic sensory processing associated with movement to investigate this possible dissociation. There was no significant difference between ASD and DCD on a standardised motor battery (Movement Assessment Battery for Children 2nd edition: MABC-2), with 70% of children with ASD showing motor difficulties within the clinical range on tasks such as timed manual dexterity tasks and balance. Similarly, children with ASD and poor motor skills were indistinguishable from children with DCD on a number of basic motor tasks manipulating visual and proprioceptive cues. These tests included spatial location matching, reaching, goal-directed movements towards proprioceptively-defined targets, and the rubber hand illusion. Children with poor motor skills with a diagnosis of either ASD or DCD seemed to either rely more heavily on visual cues, or behaved in a similar way to typically developing (TD) children. In the spatial location matching task, children with ASD and spared motor skills showed a tendency to give more weight to proprioceptive cues, however too few children with ASD and spared motor skills took part in other tasks to fully investigate cue weighting in this subgroup. Mirroring the overlap in social and motor skills in the clinical groups, a study of the relationship between perceived social and motor ability in a large sample of TD children highlighted the related nature of these developmental domains in typical development. It is concluded that motor deficits in ASD are not ASD-specific but are instead indicative of an additional diagnosis of DCD. This is supported by the recent change to diagnostic criteria.

## **Acknowledgements**

Thank you to Rob for all his help along the way, and for the motivational mp3 rewards during write-up. Thanks also to Bonnie for her feedback on draft chapters. Thank you to Anne O'Hare, Sue Davidson and the OT department for all of their help with NHS recruitment, and thank you to all of the schools, the Skool's Out after school club, hospitals, clinics, families and children who helped with and took part in the studies. Thanks to Alasdair, Mum and Dad for listening to more than their fair share of ethics- and recruitment-related moans along the way, and thanks Dad for building all of the apparatus for me! Finally, thank you to the Kerr-Fry Bequest for funding my PhD and the Patrick Wild Centre for funding one of the studies.

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# List of Abbreviations

AC- Aiming and Catching  
AD- Autistic disorder  
ADHD- Attention Deficit/Hyperactivity Disorder  
APA- American Psychiatric Association  
AQ- Autistic Quotient  
AS- Asperger Syndrome  
ASD- Autism Spectrum Disorder  
BOTMP- Bruininks-Oseretsky Test of Motor Proficiency  
cKAT- Computerised Kinematic Assessment Tool  
CP- Cerebral Palsy  
DCD- Developmental Coordination Disorder  
DCDQ- Developmental Coordination Disorder Questionnaire  
DCDQ'07- Developmental Coordination Disorder Questionnaire 2007  
DN- Direct proprioception, no vision (Chapter 4: posting condition)  
DV- Direct proprioception, vision (Chapter 4: posting condition)  
DSM-IV- Diagnostic and Statistical Manual-4th edition  
DSM-IV-TR- Diagnostic and Statistical Manual-4th edition-text revision  
DSM-5- Diagnostic and Statistical Manual-5th edition  
DT- Deceleration Time  
GG- Greenhouse-Geisser  
GMDS- Griffiths Mental Development Scales  
HFA- High functioning autism  
ICD-10- International Classification of Diseases-10th edition  
IN- Indirect proprioception, no vision (Chapter 4: posting condition)  
IV- Indirect proprioception, vision (Chapter 4: posting condition)  
LD- Learning Disability  
MABC- Movement Assessment Battery For Children  
MABC-2- Movement Assessment Battery for Children 2nd edition  
MD- Manual Dexterity  
MGA- Maximum Grip Aperture  
MT- Movement Time  
OT- Occupational Therapy/ Occupational Therapist

PA- Path Accuracy  
PANESS- Physical and Neurological Examination for Soft Signs  
PDD- Pervasive Developmental Disorder  
PDD-NOS- Pervasive Developmental Disorder-Not Otherwise Specified  
PL- Path Length  
POS- Peak Orientation Speed  
PS- Peak Speed  
PP- Proprioception (Chapter 3: spatial location matching condition)  
RHI- Rubber Hand Illusion  
RT- Reaction Time  
RTM- Repetitive Timed Movements  
SRS- Social Responsiveness Scale  
TD- Typically Developing/ Typical Development  
TO- Terminal Orientation  
TOMI-HR- Test of Motor Impairment-Henderson Revision  
TPOS- Time to Peak Orientation Speed  
VP-Vision (Chapter 3: spatial location matching condition)  
VPP- Vision+Proprioception (Chapter 3: spatial location matching condition)  
WASI- Wechsler Abbreviated Scale of Intelligence

# Chapter 1

## Introduction

This thesis investigates motor deficits in two neurodevelopmental disorders: Developmental Coordination Disorder (DCD, also commonly called dyspraxia in the UK) and Autism Spectrum Disorder (ASD). In this introductory chapter a brief description of these disorders is given, and the diagnostic issues surrounding these conditions are discussed. This is followed by a more detailed account of ASD and DCD, focusing specifically on motor abilities in the two conditions. Finally, the first question to be addressed in this thesis will be outlined.

### 1.1 Developmental Coordination Disorder

Developmental Coordination Disorder is a term used to describe fine and/or gross motor coordination and planning difficulties in children. (Note that while the term DCD is typically used to describe children, difficulties will often persist into adulthood (Kirby, Sugden, Beveridge & Edwards, 2008).) There is no single presentation of DCD, with the range and severity of symptoms being highly heterogeneous (Sugden & Wright, 1998).

The term ‘DCD’ has been both differentiated from and used interchangeably with the commonly used term ‘dyspraxia’ (Gibbs, Appleton & Appleton, 2007), which itself has a number of synonyms, including ‘childhood apraxia’ and the now obsolete ‘clumsy child syndrome’ (Colley, 2006). The current consensus is that it is unnecessary to differentiate DCD and dyspraxia, as they appear to describe the same set of diagnostic characteristics (Gibbs et al., 2007; Magalhaes, Missiuna & Wong, 2006; cf. Miyahara & Mobs, 1995, who argue that dyspraxia is a specific deficit in motor sequencing and selection which is not always apparent in DCD). Therefore in this thesis, DCD will refer to a diagnosis of DCD, dyspraxia or any of its synonyms such as childhood apraxia (excluding verbal childhood apraxia and verbal dyspraxia).

The prevalence of DCD within a mainstream school population has been estimated to be 4-5% using a standardised test of motor impairment [Movement Assessment Battery for Children: MABC, Henderson & Sugden (1992)], supporting the 5%ile di-

agnostic cut-off for this test (Wright & Sugden, 1996). Highlighting the effect that diagnostic procedure has on reported prevalence rates, Lingam, Hunt, Golding, Jongmans & Emond (2009) report a prevalence of only 1.8% using an abbreviated MABC (using the 5%ile as a diagnostic cut-off), with an additional 3.2% having ‘probable DCD’. It should be noted however that the former refers to children in Singapore, while the later refers to children in the UK. Culture may have an effect on prevalence, as prevalence has been found to be significantly higher amongst children in Greece (19%) compared to Canada (8%), using the same test battery in both countries (Tsiotra, Flouris, Koutedakis, Faught, Nevill, Lane & Skenteris, 2006).

DCD is often found to be comorbid with other neurodevelopmental disorders, including dyslexia, dysgraphia, Attention Deficit/Hyperactivity Disorder (ADHD) and ASD (Polatajko (1999) p123; Rasmussen & Gillberg (1999) p142). Indeed, ‘pure’ cases of DCD, in which the only symptoms are in the motor domain, are thought to be relatively rare (Peters & Henderson, 2008). Of primary interest in this thesis is the suggestion that the prevalence rate within the autistic population is relatively high (Gillberg & Kadesjo, 2003). Demonstrating the coexistence of DCD and ASD, Kadesjo & Gillberg (1998) found that children in their DCD sample had on average 3 of the 19 symptoms of Asperger’s Syndrome (AS: a disorder on the Autism spectrum), while a group of children without DCD had almost no symptoms that feature in AS (on average 0.1 of 19 symptoms). It should be noted however that Gillberg and Kadesjo’s (2003) statement that the instance of comorbid ASD and DCD is relatively high highlights the first of many problems in diagnosing these conditions: DSM-IV (Diagnostic and Statistical Manual of Mental Disorders: American Psychiatric Association (APA), 2000) diagnostic criteria state that a comorbid diagnosis of ASD and DCD should not be given. In practice however this does not seem to be universally adhered to. This problem of comorbidity in the diagnostic procedure is discussed in Section 1.3.2.

### **1.1.1 Identifying and diagnosing DCD**

#### **1.1.1.1 Origins of the DCD diagnosis**

DCD has previously had a number of names, with each neither mutually exclusive, nor describing the same set of symptoms [Polatajko (1999) in Whitmore, Hart & Willems (1999), p121]. A record of the most commonly used terms from 1937 to 1995 was compiled by Polatajko (ibid, p120) and this table is reproduced in Table 1.1.

The International Consensus Meeting on Children and Clumsiness in 1994 (Polatajko, Fox & Missiuna, 1995) concluded that the previously used term ‘clumsy’ was deprecatory and instead suggested the use of the term ‘Developmental Coordination Disorder’ or ‘DCD’. The adoption of a single diagnostic label was hoped to aid cross-disciplinary research, and researchers and clinicians were urged to use the term ‘DCD’ (Whitmore et al. (1999), p121). The set of symptoms now termed ‘DCD’ has evolved as a derivative of Minimal Brain Dysfunction, which fractionated into three main dis-

Table 1.1: Terms denoting DCD, compiled by Polatajko (1999)

Term	Reference
Apraxia/Agnosia/Apraxic ataxia	Orton (1937) Walton, Ellis & Court (1962) Gubbay (1975)
Clumsy child/Clumsy child syndrome/ Clumsiness	Orton (1937) Walton et al. (1962) Dare & Gordon (1970) Gubbay (1975) Keogh, Sugden, Reynard & Calkins (1979) Gordon & McKinlay (1980) Henderson & Hall (1982) Hulme & Lord (1986) Henderson (1987) Cratty (1994)
Deficits in Attention, Motor Control and Perception (DAMP)	Gillberg & Gillberg (1989)
Developmental dyspraxia/Apraxia	Gubbay (1975) David, Deuel, Ferry, Gascon, Golden, Rapin, Rosenberger & Shaywitz (1981) Ayres (1985) Cermak (1985) Dewey (1995)
Developmental output failure	Levine, Oberklaid & Meltzer (1981)
Developmental coordination disorder	American Psychiatric Association (APA) (1987) Henderson, Rose & Henderson (1992) American Psychiatric Association (APA) (1994) Hoare (1994) Missiuna (1994) Mon-Williams & Wann (1994) Rosblad & von Hofsten (1994) Polatajko, Macnab, Anstett, Malloy-Miller, Murphy & Noh (1995) Willoughby & Polatajko (1995)
Motor delay	Henderson (1986)
Motor coordination problems/difficulties	Cratty (1986) Roussounis, Gaussen & Stratton (1987)
Motor learning difficulties	McKinlay (1987)
Movement difficulties	Bouffard, Watkinson, Thompson, Causgrove Dunn & Romanow (1996)
Perceptual motor dysfunction/deficits	Gordon & McKinlay (1980) Laszlo & Bairstow (1989) Clark, Mailloux, Parham & Primeau (1991)
Physically awkward child	Wall, Reid & Paton (1990)
Sensory integrative dysfunction	Ayres (1972)
Visuo-motor disabilities	Dare & Gordon (1970)

orders: DCD, ADHD and Learning Disability (LD), with DCD often associated with comorbid ADHD and LD (Whitmore et al. (1999), p122). Kaplan, Wilson, Dewey & Crawford (2002) report a 50% comorbidity rate of LD or ADHD and DCD; Sugden & Wann (1987) report a comorbidity rate of 29-33% for LD and DCD; and Kavale & Nye (1985) report from a meta-analysis of 1077 studies that 70% of cases of LD are associated with comorbid perceptual-motor deficits. These figures highlight that the disorder is commonly associated with a number of other specific learning difficulties, all of which have a high comorbidity with ASD [Rasmussen & Gillberg (1999) in Whitmore et al. (1999), p143]. The co-occurrence of DCD and ASD symptoms is of central interest in this thesis.

#### 1.1.1.2 DSM and ICD diagnostic criteria for DCD

DCD is listed in the DSM-IV-TR<sup>1</sup> (Diagnostic and Statistical Manual of Mental Disorders, 4th edition, text revision: APA, 2000) and is given as a diagnosis if the following criteria are satisfied:

*Criterion A:* There is a marked impairment in the development of motor coordination. This is evaluated using a norm-referenced test, of which there is currently no widely accepted ‘gold-standard’ (Hill & Barnett, 2011).

*Criterion B:* Impairment significantly interferes in both academic and daily life. (See Polatajko (1999), in Whitmore et al. (1999), p129, for detailed vignettes describing common difficulties, and the impact on daily life. Examples include difficulties dressing, using cutlery, writing and learning to ride a bike.)

*Criterion C:* Coordination deficits should not be due to any general medical condition and the criteria for Pervasive Developmental Disorder (PDD) are not met. (Note that the exclusion of PDD excludes any autism diagnosis.)

*Criterion D:* If there is coexisting mental retardation then coordination difficulties should be more extensive than those expected.

The ICD-10 (World Health Organisation, 1992) lists the following criteria for a diagnosis of DCD:

Scores on a standardised test of fine/gross motor coordination (e.g. the MABC) should be at least 2 standard deviations from the average score for age-matched children. Again, deficits should significantly interfere with daily life and there should be no neurological disorder. The ICD states that a diagnosis of DCD should not be given if the individual has an IQ below 70 (this would likely exclude children at the lower end of the autism spectrum).

---

<sup>1</sup>The main question in this thesis was based on DSM-IV criteria as the DSM-5 was not yet published. The DSM-5 (Diagnostic and Statistical Manual of Mental Disorders, 5th edition: American Psychiatric Association, 2013) has changed the relationship between DCD and ASD and will be discussed at a later point.

### 1.1.1.3 Examining the diagnostic criteria

The diagnostic criteria set out in both the DSM and ICD have met with some criticism. This first criticism involves the nature of motor difficulties. Firstly, both DSM and ICD criteria stipulate that motor problems should not be attributable to any distinguishable neurological condition, such as Cerebral Palsy (CP). Symptoms present are therefore neurological soft signs [motor signs indicative of non-specific cerebral dysfunction (Dazzan & Murray, 2002)]. These subtle signs of a movement abnormality include abnormal movements and reflexes, awkwardness, clumsiness and poor coordination, dysfunction in the regulation of muscle tone and impaired voluntary movement (Cratty, 1994; Sugden & Keogh, 1990; Gustadsson, Svedin, Ericsson, Linden, Karlsson & Thernlund, 2010). The significance of some of these signs is debatable however, as one or more will often be seen in a child with no notable motor problems (Cratty, 1994; Sugden & Keogh, 1990; Hall, 1988).

Secondly, there has been discussion of movement planning and execution deficits (Kirby, Sugden & Edwards, 2010). These are not differentiated in diagnostic criteria, and this is further confused by the (still) common use of the term ‘dyspraxia’, argued by Miyahara & Mobs (1995) to be a motor planning difficulty distinct from DCD. More recently, others have argued that this is not the case and that DCD and dyspraxia should either be used interchangeably (Gibbs et al., 2007), or the term ‘dyspraxia’ abandoned all together [Polatajko (1999) in Whitmore et al. (1999), p121]. This is supported by current diagnostic criteria, as the DSM does not include dyspraxia and the ICD gives the same diagnostic criteria for both DCD and dyspraxia.

Another problem arises when considering the stipulation that motor performance must deviate significantly from the expected average performance of typically developing (TD) children. This stipulation, found in both DSM and ICD criteria, relates very differently to each of the three schools of thought regarding the nature of DCD. Hall (1988) suggests that DCD is merely an expression of biological variance, highlighting that a normal distribution of ability across a population would predict both very high ability children and very low ability children, with the majority falling somewhere in-between. This school of thought would reject the notion that those whose skills are significantly below average have a motor disorder, but are proof of a normal distribution of ability. It would therefore reject the use of diagnostic cut-off points in motor assessments derived from normative TD data.

A second school of thought, the maturational delay hypothesis, posits that in most cases children will catch-up on skills that were previously below the level expected. This would suggest that children who meet the criterion of performance below average for their age will at some point no longer fit this criterion. This suggests that the disorder is only evident in children, an idea supported by some reports of the spontaneous resolution of motor problems with increasing age (Knuckey & Gubbay, 1983; Roussounis et al., 1987). However there is ample evidence to support ongoing problems in adults

with DCD (Kirby et al., 2008; Hill & Barnett, 2011; Losse, Henderson & Elliman, 1991; Cantell, Smith & Ahonen, 1994).

The third school of thought is that DCD symptoms are atypical and will not be corrected with time, so the condition should be considered a real disorder in its own right (Henderson, 1994). Assuming that this third school of thought is most accurate, the question driving this thesis is the nature of the relationship this distinct disorder has with ASD, which has been found to share a number of motor symptoms but is not, according to diagnostic criteria, a valid comorbid condition. The exclusion criteria regarding PDD has previously been met with criticism, with the Leeds Consensus (Sugden, 2006) stating that this exclusion criterion is inappropriate, and it should be considered that ASD and DCD can be comorbid. This stance is now reflected in the new DSM criteria (APA, 2013) which lists ASD as a possible comorbid diagnosis for DCD.

#### 1.1.1.4 Diagnostic tools

There are a number of diagnostic tools that can be used to diagnose DCD based on the criteria described above, with no one test generally recommended over the other. The unofficial ‘gold standard’ (in so much that it appears to be the test most commonly used by practitioners in the UK and is often used in research) has become the MABC, although other tests, such as the Bruininks-Oseretsky Test of Motor Proficiency (BOTMP: Bruininks, 1992), can be used both in clinical practice and research and is widely used in the US (Crawford, Wilson & Dewey, 2001).

One problem with the lack of regulations regarding diagnostic tools is a lack of clarity over the extent to which the different tools are measuring the same characteristics. Croce, Horvat & McCarthy (2001) report strong correlations between the MABC and BOTMP in a group of children aged 5-12 ( $n=20$  each for ages 5-6, 7-8, 11-12 and  $n=48$  for ages 9-10), with  $r = 0.9$  in the oldest age group. This strong correlation goes against Henderson and Sugden’s (1992) suggestion that the two tests should not agree strongly as the BOTMP was designed to assess a wider range of motor abilities (dexterity, balance, strength, agility etc.) while the MABC was developed primarily for the identification of motor difficulties (see also Tan, Parker & Larkin, 2001). While this difference in initial test purposes was not reflected in the correlation between the two tests, Croce et al. (2001) did report that the BOTMP was harder to administer with children with attentional difficulties. Wiart & Darrah (2001) also suggest that the BOTMP is inappropriate for use with children with attentional or intellectual disabilities, as the instructions tend to be more complex than those in the MABC.

Despite the high correlation reported by Croce et al. (2001), it has been suggested that the two tests tend to identify different children (Larkin & Rose, 2005), and poorer correlations between the tests have been reported. For example, Henderson & Sugden (1992) report a correlation of only  $r = 0.53$  for a group of 4-12 year olds ( $n=63$ ), and

Cairney, Hay, Veldhuizen, Missiuna & Faught (2009) found clear disagreement between the MABC and BOTMP when using the 5%ile cut-off for both batteries. In a group of 24 children identified as falling at or below the 5%ile on the BOTMP, 63% were also found to lie in the lower 5th centile on the MABC, however 25% scored between 6-15%ile on the MABC (the borderline range), and 13% scored in the normal range on the MABC. Had these children been tested twice on the same battery the test-retest scores would be expected to correlate very highly:  $r=0.89$  for the BOTMP (Bruininks, 1978) and  $r=0.95$  for the MABC (Croce et al., 2001). It has been suggested that differences in both the structure and administration requirements of the two batteries may in part be responsible for low agreement (Crawford et al., 2001).

#### **1.1.1.5 What exactly is DCD: Is everyone on the same page?**

An issue very important to diagnosis is the understanding and perceptions of teachers and other professionals in contact with children who are later diagnosed with DCD. As there are so many possible presentations of DCD, it is interesting to note that teachers report that they would be more concerned and likely to intervene if a child in their class demonstrated poor gross motor abilities (in a non-disruptive manner) than if a child showed fine motor deficits or motor deficits were accompanied by disruptive behaviour (Rivard, Missiuna, Hanna & Wishart, 2007). Teachers' perceptions of typical motor skills for boys and girls also influence which children they identify as having significant motor difficulties (Rivard et al., 2007). With such biases, it has been found that within a sample of 32 primary school-aged children with DCD, only eight (25%) were identified by their class teacher as having motor difficulties (compared to 15 (47%) by physical education teachers), using a motor skills questionnaire (the MABC checklist: Henderson & Sugden, 1992) (Piek & Edwards, 1997).

A related question concerns the contention surrounding the different labels used to describe children with DCD. Peters, Barnett & Henderson (2001) gathered definitions of 'clumsy', 'dyspraxia' and 'Developmental Coordination Disorder' from UK-based medical doctors, speech and language therapists, physiotherapists, occupational therapists (OTs) and teachers (teaching in primary and secondary special and mainstream schools). As expected, some respondents were more familiar with these terms than others, with each group of professionals tending to give definitions focusing on the concerns most relevant to their profession (e.g. speech and language therapists describing dyspraxia with a focus on articulatory deficits). Despite growing consensus that DCD is now the preferred term for dyspraxia and associated disorders (as it was at the time of the survey), 32% of the 234 respondents were unable to accurately define DCD and only 7% said that 'DCD' and 'dyspraxia' were synonymous terms. Many of the less obvious facets of the disorder, such as motor planning, were not included in definitions.

The confusion among health and education professionals highlights a problem that is also common in the research literature, with different defining criteria often moulded

to reflect research questions. With such different understandings of the nature of DCD, it is unsurprising that definitions used in experimental studies differ in much the same way as in education and healthcare settings. Geuze, Jongmans, Schoemaker & Smits-Engelsman (2001) conducted a review of 164 articles published between 1980-1999 with keywords including 'DCD', 'dyspraxia' and 'perceptual-motor problems'. The diagnostic criteria for the clinical group in each article were compared with the DSM-IV diagnostic criteria for DCD. It was found that 74% of studies satisfied Criterion A: motor impairments were evaluated using normed-tasks that relate to normal daily activities. The common use of the MABC (50% of studies) satisfies this requirement, however, like all currently available test batteries, it is unable to provide a full account of all facets of motor functioning. While the vast majority of reviewed articles did explicitly address Criterion A, the vague nature of the diagnostic criteria makes it unclear which tasks should be used to give an adequate assessment of the necessary facets of motor functioning. Also, the question of quantifying the level of impairment on these tests is contentious. The majority (97%) of studies used the 15th percentile as a cut-off for DCD, however as this is an arbitrary (albeit a common) cut-off, it is unclear whether or not it is the most appropriate: children scoring slightly above the cut-off may still experience marked motor difficulties which impact on daily life, an important aspect of the diagnostic criteria. The pattern of impairment is also questioned: does motor impairment, as measured by tests like the MABC, need to be evident in more than one domain of motor functioning (e.g. gross motor skills and fine motor skills)? The MABC is failed if three of the eight tests are failed, making it possible to fail the battery with clinically impaired ability in only one dimension (e.g. manual dexterity or balance, each of which consist of three tasks). Children who fail in this manner likely belong to a DCD subgroup.

While Criterion A is relatively well adhered to, Criterion B (the stipulation that deficits detailed in Criterion A have a marked impact on daily life and academic achievement) is less rigorously tested in experimental studies. Sixty percent of studies reviewed assembled their DCD group from children referred to services such as occupational therapy (OT), suggesting that they assumed that the referral was sufficient to indicate that Criterion B was satisfied. Few studies detail the extent to which daily and academic life is affected, nor do they detail the areas most affected. Henderson & Barnett (1998) argue that Criterion B is difficult to properly address, especially as it seems to rely on the assumption of a causal relationship with Criterion A. Such a causal relationship requires verification through evidence from longitudinal study, which is currently unavailable. The relationship between the two is also noted to be age dependent, with negative impact on daily life becoming apparent only some years after Criterion A is satisfied (Henderson & Barnett, 1998).

Criteria C and D (referring to existing medical conditions and mental retardation respectively) are also difficult to assess and different definitions and standards are used throughout the experimental studies reviewed. Geuze et al. (2001) conclude that it is

most often the case that experimental studies assessing DCD do not strictly adhere to the DSM diagnostic criterion for the condition, with exclusion criteria particularly poorly adhered to. Some facets will be recorded, while others perhaps are inferred (e.g. the assumption that children in mainstream education will have an IQ within the normal range). This suggests that caution should be exercised when reviewing the literature as the DCD groups could potentially differ from one another quite substantially, making generalisation of findings problematic.

A final aspect of DCD which seems to cause confusion is DCD in adults. As yet, there appear to be no standardised behavioural assessments of DCD/dyspraxia for adults, and diagnostic criteria focus exclusively on children. The only formal diagnostic tool for adults is in the form of a checklist: the Adult Developmental Coordination Disorder/Dyspraxia Checklist, developed by Kirby, Edwards, Sugden & Rosenblum (2010). From speaking with an educational psychologist responsible for diagnosing specific learning difficulties in university students, it is clear that when it comes to adults with motor difficulties not everyone is on the same page. In this case, no tests of dyspraxia per se are actually used to give students a ‘dyspraxic profile’. Students must show difficulties in organisation, planning and coordination in an IQ test and writing tests. This procedure is far removed from the standard DCD diagnostic procedure for children, makes no attempt to satisfy DSM or ICD criteria, and does not make use of the adult DCDQ. This highlights how ill-defined DCD groups can be, and how loose the use of ‘DCD’ and associated terms is in some professional settings.

### **1.1.2 Literature review of studies investigating motor skills in DCD**

A number of studies have addressed motor skills in DCD, from basic visuomotor skills and manual dexterity to whole-body skills such as postural control. Some of these studies are discussed below, split into basic visuomotor and fine motor skills (those relying mainly on hand-eye coordination) and gross motor skills, which involve the whole body.

#### **1.1.2.1 Basic visuomotor and fine motor skills**

##### **1.1.2.2 Pointing**

Pointing tasks are a simple way to assess basic visuomotor ability. Every reaching or pointing action is made up of an acceleration (ballistic) phase and a deceleration phase. The acceleration phase (i.e. time zero to peak velocity) is a feedforward phase in that the action is programmed before it is initiated and is not adjusted. The deceleration phase follows, in which the movement is under feedback control, so it can be modified and fine-tuned (Woodworth, 1889). An online correction task, in which a target jumps during a reaching movement, is often used to investigate movement planning and execution (e.g. Veyrat-Masson, Briere & Proteau, 2010; Gritsenko, Yakovenko & Kalaska,

2009). The movement is planned when the target is straight ahead, so the movement must be corrected online (mid-reach) once the target has jumped to the side. Children with DCD ( $n=22$ , 7-13 years) have been found to show similar peak speed (the end of the feedforward phase of the movement) to TD, however the deceleration phase was significantly longer (Plumb, Wilson, Mulroue, Brockman, Williams & Mon-Williams, 2008). A specific deficit in the deceleration phase could suggest a deficit in the use of sensory feedback for online correction. However as children consistently spent longer in the deceleration phase even when the target remained stationary and large corrections were not necessary, the authors suggest that online correction is intact in DCD and the problem is a general slowness (a feedforward problem). However, the specific difference in the deceleration phase seems more likely to reflect deficient feedback control, as even movements to stationary targets require minor correction as the hand nears the target.

The details of this study give a somewhat incomplete picture. Analysis was insufficient, with no spatial measures (meaning we have no indication of the extent to which each group corrected movements) and insufficient details of acceleration and deceleration in baseline conditions in which the target did not move. These details could provide further support for the authors' 'general slowness' conclusion. From this study at least, it seems that the portion of aiming movements that are especially reliant on visual and proprioceptive feedback are more drawn out in DCD children when compared to TD counterparts (see also Zoia, Castiello, Blason & Scabar, 2005). Hyde & Wilson (2011) also report a longer deceleration phase coupled with a more typical acceleration phase in children with DCD ( $n=13$ , 8-12 years) during an aiming task. It has been suggested that DCD children might be slower to complete corrected movements as they need to foveate a target (providing ocular proprioceptive cues) before directing a hand movement to it, and tend to temporally couple hand and eye movements less tightly than TD (Wilmot, Wann & Brown, 2006). This study again found no significant differences in the feedforward portion of movement, with difficulty stemming from feedback control. These studies demonstrate motor slowness and inaccuracy in aiming in DCD, in line with diagnostic criteria, and also suggest that DCD might be associated with difficulties in correcting movements using current sensory information.

### **1.1.2.3 Action planning**

In order to complete any action, including those described above, it is necessary to initially plan the action. The action is then executed according to that plan, while taking into account new information available during the action. These planning and execution elements have been investigated by Kirby et al. (2010) in a study involving a whole-body action, as children played a 'river crossing game'. The aim was to get from one side of the 'river' (a school gym hall) to the other by placing and standing on mats ('stepping stones'). In order to assess both their planning abilities and execution abilities, children first had to place the mats on the floor in a way that would allow them to

use as few as possible but still manage to cross the river. The children then crossed the river using the stepping-stones they had placed, allowing for an examination of their ability to execute planned movements. Two groups of children (11 DCD and 28 TD) aged 9 to 11 were tested. None of the children had a diagnosis of ADHD (a disorder known to affect action planning) and all were deemed to be of normal intelligence. After three attempts to cross the river, the mats could be moved to reflect any adjustments to the spatial representation of the problem after the initial trials. There was a significant difference in the spatial positioning of the mats by the two groups, with TD children placing mats further apart than the DCD group. This is partly explained by a moderate correlation between the child's height and the distance between mats in the TD group and a relatively weaker (nonsignificant) correlation in the DCD group. This suggests that the DCD group were perhaps less aware of their stride capability, while TD children were generally able to determine how far apart mats could be for them to comfortably reach them. It was noted that the children in the DCD group tended to focus on the instruction to 'not fall in' and decided to place more mats closer together to ensure they were more likely to succeed. It was concluded that DCD children were less able to plan their movement efficiently, and this may be due to a deficit in creating a visual representation of the problem space (Wilson, 2005). They were also noted to produce visibly awkward movements. Comparing these findings to those of the pointing task employed by Plumb et al. (2008) (described in the previous section), it seems that the slower deceleration phase found in reaching movements might be explained by the 'play it safe' approach seen here, although in neither case was this approach successful in producing accurate or fluid movements such as those seen in TD.

#### **1.1.2.4 Gross motor skills**

#### **1.1.2.5 Balance, postural control and postural knowledge**

Balance is one of the three types of motor skills explicitly tested in the MABC. Balance, and the related issue of postural control and postural knowledge have been assessed in a number of ways.

The 'swinging room' paradigm (Lee & Aronson, 1974) has been used extensively to examine the sensory cues necessary for balance, in both clinical and typical groups. Subjects stand on a stable floor, with four suspended walls enclosing the space. The walls are moved to simulate the visual feedback that subjects would receive if they were swaying back and forth. In neurotypical subjects, this false visual feedback prompts them to sway their body to compensate for the apparent motion: they try to remain in an upright position. From this we can say that these subjects used visual information to help gauge their current posture. However, since there was no motion to compensate for, a number of subjects were reported to fall backwards as they compensated for

the apparent motion (Lee & Aronson, 1974). When this paradigm was used to assess postural control in children with DCD ( $n=6$ , 10-12 years) it was found that there were two subgroups within the DCD group: one group fell over; the other maintained their balance (Wann, Mon-Williams & Rushton, 1998). This suggests that there may be sub-types of DCD and it is likely that those children who remained standing would be differentiable from other DCD children in the balance component of the MABC or other standardised test batteries. However the sample size is too small to conclude with any certainty that subgroups exist. Results from the swinging room paradigm have illustrated the need for both vision and body awareness for balance. Indeed, body awareness (proprioception) is usually necessary for any motor task. The differential role of vision and proprioception in motor tasks in those with DCD and ASD will be explored in Chapters 3 and 4.

Balance has also been assessed by examining gait, with DCD children tending to show atypical gait patterns (Woodruff et al, 2002). It has been found that when compared to TD children matched for age, weight and stature, children with DCD ( $n=10$ , 6-8 years) walk with shorter, more frequent steps (Deconinck, Clercq, Savelsbergh, Coster, Oostra, Dewitte & Lenoir, 2006a). While the pattern is normal and rhythmic, the shortening of the movements and increased frequency deviates from the typical pattern. It is suggested that this immature gait pattern, often seen in very young children (Sutherland, Olshen, Biden & Wyatt, 1988), is an adaptation technique to compensate for poor balance control (Deconinck et al., 2006a). Increased postural sway in a neutral standing position also demonstrates clear postural control deficits in DCD (Geuze, 2003; Przysucha & Taylor, 2004; Wann et al., 1998).

Postural control has also been examined via postural muscle activity using electromyography (EMG) (Johnston, Burns, Brauer & Richardson, 2002). This study found that during a rapid voluntary movement of the arm, children with DCD ( $n=32$ , 8-10 years) had significantly different postural muscle activation in the trunk muscles than those without DCD. While movements completed by the TD group were characterised by anticipatory muscle activations, the DCD group's movement was characterised by later muscle activation of a more reactive nature. The DCD group was also significantly slower to perform the basic arm movement, both for reaction time (RT: time to initiate the movement) and MT (completing the movement). Atypical muscle timings in DCD have also been found in the leg and trunk muscles (Steele, 1994).

#### **1.1.2.6 Catching**

As suggested by the inclusion of catching tasks in a number of movement assessment batteries, children with DCD have been found to be significantly less proficient in catching and throwing than TD children. Astill & Utley (2006) found that children with DCD ( $n=8$ , 7-8 years) made a number of small movements (multiple acceleration and deceleration phases) rather than one single movement, when catching a ball two-

handed. This would have resulted in a very jerky, awkward movement, as described in diagnostic criteria and clinical observations (Williams, 2002). The DCD group were also described as showing inflexibility in movement, and tended to rely on only a few motor strategies (e.g. performing the same movement simultaneously with each arm), even if the strategies had proved unsuccessful. A similar study found that children with DCD aged 7-8 ( $n=8$ ) caught significantly fewer balls than age-matched TD children, showed smaller ranges of motion and again tended to tightly couple the movement of their limbs so as to reduce the degrees of freedom involved in the action (Utley, Steenbergen & Astill, 2007).

Using a perceptual paradigm in which children watched a video showing a child throw a ball towards the camera, children with DCD ( $n=40$ , 5-7 years) have also been found to be significantly worse than TD in correctly identifying whether the ball should be caught to the right or left. This uncertainty was evident throughout the viewed movement (Lefebvre & Reid, 1998). This suggests that difficulties in catching in DCD may in part be due to difficulties in predicting where the body needs to be in order to catch the ball. It is concluded from these studies that both motoric and perceptual skills necessary for ball skills are deficient in children with DCD.

#### **1.1.2.7 DCD summary**

As can be seen from the description of these previous studies and findings, a broad range of motor skills appear to be deficient in DCD, however there are some inconsistencies, e.g. the status of online correction abilities in DCD. The main observation though is the lack of studies looking at multiple types of motor skills in the same participant cohort. As it stands, the picture appears to show motor difficulties in almost every aspect of movement, however systematic investigation of this is lacking. A clearer picture would be gleaned from studies involving a number of different motor skills in a single well-defined cohort.

## **1.2 Autism Spectrum Disorder**

ASDs are neurodevelopmental conditions affecting around 1-2% of the population (Baron-Cohen, Scott & Allison, 2009; Baird, Simonoff, Pickles, Chandler, Loucas, Meldrum & Charman, 2006; Blumberg, Kogan, Schieve & Jones, 2013) with major associated deficits classically represented as a triad of behavioural impairments (Wing & Gould, 1979). These impairments are broadly defined as: impaired social interaction; restricted, repetitive and stereotyped behaviour; and communication deficits (APA, 2000). The autism spectrum encompasses a variety of different presentations of this triad of impairments. A hugely diverse and heterogeneous clinical population is represented as the spectrum spans from low-functioning autism, through to AS, high-functioning autism (HFA) and Pervasive Developmental Disorder-Not Otherwise

Specified (PDD-NOS)<sup>2</sup>.

While primarily a social deficit, motor components are also important, although this is not immediately apparent from current<sup>3</sup> diagnostic criteria. These criteria will now be discussed, with a focus on the role of motor deficits in diagnostic procedure. The prevalence of motor deficits in ASD will also be discussed, followed by a review of literature investigating various motor skills. Imitation as a motor skill will be discussed briefly and expanded on in Chapter 2.

### **1.2.1 Recognition of motor deficits in ASD in early accounts of the disorder**

Historically, autism has been associated with motor difficulties, with Kanner (1943) and Asperger (1944) both detailing motor slowness and clumsiness in their seminal observations of classic autism and AS respectively. These observations are still made in more recent literature (Klin, 2006) and parents and carers often recount instances of clumsiness and motor difficulties beyond those expected in developmental delay (Miller & Ozonoff, 2000). However, despite recognition of motor deficits in early reports of autistic disorders, they were, and currently still are, overshadowed by the traditional triad of impairments.

### **1.2.2 Diagnosing ASD: The role of motor impairments across the spectrum**

Currently, motor deficits are not a major part of the diagnostic criteria for ASD and are not routinely assessed. There appears to be uncertainty as to the universality of motor deficits in the autistic population, with empirical research at times appearing at-odds with the diagnostic criteria. The diagnostic criteria in the DSM-IV and ICD will briefly be outlined and this will be followed by a discussion of the literature addressing questions of prevalence and generality across the spectrum. Note that the DSM and ICD list definitions for different conditions on the autistic spectrum, with autistic disorder appearing uniquely in the DSM, and childhood autism and atypical autism appearing only in the ICD.

The DSM-5 was not published when the majority of work in this thesis was conceptualised and conducted. None of the participants involved in the present studies or those reviewed from the previous literature were diagnosed using these new criteria. For these reasons the DSM-5 will only be described briefly.

#### **1.2.2.1 DSM-IV-TR criteria**

##### Autistic Disorder (AD)

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<sup>2</sup>The DSM-5 has collapsed all of these conditions into a single disorder: Autism Spectrum Disorder.

<sup>3</sup>The DSM-IV was the current version at the time of writing.

At least six of the following criteria must be met for a diagnosis of Autistic Disorder to be given:

*Criterion A:* Impaired social interaction, manifested by at least two of the following: impaired use of nonverbal behaviours; failure to develop appropriate relationships with peers; lack of spontaneous sharing of experience; failure to show emotional or social reciprocity.

*Criterion B:* Impaired communication, manifested by at least one of the following: a delay or lack of spoken or gestural communication; inability to sustain conversation; stereotypies of language; lack of imitative and pretend play.

*Criterion C:* Restricted and stereotyped behaviours, manifested as intense and focused preoccupation with restricted patterns of interest; strict adherence to routine and rituals; stereotyped and repetitive atypical motor mannerisms; a preoccupation with details and parts of objects.

### Asperger's Disorder

The DSM-IV-TR gives the following criteria for a diagnosis of Asperger's Disorder (or Asperger's Syndrome), assuming a certain number of the criteria are met:

*Criterion A:* Impaired social interaction, manifested by at least two of the following: impaired use of multiple nonverbal behaviours to successfully regulate social interactions; failure to develop appropriate relationships with peers; lack of spontaneous sharing of experiences; lack of emotional and social reciprocity.

*Criterion B:* Restricted repetitive and stereotyped interests and behaviours, manifested by at least one of the following: intense preoccupation with stereotyped interest; inflexibility with regards to routines and rituals; stereotyped and repetitive motor mannerisms; a preoccupation with parts of objects at the expense of attending to the whole.

*Criterion C:* Impairments and difficulties have a significant adverse effect on daily living.

*Criterion D:* Language was not significantly delayed.

*Criterion E:* There is no significant cognitive delay, with the exception of difficulties in social interaction.

*Criterion F:* Criterion is not met for any other pervasive developmental disorder or schizophrenia.

### Pervasive Developmental Disorder-Not Otherwise Specified

The DSM-IV-TR states that a diagnosis of PDD-NOS will be given in cases where not all symptoms for autistic disorder are present. PDD-NOS therefore includes atypical autism. There are no guidelines regarding the number of symptoms necessary for diagnosis.

### 1.2.2.2 DSM-5 criteria

The DSM-5 now only lists Autism Spectrum Disorder (ASD), in place of each individual autism diagnosis listed in the DSM-IV. Of importance with relation to motor deficits, ASD and DCD are now listed as possible comorbid conditions. Although ASD and DCD may now be diagnosed together, the reason for this change is unclear. There are very few comparisons of motor deficits in ASD and DCD in the psychology literature, and the same seems to be true in the occupational therapy and physiotherapy literature. It is possible that it was simply prompted by reports from clinicians of frequent motor deficits in ASD. This is certainly possible given the reported prevalence rates, which are discussed in Section 1.2.3 below.

### 1.2.2.3 ICD-10 criteria

#### Childhood Autism

A diagnosis of Childhood Autism would describe a child with a PDD characterised by the following:

- a) By the age of 3 years, development is impaired or abnormal in language, social interaction or symbolic play;
- b) There is evidence of the triad of impairments (social reciprocity; communication; repetitive and stereotyped behaviours).

#### Asperger's Syndrome

A diagnosis of Asperger's Syndrome is reserved for those who meet the criteria for childhood autism, but there is no general delay in language development and cognitive development is unaffected. Those with AS tend to exhibit 'marked clumsiness'.

#### Pervasive Developmental Disorders (PDD)

A diagnosis of PDD is given when there are qualitative abnormalities in reciprocal social interactions and communication, and there is a restricted and stereotyped repertoire of interests and activities.

#### Atypical autism

A diagnosis of atypical autism would describe a child with a PDD that does not meet all of the diagnostic criteria for a diagnosis of childhood autism. Either the onset of symptoms is beyond age 3, or there are impairments in some but not all of the triad of impairments.

#### 1.2.2.4 Criteria summary

The obvious lack of clear criteria regarding motor deficits in ASD in all but the ICD criteria for AS suggests that they are not considered to be an important diagnostic marker<sup>4</sup>. This is despite growing empirical work supporting Kanner (1943) and Asperger's (1944) original observations of motor clumsiness and slowness in ASD, and links between motor deficits and common autistic symptomatology such as imitation deficits (detailed in Chapter 2). It has recently been suggested that repetitive and stereotyped behaviours referred to in diagnostic criteria may be a manifestation of general motor impairment (Ravizza, Solomon, Ivry & Carter, 2013), however this possible link has not been thoroughly investigated. Using a repetitive tapping task, Ravizza et al. (2013) found that increased variability in this basic motor task tended to coincide with an increased severity of repetitive behaviours such as hand flapping. However, this study used a very basic measure of motor ability, concerned primarily with producing temporally accurate movements. The relationship between broader motor ability and repetitive behaviours is unclear. Throughout this thesis 'motor deficits' refers to motor coordination difficulties, rather than behavioural idiosyncrasies such as repetitive and stereotyped movements.

### 1.2.3 How prevalent are motor deficits in ASD?

#### 1.2.3.1 Do motor deficits differentiate AS from HFA/AD or do they unite the spectrum?

The prevalence of motor deficits in ASD is not clearly documented. The question is further confounded as diagnostic criteria for AS include motor deficits (in the ICD-10), although they are not necessary for a diagnosis (Manjiviona & Prior, 1995), with the traditional triad of impairments carrying more weight. Suggestion from some researchers (e.g. Tantum, 1988) and the presence of motor deficits in the ICD-10 diagnostic criteria for AS and the absence of such criteria for autism led some researchers to the assumption that individuals with AS should not be mixed with individuals with other ASD diagnoses in studies of motor skills. From this, a question has arisen as to whether motor deficits might differentiate AS and autism<sup>5</sup>.

A number of studies and clinical observations have, however, suggested that motor deficits may be apparent in all forms of ASD (e.g. Seal & Bonvillian, 1997; Page & Boucher, 1998; Maurer & Damasio, 1982). Using the Test of Motor Impairment-Henderson Revision (TOMI-HR), Manjiviona & Prior (1995) found that AS ( $n=12$ ) and HFA ( $n=9$ ) children are not differentiable in terms of motor function. Using the BOTMP, Ghaziuddin, Butler, Tsai & Ghaziuddin (1994) also directly compared AS

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<sup>4</sup>This is still the case in the DSM-5, however DCD is listed as a possible comorbid condition.

<sup>5</sup>This is now a less relevant point from a clinical perspective, since the DSM-5 collapsed all previous autistic spectrum diagnoses into a single diagnosis, however it is still of interest in research considering the incidence of motor deficits in ASD.

( $n=11$ , 9-19 years) and HFA ( $n=9$ , 7-17 years) and again found that motor deficits did not differentiate children with these apparently distinct diagnoses. There were no significant differences in any of the individual subtests for gross motor, fine motor or upper limb functioning across the 2 ASD groups, providing further evidence of motor deficits across the spectrum, regardless of specific diagnosis. The authors do not detail analyses of differences between subtests within the ASD group as a whole, so it is not possible to ascertain the pattern of difficulties in motor skills. However, the test items do not seem to be heavily reliant on child-experimenter interactions or the understanding of social cues, suggesting that basic motor problems in ASD are not artefacts of social deficits and misunderstandings of social cues in testing situations.

It is now largely accepted that when assessing motor deficits in ASD, participants with AS and autism (high or low functioning) can be considered as a single group. Reported prevalence of motor deficits in ASD, and where applicable in ASD sub-diagnoses, will now be discussed.

#### **1.2.3.2 Prevalence rates across the autistic spectrum**

The reported prevalence of motor impairment in ASD varies considerably from one test of motor skill to another (and consequently from one study to another). For example, Ming, Brimacombe & Wagner (2007) found that, according to a doctor's diagnosis of apraxia, 34% of their sample of children with ASD ( $n=154$ ) exhibited motor apraxia, with a tendency for this to be more prevalent in younger children (2-6 years) than older children (7-18 years) ( $p = 0.06$ ). The children in this study had a diagnosis of AD ( $n=74$ ), PDD-NOS ( $n=70$ ) or AS ( $n=10$ ), as described in the DSM-IV. There was no significant association between exact diagnosis and presence of motor deficits, providing further support to the assertion that motor deficits are not restricted to AS, contrary to ICD criteria. Screening for motor deficits using the MABC (a standardised test discussed in more detail in Chapter 2), Green, Charman, Pickles, Chandler, Loucas, Simonoff & Baird (2009) found a considerably higher prevalence of four out of five in 9-10 year old ASD children ( $n=101$ ) with mixed diagnoses (i.e., autistic diagnoses across the spectrum). These children scored in the lower 5th percentile, with a further 9.9% falling between the 6th and 15th percentile, suggesting borderline motor deficits. These findings were irrespective of exact diagnosis and IQ, suggesting that poor motor ability is not reserved only for lower functioning ASD children [although a positive significant correlation between motor deficits and IQ has been reported by Green et al. (2009)]. As balance board was one of the two worst performed tasks, this again suggests that motor difficulties are based on a fundamental motor problem, rather than manifesting through a social deficit whereby children do not understand the requirements of the task due to a reliance on tester/child interaction (e.g. ball catching).

A slightly lower prevalence has been reported using the TOMI-HR (the predecessor of the MABC) with children aged 7-17, although again there was no significant difference between prevalence in AS ( $n=12$ ) and HFA ( $n=9$ ): 50% prevalence of motor deficits in AS and 66.7% in HFA (Manjiviona & Prior, 1995). Additionally there was no clear pattern of relative difficulty between the 3 test subscales (manual dexterity, ball skills, and balance). These results again support the suggestion that motor deficits are found across the autistic spectrum, although with relatively small sample sizes and a larger age range than Green et al. (2009) (7-17 years, mean age 11 years), the reported prevalence could be misleading.

Other high prevalence estimates include that reported by Klin, Volkmar, Cicchetti & Rourke (1995), who report a 90% prevalence in AS and HFA using the diagnostic criteria for Nonverbal Learning Disabilities, which include motor and visuo-motor deficits; and Gillberg (1989) reported 83% prevalence in AS using the gross motor scale from the Griffiths Mental Development Scales (GMDS, Griffiths, 1970). Gillberg (1989) reported that children with AS had relatively deficient motor ability, walked in a stiff, awkward manner, without swinging the arms, and appeared generally uncoordinated and clumsy [see also Ghaziuddin, Luke & Ghaziuddin (1992)].

While the studies detailed so far have found relatively high instances of motor deficits in ASD, Gillberg, Ehlers, Schaumann, Jakobsson, Dahlgren, Lindblom, Bagenholm, Tjuus & Blinder (1990) cited a gross motor deficit in just two children from a group of 18, using the GMDS, in stark contrast to Gilberg's (1989) prevalence using the same battery. However, the former study is quite unusual in that it tested autistic children under three years of age, while the majority test school-aged children.

While the incidence of motor deficits seems superficially higher in AS relative to HFA/AD in some instances, this may be due to factors associated with working with lower functioning individuals, such as language and IQ difficulties, and perhaps also motivation and willingness to participate.

It is clear that the exact prevalence of motor difficulties in ASD is difficult to assess. Previous studies have used a number of different standardised and non-standardised tests, with some testing very basic motor skills and others testing more difficult tasks involving a sequence of actions and imitation. What is clear however is that ASD, across the spectrum, involves a significant level of motor impairment. In all of the studies detailed above, ASD subjects were not children with comorbid DCD/dyspraxia diagnoses. An interesting question then is how these DCD-like deficits compare to those deficits exhibited by children with DCD. Green et al. (2009) have suggested that, based on standardised tests of movement, there are generally more similarities than differences between motor deficits in ASD and DCD. There are however comparatively few studies assessing motor skills in ASD and DCD using the same paradigm or test. These comparative studies are detailed in Section 1.3.1.

## 1.2.4 Literature review of studies investigating motor skills in ASD

The frequent coexistence of ASD and DCD-like symptoms calls into question whether these two deficits are mutually exclusive, as diagnostic criteria suggest. Just as research has investigated a number of diagnostic characteristics of children and adults with DCD, there is also a growing literature addressing the question of the specific nature of motor difficulties in ASD. These studies have assessed motor skills using standardised movement assessment batteries and praxis tests, and a number of other experimental paradigms similar to those used to investigate motor skills in DCD. A number of these studies have already been detailed in the previous section on prevalence rates. A selection of these studies will now be discussed in more detail, alongside other studies assessing various motor skills in ASD.

### 1.2.4.1 Studies using standardised motor batteries

Key details of the studies discussed in this section are given in Table 1.2. Of those studies using batteries assessing both fine and gross motor skills, the majority do not report greater impairment in one or the other (Green et al., 2009; Manjiviona & Prior, 1995; Jansiewicz, Goldberg, Newschaffer, Denckla, Landa & Mostofsky, 2006; Liu, Hamilton, Davis & Garhy, 2014), although Kopp, Beckung & Gillberg (2010) did find that gross motor deficits were more apparent than fine motor deficits.

While a number of the findings can be taken at face value, some deserve further discussion. It is clear that children with ASD have marked difficulties on a range of tasks compared to TD children, however in some cases social difficulties may exacerbate the situation. Using an apraxia battery, Dowell, Malone & Mostofsky (2009) found that the ASD group were significantly impaired in producing gestures, however the social nature of the task makes inferences about underlying motor deficits problematic. Subjects also completed a postural knowledge test and ASD children were significantly worse at identifying intransitive gestures (symbolic gestures without tools) compared to TD children, but were as able as TD to identify transitive gestures. This highlights a selective deficit in knowledge of gestures that require more social understanding, again suggesting a cautious interpretation of inability in gesture tests. This indirect effect of social difficulties in some motor assessments is also seen in a gross motor deficit identified by Pan, Tsai & Chu (2009). In this case two thirds of children with ASD failed a ‘galloping’ task (similar to skipping), compared to only 5% of the TD group. This is likely in part due to difficulties in coordinating the movement and maintaining balance, but it is possible that the impairment may be confounded by social difficulties in understanding the strange request, or a reluctance to complete an action that seems nonsensical.

Table 1.2: Studies using motor batteries to assess motor skills in ASD

Author	Clinical groups	Ages	Tests	Main findings
Dowell et al. (2009)	ASD (n=37)	8-13 years	PANESS (RTM), Florida Apraxia Battery	ASD significantly poorer performance than TD in both tasks.
Green et al. (2009)	ASD (n=11)	6-10 years	MABC	Performance is poor throughout the battery.
Jansiewicz et al. (2006)	AS (n=15), HFA (n=16)	6-17 years	PANESS	Performance is poor throughout the battery.
Kopp et al. (2010)	AD (n=14 3-6 years, n=10 7-16 years), AS (n=1 3-6 years, n=5 7-16 years), PDD-NOS (n=4 3-6 years, n=5 7-16 years), Childhood Disintegrative Disorder (n=1 3-6 years). Comorbid ADHD in 95% of 7-16 year olds	3-6 years (pre-school), 7-16 years (school-aged)	Cailler-Asuja Scale, MABC, EB-test, MNP assessment, TGMD-2	25% of school-aged children and 80% of pre-school children meet DCD criteria. Gross motor more problematic although deficient performance across all tasks.
Liu et al. (2014)	ASD (n=21)	5-10 years	TGMD-2	Performance is poor throughout the battery. Significantly worse than TD on object control, locomotor, and overall gross motor quotient.
Manjiviona & Prior (1995)	AS (n=12), HFA (n=9)	7-17 years (mean=11)	TOMI-HR	Performance is poor throughout the battery.
Pan et al. (2009)	ASD (n=28), ADHD (n=29)	6-10 years	TGMD-2	Performance is poor throughout the battery. Significantly worse than TD on object control, locomotor, and overall gross motor quotient. Significantly worse than ADHD on object control and locomotor subtests.

RTM: Repetitive timed movements (a fine motor skill subtest from the Physical and Neurological Examination for Soft Signs (PANESS)).

#### 1.2.4.2 Fine motor skills

Using a modified version of the Fitts' pointing task (Fitts, 1954), in which subjects make repetitive pointing movements to targets in sequence, Papadopoulos, McGinley, Tonge, Bradshaw, Saunders & Rinehart (2012) found that HFA children ( $n=19$ , mean age 9 years) had comparable movement times (MT) to TD, but made significantly more errors and had more variable end-point accuracy. Kinematic variables other than MT were not analysed in this case so it is not possible to ascertain whether the feedforward and feedback phases of the movement were also similar between groups. Using a similar paradigm in adults with ASD ( $n=9$ ), Glazebrook, Elliot & Lyons (2006) found that the ASD group was significantly slower to initiate movements towards targets and this was significantly correlated with IQ, with lower IQ subjects taking longer to prepare movements. Movement execution was also significantly slower in ASD, as was peak acceleration, peak velocity and time to peak velocity, although this slowing was unrelated to IQ. Similar results were reported in a pre-cued pointing task (Glazebrook, Elliot & Szatmari, 2008), and in some conditions lower nonverbal IQ was associated with longer RTs (time to plan and initiate the movement). In this case MT was longer in ASD when aspects of the required movement were pre-cued, suggesting possible over-planning and then difficulty executing the planned movement when predictions were incorrect. From these studies we can conclude that fine motor skills associated with pointing appear to be deficient, although pinpointing the deficit is difficult. There appears to be disagreement as to which aspects of these actions are deficient: planning, execution, or both. This is discussed in more detail in Section 1.2.4.4.

#### 1.2.4.3 Gross motor skills

Gross motor deficits seem to be relatively common in ASD. Klin et al. (1995) reported such deficits in 63% of HFA children ( $n=19$ , mean age 15 years) and 100% of children with AS ( $n=21$ , mean age 16 years), based on observations written in medical records. Similar to findings in DCD, gait patterns have been found to differentiate ASD and TD. Weiss, Moran, Parker & Foley (2013) reported that young adults with severe autism ( $n=9$ , 16-22 years) walked more slowly than TD, took shorter strides, spent longer in a stance position (with both feet on the ground) and swung their arms less while walking. Another study using predominantly lower functioning children with ASD ( $n=21$ , 3-10 years) also found shorter strides (controlling for the child's height) and longer stance phase, as well as a tendency to walk flat-footed or walking on the toes rather than letting the heel hit the ground first (this latter feature was significantly correlated with IQ, tending to occur in lower IQ participants) (Vilensky, Damasio & Maurer, 1981). These results support Gillberg's (1989) account of unusual gait in AS. Children with ASD ( $n=21$ , 9-14 years) have also been found to have less stable posture when standing still on two feet, showing more sway than TD children. Such sway was not specific to younger children, as was the case in TD (Memari, Ghanouni, Gharibzadeh,

Eghlidi, Ziaee & Moshayedi, 2003). From these studies we can conclude that gross motor skills related to postural control and gait are deficient in ASD (however see Calhoun, Longworth & Chester, 2011).

#### 1.2.4.4 Action planning

As discussed previously with reference to fine motor skills, there is some evidence for action planning deficits in ASD. Hughes (1996) found that children with ASD ( $n=36$ , mean age 12 years) had difficulty in planning the appropriate grasp (overhand or underhand) in a rod placing task. Children with ASD ( $n=12$ , mean age 7 years) have also been found to have difficulties modulating the first step of a multi-step reach according to varying stimulus size (Fabbri-Destro, Cattaneo, Boria & Rizzolatti, 2009). In the latter case, children with ASD were able to complete the task but seemed to approach it as disjointed components and did not plan the task holistically. However, using a simpler reach-to-grasp task, Mari, Castiello, Marks, Marraffa & Prior (2003) found that manipulations of the object size and distance had similar effects on children with ASD ( $n=20$ , 9-13 years) and age-matched TD. When looking at within-group differences, those ASD children with an IQ  $<70$  were significantly slower than their higher functioning counterparts, mirroring Glazebrook et al.'s (2006) IQ effect. In Mari et al.'s (2003) task grasping occurred at 72% of movement duration in lower IQ children, compared to 15% for higher IQ children, showing that lower IQ children were not able to adopt an appropriate grasp until late in the movement. This may be due to difficulties performing different components of a movement together, making the action more piecemeal, as was observed by Fabbri-Destro et al. (2009). It should be noted though that all children with ASD were affected by manipulations of the object (and therefore manipulations of the required action plan) in the same way as TD children. This means that the plan was perhaps less efficient (although ultimately accurate) or was less well executed in ASD.

Differences between Fabbri-Destro et al. (2009) and Mari et al. (2003) may be due to the additional step in the former making the task more demanding. Mari et al. (2003) required subjects to reach to and lift an object, while Fabbri-Destro et al. (2009) required subjects to lift and transport the object.

Age and/or specific ASD diagnosis has been found to affect motor planning in ASD, with older children with AD comparable to controls, and younger HFA children significantly slower to initiate movements (Rinehart, Bellgrove, Tonge, Brereton, Howells-Rankin & Bradshaw, 2006). The authors interpret this as a sign of slowed motor planning. They also found that in an HFA group, time in the deceleration phase was not modulated by target expectancy: when targets were in an expected location, they spent longer in the deceleration phase. This reliance on the feedback portion of a pre-planned movement to a primed location suggests the action was poorly planned. Children with AD were not different from TD in any respect on these tasks, however this

group was significantly older than the HFA group (on average 4 years older), cautioning against an interpretation of these results as evidence of an AD/HFA dissociation.

#### **1.2.4.5 ASD summary**

As with the literature on motor difficulties in DCD, there does not appear to be a clear picture of relatively deficient and spared areas of motor functioning in ASD. While some studies suggest that gross motor deficits are more common than fine motor deficits, this pattern is certainly not clear enough to suggest that fine motor skills are spared to any significant extent. The lack of a clear pattern could be explained by the use of varying methodologies and standardised and non-standardised tests; it could also be due to the heterogeneous nature of the disorder. It is also clear from the literature on motor functioning in ASD that motivation, cooperation and social difficulties also pose problems for researchers aiming to assess motor functioning in isolation (e.g. Page & Boucher, 1998). The issue of motor deficits impacting imitation skills is closely linked to this problem and is discussed in Chapter 2.

### **1.3 Comorbidity between ASD and DCD**

#### **1.3.1 Comparing ASD and DCD directly**

Comparing findings from the studies detailed above, there appears to be little differentiating motor impairments in ASD and DCD. Both seem to have difficulties planning movements, tend to have slower movements than TD, and gait and postural stability also differentiate both groups from TD. However this conclusion is not directly evidenced as the studies detailed above all use different paradigms and motor batteries, and subject groups vary widely in age and IQ, both of which have been found to affect motor ability (e.g. Glazebrook et al., 2006).

Few studies make a direct comparison between motor skills in ASD and DCD. In one such study, Dewey, Cantell & Crawford (2007) found that children between 5-18 years with either ASD or DCD were significantly impaired relative to both TD and ADHD on the short-form BOTMP. This highlights the known problem of comorbidity and symptom overlap (Rasmussen & Gillberg (1999) p142). Interestingly, post hoc analysis showed that performance in the ASD group was the worst of all clinical groups, significantly worse than the DCD and DCD+ADHD groups. However, despite the ASD group appearing the most impaired group, 41% of the group did not meet diagnostic criteria for motor impairment set out in the short form BOTMP. As discussed previously, there is not perfect agreement between motor batteries so some of these children may have met DCD criteria if a different test or a combination of diagnostic tools were used. Unfortunately, there was no reported analysis of fine- compared to gross-motor skills within and between groups, so it is not possible to form a profile of motor skills for either group.

Similar findings are reported using the MABC, with children with AS and DCD (6-10 years) not significantly different on overall test score (Green, Baird, Barnett, Henderson, Huber & Henderson, 2002). In this case however AS did score significantly lower than DCD in the ball skills component, although the underlying cause of this difference is unknown. While it may be due to the unique motoric components of the ball skills tasks relative to manual dexterity and balance, the authors suggest that extraneous social factors may be responsible for the poorer performance in AS. Children with AS may be less well practised in ball skills as ball games are generally played with other children, and such social play is often avoided. It could also be the case that due to the heterogeneous nature of both conditions, small sample size ( $n=11$  and 9 for AS and DCD) created an artificial group difference.

### 1.3.2 Comorbidity or coincidence?

Comorbid conditions refer to distinct chronic conditions which coexist in an individual. For conditions to be comorbid the presence of one may not preclude the other, as was the case for ASD and DCD in the DSM-IV. The conditions are not one and the same but are likely to occur together, perhaps suggesting an increased susceptibility to certain kinds of conditions, such as the clustering of common neurodevelopmental disorders such as ASD, DCD, ADHD and dyslexia (Rasmussen & Gillberg, 1999). The exact meaning of comorbidity is often not defined in the relevant previous literature. As is often the case in such literature, throughout this thesis ‘comorbidity’ refers to the co-occurrence or concomitance of multiple conditions in a single individual. The question of whether this co-occurrence reflects shared causal origins of two conditions, or two truly distinct conditions (as Feinstein (1970) suggests should be the case for comorbidity), is the central question here.

The presence of motor deficits in ASD can complicate the diagnosis process. As the presence of motor deficits is included in the diagnostic criteria but is not necessary for a diagnosis, those children or adults presenting with autistic traits and coordination and motor problems are not well catered for. A judgement call has to be made about the severity of both the autistic traits and the motor issues and then the most severe difficulty is used for diagnosis. Note that even if motor development is severely affected, a DCD diagnosis is automatically ruled out if autistic traits are severe enough for any PDD diagnosis<sup>6</sup>. The difficulty is, motor deficits are apparent in ASD, and there is some evidence of social deficits in DCD (Jarus, Lourie-Gelberg, Engel-Yeger & Bart, 2011; Chen, Tseng, Hu & Cermak, 2009), making it very difficult to know where the cut-off lies. Additionally, in cases where children with DCD have social difficulties, it is unclear whether they are brought about by reduced peer contact due to the emphasis on physical play in the playground, or whether social difficulties were preexisting and

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<sup>6</sup>This is no longer the case since the introduction of the DSM-5, although was the case for all of the subjects in empirical work detailed in this chapter and in studies detailed throughout this thesis.

are independent of their motor difficulties. If there are no differences between motor deficits in ASD and DCD, as appears to be the case, and if ASD-like social deficits are often found to some extent in children with DCD, this would suggest that the two conditions could form two intersecting spectra, with the majority of ASD and DCD children likely to have symptoms from both the social dysfunction and motor dysfunction spectrum. It is important then to thoroughly test motor skills in a way that separates them as much as possible from social skills (e.g. avoiding cooperative ball games). If deficits are detected and the areas of difficulty appear to follow the same pattern in ASD and DCD this suggests that lower-level skills, such as basic sensory components of movement (vision and proprioception), should be investigated. This will give us a clearer understanding of the mechanisms underlying deficits already detected. If further investigation were to show no differences in ASD and DCD on high-level motor tasks (such as model building, throwing etc.) or more basic sensory cue processing, this would suggest that diagnostic criteria for ASD and DCD should converge, and certainly the status quo of the two being mutually exclusive (as dictated by DSM-IV criteria) should be reconsidered.

## 1.4 Chapter 1 conclusions

This introductory chapter has described two neurodevelopmental disorders, ASD and DCD, with a particular focus on motor skills. The diagnostic criteria in current and recent editions of the DSM and ICD have also been discussed. It was noted that motor deficits in ASD are not well reflected in these criteria: those with ASD are not able to obtain a DCD diagnosis (using DSM-IV criteria), and motor deficits are sidelined by the traditional triad of impairments. The reported prevalence of motor deficits in ASD (and ASD subgroups) was discussed: motor deficits appear to be common in ASD, irrespective of exact diagnosis, cognitive functioning or autism severity (Dewey et al., 2007), however prevalence estimates vary widely.

While a number of studies have assessed various aspects of motor functioning in ASD and DCD, it is difficult to understand how the difficulties in one group compare to difficulties in the other. Direct comparison in the literature appears to be sparse, and generalising across the many different tasks, research questions, age ranges, diagnoses and other study variables is difficult. It is not possible to create a clear profile of motor deficits in each group in a meaningful, comparative way from the existing literature.

Finally, the issue of ASD/DCD comorbidity was discussed and it was suggested that the high rate of comorbidity of ASD and DCD perhaps suggests that the diagnostic criteria set out at the start of the chapter need to become less mutually exclusive<sup>7</sup>.

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<sup>7</sup>This concern has now been addressed in the DSM-5.

## 1.5 Outline of thesis

The primary question of this thesis is whether motor deficits, which are clearly evident in both clinical groups, are characteristically different or not. Due to the lack of a clear profile of motor deficits in ASD relative to DCD, as a starting point for this thesis a number of different motor tasks will be administered to adults and children with ASD and DCD in order to seek a profile of motor functioning. Profiling will be based primarily on two batteries: a computerised visuomotor battery and a standardised motor battery (the widely used MABC-2: Henderson, Sugden & Barnett, 2007). A motor imitation task will also assess motor skills in action reproduction. As these two clinical groups are known to be highly heterogeneous, it may be that no clear pattern is found, however performance on the tasks will still inform future studies. Some of the participants included in the profiling study reported in Chapter 2 also take part in follow-up experiments, which allows their performance to be considered relative to their observed motor ability measured in the profiling study.

This thesis now describes a number of studies:

- Chapter 2 reports an examination of basic motor skills and imitation skills in adults and children with ASD, DCD and TD.
- Chapter 3 is the first of three chapters investigating the use of visual and proprioceptive cues in ASD, DCD and TD. It investigates sensory cue processing in a spatial location matching task and a reaching task following visual displacement of the hand.
- Chapter 4 investigates visual and proprioceptive contributions to perception and action in an orientation matching task and posting task.
- Chapter 5 investigates the role of proprioceptive acuity in a visuo-tactile body illusion in neurotypical adults and children with ASD, DCD and TD.
- Chapter 6 reports a study assessing the relationship between social and motor development in TD children.
- Chapter 7 presents general conclusions and scope for further research.

## Chapter 2

# Profiling motor skills in ASD and DCD

There is no clear understanding of the nature of motor deficits in ASD relative to DCD. Therefore this chapter presents a direct comparison of these groups on a series of tasks assessing various high-level and basic visuomotor skills. This study's primary aim is to provide a profile of motor skills in ASD and DCD groups. A secondary aim is to provide a comparison of two tests of motor skill: the debatable 'gold standard' MABC-2 and a new, currently non-standardised tool (the computerised Kinematic Assessment Tool: cKAT)<sup>1</sup>. The study includes both adult and child groups and results from these will be reported and discussed separately, followed by a general discussion.

### 2.1 Aim 1: Profiling motor skills and drawing comparisons between subject groups

In Chapter 1 a number of studies that have used tests of motor ability with either ASD or DCD groups were discussed. However, a weakness in the current literature is that few have provided detailed accounts of performance. It is difficult to determine from the literature if and where points of difference lie in the profiles of ASD and DCD individuals on tests such as the MABC.

The first study in this thesis aims to provide a more in-depth profile of motor skills in age-matched groups of adults and children with ASD and DCD. This will be accomplished by using two test batteries and a motor imitation task. This approach allows a broad profile of ability to be investigated (capturing possible sub-groups within each clinical group) and also allows for the comparison of two very different motor test batteries: one standardised battery testing both fine and gross motor ability against age-norms, and one as-yet unstandardised test assessing basic visuomotor processing.

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<sup>1</sup>At the time of testing the version of the battery used was not standardised: the latest version of the battery is currently in the process of standardisation.

### 2.1.1 MABC-2

The MABC (or MABC-2) is the most prominent standardised tool used in assessing DCD in the UK and Europe (Blank, Smits-Engelsman, Polatajko & Wilson, 2012) and is commonly used in research. The MABC-2 is made up of eight subtests, divided into three components: manual dexterity (MD), aiming and catching (AC), and balance. The battery provides age appropriate tests in three ages-bands for children aged 3-16 years (3-6, 7-10, and 11-16 years). Performance on each subtest is compared to age-appropriate norms and from this percentile rank scores can be calculated for each component, and for the battery as a whole. Generally the 15th percentile identifies children with a probable impairment whose motor skills should be monitored, and the 5th percentile is used to identify children with a significant motor deficit.

The present study uses the 15th percentile as a cut-off, following advice from clinicians working with children with DCD and in line with the majority of research (e.g. Zwicker, Yoon, Mackay, Petrie-Thomas, Rogers & Synnes, 2013; Johnston et al., 2002; Sinani, Sugden & Hill, 2011; Pannekoek, Rigoli, Piek & Barrett, 2012; Geuze et al., 2001). The frequent use of the 15th percentile is supported by academic associations (Blank et al., 2012) and there is supporting clinical evidence that children with a firm diagnosis of DCD can fall between the 5th and 15th percentile on the MABC: Missiuna, Gaines, McLean, DeLaat, Egan & Soucie (2008) found that 20 of 88 children with OT-diagnosed DCD were between the 6th and 15th percentile. Although in the minority, it should be noted that some researchers favour the more stringent 5th percentile (Sugden, 2006; Dunford, Street, O'Connell, Kelly & Sibert, 2004).

MABC-2 subtests are described below and in Tables 2.1-2.3. For each task children are given a demonstration and practice trials before two test trials are carried out. Only the score from the most successful test trial is used.

- *Manual dexterity*: These are all timed tasks involving hand-eye coordination. Manual dexterity tests for each age group are detailed in Table 2.1.
- *Aiming and Catching*: These tasks involve throwing and catching either a bean-bag or tennis ball. None of the tasks are timed. Each practice trial involves five throws/catches and each test trial involves ten. Aiming and catching tests for each age group are detailed in Table 2.2.
- *Balance*: These are tests of gross motor skills and postural stability. One of these tasks is timed. Balance tests for each age group are detailed in Table 2.3.

### 2.1.2 Previous findings using the MABC with ASD and DCD groups

A number of studies assessing motor skills in ASD and DCD using the MABC have been detailed in Chapter 1. In general, those testing ASD groups have found high

Table 2.1: Manual dexterity subtests for each age bracket

Age band	Task	Instruction	Time limit
Young	Coin posting	Place coins into a box through a slot in the top of the box. This is done separately for the dominant and non-dominant hand.	As quickly as possible.
	Threading beads	Thread a lace through a number of plastic beads.	As quickly as possible.
	Drawing trail	Draw through a path while attempting to stay in the lines.	None.
Middle	Placing pegs	Place 12 small rounded pegs into a pegboard. This is done separately for the dominant and non-dominant hand.	As quickly as possible.
	Threading lace	Thread a lace through a line of holes in a ruler.	As quickly as possible.
	Drawing trail	Draw through a path while staying within the lines. The two sides of the path are closer together than in the drawing trail used in age band 1.	None.
Old	Turning pegs	12 pegs (inch long sticks with a yellow half and a red half) are placed in holes on a board. All pegs show the same colour. Using one hand, children must turn each peg around using their fingers and place them back in the pegboard so that the previously hidden colour is now showing. This is done separately for the dominant and non-dominant hand.	As quickly as possible.
	Triangle building	A triangle is constructed (using a reference model) from 3 straight lengths of plastic, with a nut and bolt securing each corner. Only the required pieces are given and once an item is picked up it cannot be put down again.	As quickly as possible.
	Drawing trail	Draw through a path while attempting to stay in the lines. Lines are closest together in this path compared to the paths used for age band 1 and 2.	None.

Table 2.2: Aiming and catching subtests for each age bracket

Age band	Task	Instruction	Time limit
Young	Catching bean-bag	Children throw a beanbag in the air directly in front of them and must catch it with both hands.	None.
	Throwing bean-bag onto mat	Children throw a beanbag (with two hands) onto a mat on the floor, 1.8 metres in front of them. The child should aim for any part of the mat.	None.
Middle	Catching two hands	Using two hands, the child throws a tennis ball at the wall from a distance of 1.5 metres and catches it. The ball may bounce on the floor once. One proper trial involves 10 throws.	None.
	Throwing bean-bag onto mat	Children throw a beanbag (with one hand) onto a mat on the floor, 1.8 metres in front of them. The child should aim for a large circle (10 cm diameter) in the centre of the mat. One proper trial involves 10 throws.	None.
Old	Catching one hand	The participant stands 1.5 metres from a wall and throws a tennis ball at the wall and catches it with one hand. The ball may not bounce and must be caught cleanly with one hand, without bringing the ball into the body to cushion it. Each hand is given two proper trials, with one trial involving 10 throws. The ball should always be caught with the same hand that threw it. This is done separately for the dominant and non-dominant hand.	None.
	Aiming at wall target	The participant must throw a tennis ball (with one hand) at a large red circle (25.5 cm in diameter) placed just above their head-height, from 2.5 metres away. The participant may choose which hand to use and is free to change hand used at any point. As above, one trial constitutes 10 throws. This is done separately for the dominant and non-dominant hand.	None.

Table 2.3: Balance subtests for each age bracket

Age band	Task	Instruction	Time limit
Young	One leg balance	Children stand on a mat on the floor on one foot and aim to maintain this position for 30 seconds. This is done separately for the dominant and non-dominant foot.	30 second target.
	Walking on toes	Children must walk forwards on their toes along a 4.5 metre line, ensuring that their heels are raised.	None.
	Two foot jumping	Children jump with both feet along a series of mats in a straight line in front of them. They must move from mat-to-mat without jumping on mats more than once and without jumping outside of the mats.	None.
Middle	One board balance	The child stands on a small balance beam with one foot and should hold the position for 30 seconds. This is done separately for the dominant and non-dominant foot.	30 second target.
	Walking heel-to-toe forwards	Children must walk forwards along a 4.5 metre line, ensuring that their heel of one foot and toes of the other are together as they walk along the line.	None.
	Hopping	The child hops along a series of mats straight in front of them. They must move from mat-to-mat without jumping on mats more than once and without jumping outside of the mats. The same foot is used for a whole trial. This is done separately for the dominant and non-dominant foot.	None.
Old	Balance board	Balance with feet one in front of the other, heel and toe touching, on a small balance beam (narrower than that used for age band 2). It is raised 3.5 cm from the floor and the area which can be stood on is 10 cm wide and 64 cm long. This position is to be held for 30 seconds.	30 second target.
	Walking heel-to-toe backwards	Walk backwards along a 4.5 metre line, ensuring that the toe of the back foot always touches the heel of the front foot when walking.	None.
	Zig-zag hopping	Mats are placed on the floor in a zigzag pattern, with two corners touching (like the black spaces in two adjacent rows on a chess board). The child must hop (with the same foot throughout each trial) sequentially from one mat to the other. They must move from mat-to-mat without hopping on mats more than once and without hopping outside of the mats. Each trial is completed with one foot only. This is done separately for the dominant and non-dominant foot.	None.

instances of clinical motor deficits (Green et al., 2009). Few studies detail performance in DCD groups in their own right and tend to use the MABC only as part of a DCD identification process (e.g. Johnson & Wade, 2009; Astill & Utley, 2006; Coleman, Piek & Livesey, 2001). Others simply state that children with DCD are outperformed by TD throughout the battery (Engel-Yeger, Hanna-Kassis & Rosenblum, 2012). A rare comparison of MABC performance in ASD and DCD found greater deficits in ball skills in the ASD group, although this may be confounded by small sample size (11 ASD, 9 DCD) and extraneous social factors associated with ball games (Green et al., 2002). There is no clear picture of relative areas of strength and weakness in ASD compared to DCD using the MABC.

### **2.1.3 Can the MABC be a true gold standard?**

One aim of the present study is to compare the MABC-2 with a new visuomotor battery (cKAT). The MABC has been described by some as the “de facto gold standard” (Mon-Williams & Tresilian (2006), p341), however this may be due to its relatively more common usage rather than its greater power as a diagnostic tool. Indeed, its creators suggested that no tool available at the time, including the MABC, should be considered a ‘gold standard’, as none possessed enough supportive data (Henderson & Barnett, 1998).

Venetsanou, Kambas, Ellinoudis, Fatouros, Giannakidou & Kourtessis (2011) have returned to this question, to investigate whether the MABC should now be called the ‘gold standard’ of motor assessment tools. The authors note that when compared with other already well-established tests, it becomes apparent that different tools identify different children. Crawford et al. (2001) reported around 80% agreement when comparing the MABC with the BOTMP and the Developmental Coordination Disorder Questionnaire (DCDQ: Wilson, Dewey, Crawford & Kaplan, 2000), however agreement with other measures is considerably lower. Rodger, Watter, Marinac, Woodyatt & Ziviani (2007) found that only 35 of 60 children (58%) identified as motor impaired on the Neurodevelopmental Physiotherapy Assessment (Watter, 1996) were outwith the normal range on the MABC. Similarly, the MABC has been found to identify DCD in only 25 of 60 children (42%) referred to physiotherapy and physical therapy for motor difficulties (Watter, Rodger, Marinac, Woodyatt, Ziviani & Ozanne, 2008). Smits-Engelsman, Henderson & Michels (1998) found that 44 of 74 children (59%) referred for motor assessment fell below the 15th percentile on the MABC, and those with handwriting difficulties were often overlooked. Finally, Waelvelde, Peersman, Lenoir & Engelsman (2007) report an even poorer agreement (29%) between the MABC and the Peabody Developmental Motor Scales-2 (Folio & Fewell, 2000). Venetsanou et al. (2011) conclude that multiple tests of motor ability should be used in parallel when diagnosing DCD, as the MABC alone cannot reliably identify all children with clinically relevant motor impairment. It is still not clear why the MABC has been widely

adopted as a substitute ‘gold standard’.

The present study uses the MABC-2 for the following reasons: it will allow for performance of children in clinical groups to be compared with any previous MABC assessments detailed in medical records; and it is the most commonly used test in previous research and clinics in the UK.

#### **2.1.4 cKAT: a future gold standard?**

The computerised Kinematic Assessment Tool (cKAT: Culmer, Levesley, Mon-Williams & Williams, 2009) is a newly developed tool. Its efficacy has not yet been tested relative to diagnostic tools, nor has standardisation data been published. The battery was developed with the aim to make “a sophisticated tool...widely accessible to the scientific and clinical community”. The developers suggest it as a better alternative to more low-tech and limited tasks such as the MABC tracing task to assess basic facets of visuomotor control with a focus on kinematic variables. The battery uses a touchscreen tablet laptop with a stylus and does not require motion tracking systems such as those commonly used in the lab to gather kinematic data. This makes the test portable and easy to use in settings such as schools and hospitals, which may lack the space for larger motion tracking set-ups. The version of the battery used here includes three tasks assessing tracking and tracing ability. Kinematic and accuracy data are collected for each task.

As well as including performance on the cKAT battery in the motor profile for each group, the secondary aim of this study is to compare this touchscreen-based test with the MABC-2, to investigate whether basic kinematic properties of visuomotor control can differentiate those with and without motor difficulties. If a narrower test (cKAT) manages to pick out individuals with DCD, regardless of which motor domain they find most problematic, this approach may be able to replace motor batteries with fine- and gross-motor components as a screening instrument for DCD. The comparison of test efficacy between age groups should also prove interesting, as there is presently no test designed specifically to screen for DCD in adults, and cKAT may be a viable new option.

The MABC-2 and cKAT will give a detailed profile of fine-, gross-, and visuo-motor skills, without a potentially confounding reliance on subject-experimenter interaction. (This lack of interaction is particularly important for the ASD group.) A motor imitation task will also add breadth to the profile built up for each group. Performance in the motor imitation task will also be considered relative to general motor ability as measured by the two motor batteries. A review of recent literature on imitation in ASD and DCD and its relevance to motor ability is given in the next section.

As each task in the MABC, cKAT and imitation task has slightly different requirements, even if they are within the same broad domain (e.g. fine motor), it should be possible to pinpoint which kinds of tasks highlight any points of difference between

groups. This will inform future studies in this thesis which will examine any underlying task components found to differentiate groups.

## 2.2 Imitation

Imitation can be considered from a number of perspectives: motor, cognitive and social. For this reason imitation ability is of interest in both ASD and DCD as both groups have varying degrees of social and motor impairment. Imitation has received little attention in the DCD literature but has been a popular area of ASD research, although it is often considered from a social or cognitive perspective. Imitation from a motor perspective has received relatively little attention in the ASD literature. The imitation task used in the motor profiling battery in this study focuses on imitation as a more fundamental motor ability. It will be contrasted with general motor performance in each group and results will perhaps add to the current understanding of imitation ability in ASD and to the small literature on imitation in DCD.

### 2.2.1 Understanding imitation

Imitative behaviour is typically used in social interactions in order to facilitate communication with others, and is also utilised in learning and play. Anyone who has played ‘Simon Says’ as a child has purposefully imitated: Another child said “Simon says do this” and jumped on the spot, and you will have likely jumped on the spot as well. Many people claim that autistic individuals have problems with imitation (Hammes & Langdell, 1981; Sigman & Ungerer, 1984; Ohta, 1987; Stone, Lemanek, Fishel, Fernandez & Altemeier, 1990; Rogers, Bennetto, McEvoy & Pennington, 1996), however there is evidence to the contrary (e.g. Bird, Press & Heyes, 2007; Ingersoll, Schreibman & Tran, 2003; Whiten & Brown, 1998).

The lay “Simon says” definition of imitation given above is not representative of the full conceptualisation of imitative behaviour. Delineation of the term is essential, and this has been highlighted as one of the major problems in the burgeoning area of research into imitation in ASD (Sevlever & Gillis, 2010). Once different types of imitation are considered, it becomes clear that the once popular opinion that autistic individuals cannot imitate is an over-generalisation, and the problem is much more complicated.

Imitation is a multifaceted concept and the majority of studies have approached it from either a social or cognitive perspective, and within each perspective use varying definitions of the term (Sevlever & Gillis, 2010). Each definition then leads to certain tests and paradigms being used, and this variation in studies produces anything but a unified answer to the question of possible imitation deficits in ASD. Sevlever & Gillis (2010) describe four types of imitation: stimulus enhancement, emulation, mimicry and true imitation. These are described in Table 2.4.

Table 2.4: Types of imitation, as defined by Sevillever &amp; Gillis (2010)

Imitation type	Definition
Stimulus enhancement	Imitation triggered by attention being captured by a stimulus. Example: a friend takes a slice of cake and you do the same.
Emulation	The end goal of an action is imitated, but the means by which the goal is achieved is not copied. Example: a friend picks up a slice of cake and you take a mouthful of cake with a fork.
Mimicry	Imitation of both end goal and means. Some argue that this exact replication replaces an understanding of the intention and is an ‘empty’ action (Tomasello, 1990). Example: a friend taps the table twice then picks up a slice of cake and you do exactly the same.
True imitation	Involves the underlying processes of emulation and mimicry. Described by Whiten & Ham (1992) as the likelihood that a novel action will be performed in a certain way after demonstration, compared to the likelihood the same movement would be produced without demonstration.

The circumstances in which we imitate often dictate the kind of imitation we do: purely social circumstances will often call for different imitation than more functional contexts. For example social imitation such as the mirroring body language, compared to emulation used when opening a box with a latch similar to one you have used before. Tasks not involving objects (for example copying hand movements such as waving) are termed gestures, while those tasks involving an object are termed action-on-object imitation. Gestures can be seen as more prosocial, as the focus of the imitation is another person. They are further defined as being either meaningful (such as waving-synonymous with hello and goodbye) or non-meaningful (such as tapping your index finger on the table) (Williams et al, 2004). Meaningful gestures and object-oriented imitation are termed goal-directed (Sevillever & Gillis, 2010), although it is clear that the former is goal-oriented in a more social way than the latter. Research focusing on various aspects of imitation in ASD are described in the next section.

### 2.2.2 Can people with ASD imitate?

This review describes studies investigating various aspects of imitation, including the propensity for spontaneous imitation, the effect of meaning in imitated gestures, imitating kinematics, and finally an overview of the effect of social motivation on imitation in ASD.

### **2.2.2.1 Spontaneous versus elicited imitation**

Neurotypicals will often spontaneously imitate and mimic others, for example, mirroring movements and body language of someone sitting opposite them [the Chameleon Effect: Chartrand & Bargh (1999)]. Imitation is often socially advantageous as it increases our affiliation with others (Lakin, Jefferis, Cheng & Chartrand, 2003). Given the lack of social motivation in ASD (Chevallier, Kohls, Troiani, Brodtkin & Schultz, 2012), it is unsurprising that some have noted a failure to engage in such spontaneous imitation (Ingersoll & Gergans, 2007; Hobson & Lee, 1999). However to say that there is a deficit in spontaneous imitation seems incorrect: some of the idiosyncrasies of ASD suggests that these individuals, broadly speaking, have the ability to imitate (or at least emulate) spontaneously, i.e. without explicit instructions. This is evident from reports of echopraxia and echolalia in which movements or phrases are repeated very precisely (Charman & Baron-Cohen, 1994; Lee, Mikesell, Joaquin, Mates & J. H, 2009). This clearly shows an intact ability to match another's actions, but it tends not to be socially motivated and may lack insight of the action being performed (Hamilton, 2008).

Additionally, a number of studies provide evidence of more typical intact spontaneous imitation outside of echopraxia. For example, Bird et al. (2007) found typical automatic imitation of hand actions in adults with ASD (i.e. faster action reproduction following a congruent primer). While not socially appropriate or advantageous, this behaviour is qualitatively different from imitation associated with echopraxia. More importantly, natural kinds of spontaneous imitation have also been found to some degree in young children with autism (Brown & Whiten, 2000). Interestingly, when imitation is elicited by experimenters there is little evidence of a marked deficit in imitation fidelity (e.g. Beadle-Brown, 2004; Beadle-Brown & Whiten, 2004).

### **2.2.2.2 Meaningful versus meaningless: does meaning aid imitation?**

Just as explicit instruction seems to aid imitation in ASD, there is evidence of a facilitatory effect of meaning in the imitation of gestures in ASD (Williams, Whiten & Singh, 2004; Wu, Chiang & Hou, 2011; Aldridge, Stone, Sweeney & Bower, 2000; Rogers et al., 1996). Aldridge et al. (2000) found that young children with ASD ( $n=10$ , 2-4 years) were able to judge intention and complete an unfinished action, but were unable to complete a meaningless gesture, in which intention was not obvious. This finding could be interpreted as evidence that children with ASD can only emulate a perceived end-goal (intention); alternatively it could highlight an intrinsic insistence on meaning by ASD subjects in imitation tasks. Deficits in imitating meaningful actions in a pantomime manner compared to imitating with tools (Hammes & Langdell, 1981) could also point to an insistence on purposeful actions: If you were a child with a very literal way of thinking, would you see the point in hammering an imaginary nail with an imaginary hammer?

Williams et al. (2004) suggest that the use of meaning to guide successful imitation may be related to increased language competence (and therefore age). However, previous studies reporting deficits in meaningless actions coupled with typical imitation of meaningful actions (e.g. Wu et al., 2011) have involved very young children (2-4 years), casting doubt over the role of language development.

There appears to be a general consensus that meaningful actions are easier for children with ASD to imitate than meaningless actions, however the reasons for this are unclear. The reasons for imitating nonsensical or pointless gestures may be apparent to TD children, who understand the game-like nature of the tasks, although children with ASD might find the social aspect of the strange request difficult to understand.

### **2.2.2.3 Imitating kinematics**

More relevant to imitation as a motor skill is the imitation of the style of movement. Just as higher-level imitation is often reported to be deficient in ASD, so too is the basic style and kinematics of imitated actions (Hobson & Hobson, 2008; Hobson & Lee, 1999).

Gowen (2012) suggests that typically we have a flexible approach to imitation: we emulate when there is a clear goal, as the style is incidental to achieving this goal; but we mirror the action (including its kinematics) more closely when the goal is unknown. Those with ASD might be less attuned to kinematic properties of movements (perhaps due to underlying motor difficulties), resulting in specific deficits in imitating meaningless actions, or those with no visual goal as described above. However Wild, Poliakoff, Jerrison & Gowan (2012) found that adults with ASD tended not to imitate speed in either a goal-directed or meaningless action, while neurotypical adults did modulate their speed for meaningless actions. Importantly however, in each case there was no explicit requirement to copy the speed or general style of the action. The instructions to subjects simply stated “watch the video clip carefully and then copy what you saw as best you can” (Wild et al., 2012, p1742). This is important as it has already been reported that those with ASD may not imitate spontaneously, but are able to when the requirement is explicitly stated. Additionally, with the high incidence of motor difficulties in ASD, it is possible that poor imitation of kinematics may be due to motor difficulties. Wild et al. (2012) rule out this possibility in their study as the groups were equivalent on a basic tracing task, however this is insufficient evidence to rule out a role of motor impairment. In order to rule out the role of motor difficulties it is necessary to have a more complete picture of subjects’ motor ability.

There appears to be a general consensus that style is less-well imitated in ASD, but comparatively little work has been conducted in this area. Results may be affected by aspects of the task, such as the type of gesture being imitated, and the way in which imitation is elicited. The effect of motor ability on imitation in ASD requires further investigation.

#### **2.2.2.4 Social motivation**

The prevailing theories in autism research for some time have focused on a primary deficit in social cognition (e.g. Baron-Cohen, Leslie & Frith, 1985). The idea that dysfunction in social motivation could account for social deficits is a relatively new one (Chevallier et al., 2012), although is supported by a similar dissociation between ability and propensity for empathy often seen in ASD (Keysers & Gazzola, 2014). Chevallier et al. (2012) describe social motivation as a set of biological mechanisms and psychological dispositions that promote social orienting, social reward and maintaining of social relationships. Examples of the behavioural aspects of social motivation include the speeded attentional response to socially rewarding stimuli, such as faces (Fletcher-Watson, Findlay, Leekam & Benson, 2008), demonstrating that objects afforded social importance are prioritised. As discussed previously, ingratiating behaviours such as social mimicry are further demonstrations of an innate desire to maintain good social relationships.

The picture in autism is strikingly different in a number of areas. From an attentional perspective, there is no bias towards orienting to social stimuli, either in visual or auditory attention (Riby & Hancock, 2008; Klin, Jones, Schultz, Volkmar & Cohen, 2002; Ceponiene, Lepisto, Shestakova, Vanhala, Naatanen & Yaguchi, 2003). The enjoyment of social interaction for its own sake is also diminished (Chevallier et al., 2012; Krantz & McClannahan, 1993; Liebal, Colombi, Rogers, Warneken & Tomasello, 2008). It should be noted that some studies have found increased levels of loneliness in ASD (e.g. Mazurek, 2014), so it is likely that age, functioning and individual differences in personality will have some effect on the drive for recreational social interaction.

It seems likely that deficits in spontaneous imitation and imitation of meaningless gestures could be explained by a lack of social motivation. There is already evidence of intact imitation when it is elicited, but a failure to spontaneously imitate, pointing to a motivational deficit when imitation is purely social (Whiten & Brown, 1998). Spontaneous imitation offers no reward that is of interest to those not socially motivated. Similarly, imitating an action with no clear purpose, beyond pleasing the experimenter, will likely perplex those not driven by social motivation.

#### **2.2.2.5 Imitation in ASD summary**

The status of imitation skills in ASD is unclear, and the cause of reported deficits is difficult to establish, although motivational and motor deficits seem likely to play a role. It is important to note that even when studies report a deficit in imitation (relative to controls), it is often the case that imitation in ASD is not entirely absent or unsuccessful when appropriately motivated. One clear example is preference for imitation when sensory rewards are offered instead of social rewards (Ingersoll et al., 2003; Roeyers, Oost & Bothuyne, 1998). This, alongside increased difficulties in imitating meaningless actions (Williams et al., 2004), and spontaneously using imitation (Ingersoll & Gergans,

2007) suggests that a motivation deficit may explain a large part of the imitation ‘deficit’ previously reported in ASD. Findings of difficulties in imitating kinematics also point to a possible role of motor deficits.

### 2.2.3 Imitation in DCD

Compared to a wealth of literature on ASD, there are relatively few studies investigating imitation in DCD. However, studies that have been conducted seem to suggest that imitation skills are underdeveloped or deficient in DCD. Compared to age-matched controls, 9-11 year old DCD children have been found to be less able to imitate transitive and intransitive gestures. This was only true for imitation of gestures (matching motor output to visual input), with younger TD children (5-6 years) outperforming those with DCD. The ability to perform gestures following a verbal command or through tool use was not deficient in DCD (Sinani et al., 2011). Conversely however, Zoia, Barnett, Wilson & Hill (2006) found deficient pantomime imitation of meaningful gestures alongside deficient imitation using tools and following verbal command. Hill, Bishop & Nimmo-Smith (1998) report that children with DCD ( $n=11$ , mean age 9 years) commit more gestural errors (such as errors in gesture orientation) in both transitive and intransitive gestures than TD children, but are comparable to children with Specific Language Impairment, which is linked to ASD (Leyfer, Tager-Flusberg, Dowd, Tomblin & Folstein, 2008). In this case the imitation was attempted correctly, but executed poorly, highlighting that the problem is not one of conceptualisation but of sequencing the necessary movements.

### 2.2.4 Comparing imitation in ASD and DCD

Comparing children with ASD and DCD in imitation skills will help investigate the role of motor deficits in imitation ability in ASD. It seems likely from the literature on each group individually that there is some degree of overlap in imitation difficulties in these groups. However, it appears that a direct comparison of ASD and DCD has not received much attention. Dewey et al. (2007) compared imitation of meaningful gestures in children and adolescents with DCD and ASD (with motor deficits), and found an imitation deficit only in the ASD group. This would suggest that imitation deficits in ASD are not simply a manifestation of motor impairments. It should be noted however that this was true both for imitation (‘do as I do’) and gesturing following a verbal command, so the deficit is not specific to imitating per se. Similarly, Green et al. (2002) found that both children with AS and DCD (6-10 years) had difficulty producing meaningful gestures to verbal command, and imitating meaningless gestures, but performance in the AS group was significantly worse in both cases. Interestingly, motor performance on the MABC was correlated with imitation and gesturing ability in the AS group. It appears that motor difficulties in ASD may contribute to imitation deficits, but there is some evidence for imitation deficits greater than those associated

with DCD.

## 2.3 Methods

This chapter details a series of tasks intended to provide a motor profile of adults and children with ASD, DCD and TD via performance on the MABC, cKAT and visuomotor imitation tasks. The majority of literature suggests a large degree of overlap in motor difficulties in a variety of tasks, but direct comparisons are rare. Additionally, relative strengths and weaknesses within each group are difficult to determine from the existing literature. The adult and child studies will be reported and discussed separately (with the exception of the procedure, which is almost identical for both), followed by a short general discussion.

### 2.3.1 Subjects (adults)

Adults with a diagnosis of either an ASD<sup>2</sup> ( $n=10$ , aged 19-51 years: median=25.5) or DCD ( $n=17$ , aged 18-33 years: median=22) were recruited, alongside neurotypical/typically developed (TD) adults ( $n=20$ , aged 21-30 years: median=23). Groups were age- and IQ-matched ( $H(2) = 4.39, p = 0.111$  and  $H(2) = 0.20, p = 0.907$ ). All subjects gave written consent and were reimbursed £10 for participation. All testing took place within the University of Edinburgh.

ASD subjects were recruited through the University of Edinburgh's Disability Office and Number 6 (a local service for adults with HFA or AS). Those recruited through the University had a diagnosis of some form of ASD, verified by a healthcare practitioner (GP, psychologist etc.). Subjects recruited through Number 6 had their diagnosis of either AS or HFA verified by the Number 6 organisation, which requires written confirmation from a relevant medical practitioner.

Recruitment of DCD subjects was also through the Disability Office. Students with DCD had a childhood diagnosis of DCD and/or were described by an Educational Psychologist as having a 'dyspraxic profile' when they entered university<sup>3</sup>. As the majority of individuals with DCD are typically not re-assessed after childhood outside of university situations such as these, this group of individuals is as well defined as could be expected in an adult population.

TD subjects were recruited by word-of-mouth and an email advert, and tested by Lorcan Kenny as part of a Master of Science qualification. (The adult study detailed

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<sup>2</sup>An ASD is any diagnosis under the umbrella term. In line with new DSM-5 criteria, from this point the term 'ASD' will be used to describe subject groups in the present studies made up of subjects with various diagnoses on the autistic spectrum. When describing previous literature the diagnostic term used by individual authors will be used.

<sup>3</sup>On entering university, students with DCD diagnoses or those expressing concern with coordination/handwriting difficulties were required to complete the Wechsler Adult Intelligence Scale (WAIS) and writing tests. Those who show difficulties in motor, planning or organisational aspects of these tests are described as having a dyspraxic profile.

in this chapter is the only study in which testing was split between multiple experimenters.) None of the TD subjects reported having any previous diagnosis of DCD, ASD, ADHD or dyslexia.

## **2.3.2 Procedure (adult and child)**

### **2.3.2.1 Questionnaires**

Questionnaire data were requested from each subject. Adults were asked to complete the Autism-spectrum Quotient (AQ: Baron-Cohen, Wheelwright, Skinner, Martin & Clubley, 2001) and have someone complete the Social Responsiveness Scale (SRS: Constantino, 2005) on their behalf. Both questionnaires are measures of autistic traits. Parents/carers of children who took part were asked to complete the SRS and the latest version of the DCDQ (DCDQ-07: Wilson, Crawford, Green, Roberts, Aylott & Kaplan, 2009), to give an indication of the perceived incidence of ASD and DCD symptoms respectively.

### **2.3.2.2 Behavioural overview**

The behavioural procedure for adults and children differs very slightly as an additional task (visual-proprioceptive matching) was created for children.

The testing location was a large, quiet room, with subjects tested one at a time. Subjects completed the MABC-2, cKAT and imitation tasks in a fixed order and were offered breaks between each of these tasks. The MABC-2 was completed first and was administered as per instructions in the test manual and described previously in Tables 2.1-2.3. The MABC-2 took approximately 30 minutes to complete. All adults completed tasks for the oldest age group (see Borremans, Rintala & J. A., 2009 and Sahlander, Mattsson & Bejerot, 2008), and all children completed tasks appropriate for their age.

For the computer-based tasks (described in Sections 2.3.2.3 and 2.3.2.4) subjects sat at a desk in front of a tablet laptop with the screen lying flat, facing upwards. It was ensured that artificial and/or natural light sources were not creating glare on the screen. For the cKAT battery, subjects were told that they would complete a series of tasks, in which they would use the stylus directly on the computer screen. They were asked to keep the stylus on the screen at all times. Brief instructions were given for the first task and subjects were told that instructions for each task would be given on-screen. For the imitation task a second laptop used to show stimuli was placed on the desk behind the tablet computer. Subjects were told that they would watch videos of an action which they should copy onto the tablet. The computer-based tasks (cKAT and imitation) took around 25 minutes to complete.

Adults then completed the 2-subtest Wechsler Abbreviated Scale of Intelligence (WASI: Wechsler, 1999) (chosen for brevity), which concluded testing. After complet-

ing the MABC-2 and computer tasks, children had completed the first of two one-hour sessions. The second session included a visual-proprioceptive matching task, detailed separately in the first half of Chapter 3, followed by the WASI. Adults generally completed all tasks in one session. Where adults opted to complete two sessions, or children one session, tasks were completed in the usual order.

### **2.3.2.3 cKAT**

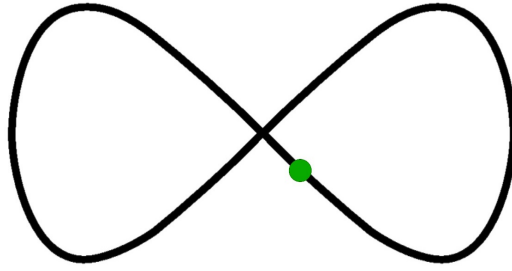
Three tests were included in the cKAT battery (see below and Figure 2.1).

- **Figure of 8 tracking:** Subjects must follow a dot around a large invisible infinity symbol. The dot accelerates throughout the trial and the aim is to keep the stylus on the dot at all times. A second condition includes a visible guide showing the path the dot will take and this guide remains on-screen throughout the trial. In each condition the dot traces the symbol 9 times.
- **Pentagram:** A dot moves between the corners of an invisible pentagram shape. The dot jumps from one corner to another 74 times. The aim is to move to the dot's new location as quickly as possible. The dot moves to the next corner as soon as the stylus is rested on the dot.
- **Path tracing:** Subjects must move the stylus through a path while trying to stay within the two boundary lines of the path. A box moves along the path every 5 seconds and subjects are asked to stay within the box while tracing the path. The box therefore acts as a means of pacing. There are two paths, each used three times.

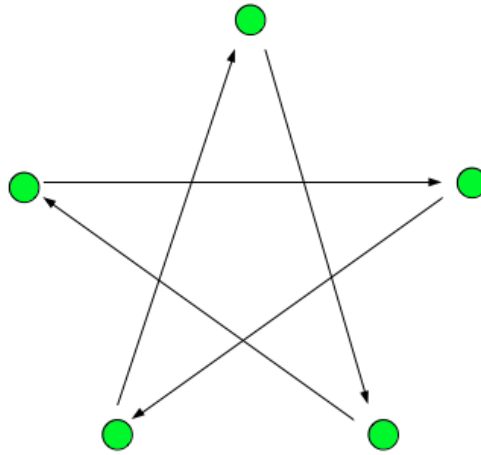
### **2.3.2.4 Imitation**

The imitation task used here is being categorised as assessing true imitation (Tomasello, Kruger & Ratner, 1993). The explicit requirement to copy all kinematic and visual properties of the stimulus (shape, size and speed) means that emulation (copying only the end result, i.e. shape) is not appropriate and is actively discouraged.

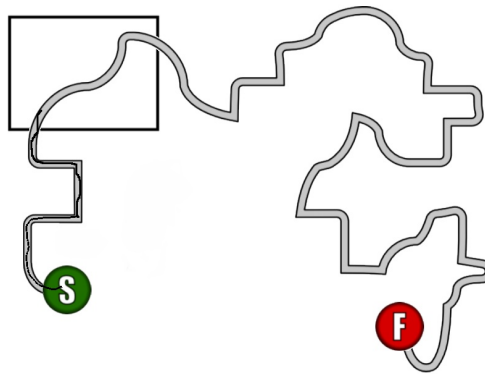
Subjects watched videos of a drawing task in two main conditions. In one condition an actor drew shapes on a tablet computer like that being used by the subject. In the second condition, the actor watched as a dot traced a shape on the tablet (see Figure 2.2). In both cases he was silent throughout and did not look up from the tablet. In both conditions the shape drawn was invisible (neither the actor's stylus nor the dot left a trail). Twenty seven unique trials were recorded in the actor condition, and these actions were then replicated exactly in the dot condition. This was achieved by replaying the actor's action (screen coordinates and kinematics) with a black dot indicating where the stylus had been. The drawing produced was always one of three shapes



(a) The tracking task with a guide.



(b) Dot movement in the pentagram task (note only one dot was visible at any one time).



(c) One of the two tracing paths.

Figure 2.1: Screenshots/illustrations of each cKAT task

(circle, triangle, square); sizes (small, medium, large); and speeds (slow, medium, fast). Shapes were drawn free-form by the actor, so each instance of a ‘small’ shape occupied a slightly different area of the screen, and similarly each instance of a ‘fast’ shape had a slightly different speed.

Subjects were asked to recreate what they had seen on the tablet computer in front of them, copying the shape, size and speed. A black dot was positioned in the bottom left corner of the screen (8 cm from the left and 5 cm from the bottom). The dot remained on the screen throughout and subjects were instructed to start and end each drawing at the dot. The pen leaving and re-entering the dot triggered data collection. Subjects were asked to keep the pen on the screen throughout the drawing. They were told that the screen would only respond to the pen, so they could lean on the screen with their arm if necessary. Due to the sensitivity of the screen to the stylus, additional ‘blank pages’ (screens with a start/end dot) were given to allow for false trials in which data collection began prematurely.

Condition order was counterbalanced within each group. Trials in the first condition followed fixed random order A and the second condition followed fixed random order B, to prevent memory of trial order from the first condition priming responses in the second condition.

## 2.4 Results (adults)

### 2.4.1 AQ and SRS questionnaire measures

Average AQ and SRS scores are shown in Table 2.5. AQ data were normally distributed therefore scores were entered into a one-way ANOVA to test the effect of group. There was a statistically significant difference between groups on AQ score:  $F(2, 34) = 26.88, p < 0.001, \eta_p^2 = 0.61$  and each group differed significantly from each other, although the difference between DCD and TD would not survive a Bonferroni correction for multiple comparisons (see Table 2.6).

SRS scores were not normally distributed therefore nonparametric Kruskal-Wallis and Mann-Whitney U analyses were conducted. There was a significant effect of group on SRS scores ( $H(2) = 12.06, p = 0.002$ ). The TD group were reported to have significantly fewer autistic traits than both clinical groups:  $U = 10, p = 0.006$  for ASD and  $U = 12.5, p = 0.005$  for DCD. There was no significant difference between the two clinical groups:  $U = 16.5, p = 0.52$ .

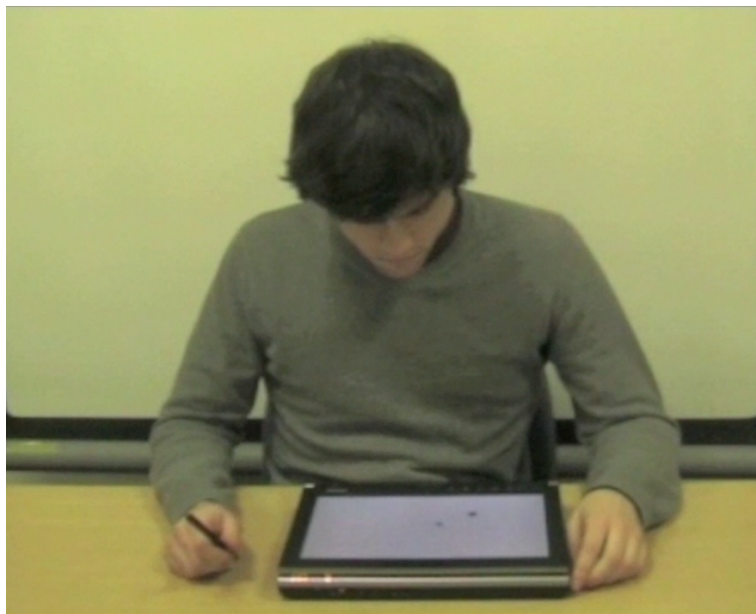
As would be expected, TD had significantly fewer autistic traits than ASD. The difference in the incidence of autistic traits in ASD and DCD was less apparent.

### 2.4.2 MABC-2

Due to differences in diagnostic history of subjects in the DCD group, it was decided that this group’s performance on the MABC-2 (the main test of motor skill) should



(a) Still from the actor condition: the actor is drawing a shape on the screen.



(b) Still from the dot condition: the actor is watching a dot move on the screen.

Figure 2.2: Stills from each condition in the imitation task

Table 2.5: Average scores for autistic trait measures

	AQ mean (SD)	SRS median (range)
TD	12.00 (4.89)	11 (0-55)
ASD	33.38 (7.67)	69.5 (16-119)
DCD	19.33 (9.95)	37 (26-109)

Figures are mean and SD for AQ scores which were normally distributed, and median and range for SRS scores which were not. AQ was available for all TD, 8/10 ASD and 9/17 DCD; SRS was available for 15/20 TD, 6/10 ASD and 7/17 DCD. A low score indicates fewer autistic traits.

Table 2.6: Results from post hoc analyses of AQ scores

	Mean difference (SE), $p$
ASD-DCD	14.04 (3.39), $p = 0.001$
ASD-TD	21.38 (2.92), $p < 0.001$
DCD-TD	7.33 (2.80), $p = 0.039$

Note the DCD-TD comparison would not survive a Bonferroni correction for multiple comparisons.

initially be investigated according to diagnostic history. This would aid in the decision as to whether to treat the childhood-diagnosis and adult-diagnosis subgroups as a single DCD group in further analyses.

Eight of the 17 subjects with DCD received a childhood diagnosis, while the remaining nine were diagnosed when they were adults (17+years) by the disability office. The procedure used by the disability office differs considerably from the typical childhood diagnostic procedure.

Data were not normally distributed therefore nonparametric analyses were used. The median percentile rank on the MABC-2<sup>4</sup> for those diagnosed as children was 37 (range=5-75), and 16 (range=5-91) for those diagnosed as adults. There was no significant difference between MABC-2 percentile ranks for the DCD subgroups ( $U = 31.5, p = 0.673$ ). For this reason all DCD adults will be included in analyses as a single group.

#### 2.4.2.1 Overall percentile rank

Preliminary analysis of MABC-2 scores was conducted by comparing overall percentile ranks between diagnostic groups. (Note these percentile ranks are based on test standardisation data, not data gathered in the present study.)

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<sup>4</sup>Note this is scored as 16 years, 11 months (the upper age limit in the MABC-2).

Data were not normally distributed therefore nonparametric analyses were used. Kruskal-Wallis analysis shows a significant effect of group on percentile rank:  $H(2) = 16.54, p < 0.001$ . This difference was further investigated using multiple Mann-Whitney U tests and it is noted when results would not survive a Bonferroni correction for multiple comparisons ( $\alpha = 0.017$ ). Performance on the MABC-2 did not differentiate ASD and DCD ( $U = 69, p = 0.418$ ). The main effect of group therefore was driven by significant differences in percentile rank between the two clinical groups and the control group:  $U = 36, p = 0.004$  and  $U = 48.5, p < 0.001$  for ASD vs. TD and DCD vs. TD respectively.

#### 2.4.2.2 Performance in each test component

As the two clinical groups were not differentiable by overall percentile on the MABC-2, analysis was re-run with the three percentile ranks for each component (MD, AC, and balance) to look for any underlying pattern of strengths and weaknesses. Percentile rank between groups for each MABC-2 component and total rank are shown in Figure 2.3.

Post hoc Mann-Whitney U were carried out to explore differences between groups in each MABC-2 component (see Table 2.7). The two clinical groups did not differ significantly on any measure. However the clinical groups did differ significantly from the TD group in the majority of components. TD significantly outperformed ASD in the AC component, and significantly outperformed DCD in all components, however the difference between TD and DCD in the balance component is not robust enough to survive correction for multiple comparisons.

A Friedman test for within-subject differences between the components for each group showed no differences in ASD or TD ( $X^2(2) = 4.67, p = 0.097$ ; and  $X^2(2) = 1.95, p = 0.377$ ). There was however a significant difference for DCD:  $X^2(2) = 8.09, p = 0.017$ . This group was relatively less impaired on the balance component compared to the AC component ( $Z = -2.39, p = 0.017$ ). There was no significant difference between AC and MD ( $Z = -1.37, p = 0.171$ ) or balance and MD ( $Z = -0.63, p = 0.530$ ).

Table 2.7: Mann-Whitney U analyses of group differences in each MABC-2 component

	MD	AC	Balance
ASD/TD	$U = 76, p = 0.307$	$U = 46, p = 0.017$	$U = 62.5, p = 0.100$
ASD/DCD	$U = 66, p = 0.359$	$U = 77.5, p = 0.711$	$U = 78, p = 0.749$
DCD/TD	$U = 74, p = 0.003$	$U = 76.5, p = 0.004$	$U = 103, p = 0.042$

#### 2.4.2.3 MABC-2 summary

The MABC-2 revealed a number of subjects with very poor motor skills in both clinical groups. The level of motor impairment in the DCD group was unrelated to diagnostic

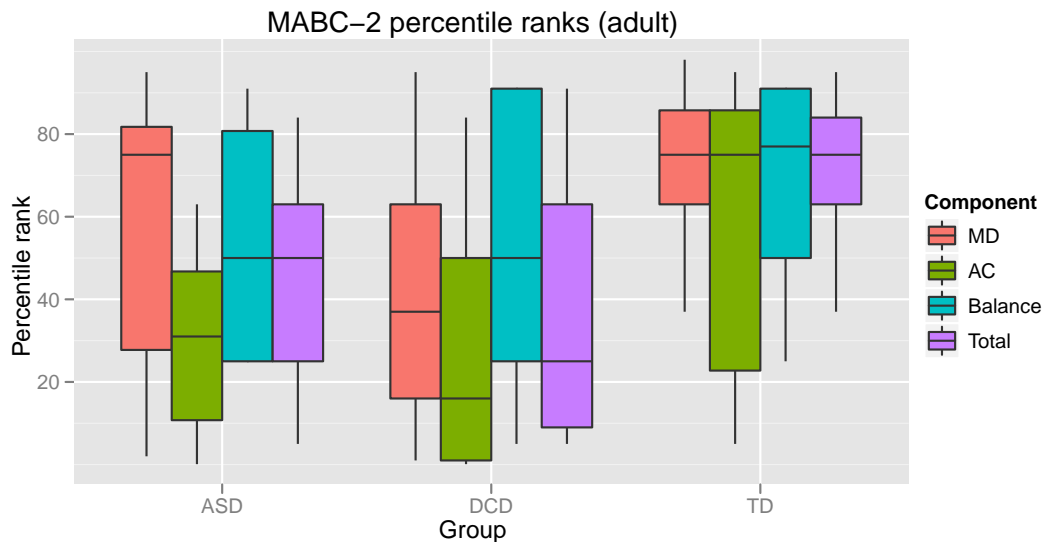


Figure 2.3: Spread of MABC-2 percentile ranks for each (adult) group. A total rank at or below the 15th percentile is outwith the typical range. Total scores for ASD and DCD are significantly worse than TD. Within the DCD group, the difference between AC and balance is significant.

route. Overall there was no significant difference in performance between ASD and DCD, although the median for each group was within the normal range, with only 20% of the ASD group and 29% of the DCD group failing the battery (<15%). If we consider each subscale, 50% of the ASD group and 53% of the DCD group fell at or below the 15th percentile for at least one subscale. Only the DCD group showed significant differences between the MABC-2 components, with poorer scores in the AC component compared to the balance component.

### 2.4.3 cKAT

Data for one ASD subject and 8 TD subjects for the pentagram and tracking tasks were not able to be used due to recording errors. Similarly, data for 6 TD subjects for the tracing task were not able to be used.

Measures used are described in Table 2.8. The majority of measures were normally distributed, therefore data were entered into a one-way ANOVA. There was no significant effect of group on any measure in any of the three tasks (see Table 2.9).

To directly assess the agreement between the two motor batteries, measures from the cKAT battery were correlated with the MABC-2 total rank using Pearson correlations (see Table 2.10). Once corrected for multiple comparisons there would be no significant correlation between any cKAT component and the MABC-2, supporting the nonsignificant effect of group on cKAT performance.

Table 2.8: cKAT variables

Measure	Definition
Reaction time (RT)	Time taken to begin moving towards a stimulus.
Movement time (MT)	Duration of the movement.
Peak Speed (PS)	Maximum velocity reached.
Deceleration time (DT)	Time from peak speed to the end of the movement.
Path length (PL)	Distance travelled throughout the movement.
Path accuracy (PA)	Accuracy of the movement compared to a reference path (the ideal movement).

The pentagram task uses all but PA, and tracking and tracing only use PA.

Table 2.9: cKAT group effects

	$F$ statistic
Pentagram RT	$F(2, 35) = 2.04, p = 0.146$
Pentagram MT	$F(2, 35) = 2.44, p = 0.102$
Pentagram DT	$F(2, 35) = 2.01, p = 0.149$
Pentagram PS	$F(2, 35) = 1.14, p = 0.333$
Pentagram PL	$F(2, 35) = 2.68, p = 0.082$
Tracking PA	$F(2, 35) = 0.16, p = 0.850$
Tracing PA	$F(2, 38) = 0.45, p = 0.638$

For pentagram and tracking  $n_{ASD} = 9$ ,  $n_{DCD} = 17$ ,  $n_{TD} = 12$ ; for tracing  $n_{ASD} = 10$ ,  $n_{DCD} = 17$ ,  $n_{TD} = 14$ .

Table 2.10: Pearson correlations between MABC-2 total score and each cKAT component

	Pearson correlation (uncorrected $p$ )
Pentagram RT	$r = -0.18(36), p = 0.268$
Pentagram MT	$r = -0.35(36), p = 0.032$
Pentagram DT	$r = -0.32(36), p = 0.052$
Pentagram PS	$r = 0.10(36), p = 0.547$
Pentagram PL	$r = -0.32(36), p = 0.049$
Tracking PA	$r = -0.32(36), p = 0.049$
Tracing PA	$r = -0.38(36), p = 0.014$

Note the  $p$ -values have not been corrected for multiple comparisons. No significant correlations survive multiple comparison correction.

### **2.4.3.1 cKAT summary**

The basic kinematic variables assessed using cKAT did not highlight any significant differences between groups. Correlations with the MABC-2 total percentile rank were weak and mostly non-significantly, suggesting that the cKAT battery is not suitable for identifying adults with general motor impairment.

## **2.4.4 Imitation**

### **2.4.4.1 Measures**

The imitation task was created for this study and is not part of the cKAT battery, although it does use the same software and hardware as cKAT. During stimulus recording, the coordinates of the pen on the screen were recorded using the cKAT software when the actor drew on the tablet. The PL and speed of the drawing were calculated from these coordinates. Subject responses were recorded in the same manner. This makes it possible to directly compare all components of the subject's and actor's<sup>5</sup> movement for each trial.

Three measures were compared between groups for PL (used here to indicate shape size) and speed in each condition: the correlation between stimuli and subject responses (to assess how closely a subject's response differs systematically according to the properties of the stimulus); constant error (signed error, calculated by subtracting model PL and speed from the subject's corresponding measures: this gives an indication of copying accuracy) and variable error (SD of errors: this gives an indication of copying precision).

### **2.4.4.2 Hypothesis**

As imitation requirements were made explicit ("copy the shape, size and speed") it is hypothesised that there will be no ASD-specific imitation deficit, in line with specific deficits in spontaneous imitation reported in previous literature. None of the dependent variables (subject/model correlation, constant error, or variable error) are expected to differ significantly between groups. Additionally it is hypothesised that there will be no differential effect of condition in the ASD group: the explicit instructions are expected to over-ride any motivational effect which would affect the more socially relevant actor condition. The two motor-impaired groups may have poorer imitation fidelity than TD, particularly in the speed component.

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<sup>5</sup>The dot condition uses the measures from the actor's movements to recreate the movement on screen, therefore a comparison between subject and dot is also possible.

### 2.4.4.3 Is motor output modulated by stimulus properties?

A correlation (Spearman's *rho*) between model and subject was calculated for each subject<sup>6</sup> for both conditions (dot and actor) separately, for both the PL (used to quantify shape size) and speed measures. These were then transformed using a Fisher's *r* to *z* transformation. Data were then entered into a 3x2x2 ANOVA, comparing group, measure and condition. Where the sphericity assumption is violated degrees of freedom are adjusted using a Greenhouse Geisser (GG) adjustment. (This is true for every ANOVA reported in this thesis.) Mean correlation coefficients are shown in Figure 2.4 (note data have been back-transformed for ease of interpretation).

There was no significant effect of group:  $F(2, 43) = 0.33, p = 0.721$ . Groups were not differentially affected by condition or measure ( $F(2, 43) = 0.96, p = 0.390$  (GG); and  $F(2, 43) = 2.17, p = 0.126$  (GG) respectively).

There was no significant main effect of condition:  $F(1, 43) = 0.66, p = 0.42$  (GG). There was however a significant main effect of measure, with PL modulated more closely than speed [ $F(1, 43) = 76.22, p < 0.001, \eta_p^2 = 0.64$ , mean difference= 0.27, SE= 0.03] and this difference was more apparent in the dot condition (condition\*measure interaction:  $F(1, 43) = 16.35, p < 0.001, \eta_p^2 = 0.28$ ). Means for this interaction effect are given in Table 2.11.

As hypothesised, there is no evidence for an ASD-specific imitation deficit, however contrary to previous findings of poor kinematic imitation, both end-goal (size) *and* style (speed) were modulated closely in both clinical groups.

Table 2.11: Mean correlation coefficients split by condition and measure

Condition	Measure	Mean (SE)
Dot	PL	1.39 (0.038)
	Speed	1.03 (0.046)
Actor	PL	1.27 (0.036)
	Speed	1.09 (0.044)

### 2.4.4.4 Constant error

Subject mean constant error for PL and speed were calculated for the dot and actor condition. Group mean errors are shown in Figure 2.5, which shows similar performance between groups. Mean constant errors were entered into a factorial analysis as outlined above. Again there was no effect of group:  $F(2, 43) = 0.52, p = 0.598$ . Additionally there was no significant effect of condition or measure, and no interaction effects (all  $p > 0.131$ , see Table A.1 in Appendix A).

<sup>6</sup>One TD subject was excluded from all imitation analyses due to recording errors. All other data were included.

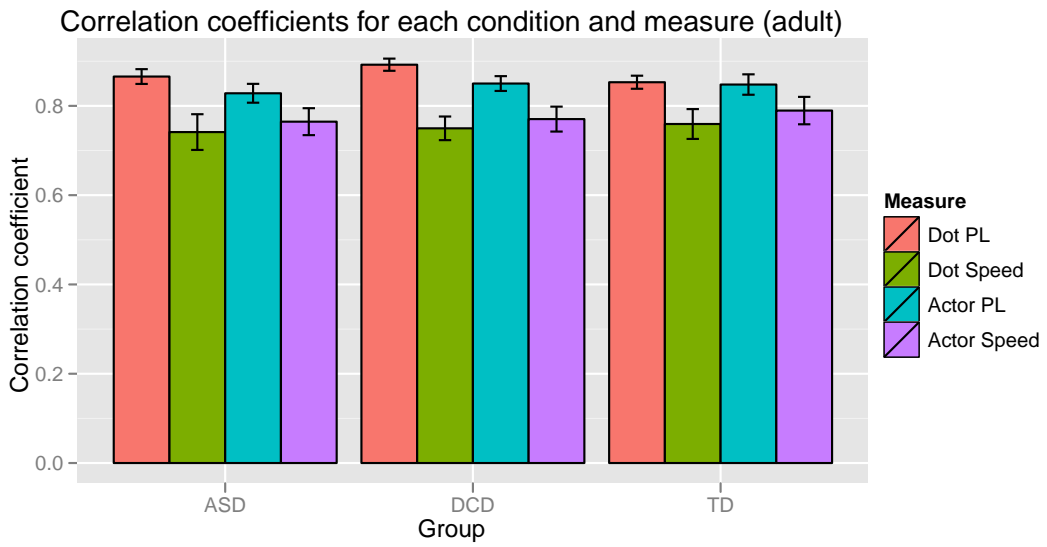


Figure 2.4: Mean correlation coefficients for each (adult) group across each imitation condition and measure. Error bars show SE. There is a significant condition\*measure interaction.

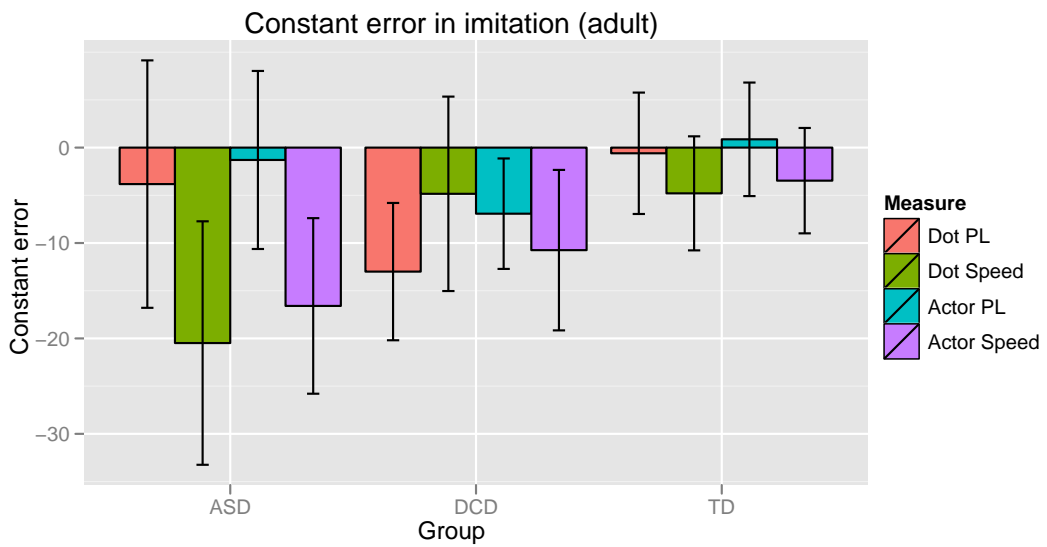


Figure 2.5: Mean constant error for each imitation condition and measure (adults). Error bars show SE. There is no significant effect of group, condition, or measure and no interaction effects.

#### 2.4.4.5 Variable error

Mean variable error for groups across each condition are shown in Figure 2.6. Variable error was analysed as above. As with accuracy, there was no significant effect of group on imitation precision:  $F(2, 43) = 0.31, p = 0.733$ . There was a significant interaction between condition and measure, with PL more variable than speed in the actor condition, and vice versa in the dot condition:  $F(1, 43) = 10.11, p = 0.003$  (GG). Means for each condition and measure are given in Table 2.12. All other within-subject and interaction effects were not significant (see Table A.2 in Appendix A).

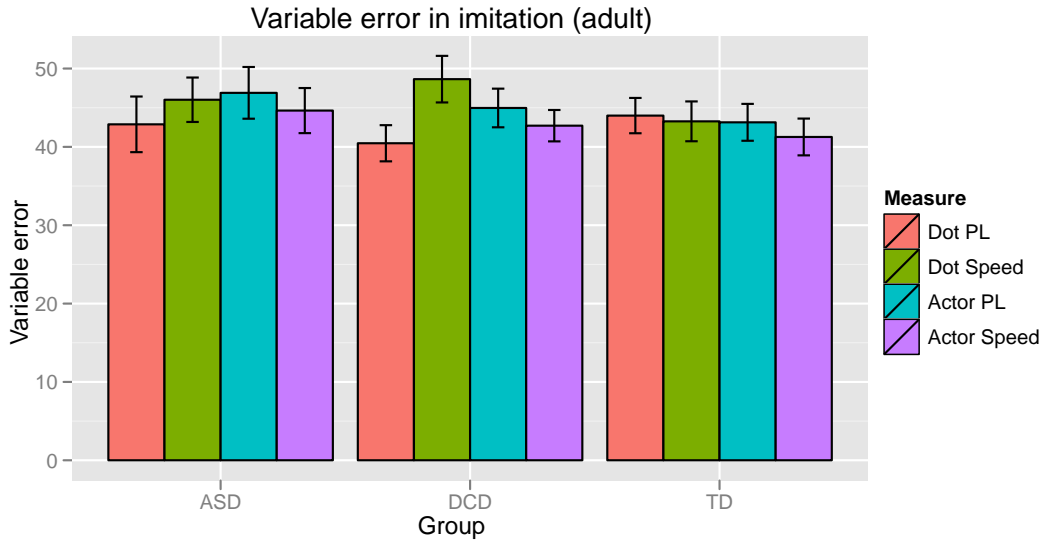


Figure 2.6: Mean variable error for each imitation condition and measure (adults). Error bars show SE. There is a significant condition\*measure interaction.

Table 2.12: Comparison of variable error for each condition and measure

Condition	Measure	Mean (SE)
Dot	PL	42.45 (1.54)
	Speed	45.97 (1.71)
Actor	PL	45 (1.57)
	Speed	1.09 (0.04)

#### 2.4.4.6 Imitation summary

As hypothesised, there was no evidence for an ASD-specific imitation deficit, and no selective difficulty in the ASD group for the actor condition. Motor difficulties did not affect imitative ability in either ASD or DCD.

## 2.5 Discussion (adults)

Adults with ASD, DCD and TD controls completed a number of tasks assessing various types of visuomotor skill. These included a standardised motor battery (MABC-2), an unstandardised computerised test of visuomotor skill (cKAT) and a computerised imitation task.

Performance on the MABC-2 revealed significant differences between TD and both clinical groups, however there was little difference between the two clinical groups, supporting Green et al. (2009). Within the clinical groups, aiming and catching was significantly poorer than balance for the DCD group, however no other points of difference were found. It is possible that similar patterns in the clinical groups were an artefact of relatively small sample sizes, combined with the heterogeneity of both disorders (Zoia et al., 2006; Bhat, Landa & Galloway, 2011). The use of a children's motor skills battery in adults has obvious limitations. Scores are usually standardised according to age, however the MABC-2 is only standardised up to the age of 16 years 11 months. However, despite this imprecise score standardisation, MABC-2 results did highlight below average motor skills in the majority of the DCD group and also significantly differentiated TD and DCD, in line with similar previous research (Borreman et al., 2009; Cousins & Smyth, 2003). While a number of subjects did perform outwith the normal range, there were exceptions, and these might be explained by the heterogeneous nature of DCD. Kirby et al. (2008) suggest that organisational skills (not assessed in the MABC-2) may be more problematic in adults with DCD, and will be more prominent than motor difficulties typically associated with DCD in childhood.<sup>7</sup> The similarity in MABC-2 percentile rank for the DCD subjects diagnosed as children compared to those with adult diagnoses provides support both for the assertion that DCD is a diagnosis that persists into adulthood (Kirby et al., 2008; Hill & Barnett, 2011) and also the argument that a diagnostic procedure for DCD in adults should be formulated. Although the diagnostic history of the DCD group was less controlled than the ASD group, the recruitment process is in line with previous research into DCD in adults (Cousins & Smyth, 2003). In the case of adults with motor difficulties, the cKAT battery is not a sufficient test of more fundamental aspects of movement. It should be noted that the three adults with the lowest scores on the MABC-2 manual dexterity subscale (which is presumably most similar to the cKAT) did not show similarly poor performance on the cKAT battery. The null effect here is not thought to be due to low power or insufficient sample size as the difference between groups did not approach significance for any measure, and the battery showed only weak and marginally significant

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<sup>7</sup>One subject in the present study was identified as having a 'dyspraxic profile' due to organisational and time management difficulties, and scored on the 91st percentile on the MABC. His profile does not seem to fit diagnostic criteria for DCD or dyspraxia set out in the DSM or ICD, although these criteria describe DCD as a childhood disorder and tend to focus on motor difficulties. The inclusion of this subject did not affect overall findings.

correlations with MABC-2 performance.

In addition to the two motor batteries, subjects also completed a computerised motor imitation task, which required subjects to copy the shape, size and speed of geometric shapes drawn by a person, or reproduced by the computer. As expected, there did not appear to be a deficit in the ASD group. Intact ability to make the required visuospatial and visuomotor transformations and similar kinematic output suggests that there is no clear imitation deficit and poor motor skills appear not to have adversely affected imitation skill. It is possible that previous imitation ‘deficits’ reported in the literature may be the product of socially-loaded tasks or a lack of motivation when there is no clear goal or reason to imitate. The use of video has likely been beneficial in this situation, as it removes the need for subject-experimenter interaction, and has previously been found to be more successful than live demonstrations (Lindsay, Moore, Anderson & Dillenburger, 2013). The nature of the actor video, in which he plays no social role, may also have positively affected the ASD group’s ability to imitate: the actor is clearly important, but his presence is not socially demanding. The ASD group’s comparable ability in both imitation conditions suggests that the translation of another’s movement into egocentric coordinates to enable imitation of the action is intact.

Superficially, the present findings do not sit well with previous reports of imitation deficits in adults with ASD (Beall, Moody, McIntosh, Hepburn & Reed, 2008; Leighton, Bird, Charman & Heyes, 2008). However, the methodology of the present study is not one that has been used previously. The design of the present study allowed for precise control of the stimuli, with kinematics recorded for each individual trial. The use of the kinematic properties of the actor’s movement in the dot condition results in a paradigm whereby it is possible to compare two movements that are kinematically identical, yet defined differently. By making explicit reference to the variables to be copied (shape, size and speed), the problem of spontaneously imitating style, previously found to be impaired (Wild et al., 2012), is negated.

The null result is again not thought to be a product of low statistical power, as it is clear from an inspection of the mean errors produced by each group across conditions that the pattern is strikingly similar between all three groups. Of course, the role of age should be considered as the use of adults in imitation tasks is relatively unusual, and age might explain the null effect of group. An alternative interpretation of the results is that imitation ‘deficits’ previously reported in children and adolescents are not in fact deficits (suggesting lack of ability) but rather a *delay* in the ability to imitate. The two could also interact, with a delay in the ability to imitate being tied to the delay in the social understanding that imitating in a game is worthwhile, even if the child does not spontaneously wish to engage in imitation. Results from the child imitation study reported in the second half of this chapter will provide further evidence to consider the role of age and the question of delay or deficit.

This study intended to build a profile of motor skills in both adults and children with ASD and DCD. The adult study has verified the presence of motor deficits in adults with ASD, which has received comparatively little attention compared to children with ASD. It has also found performance by adults with ASD and DCD to be similar on all tasks. The second half of this chapter reports findings from the profiling procedure completed by groups of children with ASD, DCD and TD. The chapter is concluded with a brief general discussion of the results from both the adult and child profiling studies.

## 2.6 Subjects (children)

Children with a diagnosis of either ASD ( $n=33$ ) or DCD ( $n=10$ ) and TD children ( $n=28$ ) were recruited to take part in the profiling study. All children were between 6 and 15 years old. Children in the TD group were aged 9-11 years (mean age 10.14,  $SD=0.85$ ), the ASD group ranged from 6 to 15 (mean 10.82,  $SD=1.91$ ) and the DCD group from 7 to 14 (mean=9.9,  $SD=2.28$ ). Groups were age and IQ matched ( $H(2) = 3.51, p = 0.173$  and  $F(2, 68) = 0.50, p = 0.608$  respectively). All but two children (one with ASD and one with DCD) were male. Some children were not able, or chose not to complete all tasks in the profiling battery: there were no significant changes to demographic information between groups as a result.

Following ethical approval from the NHS and City of Edinburgh council, children in both clinical groups were recruited through NHS Lothian (which encompasses Edinburgh, East Lothian and West Lothian), a local special school and a local mainstream primary school. All children in clinical groups had a confirmed diagnosis of either ASD or DCD (or in the latter case had been referred to paediatric services for motor difficulties, had failed the MABC (or MABC-2) <15th percentile and were eligible for a diagnosis). These diagnoses were verified either by a clinician at the Edinburgh Royal Hospital for Sick Children, or by the school who have records of medical diagnoses given by a multidisciplinary team of medical practitioners. TD children were recruited through a local primary school.

All parents gave written consent on behalf of their child. Parents of children in clinical groups also gave permission for their child's medical records to be accessed. Children were tested either in their school, in the Psychology Department at Edinburgh University or at St Johns Hospital in Livingston, West Lothian. Children were not reimbursed for participation, although travel expenses were offered to parents of children tested outside of school.

## 2.7 Results (children)

Note that the  $\alpha$  level for post hoc analyses was not corrected to account for multiple comparisons. All post hoc results in child studies will give exact (uncorrected)  $p$ -values

although it will be noted when these results would fail to reach statistical significance were a Bonferroni correction implemented. This approach is used to give the results greater transparency. This is true for all analyses detailed in this thesis from this point.

### 2.7.1 SRS and DCDQ-07 questionnaire measures

Data were not normally distributed therefore groups were compared using Mann-Whitney U tests. Median SRS and DCDQ-07 scores are shown in Table 2.13. ASD scored significantly higher on the SRS (more ASD symptoms) than both DCD ( $U = 29.5, p < 0.001$ ) and TD ( $U = 5, p < 0.001$ ). DCD scored significantly higher than TD ( $U = 18.5, p < 0.001$ ).

The DCDQ-07 also differentiated TD from both ASD and DCD ( $U = 14, p < 0.001$ ; and  $U < 0.001, p < 0.001$  respectively), with the clinical groups' scores tending to be in the clinical range. Scores in the two clinical groups were not significantly different ( $U = 98.5, p = 0.375$ ).

As expected, autistic traits and motor difficulties were evident in both clinical groups. Note that all findings remain when considering only children who completed every task (MABC, cKAT and imitation).

Table 2.13: Median SRS and DCDQ scores for each group (including any child who successfully completed at least one battery)

	SRS median (range, $n$ )	DCDQ-07 median (range, $n$ )
TD	12 (0-52, $n=28$ )	72.5 (47-75, $n=28$ )
ASD	117 (38-153, $n=29$ )	32 (21-65, $n=31$ )
DCD	59 (25-103, $n=9$ )	34 (18-46, $n=8$ )

A high score on the SRS indicates more pronounced difficulties and the inverse is true for the DCDQ. Clinical range for DCDQ-07 is: 15-46 (5-7 years); 15-55 (8-9 years); 15-57 (10-15 years). Clinical range for SRS raw scores is 70+ for males and 65+ for females.

### 2.7.2 MABC-2

Performance on the MABC-2 was first considered at the highest level, comparing pass/fail rates between the three groups (see Figure 2.7). Seventy percent of each clinical group failed the battery, while only 14% of the TD group failed. Given the 15%ile cut-off for the battery, this failure rate would be expected in TD. It should be noted that not every child who failed the battery overall failed every component: only two DCD and six ASD failed every component. MABC-2 percentile ranks for each component and total percentile rank are shown in Figure 2.8. From an initial inspection of the spread of percentile ranks it appears that there is little difference between test components within each clinical group, however children in the TD group seem to

have relatively poorer scores in the manual dexterity component compared to the two gross motor components.

MABC-2 percentile ranks were not normally distributed, therefore nonparametric analyses were performed. There was no significant difference between the two clinical groups on any measure (see Table 2.14). The ASD group was significantly worse than TD on total score, AC and balance ( $U = 136.5, p < 0.001$ ;  $U = 107, p < 0.001$ ;  $U = 151, p < 0.001$  respectively). The same was true for the DCD group relative to TD ( $U = 31, p < 0.001$ ;  $U = 60.5, p = 0.007$ ;  $U = 48.5, p = 0.002$  respectively). Neither clinical group were significantly poorer than TD in the manual dexterity component:  $U = 350, p = 0.103$  (ASD) and  $U = 90, p = 0.101$  (DCD).

In TD, performance in the manual dexterity component was significantly worse than both AC and balance ( $Z = -3.62, p < 0.001$ ; and  $Z = -3.46, p < 0.001$ ). The difference between balance and AC was not significant:  $Z = -1.24, p = 0.216$ . There were no significant differences between component scores for either clinical group (see Table 2.15).

Table 2.14: Analysis of MABC-2 percentile rank differences in ASD and DCD

	Mann-Whitney U result
Total percentile	$U = 159.5, p = 0.873$
MD percentile	$U = 134.5, p = 0.378$
AC percentile	$U = 129, p = 0.298$
Balance percentile	$U = 162, p = 0.931$

To investigate previous reports of a positive association between IQ and motor ability in ASD (Green et al., 2009), IQ standard scores were correlated with total MABC-2 rank. It was found that those children with ASD with lower IQ tended to exhibit more serious motor deficits:  $r_s = 0.57, p < 0.001$ .

### 2.7.3 MABC-2 summary

An equal proportion of children with ASD and DCD failed the battery, and neither total score nor component scores differentiated the two clinical groups. Performance in the

Table 2.15: Comparison of MABC components for each clinical group

Group	MABC component comparison	Result
ASD	AC-MD	$Z = -0.82, p = 0.412$
	Balance-MD	$Z = -0.93, p = 0.354$
	Balance-AC	$Z = -0.43, p = 0.666$
DCD	AC-MD	$Z = -0.85, p = 0.398$
	Balance-MD	$Z = 0.06, p = 0.953$
	Balance-AC	$Z = -1.18, p = 0.237$

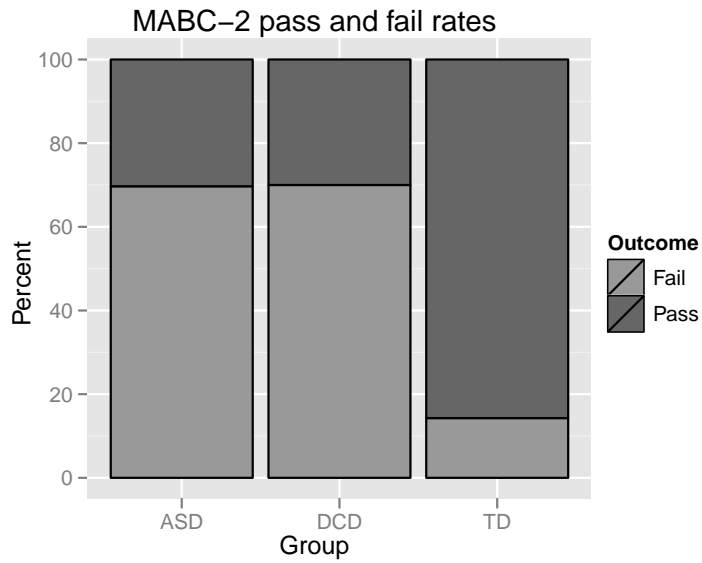


Figure 2.7: Percentage of children in each group passing and failing the MABC

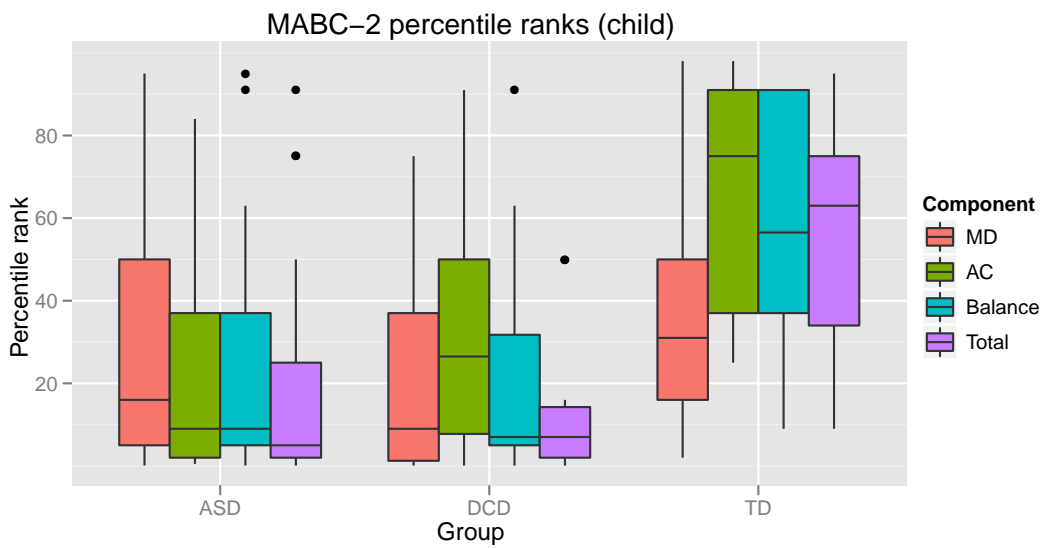


Figure 2.8: Spread of MABC-2 percentile ranks for each (child) group. MD=manual dexterity, AC=Aiming and catching. A total rank at or below the 15th percentile is outwith the typical range. TD scores are significantly higher than both ASD and DCD for all but MD. In TD scores in the MD component were significantly lower than AC and balance.

ASD and DCD groups was significantly worse than the TD group. The only measure in which the clinical groups were not significantly worse than TD was MD. There was no clear differential pattern among components in the clinical groups, although within the TD group children were significantly less proficient in MD skills relative to AC and balance.

#### 2.7.4 cKAT

Several children from clinical groups lost interest before the end of the battery and data from the final task (tracing) has been removed. One child in the ASD group chose not to complete any of the cKAT battery.

Scores from each task were entered into a one-way ANOVA to investigate the effect of group. There was a significant effect of group on tracking PA:  $F(2, 63) = 3.97, p = 0.024, \eta_p^2 = 0.11$ , however all other scores were equivalent between groups (see Table A.3 in Appendix A). Tracking accuracy was less accurate in ASD compared to TD: mean difference=0.34, SE=0.16,  $p = 0.034$ ; similarly DCD were also less accurate than TD: mean difference=0.53, SE=0.24,  $p = 0.029$ . Note that these would not survive a Bonferroni correction for multiple comparisons ( $\alpha = 0.017$ ).

As per the adult study, MABC-2 percentile rank was correlated with each cKAT measure. There were moderate correlations between MABC-2 and pentagram RT, tracking PA and tracing PA (see Table 2.17).

As more than two thirds of the ASD group failed the MABC-2, it was decided to split the ASD group according to movement ability and re-run the factorial analysis described above. First, it was confirmed that this was a viable option by comparing the DCD group to those children with ASD who failed the MABC-2. There was no significant difference between motor impaired ASD and DCD children on any measure (see Table 2.16). For this reason, those ASD children who failed the MABC-2 (<15th percentile) were placed into a group alongside all DCD children. (Although three children in the DCD group passed the battery they had all previously had motor impairment verified by a relevant healthcare professional, therefore no child was removed from the DCD group.) This new group is termed the ‘*clinical motor deficit*’ group. Those children with ASD who passed the MABC-2 were placed into a group termed the ‘*ASD pure*’ group. The TD group remains unchanged. This regrouping will be carried out throughout the thesis alongside analyses using diagnostic groups.

Comparing MABC-defined groups there was a significant effect of group on Pentagram RT:  $F(2, 62) = 6.08, p = 0.004, \eta_p^2 = 0.16$ . Group was also significantly different for path accuracy in both tracking and tracing tasks:  $F(2, 60) = 8.54, p = 0.001, \eta_p^2 = 0.22$ ;  $F(2, 60) = 5.13, p = 0.009, \eta_p^2 = 0.15$ . Non-significant results for the remaining measures are shown in Table A.4 in Appendix A.

Table 2.16: A comparison of children with ASD who failed the MABC-2 and children with DCD on each cKAT measure

	Statistic
PentRT	$t(30) = -0.28, p = 0.785$
PentMT	$t(30) = 0.54, p = 0.595$
PentDT	$t(30) = 0.91, p = 0.369$
PentPS	$t(30) = -1.26, p = 0.217$
PentPL	$t(30) = -0.23, p = 0.821$
Track PA	$t(28) = -0.01, p = 0.991$
Trace PA	$t(27) = 0.05, p = 0.965$

Post hoc analyses are summarised in Table 2.18. They show that the clinical motor deficit group had a significantly slower RT in the pentagram task than both ASD pure and TD (mean difference=0.06s, SE=0.02,  $p = 0.002$ ; and mean difference=0.03s, SE=0.15,  $p = 0.026$ ), however the comparison with TD would not survive correction for multiple comparisons. Additionally the differences are less than one tenth of a second in each case so are of a relatively small value given the tasks take approximately 75 seconds to complete. There was no significant difference between ASD pure and TD (mean difference (ASD pure-TD)=0.03s, SE=0.02,  $p = 0.154$ ). Differences in tracking accuracy were driven by significantly less accurate tracking in the clinical motor deficit group compared to ASD pure and TD: mean difference=0.64, SE=0.21,  $p = 0.003$ ; and mean difference=0.53, SE=0.15,  $p = 0.001$ . There was no significant difference between ASD pure and TD: mean difference (ASD pure-TD)=-0.11, SE=0.21,  $p=0.596$ . In tracing accuracy, the motor deficit group was less accurate than TD and ASD pure, however the latter difference would not survive correction for multiple comparisons: mean difference=0.33, SE=0.11,  $p = 0.005$ ; and mean difference=0.32, SE=0.15,  $p=0.033$  (both uncorrected). These differences are in line with the MABC-2/cKAT correlations detailed above. Accuracy in ASD pure and TD groups was not significantly different: mean difference (ASD pure-TD)=0.01, SE=0.15,  $p=0.971$ .

Table 2.17: Spearman correlations for MABC-2 percentile rank and each cKAT measure

	Spearman's result (uncorrected $p$ )
MABC/pentagram RT	$r_s(67) = -0.46, p < 0.001$
MABC/pentagram MT	$r_s(67) = -0.31, p = 0.010$
MABC/pentagram DT	$r_s(67) = -0.22, p = 0.078$
MABC/pentagram PS	$r_s(67) = 0.06, p = 0.650$
MABC/pentagram PL	$r_s(67) = -0.30, p = 0.014$
MABC/tracking PA	$r_s(64) = -0.49, p < 0.001$
MABC/tracing PA	$r_s(64) = -0.48, p < 0.001$

Table 2.18: Significant post hoc comparison findings for cKAT tasks

Task (measure)	Post hoc finding
Pentagram (RT)	Clinical motor deficit significantly slower than ASD pure.
Tracking (accuracy)	Clinical motor deficit significantly less accurate than ASD pure and TD.
Tracing (accuracy)	Clinical motor deficit significantly less accurate than TD.

### 2.7.5 cKAT summary

The pentagram task was largely unsuccessful in identifying motor difficulties. Ability in tracking and tracing tasks was more reliably related to MABC-2 performance and diagnostic group, however correlations with the MABC-2 were weak and differences between groups were of a relatively small magnitude.

### 2.7.6 Imitation

Unlike the MABC-2 and cKAT, the purpose of the imitation task is not to identify children with motor impairment, but to compare groups. For this reason children in the TD group who failed the MABC-2 are not included in the analysis reported below, as it was not clear if they would fit criteria for DCD if a diagnosis was sought. The exclusion of these children had no effect on the main results of any of the analyses reported. Two children with ASD chose not to complete the imitation task.

#### 2.7.6.1 Is motor output modulated by stimulus properties?

Spearman's *rho* correlations were calculated for each subject and were Fisher's *r* to *z* transformed as described previously for adults. These data were entered into a 3x2x2 ANOVA, comparing group on measure (PL/speed) and condition (actor/dot). Mean correlation coefficients are shown in Figure 2.9 (note these data have been back-transformed).

There was a significant effect of group ( $F(2, 62) = 4.83, p = 0.011, \eta_p^2 = 0.14$ ), with significantly greater subject/model correlations in the TD group compared to both ASD and DCD [mean difference=0.20, SE=0.08,  $p = 0.012$ ; and mean difference=0.28, SE=0.11,  $p = 0.011$  for ASD and DCD respectively]. There was no significant difference between ASD and DCD: mean difference (ASD-DCD)=0.08, SE=0.10,  $p = 0.426$ .

There was a significant effect of measure, with response and model better correlated for PL than speed:  $F(1, 62) = 87.35, p < 0.001, \eta_p^2 = 0.59$  (GG), mean difference=0.25, SE=0.03. There was also a significant interaction between condition and measure:  $F(1, 62) = 6.84, p = 0.011, \eta_p^2 = 0.10$  (GG) (see Table 2.19). Speed tended to be less well modulated than PL, particularly in the dot condition, mirroring findings in the

Table 2.19: Mean (SE) z-transformed correlation coefficients for each condition and measure

Measure	Condition	Mean (SE)
PL	Dot	1.00 (0.05)
	Actor	0.88 (0.04)
Speed	Dot	0.70 (0.05)
	Actor	0.70 (0.04)

adult study. The difference between measures was less pronounced in the actor condition. All other main effects and interaction effects were not statistically significant (see Table A.5 in Appendix A).

#### Analysis using MABC-defined groups

As with cKAT, analysis was re-run with redefined groups (clinical motor deficit, ASD pure, and TD). Non-significant differences between ASD < 15% on the MABC-2 and DCD confirmed that entering all of these children into a clinical motor deficit group was feasible (see Table A.6 in Appendix A). Mean correlation coefficients are shown in Figure 2.10. The effect of measure and the condition\*measure interaction follow the same pattern as above. Modulation was better for PL than speed:  $F(1, 62) = 101.17, p < 0.001, \eta_p^2 = 0.62$ , mean difference=0.26, SE=0.03. The condition\*measure interaction ( $F(1, 62) = 6.43, p = 0.014, \eta_p^2 = 0.09$ ) highlights that modulation of PL was more sensitive to condition than the modulation of speed, which seemed to be modulated similarly regardless of condition (see Table 2.20). Again there was no significant main effect of condition:  $F(1, 62) = 0.52, p = 0.474, \eta_p^2 = 0.01$ .

Again, there was a significant effect of group ( $F(2, 62) = 7.60, p = 0.001, \eta_p^2 = 0.20$ ), driven by significantly better modulation in TD compared to the clinical motor deficit group: mean difference=0.27, SE=0.07,  $p < 0.001$ . There was no significant difference between ASD subjects with spared motor abilities (ASD pure) and TD: mean difference (ASD-TD)= -0.04, SE=0.10,  $p = 0.678$ . Modulation was better in ASD pure compared to the clinical motor deficit group (mean difference=0.23, SE=0.10,  $p = 0.022$ ), however this difference would fall just outside the 5%  $\alpha$  criterion if corrected for multiple comparisons using a Bonferroni adjustment ( $\alpha = 0.017$ ). As with the previous analysis using diagnostic groups, there was no significant interaction between group and either measure or condition:  $F(2, 62) = 1.40, p = 0.255$  and  $F(2, 62) = 0.08, p = 0.927$ .

These results suggest that those with ASD without pronounced motor difficulties do not show a reliable deficit in their ability to modulate motor output according to specific stimulus properties.

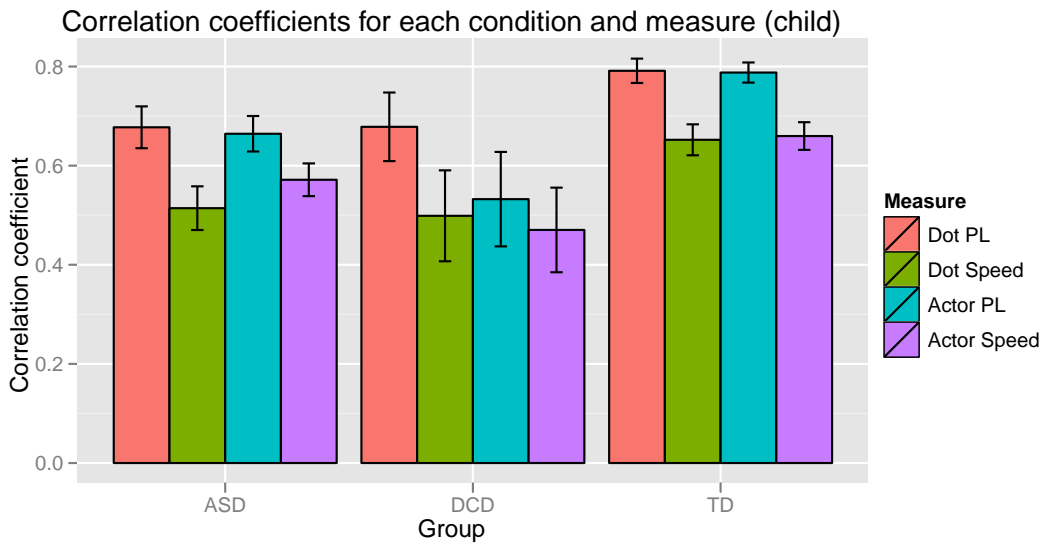


Figure 2.9: Mean correlation coefficients for each condition in the imitation task between diagnosis-defined groups. Error bars show SE. Coefficients in ASD and DCD are significantly lower than TD, and there is a significant condition\*measure interaction.

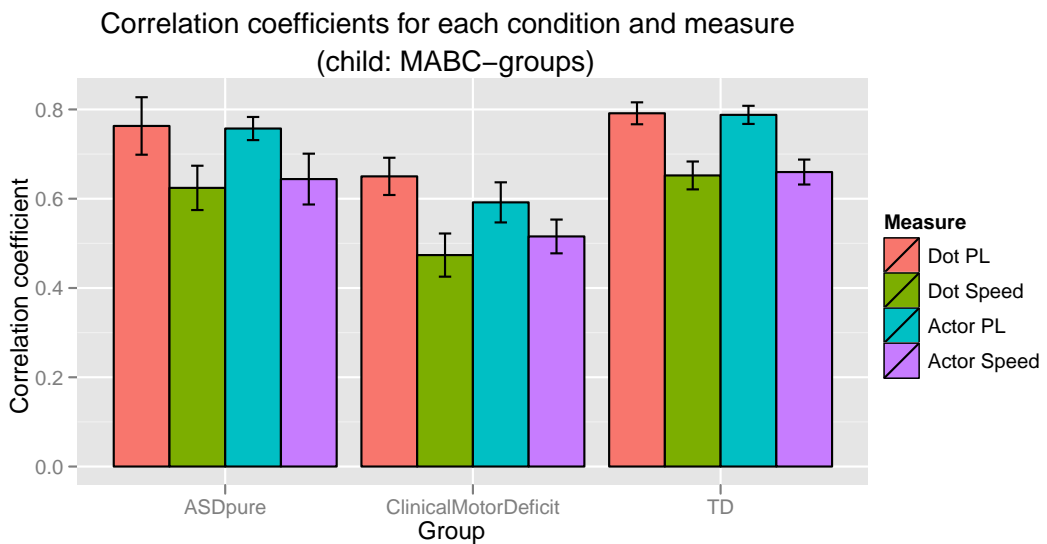


Figure 2.10: Mean correlation coefficients for each condition in the imitation task with groups split according to MABC-2 performance. Error bars show SE. Coefficients in the clinical motor deficit group are significantly lower than TD.

Table 2.20: Mean (SE) z-transformed correlation coefficients for each condition and measure (MABC-2-defined groups)

Measure	Condition	Mean (SE)
PL	Dot	1.05 (0.05)
	Actor	0.96 (0.04)
Speed	Dot	0.72 (0.05)
	Actor	0.76 (0.04)

### 2.7.6.2 Constant error

Median constant error<sup>8</sup> (subject PL and speed minus model PL and speed) was calculated for each subject to give an indication of accuracy, and was entered into a 3x2x2 ANOVA as outlined above. Constant errors are shown in Figure 2.11.

Constant error did not differ significantly between groups:  $F(2, 62) = 1.31, p = 0.278$ . However, there was a significant interaction between group and measure:  $F(2, 62) = 3.14, p = 0.05, \eta_p^2 = 0.09$  (GG), likely due to the DCD group being noticeably more accurate in copying speed compared to copying PL. The same is also true for the TD group, while accuracy in the ASD group did not seem to differ greatly between PL and speed (see Table 2.21). All other main effects and interaction effects were not statistically significant (see Table A.7 in Appendix A).

#### Analysis using MABC-defined groups

Again, DCD and motor-impaired ASD did not differ significantly (see Table A.8 in Appendix A), therefore these children were entered into a single ‘clinical motor deficit’ group as described previously. Mean constant error for MABC-defined groups are shown in Figure 2.12. Again there was no significant effect of group on constant error ( $F(2, 62) = 0.93, p = 0.401$ ). In this case there was no group\*measure interaction ( $F(2, 62) = 1.45, p = 0.242$ ). All other non-significant main and interaction effects are given in Table A.9 in Appendix A.

Table 2.21: Group\*Measure mean (SE) for constant error

Group	Measure	Mean (SE)
ASD	PL	-13.47 (6.98)
	Speed	-11.47 (5.73)
DCD	PL	13.66 (12.29)
	Speed	-4.28 (10.08)
TD	PL	-19.21 (7.93)
	Speed	-8.67 (6.51)

<sup>8</sup>Median was used to minimise the effect of any outliers due to technical difficulties with the software (these problems were less apparent in the adult study in which the mean was used).

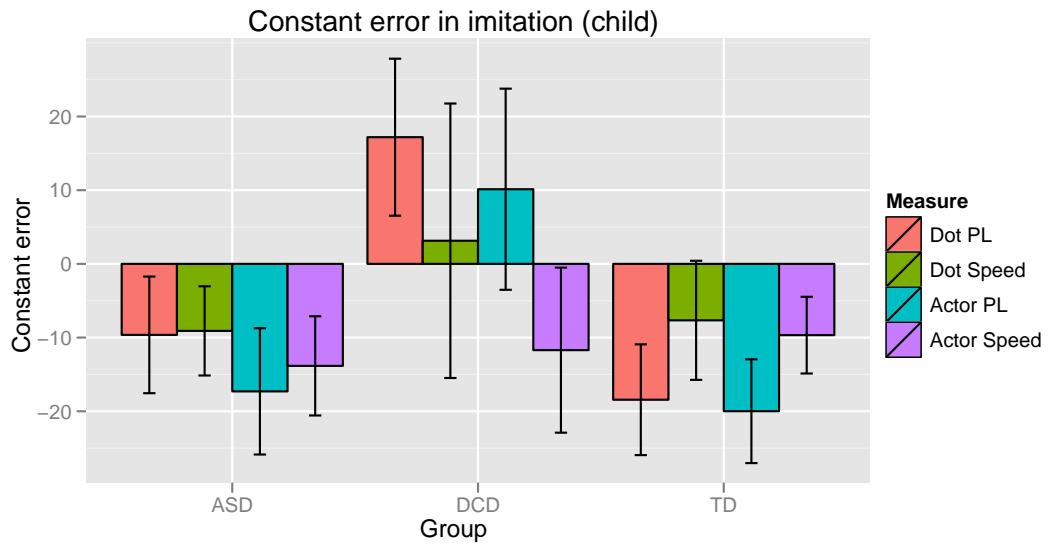


Figure 2.11: Median constant error across each condition and the three diagnostic groups. Error bars show SE. There is a significant group\*measure interaction.

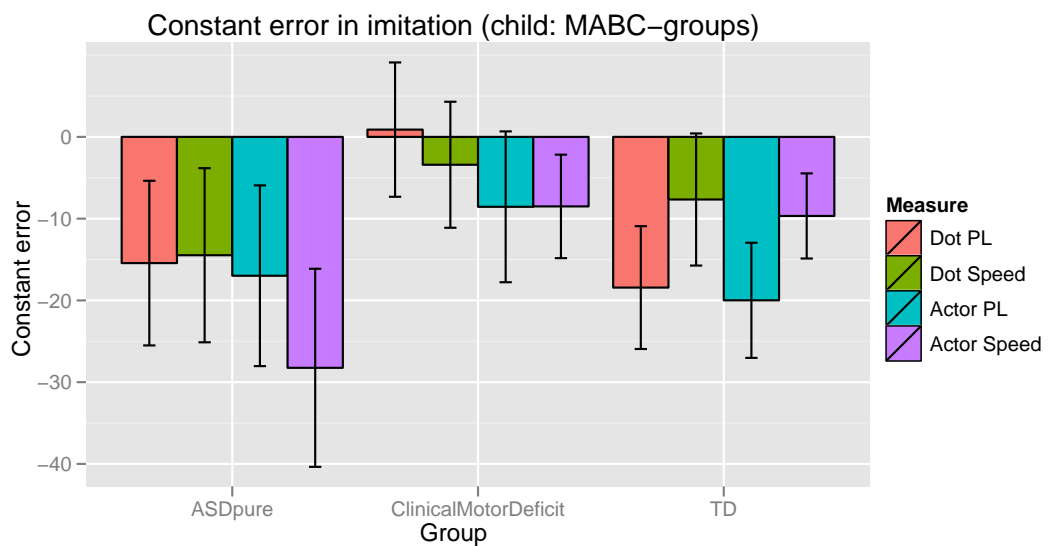


Figure 2.12: Median constant error across each condition and the MABC-defined groups. Error bars show SE. There are no significant effects.

### 2.7.6.3 Variable error

In order to assess precision, the standard deviation of PL and speed was calculated for each subject in each condition. Mean variable error is shown in Figure 2.13.

Diagnostic groups differed significantly ( $F(2, 62) = 4.81, p = 0.011, \eta_p^2 = 0.13$ ), with the TD group producing significantly less variable responses than both ASD and DCD: mean difference=9.07, SE=3.92,  $p = 0.024$ ; and mean difference=15.38, SE=5.43,  $p = 0.006$ . (Note that the difference between TD and ASD is not robust enough to survive a Bonferroni correction.)

Within-groups there was a significant condition\*measure interaction:  $F(1, 62) = 14.53, p < 0.001, \eta_p^2 = 0.19$ . As with the adult study, action reproduction in the dot condition tended to be more variable when copying speed compared to PL and this pattern was reversed in the actor condition (see Table 2.22). All other main effects and interaction effects were not statistically significant (see Table A.10 in Appendix A).

#### Analysis using MABC-defined groups

Again, DCD and motor-impaired ASD did not differ significantly (see Table A.11 in Appendix A), therefore these children were entered into a single ‘clinical motor deficit’ group. When ASD pure, clinical motor deficit and TD were compared, the main effect of group remained:  $F(1, 62) = 8.14, p = 0.001, \eta_p^2 = 0.12$  (see Figure 2.14). The motor deficit group was significantly more variable than both the TD group (mean difference=13.92, SE=3.75,  $p < 0.001$ ) and the ASD pure group (mean difference=13.59, SE=5.56,  $p = 0.017$ ). Precision in the ASD pure and TD group was not significantly different [mean difference (ASD pure-TD)=-0.33, SE=5.19,  $p = 0.950$ ]. The condition\*measure interaction also remained ( $F(1, 62) = 12.90, p = 0.001, \eta_p^2 = 0.17$ ). No other main effects or interaction effects were statistically significant (see Table A.12 in Appendix A).

Table 2.22: Condition\*Measure mean (SE) for variable error

Condition	Measure	Mean (SE)
Dot	PL	59.66 (2.64)
	Speed	62.54 (2.79)
Actor	PL	63.95 (2.13)
	Speed	58.67 (2.10)

### 2.7.6.4 Imitation summary

Children with ASD with spared motor skills appear to have an intact ability to imitate at a level similar to TD children. Those with motor difficulties perform at a level comparable to DCD, suggesting a visuomotor deficit rather than an imitation deficit per se.

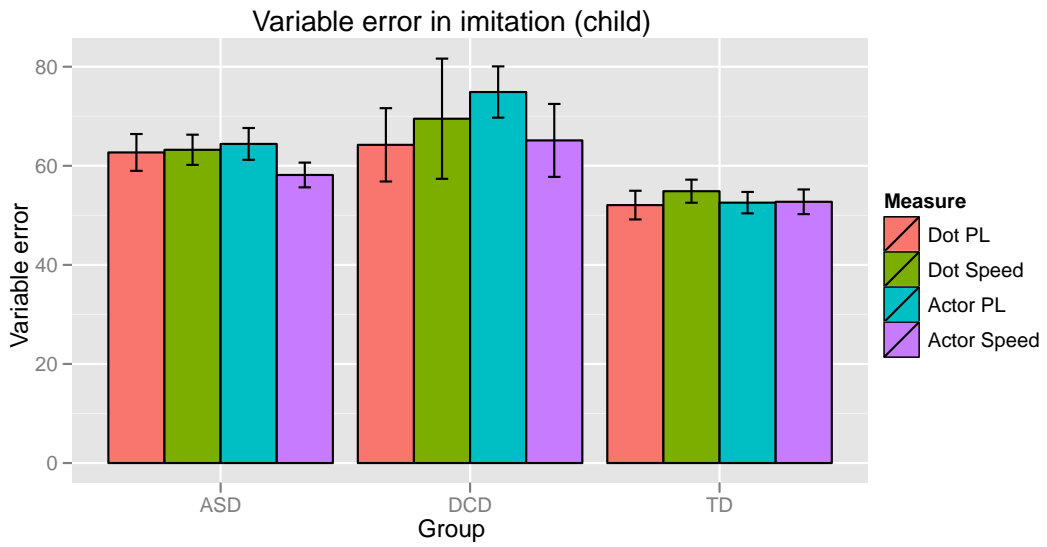


Figure 2.13: Variable error across each condition between diagnosis-defined groups. Error bars show SE. Variable error in the DCD group is significantly higher than ASD and TD. There is also a significant condition\*measure interaction.

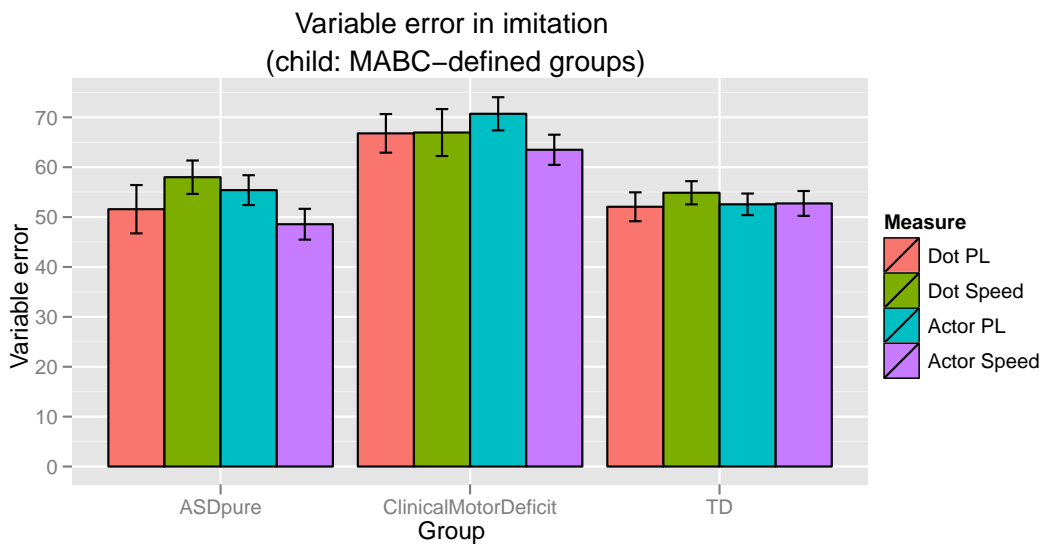


Figure 2.14: Variable error across each condition between MABC-defined groups. Variable error in the clinical motor deficit group is significantly higher than ASD and TD. There is also a significant condition\*measure interaction.

## 2.8 Discussion (children)

As with the adult study reported previously, children with ASD, DCD and TD completed the MABC-2, cKAT and an imitation task to provide a profile of motor skills. Relative strengths and weakness between clinical groups were not found using high-level tasks in the MABC-2. However MABC-2 performance did uncover a high incidence of motor skills outwith the normal range in children with ASD (23 out of 33). This is in line with results from Green et al. (2009), who found 89.1% of an ASD group ( $n=101$ ) scored at or below the 15th %ile. A narrower age range (10-14 years) in the latter study may explain the slight discrepancy in failure rates. From discussions with clinicians, motor difficulties of this magnitude are to be expected in ASD and is unlikely to be an effect of sampling bias<sup>9</sup>.

The IQ effect reported by Green et al. (2009) was replicated here, with ASD children with lower IQ showing more prominent movement difficulties, which they would interpret as evidence for broader neurological dysfunction. The effect of IQ seems unclear however, with some studies finding more severe motor impairment in AS compared to autism (generally lower functioning than AS), while others have found no difference between different ASD subgroups (e.g. Ghaziuddin et al., 1994; see also Bhat et al., 2011). Previous findings of more difficulties in ball skills in ASD compared to DCD is not replicated here, although the initial finding was based on a relatively small sample of 11 ASD and 9 DCD (Green et al., 2002).

The results from the TD group suggest that the battery was administered correctly, with 14% of the group falling below the 15th percentile. While it is expected that some TD children will fail the battery and have no motor disorder, these children were excluded from imitation analysis (in which group comparisons were of primary importance) as DCD in these cases cannot be ruled out without further investigation from a clinician.

While the MABC-2 was able to differentiate clinical groups from the TD group, the computerised battery was again less sensitive to motor differences. Two of the three tasks (tracing and tracking) did show some differences between groups, although it is likely that there is a large amount of error in this data due to restrictions in the cKAT software. In the tracing task a number of children in the clinical groups lifted the pen from the screen to adjust their grip on the stylus. As the stylus was registered by the touchscreen up to an inch above the screen this meant that these movements were incorrectly recorded as a response, thereby affecting path accuracy measures. This problem was more apparent in the clinical groups as a number of children with motor difficulties had difficulty gripping a pen and had received or were receiving OT and physiotherapy interventions to help with their handwriting. A related problem was the occlusion of the target by the arm in the tracking tasks, or the occlusion of the end of the tracing path. This led some children to either lift their arm from the screen

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<sup>9</sup>Parents were aware of the nature of the study when consenting to their child taking part.

or adopt an unnatural posture so as not to block the screen with their arm. This unnatural posture and the requirement to keep the pen on the screen at all times was a major source of concern for a team of OTs when shown the cKAT battery. Culmer et al. (2009) state that cKAT should be a more sensitive and useful tool for assessing pen control than the limited MABC tracing tasks, however these practical restrictions suggest otherwise. It is suggested that initial experimental validation of cKAT should have involved more varied subject groups (only 12 typical adults were involved) and should have included all tasks intended for use in the final battery. The pacing box feature of the tracing task was not tested in the experimental validation and was a particular problem in the present study: a number of children became bored during the battery of tests, particularly the tracing task as there were a number of repetitions and many became frustrated by reminders to stay inside the pacing box. This instruction was rarely adhered to, particularly by those in the ASD group. An in-game prompt or consequence may be beneficial, particularly if the task is to be used with younger children or those with attentional difficulties. Such changes would make the task more game-like and would likely increase engagement in an otherwise tedious task.

The finding of slower reaction times in the clinical groups in the pentagram task is unsurprising, as a number of these children asked for clarification before starting each task due to difficulties understanding the on-screen instructions. This will also have affected tracking and tracing accuracy, as a number of children asked for clarification once data collection had already been triggered, thereby artificially decreasing accuracy. This was seen particularly in the ASD group. For these reasons the results from the cKAT battery should be considered with caution. The cKAT battery may be useful in initial screening for motor difficulties in children in the future, however the practical limitations identified during testing would need to be addressed first if the tool is to be used with similar groups.

Finally, as hypothesised, findings from the imitation task reveal no evidence for an ASD-specific imitation deficit and no group by condition interaction. It did however uncover a clear role of motor ability in basic imitation skill in both clinical groups. While some children with ASD did perform significantly worse than TD, once the groups were rearranged into MABC performance-defined groups it was found that those with spared motor skills were comparable to TD. This supports previous findings of intact imitation in ASD (Beadle-Brown & Whiten, 2004; Beadle-Brown, 2004; Ingersoll et al., 2003). It is possible that deficits may have been found in a more socially-oriented task, as has been found previously. Results suggest that previous deficits may have been due to motor deficits rather than more classic autism symptoms. The previous findings of increased autism severity correlating with more pronounced deficits (e.g. Rogers, Hepburn, Stackhouse & Wehner, 2003; Zachor, Ilanit & Itchak, 2010) can be explained speculatively as being caused by motor deficits, as typically those lower functioning children had more pronounced motor difficulties in the present study. With some differences between ASD and TD it is possible that there is a slight delay in motor

reproduction skills, as the adult study showed no differences, however the adult ASD group was less affected by motor difficulties, again suggesting that in this case motor difficulties likely played an important role in imitation ability.

Stewart, McIntosh & Williams (2013) completed an almost identical imitation task to the one described in the present study with 11-17 year olds with ASD and TD. Using the same set of stimuli, they found that ASD were significantly worse in the actor condition for both PL and time measures. This study has some major methodological flaws however: the conditions were not counterbalanced, with the actor condition always being completed first. With clinical groups often taking longer to settle into tasks it is important to counterbalance so that this effect is not consistently associated with one condition. Secondly, there is no analysis of speed, only duration. The two are not interchangeable, as speed is calculated relative to PL, and duration is a standalone property. Also, subjects were explicitly instructed to copy the *speed* of the drawing, not the duration. Finally, there are no details regarding measured motor ability, and despite the DCDQ-07 being completed for all children, these data were not used to assess the impact motor difficulties might have had on imitation ability. Given the finding of the current study, which has a larger sample size, a clinical control group and a number of motor skill measures, it is suggested that the apparent deficit reported by Stewart et al. (2013) may be an artefact of these methodological shortcomings.

The imitation task had one minor limitation: the laptop being used was very sensitive to the stylus, resulting in a number of false trials (trials triggered prematurely). This happened most frequently in the clinical groups, as these children often had attentional issues and would frequently play with the stylus close to the screen before they were ready to draw. This was discouraged and when necessary the child's arm was held on the table until given a signal. These false trials resulted in a slight delay between some trials, although extra 'blank pages' were given so in the majority of cases all trials were completed. While this limitation did not directly affect results, it did result in a lengthier testing session for a number of children in the clinical groups.

The overall profile of motor skills for ASD and DCD does not differ greatly. Those ASD children with spared motor skills were comparable to TD in all aspects of movement assessed here (fine, gross, visuomotor and imitation). Given that there were no clear differences on any measure between ASD children who failed the MABC-2 and those with DCD, it appears that motor deficits in ASD may not be ASD-specific, but reflect an additional diagnosis of DCD. This supports the change to diagnostic criteria in the DSM-5, which now allows for comorbid diagnosis of ASD and DCD. Questionnaire measures of autistic and DCD traits differentiated the two clinical groups from TD, but ASD and DCD showed a large degree of overlap, particularly in motor skills. SRS responses for children with DCD showed they exhibited significantly more autistic traits than TD children, but significantly fewer than ASD children. This supports findings that children with DCD tend to have more AS symptoms than TD (Kadesjo & Gillberg,

1998) and further supports the large degree of overlap in performance on the three motor tasks.

## 2.9 General discussion

This chapter has reported motor profiling in children and adults with ASD, DCD and TD, which found motor deficits in ASD and DCD to be broadly similar. There was no significant difference in MABC-2 scores between ASD and DCD in either age group, however the motor skills in both clinical groups were significantly poorer than the TD group in both age groups. The cKAT battery was not successful in identifying motor deficits in adults with ASD and DCD, even those who had failed the MABC-2. It was slightly more sensitive to difficulties in children, although practical limitations in the battery and only weak-moderate correlations with the MABC-2 warrant a cautious interpretation of any significant results in the child study. The imitation task found that motor deficits in ASD and DCD did not adversely affect visuomotor imitation in adults, as the clinical groups performed comparably to TD. Similarly, there was little compelling evidence for an ASD-specific imitation deficit in children, however the motor deficits in the ASD group did seem to hamper imitation ability, although not to a level below children with DCD. Those children with ASD with spared motor skills showed no deficit relative to TD.

It appears that in order to locate any point at which the two clinical groups' motor ability deviates it may be necessary to look at the components of movement more closely, rather than focus on full movements. While the MABC-2 is useful for identifying those with motor impairment, it does not give any indication of the reasons underlying the difficulty. It is possible that different underlying problems are responsible for the deficits in each group. This will be explored further in the following three chapters, which investigate a possible double dissociation in the use of visual and proprioceptive cues in perception and action in the two clinical groups.

## Chapter 3

# Vision and proprioception in perception and action

This chapter reports two tasks assessing the use of visual and proprioceptive cues in perception and action in children with ASD, DCD and typical development. The study of the sensory components of movement was prompted by the lack of any clear difference in high-level motor skills emerging from the profiling study reported in the previous chapter.

Under normal circumstances, motor actions tend to rely on visual and proprioceptive feedback. Visual feedback allows you to monitor the action relative to extrinsic landmarks, while proprioception allows actions to be monitored relative to an internal sense of body positioning: it is proprioception that lets you do the actions to the ‘YMCA’ without watching your arms.

Previous literature suggests that there may be a double dissociation in the way those with ASD and DCD spontaneously use these two kinds of sensory feedback. ASD appears to be associated with an increased reliance on proprioception, at the expense of visual cues, while DCD appears to be associated with a relative reliance on visual cues. However, it appears that the two groups have not been compared directly in a single study. A review of the literature on cue weighting in perception and action in the two groups is given below, followed by results of two experiments assessing visual and proprioceptive weighting in ASD and DCD. The way in which these results interact with MABC-2 scores from the study detailed in Chapter 2 will also be discussed.

### 3.1 Comparing the roles of vision and proprioception in perception and action in ASD

The following review will be split as follows: studies in which proprioception is actively manipulated will be described first, followed by studies which only manipulate visual feedback, and finally studies which manipulate the presence or absence of both visual

and proprioceptive cues to assess the relative use of each type of cue.

### 3.1.1 Altering proprioception

The use of proprioception can be difficult to measure, as it is much more difficult to perturb or eradicate proprioceptive feedback than visual feedback. Muscle and tendon vibration seems to effectively disturb proprioceptive feedback (Palluel, Aspell & Blanke, 2011; Vaugoyeau, Hakam & Azulay, 2011), however no studies using this method with either ASD or DCD appear to have been published. Cold temperatures applied to a limb has also been found to dampen proprioceptive sensitivity in some cases, although this is not a universal finding (Surenkok, Aytar, Tuzun & Akman, 2008; Wassinger, Myers, Gatti, Conley & Lephart, 2007; Costello & Donnelly, 2010; cf. Dover & Powers, 2004; Costello, Algar & Donnelly, 2007). Again this technique has not been used with ASD or DCD.

The use of a force field applied to a tool has been more robustly verified as an effective way of altering proprioceptive feedback, and has been used to investigate proprioceptive reliance in ASD (Larson, Bastian, Donchin, Shadmehr & Mostofsky, 2008; Mostofsky, Izawa, Penky, Marko, Dowell & Shadmehr, 2010; Haswell, Izawa, Dowell, Mostofsky & Shadmehr, 2009). When looking at full body postural control, some studies have attempted to perturb or remove proprioceptive feedback, although it is often the case that these manipulations are not adequate. Both force field techniques and proprioceptive manipulation during full body postural control are discussed below.

Using a tool that behaves in a way that is incongruent with the force you exert on it is one of the more effective ways to perturb proprioceptive feedback: the arm feels as though it is in a position at odds with the position you expected it to be in. Using this method, children with ASD ( $n=14$ , mean age 10 years) have been found to generalise learning from the tool in intrinsic (proprioceptive) coordinates more effectively than using external (visual) coordinates (Haswell et al., 2009). Additionally, generalisation in intrinsic coordinates was *better* than that of age-matched TD controls (Haswell et al., 2009) and ADHD and TD controls (Mostofsky et al., 2010). Learning was considered to use intrinsic coordinates when the trained movement and target movement involved the same joint rotations, while learning using extrinsic coordinates involved the same hand motion, but different joint rotations. Haswell et al. (2009) suggest that in ASD, internal models (our predictions of the sensory outcome of motor commands) are built more heavily on the association between proprioception and self-generated motor commands, than they are between visual information and self-generated motor commands. This is thought to reflect the use of overabundant short-range connections in the brain, between MI and the somatosensory cortex. This increased proprioceptive reliance was associated with both increased autism severity, and decreased motor skills (measured by the PANESS) (Haswell et al., 2009). It is not clear whether those children with more

pronounced ASD symptoms tended to also have more pronounced motor difficulties, although previous research suggests this may be the case (MacDonald, Lord & Ulrich, 2014; Hilton, Wente, LaVesser, Ito, Reed & Herzberg, 2007; cf. Zachor et al., 2010). It should be noted however that Larson et al. (2008) failed to provide evidence for a proprioceptive bias in ASD ( $n=21$ , 8-13 years) using a similar force field paradigm.

Further evidence of intact use of proprioceptive feedback in HFA was provided by Mostofsky, Bunoski, Morton, Goldberg & Bastian (2004) in a task which manipulated the weight of two balls, while keeping constant their visual appearance (e.g. a light ping-pong ball and a heavier squash ball). The only difference was the force each ball exerted on the arm when it was caught. Subjects were required to catch the two balls while keeping both hands within a window frame. The change in upward arm movement over trials in response to the unexpectedly heavier ball was measured as an indication of the use of proprioceptive feedback to ensure that the arm remained inside the frame. Children with ASD ( $n=8$ , 8-13 years) were able to adapt to the unexpectedly heavier weight as well as TD children, suggesting again that the use of proprioceptive feedback in action is intact in ASD. In contrast with the above study, there was no evidence of an over-reliance on proprioception compared to TD in this case.

Some studies also perturb proprioception to assess balance skills, although the methods employed are questionable. For example, Molloy, Dietrich & Bhattacharya (2003) perturbed proprioceptive cues by asking children to stand on foam. They found that static balance of children with ASD ( $n=8$ , 5-12 years) was more affected by the removal of vision than by the ‘altering’ of proprioception via a foam platform. The authors therefore suggest that visual feedback is the most pertinent feedback available to children with ASD. However, the validity of the method used to perturb proprioception is much less than that of the examples above: standing on foam merely puts the subject off balance, rather than altering the proprioceptive representation of the body. For this reason the conclusion drawn may not be appropriate.

Studies altering proprioceptive feedback show clear proprioceptive bias in ASD in some cases, or proprioceptive processing similar to TD. A proprioceptive bias has not been uncovered in postural control, suggesting that a proprioceptive bias may be specific to certain types of actions such as reaching (Haswell et al., 2009). Alternatively, methods used may be insufficient to assess sensory bias in postural control. The relative roles of vision and proprioception in postural control is further discussed in Section 3.1.2.2.

### **3.1.2 Altering visual feedback to assess visual/proprioceptive weighting**

As altering proprioceptive feedback is difficult without the correct equipment, studies often only alter visual feedback. By perturbing or removing visual feedback and measuring how this affects performance, it is possible to measure the influence of vision,

and consequently to infer the role proprioception.

### **3.1.2.1 Prismatic displacement**

Using a prism to displace the visual field creates a conflict between the visual location of a target in space relative to the felt location of the body in space. This skewed frame of reference makes us misreach for a target until the representation of the body is realigned with the displaced visual field or vice versa. Children with ASD ( $n=21$ , 8-13 years) have been found to adapt to prismatic displacement at a similar rate to TD children (Larson et al., 2008). Children threw a ball with normal vision, displaced vision, and then normal vision. There was a similar progression from missing the target at the start of the second normal condition to realigning the body with the displaced target and hitting it. Masterton & Biederman (1983) also used prisms to alter visual feedback in a task involving moving objects between locations. They found that an ASD group ( $n=11$ , 7-15 years) showed generalisation to the opposite hand from the one used before the prismatic adaptation phase. This kind of generalisation was not found in a TD group or children with mental retardation, and suggests that the ASD group relied heavily on proprioceptive feedback to modulate their movement toward the target, as evidenced by their successful cross-limb generalisation. Findings from tasks involving prismatic displacement support the findings of spared or enhanced proprioceptive processing observed in tasks involving proprioceptive perturbations (e.g. Haswell et al., 2009).

### **3.1.2.2 Vision for postural control**

Just as displacing the visual field can affect the felt position of the body relative to shifted visual cues, it is also possible to perturb the sense of proprioception by visually inducing a sense of motion. The swinging room paradigm (Lee & Aronson, 1974) shows that by moving the walls of a room back and forth while keeping the floor steady, subjects will experience visually-induced motion and sway their bodies to maintain balance, even though the floor is steady. Using a similar paradigm, Gepner, Mestre, Masson & de Schonen (1995) assessed postural stability in young children (4-7 years) with ASD by means of various visual conditions. Subjects stood on a stable force plate which recorded their postural sway, and were instructed to stand still with their arms at their side and either watch the screen in front of them, or close their eyes. When visual feedback was provided, this was either unperturbed feedback (no induced movement), or six levels of patterns inducing a sense of movement. TD children responded to the visual motion cues and swayed their body, showing that their sense of current body positioning was being altered by the visual feedback. ASD children on the other hand did not, suggesting that their sense of body position is built more strongly on proprioceptive cues. It should be noted that the clinical sample size in this study was very small, comprising only five children with ASD. For this reason generalising across the

autistic spectrum from this study is difficult: more studies using this sort of task are necessary to fully investigate cue weighting in postural control. However these findings do seem to be in agreement with the studies described above.

The studies reviewed so far have all perturbed sensory cues and the majority have found that children with ASD have at least an intact ability to use proprioceptive cues to guide actions. In some cases the ability to use proprioception was relatively enhanced.

### 3.1.3 Assessing visual and proprioceptive benefit and acuity

It is not necessary to perturb a sensory cue in order to assess its importance. By comparing performance on a task with feedback from only one modality to the same task with a combination of the two, it is possible to assess the efficiency with which each kind of cue is utilised (and the benefit of having multiple cues). Ranta & Mostofsky (2011) assessed the differential use of visual and proprioceptive feedback using a maze task, in which subjects traced a maze with a stylus while blindfolded. They were then asked to identify the maze they had traced, using either vision, or vision and proprioception. For both children with ASD ( $n=22$ , 8-13 years) and age-matched TD controls ( $n=23$ ), identification success was greater when proprioceptive feedback (feeling their way through the maze) was allowed alongside visual feedback, compared to visual identification alone. The increase in success rate between groups appears sizeable: 40% increased success rate for ASD compared to 15% for TD, however this difference did not reach statistical significance. Even this indication of a possible group effect<sup>1</sup> suggests that proprioceptive feedback is utilised more effectively than visual feedback by children with ASD. This is further supported by the finding that children scoring highly on an autistic social interaction scale (and therefore deemed to show more severe autism symptoms) tended to benefit more from proprioceptive feedback ( $p < 0.1$ ). This finding appears to mirror that of Haswell et al. (2009), who found that motor learning was proprioceptively driven in ASD children, particularly in those with more pronounced social deficits. It would be of interest to know whether the visual-proprioceptive condition was easier than the visual condition because of the use of proprioception, or merely because it allows for an integration of the two cues. Obviously a greater success rate due to sensory integration is not the same as a greater success rate attributed to the increased reliance on proprioceptive information.

Comparable proprioceptive acuity in adults with ASD relative to TD has also been reported in a limb position matching task (Fuentes, Mostofsky & Bastian, 2011). This is a more direct test of proprioceptive acuity in which subjects either moved a visual stimulus to match the orientation of their hidden arm, or moved the arm to match the visual stimulus. In this case the ASD group had significant motor and sensory deficits

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<sup>1</sup>It is not possible to ascertain how strong a trend this is as the study is published in conference proceedings without full details of the results.

according to the PANESS and the Adult Sensory Profile, although still appear to have had an intact sense of their proprioceptive positioning when vision of the limb was removed.

The majority of the studies described so far suggest a relative dependence on proprioceptive over visual feedback in those with ASD, using both sensory perturbation and sensory deprivation measures. Anecdotal evidence also further supports this suggestion. For example, Ricks & Wing (1975) comment that when teaching autistic children motor skills, ‘do as I do’ visual instructions appear not to work. They suggest that “the only way to teach motor skills to a young autistic child (is) to move his limbs through the action which is required” [Ricks & Wing (1975), p. 204]. Considering the spontaneous use of sensory feedback, it has also been found that those with ASD spend relatively more time in tactile exploration tasks than in visual exploration tasks (Schopler, 1965). However with known sensory sensitivities in autism, such as hyposensitivity or hypersensitivity (craving or avoiding stimulation) to touch and some aspects of visual stimulation (Stewart, Russo, Banks, Miller & Burack, 2009), it is difficult to ascertain the reasons for spontaneous tactile exploration in some children with ASD.

The consensus seems to be that ASD is associated with a reliance on internal representations of the body. However one study has suggested that motor impairments are actually caused by a proprioceptive deficit. This study is discussed in the following section.

### **3.1.4 A counter argument**

While the majority of studies seem to suggest a bias toward proprioceptive cues over visual cues, Weimer, Schatz, Lincoln, Ballantyne & Trauner (2001) suggest that motor problems in AS may actually be caused by a deficit in the use of proprioception. They reached this conclusion after administering a number of fine and gross motor tasks and observing two significant findings: significantly poorer one-leg balance without vision compared to TD, and significantly poorer performance on a finger-thumb apposition task. In the former task however the difficulties in the no-vision condition, which importantly was specific to the non-dominant foot, could simply be because the task was harder, and not directly due to the absence of vision (Cherng, Hsu, Chen & Chen, 2007). Performance in the finger-thumb apposition task were said to support the proprioceptive deficit hypothesis as children were observed to watch their hands more than TD children, yet still were significantly slower. The authors suggest that they did this to try to overcome a proprioceptive deficit, but were unable to use the visual information to improve their speed and accuracy. However rather than pointing to a proprioceptive deficit, this seems to suggest a visual deficit.

The authors conclude that tasks that were more centrally reliant on proprioception highlighted significantly poorer performance in the AS group. However, at no point

was proprioception actively manipulated, and the one task in which vision was manipulated has already been questioned. It is suggested then that this study does not adequately manipulate vision and proprioception in order to justify their claim of a primary proprioceptive deficit in AS. Additionally, the AS group was slower and less able on every task (e.g. 8.7 seconds slower than TD in a manual dexterity task), even on tasks which the authors would be unlikely to describe as being centrally reliant on proprioception.

Additional cautionary notes concern the sample of subjects involved in the study. A group of 10 children and young adults (9-19 years) were compared to age-matched controls. The tasks which significantly differentiated groups were not age-normed, so the wide age range becomes problematic. Calculating mean performance in groups with such wide age ranges and so few children of similar ages is not standard procedure, particularly when performance has not been standardised to account for age differences, as is the case in the MABC. To put the age range into context, the AS group is made up of: one child who would typically be mid-way through primary school; one in the final year of primary school; three who would be in the third year of secondary school; one each in the fourth and fifth year of secondary school; and two who could be in the second or third year of an undergraduate degree course. Given the heterogeneity of the condition, this kind of variation within the group is a concern, as performance on these kinds of tasks develops with age up to at least 16 years of age (which is the point up to which the MABC-2 is standardised according to age). In addition to the large variation in age, the clinical group had a host of other medical conditions, primarily Attention Deficit Disorder, although others showed neurological dysfunction, mental health problems and auditory difficulties. While the results themselves are likely reliable (and the AS group did have difficulty in a range of tasks), the conclusions drawn by the authors are not convincing, particularly in light of the frequent proprioceptive bias reported in more methodologically sound studies.

## **3.2 Comparing the roles of vision and proprioception in DCD**

The review of the literature concerning DCD follows the same structure as the previous section focused on ASD. Literature is split according to the type of manipulation used: altered proprioception; altered vision; and measures of visual and proprioceptive benefit.

### **3.2.1 Altering proprioception**

Difficulties in balance and postural control have been identified as a common feature in DCD, and the increased role of vision in maintaining balance has been highlighted (Williams, 2002). As discussed previously, the explicit perturbation of propriocep-

tive feedback is difficult to achieve, particularly when investigating full body postural control. In a paradigm similar to that employed by Molloy et al. (2003) with ASD subjects, Cherng et al. (2007) ‘degraded’ proprioceptive feedback by requiring participants to stand on foam during balance tasks. As noted earlier, this will make the task more difficult, but it seems reasonable to argue that it does not actually perturb proprioceptive feedback. This study found that DCD children ( $n=20$ , 4-6 years) were worse than TD in all conditions, but particularly in conditions where feedback (both visual and proprioceptive) was either removed or degraded. This finding with regard to proprioception is unsurprising: the DCD group had known motor problems, therefore would be expected to be worse at balancing even on a flat surface. By increasing the task difficulty this highlights their relative difficulty more. The finding that conditions with any kind of cue perturbation were more difficult for the DCD group perhaps highlights that the visual perturbation (arguably the only true perturbation) was problematic for the DCD group because visual feedback is often weighted heavily. From this study the relative role of vision and proprioception in DCD is not clear.

### **3.2.2 Altering visual feedback**

#### **3.2.2.1 Vision for postural control**

Using the swinging room paradigm described previously, Wann et al. (1998) found that children with DCD ( $n=6$ ) were more affected by distortion of visual feedback than TD children. They were also less stable with eyes closed compared to age matched (10-12 years) and nursery aged children. With normal vision the DCD group was as stable as the TD group. These findings suggest that children with DCD may be more reliant on external visual cues in maintaining postural control. This finding sits directly opposite results from Gepner et al. (1995) who used a similar task with children with ASD, revealing a lack of reactivity to visually induced motion. Slower, more cautious steps while walking in the dark also point to an increased reliance on vision for postural control in DCD (Deconinck, Clercq, Savelsbergh, Coster, Oostra, Dewitte & Lenoir, 2006b). It should be noted however that a visual bias for postural control has not always been reported (Przysucha & Taylor, 2004).

#### **3.2.2.2 Reaching tasks**

When reaching for an object the maximum width of our grasp (maximum grip aperture: MGA) is determined by visual and proprioceptive information of the hand’s current shape, relative to the visual shape of the object. Children with DCD ( $n=9$ , 7-9 years) have been found to form a less accurate MGA than TD children when visual feedback of the reach-to-grasp movement is removed (Biancotto, Skabar, Bulgheroni, Carrozzi & Zoia, 2011). While the removal of vision affected the movement time of both groups, only the DCD group seemed unable to appropriately scale their grip from the remembered size of the target. This suggests that online visual feedback is vital to children

with DCD, as remembered visual information is less informative.

Another reaching task, in which children moved beads between cups, found that restricting visual feedback of the cups and/or hands did not differentially affect TD and DCD children, although those with DCD were significantly slower and less accurate than TD overall (Rosblad & von Hofsten, 1994). The authors suggest that this shows that DCD is not characterised by an increased reliance on vision, but by only comparing the effects on the DCD group of the visual manipulation with the effects on a TD group, this only shows that DCD is not characterised by an above average sensitivity to visual information. Of interest, one of the ten children with DCD was noted to have marked difficulties in the task when visual feedback was removed. His movement time increased threefold and he groped for the cups until knocking into one. This pattern of behaviour was also seen in some of the other children with DCD although to a lesser extent. There appears to be some evidence for a visual bias in reaching, however previous findings suggest that the effect of the removal of vision may be sensitive to individual differences within DCD.

### **3.2.3 Assessing visual and proprioceptive benefit and acuity**

It has been suggested that proprioceptive information may be processed more slowly in DCD compared TD, although the speed of processing visual cues is not impaired (Smyth & Glenross, 1986; see also van der Meulen, van der Gon, Geilen, Gooskens & Willemse, 1991). Additionally, Skorji & McKenzie (1997) found that children with motor difficulties (<5th percentile on the TOMI although not diagnosed with DCD) rehearsed movements in visuospatial terms more than kinaesthetic terms, and this visual bias was greater than that seen in TD children. Rehearsal mode was investigated by introducing visual or kinaesthetic interference during a 15 second delay period before children repeated a modelled sequence of simple movements. There has also been a suggestion that kinaesthetic training could improve performance on certain motor tasks (e.g. Laszlo & Bairstow, 1985; Laszlo, Bairstow, Bartrip & Rolfe, 1988). Training included proprioceptive acuity training (judging when the two hands are level) and proprioceptive perception and memory training (returning a proprioceptively-defined shape to its previous orientation). However the kinaesthetic tests have since been found not to correlate significantly with motor ability and the training's efficacy has been questioned (Sugden & Wann, 1987).

Studies using perceptual matching tasks to investigate visual and proprioceptive benefit in DCD seem to generally suggest a reliance on visual cues associated with motor impairment (Smyth & Mason, 1998; Rosblad & von Hofsten, 1992; Sigmundsson, Whiting & Ingvaldsen, 1999; cf. Mon-Williams, Wann & Pascal, 1999). A version of the matching task used in these studies is used in the present study and is detailed in full in Section 3.4, alongside previous findings related to DCD (the task does not seem to have been used in ASD).

Compared to the relatively strong consensus of a proprioceptive bias in ASD, there is less consensus with regards to cue weighting in DCD. Some studies find no visual bias or proprioceptive deficit in DCD (Lord & Hulme, 1988), and these authors suggest that visual perceptual difficulties may actually contribute to motor symptoms in DCD (Hulme, Biggerstaff, Moran & McKinlay, 1982; Lord & Hulme, 1987). Despite this, there is certainly evidence for a relatively heavy reliance on vision in a number of different tasks.

### **3.3 Vision and proprioception in ASD and DCD: a double dissociation?**

There is a growing body of research devoted to investigating visual and proprioceptive processing in ASD and DCD, however no studies have yet compared the two groups directly. The consensus in the ASD literature appears to be that individuals with ASD tend to be better at making use of proprioceptive cues, while less able to use visual feedback. This is supported by behavioural studies, and Haswell et al. (2009) suggest it is also consistent with the overabundance of short-range connections in the brain in ASD, connecting MI and the somatosensory cortex. Conversely, some DCD literature seems to suggest that DCD might be associated with an increased reliance on vision, although the picture is comparatively less clear, possibly due to the relative lack of relevant studies and the difficulties in recruiting large samples of children with DCD. However, related literature on acquired apraxia suggests a bias toward visual information (Jax, Buxbaum & Moll, 2006; Sirigu, Daprati, Pradat-Diehl, Franck & Jeannerod, 1999; Ietswaart, Carey & Sala, 2006). This perhaps strengthens the suggestion that DCD is characterised by a visual bias, as the disorders share a number of characteristics. Additionally, there is some anecdotal evidence for such a bias, with Wann (1987) suggesting that an immature handwriting style (common in DCD), which is characterised by slow, jerky movements, may be a mechanism used to increase the amount of visual feedback and control available when writing. There appears to be evidence of a possible double dissociation in visual and proprioceptive cue weighting in ASD and DCD.

The proposed double dissociation could be framed more explicitly in terms of disorder-specific multisensory integration deficits. The dissociation would therefore be conceptualised as an integration deficit in ASD manifesting as an over-weighting of proprioception during visuo-proprioceptive integration; and DCD conceptualised as an integration deficit characterised by an over-weighting of visual information during visuo-proprioceptive integration. As multisensory integration across a range of modalities is common in a number of neurodevelopmental conditions (Hill, Crane & Bremner, 2012) it is possible that atypical sensory integration is a reliable marker for neurode-

velopmental conditions. A shared underlying deficit in multisensory integration would support the high prevalence of children with comorbid (or co-occurring) neurodevelopmental disorders.

Given the overlap in motor impairment reported in Chapter 2, and the apparently distinct nature of motor difficulties in ASD as suggested by DSM-IV exclusion criteria for DCD, it seems reasonable to conjecture that a double dissociation may be apparent between ASD and DCD in their use of sensory information for movement. There is evidence in favour of such a dissociation, although there is also some contradictory evidence for both ASD and DCD. Further investigation of the proposed dissociation may go towards illuminating the issues underlying motor deficits in the two groups, and help in answering the question of whether ASD and DCD really are two mutually exclusive diagnoses (as defined in the DSM-IV), even though both share motor deficits as part of their symptomatology.

The remainder of this chapter will detail two experiments investigating the relative roles of vision and proprioception using both clinical groups as the main comparison, as well as a TD control group to act as a further comparison group. The first study will use the visual-proprioceptive matching paradigm. This is described in the next section, and relevant findings from previous studies using this task are discussed. The second study will use a mirror-reach task (Holmes, Crozier & Spence, 2004). This will be described in the second half of this chapter.

## **3.4 Visual-proprioceptive matching**

### **3.4.1 Perceptual matching to assess visual and proprioceptive benefit**

The visual-proprioceptive matching task has been referred to briefly in the previous section. The most basic version of this matching task requires subjects to indicate the location of a target on a tabletop by pointing underneath the table. This means that the pointing arm is only defined proprioceptively. The sensory conditions under which the target is presented are often manipulated, and tend to include vision (VP)<sup>2</sup>, proprioception (PP) and vision-proprioception (VPP). The former and latter conditions are usually the same across studies: in the VP condition the subject proprioceptively matches the location of a target they can see but not feel; in the VPP condition the subject matches the location of a target whose location is specified by their hand, which they are able to see. The PP condition seems to vary, with some studies using a purely proprioceptive condition, in which the target is not seen at any point during the trial (Wann, 1991), while others allow a brief view of the target location so that subjects can move the target hand to the correct location ready for matching (van Beers, Sittig &

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<sup>2</sup>The 'P' refers to the pointing hand: the target is defined visually (V) and the pointing hand used to indicate it's location is only defined proprioceptively.

van der Gon, 1999; von Hofsten & Rosblad, 1988; Smyth & Mason, 1998). By defining the target with vision and proprioception separately and then together it is possible to calculate the benefit of each type of sensory cue: how much accuracy increases when locating a target which is defined visually and proprioceptively compared to a target which is only defined visually or proprioceptively.

Several studies have used the basic paradigm to demonstrate a reliance on vision in DCD. However, none of these studies give a clear-cut result. Smyth & Mason (1998) report a visual reliance in DCD, but the study involved young children (5-8 years) whose typical visual-proprioceptive sensory weighting was still likely developing [there is evidence for an over-reliance on vision in typical development up until 8 years (Nardini, Began & Mareschal, 2013)]. It is therefore difficult to know the extent to which age might have influenced cue weighting. However these findings are supported by similar findings reported by Rosblad & von Hofsten (1992), who found that end-point accuracy was heavily dependent on visual cues in children with motor impairment ( $n=29$ , 5-13 years). In this case however the motor impaired group included children with DCD, CP and spina bifida, therefore the specificity of this visual bias to DCD is unclear. Finally, Sigmundsson et al. (1999) indirectly suggest a visual bias in children with DCD ( $n=6$ , age 7) by reporting a proprioceptive deficit in a version of the task designed to compare accuracy in proximal and distal reaches. Proximal reaches were made along the vertical centre, with use of the whole arm, and distal reaches were confined to a square within reach of the hand while the arm was restrained. It should be noted that both proximal and distal conditions made use of proprioceptive feedback, therefore it is unclear that there is a complete proprioceptive deficit. However the reach that required the most movement and required proprioceptive information about a larger portion of the body did specifically adversely affect performance. Due to the lack of visual feedback in all conditions, it is not clear if this is a proprioceptive deficit alongside relatively intact or enhanced sensitivity to visual feedback, or whether both would be equally problematic. Also, children were again younger than the suggested age at which sensory weightings stabilise, therefore there may again be an additional effect of age.

While these studies all point to a visual bias in motor impaired children, Mon-Williams et al. (1999) report *more* difficulty in visually guided matching. However in this case only eight children were tested, and five of these children had strabismus. An effect of strabismus on performance was ruled out as those with and without did not differ on the task, however the sample size in these subgroups is too small ( $n=5$  and 3) for formal analyses. Additionally, the children were all between 5-7 years, so again in typical development the relative weightings of visual and proprioceptive cues would not have stabilised by this point. However a reliance on vision would still be expected.

Results appear to suggest visual reliance in DCD, however there are too many inconsistencies and concerns associated with the studies to draw any firm conclusions. The task does not appear to have been used with children with ASD, although it would

be hypothesised that these children would show a proprioceptive benefit, in line with proprioceptive bias reported in other tasks (Haswell et al., 2009; Mostofsky et al., 2004).

### **3.4.2 Perceptual matching using prismatic displacement**

The basic paradigm can be extended to include a perturbation of visual information so that seen and felt location of the target is incongruent. Using this method it is possible to calculate a visual weighting to quantify how much visual cues are used compared to proprioceptive cues. Using the paradigm with typical adults, Mon-Williams, Wann, Jenkinson & Rushton (1997) found that proprioception played a relatively minor role in target location in a bimodal condition, as evidenced by substantial error in the direction of visual displacement, achieved with the use of a prism. The effect of the prism was much smaller when the task was repeated in a dark room with only a lit LED on the target fingertip. The present study uses a sparse background to avoid children focusing on background details, and allows vision only of the target and not the hand grasping it, although lights in the room are left on. As visual cues will not be entirely absent it is expected that proprioceptive cues will be given less weighting compared to the dark condition used by Mon-Williams et al. (1997). Conclusions from van Beers, Sittig & van der Gon (1996) also favour a bare background. They found that variance in responses in a VPP condition were less than was expected given the variance in responses in both a PP and VP condition. They suggest that smaller-than-anticipated variance in the VPP condition points to the role of extraneous visual cues from the environment and/or body in reducing variance when visual and proprioceptive cues are integrated. It is possible that children in different developmental groups might differ in their use of these extra visual cues, therefore vision will be restricted to only the target and a bare background in the present study.

The present study uses both the basic version of the task and the prismatic displacement version to investigate the proposed double dissociation in ASD and DCD, with TD children as an additional control group. If the proposed dissociation exists, it is expected that the DCD group will show poorer performance than the ASD group in the PP matching condition and better than the ASD group in the VPP condition. When using the prism to displace visual feedback, it is expected that the DCD group will be swayed more by this visual feedback than ASD, who are expected to make better use of veridical proprioceptive cues. This experiment will test the hypothesis in a perceptual task, and the experiment reported in the second half of this chapter will assess the dissociation in a reaching action.

Although children as young as 6 years of age completed this task as part of a larger battery of tasks (reported in Chapter 2), only those 8 years and older were intended for use in analysis. As has been mentioned previously, there is a large body of evidence

to suggest that until approximately 8 years of age the relative weightings of vision and proprioception are not yet stable, in the matching task (Nardini et al., 2013) and other tasks (e.g. Gori, Viva, Sandini & Burr, 2008; Hay, Fleury, Bard & Teasdale, 1994; Asaiante & Amblard, 1993; Greffou, Bertone, Hanssens & Faubert, 2008; Riach & Hayes, 1987; Shumway-Cook & Woollacott, 1985). There is also evidence of an inability to efficiently integrate visual and proprioceptive cues until this age (Chicoine, Lasseonde & Proteau, 1992; Pellizzer & Hauert, 1996). Finally, work related to adaptation to visual distortions and its prerequisite perceptual, sensory and motor abilities also highlights a switch in sensory processing at around 8 years of age (Ferrel-Chapus, Hay, Olivier, Bard & Fleury, 2002; Davidson, 1934, 1935; Gibson, Gibson, Pick & Osser, 1962; Rudel & Teuber, 1963; Oyama & Sato, 1975; Kaufman, 1980; Orliaguet, 1985; Hay & Bard, 1984).

## 3.5 Experiment 1: Visual-proprioceptive spatial location matching

### 3.6 Methods

#### 3.6.1 Subjects

All children had previously participated in tasks described in Chapter 2 and all parents gave written consent (see details in Chapter 2). One child with ASD did not complete this task due to difficulties maintaining attention during testing. Age and IQ statistics are slightly different from the previous chapter as a small number of children (1 ASD and 1 DCD) were removed from each group due to task-specific exclusion criteria<sup>3</sup>. Only data from those children 8 years and older will be included in analysis, and data from the four TD children who previously scored below the 15th percentile on the MABC-2 will not be included (as per additional cKAT and imitation analyses in the previous chapter). All but one child (a member of the DCD group) was male.

Mean MABC-2 percentile, IQ percentile, and age for those children whose data will be used are shown in Table 3.1. As with Chapter 2, groups were age- and IQ-matched:  $F(2, 63) = 0.27, p = 0.768$  and  $F(2, 63) = 1.74, p = 0.184$  respectively. There was a significant effect of group on MABC-2 percentile rank ( $F(2, 63) = 29.66, p < 0.001$ ). TD children had significantly higher percentile ranks than both ASD and DCD (mean difference=44.61, SE=6.15,  $p < 0.001$ ; and mean difference=46.29, SE=8.84,  $p < 0.001$  respectively). As with the previous study, MABC-2 scores in the clinical groups did not differ significantly (mean difference (DCD-ASD)=-1.59, SE=8.56,  $p=0.853$ ).

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<sup>3</sup>Children as young as 6 years of age completed this task as part of a larger battery of tasks (detailed in Chapter 2). As the testing session included an IQ test after this task, these younger children completed the task so that testing sessions were the same for all children.

Table 3.1: MABC, IQ and age demographics for each group

	Mean MABC percentile rank (SD)	Mean IQ percentile rank (SD)	Mean age (years (SD))
ASD ( $n=31$ )	16.60 (23.10)	46.55 (31.21)	11.00 (1.75)
DCD ( $n=9$ )	15.01 (20.44)	52.88 (37.56)	10.22 (2.17)
TD ( $n=24$ )	61.21 (22.70)	51.67 (25.68)	10.29 (0.91)

### 3.6.2 Apparatus

The apparatus is shown in Figure 3.1. The front face of the box is 50.6 cm high (62 cm high at the back), 46 cm deep and 47.5 cm wide. There is a horizontal shelf 26 cm from the base, the upper surface of which is accessible to subjects by the upper entry points marked *A* and *B* in Figure 3.1. A slider and bead are attached to the underside of the shelf in the horizontal centre, running the full length of the shelf. The bead can be moved easily from either end of the slider. The midpoint of the bead is marked, so that its exact location can be read from a measuring tape attached to the slider-casing facing the experimenter at the back of the box. Subjects have access to the slider via the lower entry points (points *C* and *D* in Figure 3.1). The upper surface of the shelf has a number of holes which house the target (a black piece of doweling). On the front face of the box, 33.4 cm from the base of the box and in the horizontal centre, a prism or clear lens is placed behind a 4 cm diameter aperture. The bottom of the aperture is covered with dark card so that only the top of the aperture (1.3 cm at the highest point) is uncovered. The front face of the box is shown in Figure 3.1; the back face is closed above the shelf (controlling the visual background) and open below, making it possible for the experimenter to watch the responding hand in each trial. Target locations were chosen so that both peripheral targets were able to be seen through the prism without appearing to have moved beyond the walls of the box and it was ensured that each child was able to see the target at each location through the prism. Figure 3.2 shows a subject using the apparatus.

### 3.6.3 Procedure

#### Condition and trial order

Testing was split into plano (plain glass with no visual distortion) and prism blocks. All participants completed plano trials first ('plano' includes the PP condition, although it had no visual feedback). All participants completed three plano blocks (VP, VPP, PP), each with nine trials [3x3 unique targets: centre, right and left (both 6.5 cm from centre)]. Block order was counterbalanced within each subject group and trial order followed a fixed random order which was repeated for each condition. All subjects then completed two VPP blocks of six trials with the prism lens and the majority completed a third block of six. In all conditions the response was made proprioceptively without

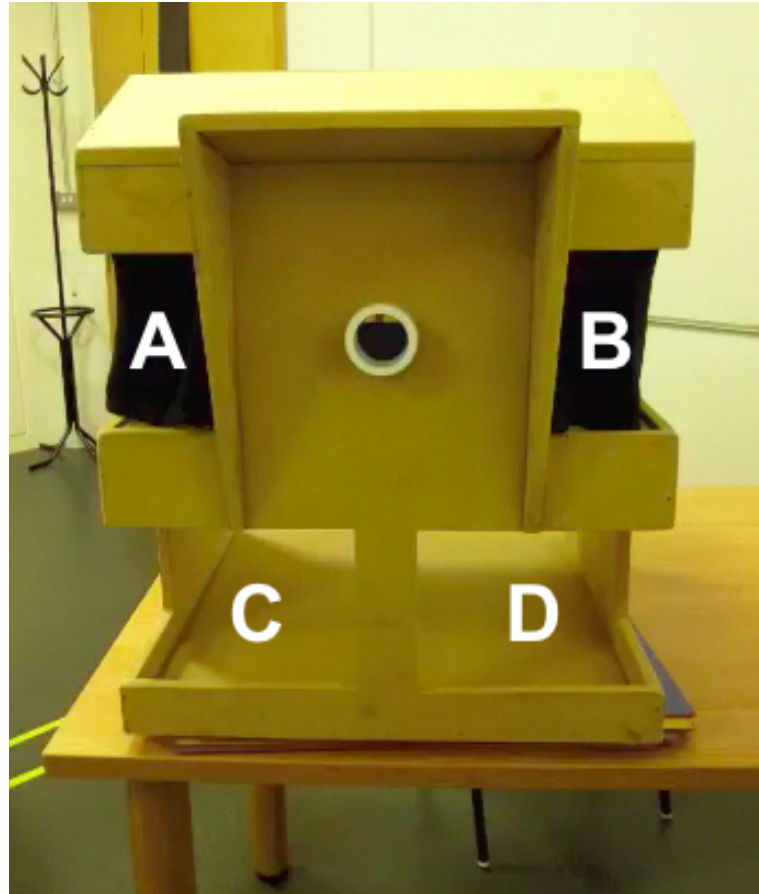


Figure 3.1: Front view of the apparatus, with the viewing aperture in the centre, two curtained entry points either side for access to the target, and two open entry points at the bottom for access to the slider and bead. A right-handed subject would use entry points A and D, a left-handed subject would use B and C.



(a) Subject looks through aperture while holding the target with the left hand.



(b) Subject holds the target with a pencil grip at the bottom.



(c) Subject moves the bead until it is underneath the target, while continuing to grasp the target and look through the aperture.

Figure 3.2: A right-handed subject completing the VPP condition with normal vision

vision of the hand moving the bead.

### **Plano trials**

Subjects sat in front of the box with the eyepiece is at eye level. They were instructed to place their dominant hand through the right or left hole in the bottom of the box and the hand was guided to the bead. The subject then looked through the eyepiece and slid the bead the length of the slider. All subjects were able to do this comfortably. Subjects were told that their task was to move the bead to directly under the target which they would either see, see and hold, or hold but not see. Subjects were asked to keep their head in the same place and look through the eyepiece at all times using only one eye, even if some of the trials did not involve vision. Subjects chose their preferred eye and were instructed not to change eyes at any point. The eyepiece was covered after every trial while the target was moved to a new location and it was ensured that the subject's hand was clear of the target area. The eyepiece was covered and the target removed and replaced even if two consecutive trials used the same target location. When the target was held, the cover was removed only once the target had been grasped. The bead was returned to the centre after each trial, although subjects were not made aware of this.

After the basic instructions subjects were then given condition-specific instructions, followed by 2 randomly assigned practice trials relevant to that condition.

*VP procedure:* A target is seen through the eyepiece. Subjects must move the bead to directly under the target. Their non dominant hand must remain outside the box.

*VPP procedure:* The experimenter guides the subject's non-dominant hand to the target and subjects are instructed to hold it at the bottom like a pencil. The eyepiece is uncovered and subjects are asked to keep their hand still on the target and, while looking through the eyepiece, move the bead to directly underneath it.

*PP procedure:* Subjects are told that they must hold the target as before, and look through the eyepiece, although this time they will not see anything as the aperture is covered from the back throughout the trial. Responses were made as above.

### **Prism trials**

Only the VPP condition was repeated with the prism lens. A 20 Diopter prism was placed behind the eyepiece in place of the plano glass. The instructions were the same as those given for the VPP plano condition. Subjects were not made aware of the switch from plano glass to the prism. Two or three blocks of six trials (1 x 6 unique trials) were completed. Prism direction and actual target location were counterbalanced in

each block (so that children who stopped after 2 blocks<sup>4</sup> had already completed fully counterbalanced trials).

The three target locations (right, centre, left) used in the plano conditions were used, alongside 2 new target locations at the midpoints between the central target and the two outer targets. The additional target locations were included so that the number of seen locations more closely matched the number of felt locations. The prism displaced targets 4.5 cm to the right or left, depending on the direction of the prism base. The outermost targets were only displaced towards the centre of the box, so that displaced vision did not extend beyond the boundaries of the box. The central target was displaced to the right and left, and the two additional locations were only displaced towards the edge of the box.

### 3.7 Results

#### 3.7.1 Recording responses

Responses were recorded by hand on the sheet shown in Figure 3.3. Motion tracking was not used as only movement endpoint was of interest and practical considerations favoured this manual approach. While it is acknowledged that this method does leave room for error, the apparatus was built so that endpoints could be easily recorded.

Figure 3.3: Recording sheet for spatial location matching

<sup>4</sup>Only one child opted to stop after two blocks.

## 3.7.2 Measures

### 3.7.2.1 Plano measures

*Absolute error:* Absolute error (error irrespective of direction) relative to target was calculated for each trial. Mean absolute error was calculated for each condition for each subject.

*Proprioceptive and visual benefit:* Proprioceptive and visual benefit were calculated to compare the effect of different plano conditions on absolute error.

Proprioceptive benefit = VP mean absolute error - VPP mean absolute error.

Visual benefit = PP mean absolute error - VPP mean absolute error.

A positive score is a benefit, meaning that the addition of the second cue aided performance; a negative score is a cost, meaning that adding the second source of sensory information hindered performance.

### 3.7.2.2 Prism measure: visual weighting

Visual weightings were calculated to investigate the perceptual weighting of the inaccurate visual information in the prism condition.

Weightings were calculated using the median constant error for each target in the VPP plano condition. As there are only three trials per target it is important not to allow extreme values (which could reflect inattention to the task) to bias the average, therefore median constant error was used.

For each target, the median constant error in the VPP plano condition was subtracted from its counterpart in the prism condition (on a trial-by-trial basis) to remove baseline (plano) error. As two new targets were introduced in the prism condition it was necessary to calculate an expected average baseline error. For prism targets immediately either side of the central target, the mean error from the two plano targets either side was used as an estimated baseline error. For example, the baseline plano error for the prism target immediately to the left of centre was the mean of VPP plano median error for the leftmost and central target.

Using the figures calculated above, the visual weighting for each trial was calculated by dividing the 'baseline-controlled error' by the signed visual displacement value (4.5 or -4.5 cm depending on the direction of the base of the prism). A mean visual weighting was then calculated for each subject.

### 3.7.2.3 Hypotheses

Assuming a double dissociation in the use of visual and proprioceptive cues in ASD and DCD, it is expected that absolute error will differentiate clinical groups as follows: greater error is expected in the ASD group in the VP condition compared to DCD; and greater error is expected in the DCD group when only proprioceptive information is available (the PP condition). Concerning cue benefit measures, visual benefit is expected to be significantly higher in the DCD group and proprioceptive benefit significantly higher in the ASD group. Finally, visual weighting in the prism condition is hypothesised to be larger in the DCD group than the ASD group.

### 3.7.3 The effect of target on error

The manipulation of target was not hypothesis-driven, but to avoid repeat-responding. Target was not fully analysed statistically, however data are shown in Figure 3.4. It is clear that there is little effect of target on response error and there does not appear to be any obvious differential effect between groups. Further analyses collapse data across targets.

### 3.7.4 Plano conditions

#### 3.7.4.1 Absolute error

Data were not normally distributed therefore mean absolute errors were entered into nonparametric analyses. Each condition was significantly different from each other (see Table 3.2): accuracy was greatest in the VP condition and poorest in the PP condition. Mann-Whitney U were used to explore group differences in each condition. Median error for each group and condition is shown in Figure 3.5. ASD were significantly less accurate than TD in the VPP condition ( $U = 241, p = 0.026$ ), however this would not survive a Bonferroni correction. The ASD group also had significantly larger errors than TD in the PP condition ( $U = 227.5, p = 0.014$ ). All other comparisons in the VPP and PP conditions were nonsignificant, and there was no effect of group in the VP condition (see Table B.1 in Appendix B).

Table 3.2: Wilcoxon pairs comparing each plano condition across all groups

Condition comparison	Post hoc result
VPP-VP	$Z = -3.42, p < 0.001$
PP-VP	$Z = -6.73, p < 0.001$
PP-VPP	$Z = -6.45, p < 0.001$

Note these results have not been corrected for multiple comparisons, however all would survive a Bonferroni correction with a new critical value of 0.017.

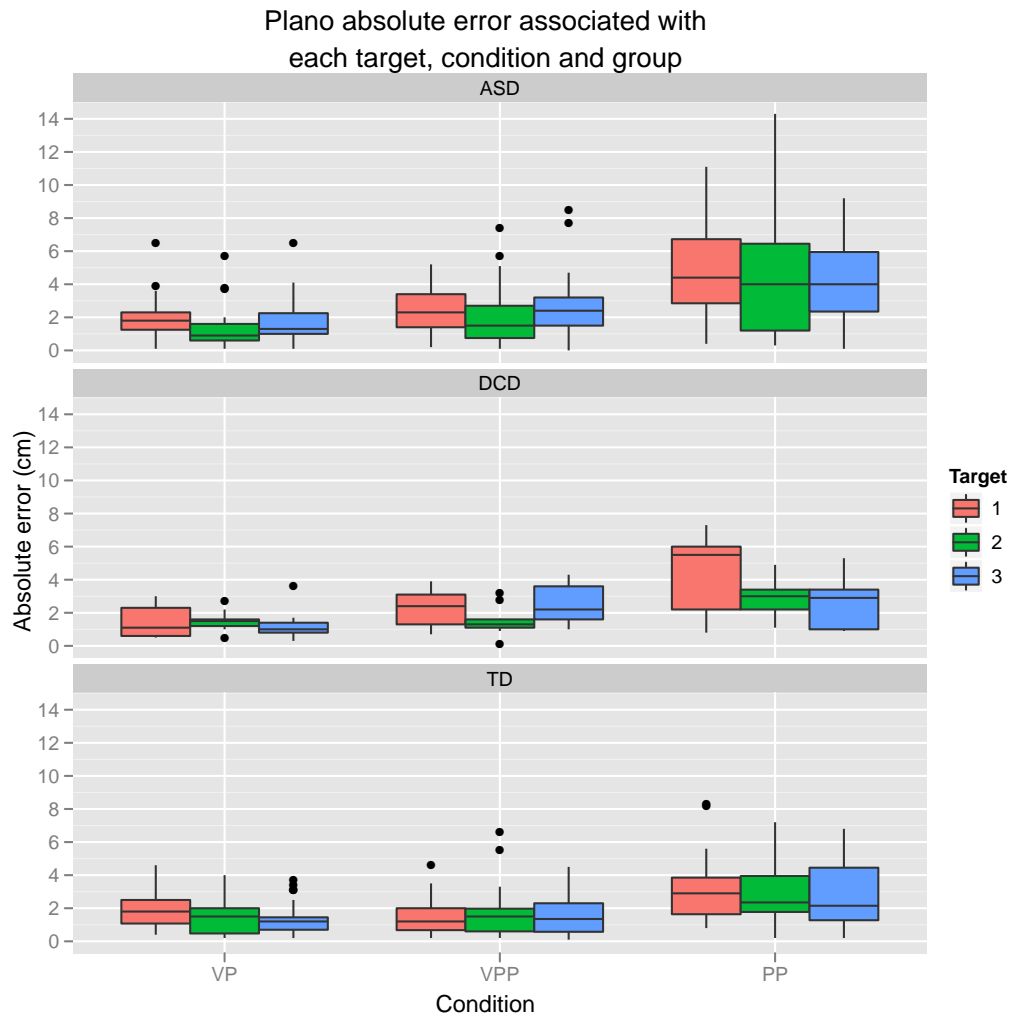


Figure 3.4: Range of median absolute errors in each plano condition for each target. Target 1 is on the subject's right, 2 is central and 3 is left. There is no clear effect of target on error in any condition or group.

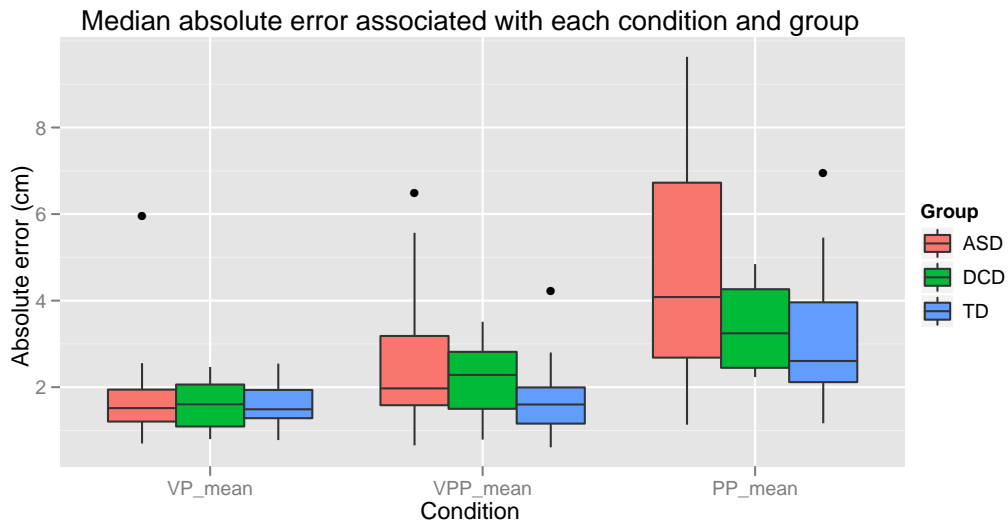


Figure 3.5: Absolute errors in each condition between groups. ASD are significantly less accurate than TD in the PP condition.

### 3.7.4.2 Proprioceptive and visual benefit

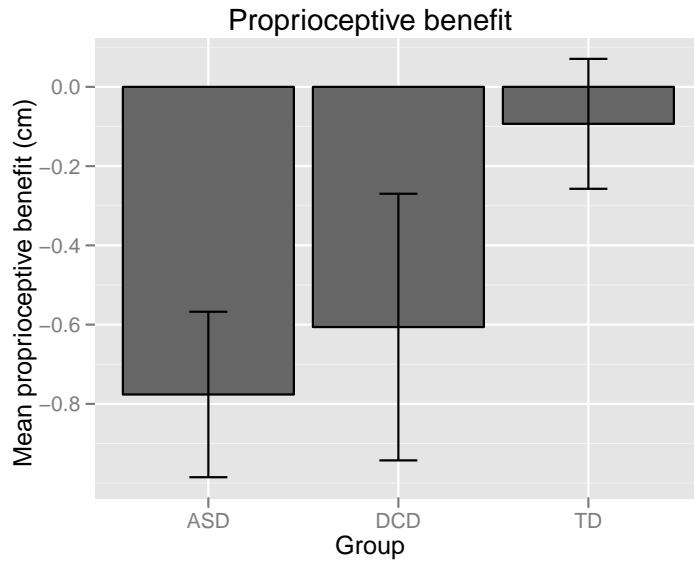
The majority of these data were normally distributed therefore visual and proprioceptive benefit were both compared between groups using a one-way ANOVA. Mean visual and proprioceptive benefits are shown in Figure 3.6. All groups show a proprioceptive cost, particularly the two clinical groups, and all groups show a visual benefit.

The effect of group approached significance for proprioceptive benefit ( $F(2, 61) = 3.09, p = 0.053, \eta_p^2 = 0.09$ ) and post hoc tests showed that the ASD group had a significantly larger proprioceptive cost than TD: mean difference=0.68, SE=0.28,  $p=0.017$ . Proprioceptive cost in the ASD group was not significantly larger than proprioceptive cost in the DCD group: mean difference (ASD-DCD)=-0.17, SE=0.39,  $p=0.669$ . Similarly proprioceptive cost in the DCD group was not significantly larger than the TD group: mean difference (DCD-TD)=-0.52, SE=0.40,  $p=0.199$ .

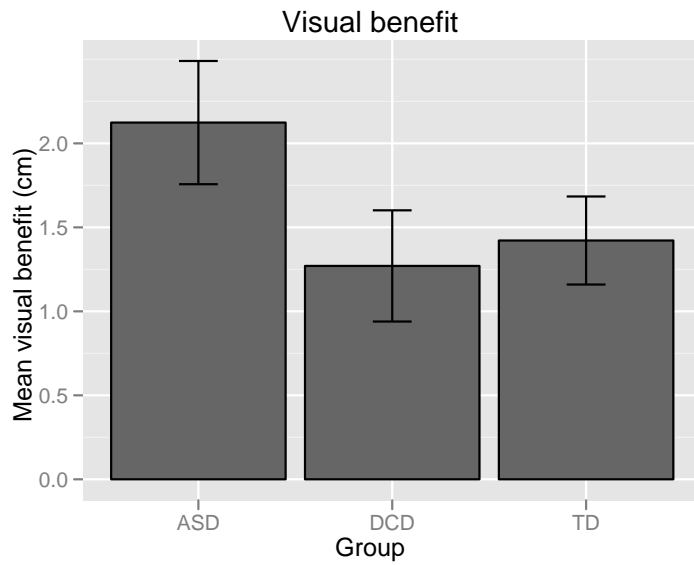
There was no significant effect of group on visual benefit:  $F(2, 61) = 1.60, p = 0.210$ . (The homogeneity assumption was violated, however Welch and Brown-Forsythe robust tests confirm the non-significant effect of group:  $F = (2, 28.35), p = 0.208$ ; and  $F(2, 55.94) = 2.23, p = 0.117$ .)

### 3.7.4.3 Plano conditions summary

When estimating the location of a target defined proprioceptively (either with or without additional visual information), the ASD group was less accurate than TD and showed a significant proprioceptive cost relative to TD. There was no significant difference between clinical groups. These findings are not in line with the proposed double



(a) Group mean proprioceptive benefit (error bars show SE).



(b) Group mean visual benefit (error bars show SE).

Figure 3.6: Mean proprioceptive and visual benefit. ASD show a significantly larger proprioceptive cost than TD. Groups are not differentiated by visual benefit.

dissociation.

### 3.7.5 Prism condition

Again, target was manipulated to avoid stereotyped responding, and choice of targets was not hypothesis-driven. To rule out the effect of target a median weighting was calculated for each subject for each target (mean weightings are shown in Figure 3.7). These data were normally distributed so were entered into a 3x6 ANOVA. There was no significant effect of target on visual weighting, and no significant interaction with group:  $F(2.92, 175.29) = 1.90, p = 0.132$ ; and  $F(5.84, 175.29) = 0.52, p = 0.792$  (both GG). Target will not be analysed further and data will be collapsed across targets.

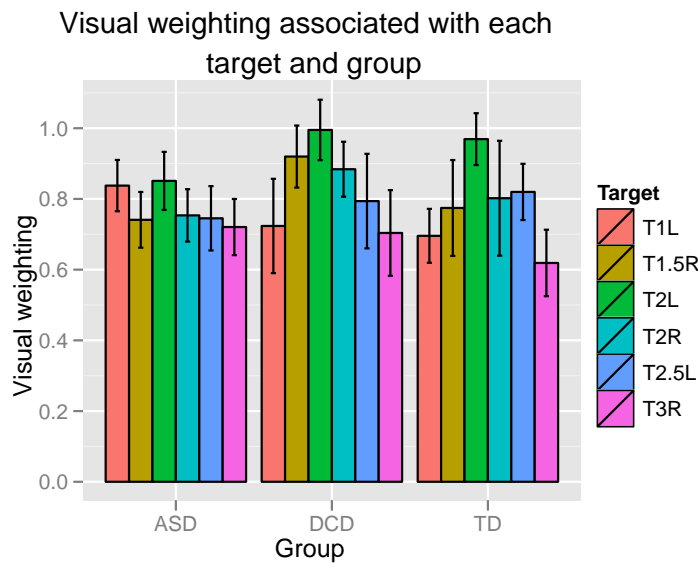


Figure 3.7: Visual weighting for each target in each group. There is no clear effect of target and no apparent interaction with group.

A visual weighting was calculated for each trial for each subject. A mean was then calculated for each subject. These means were not normally distributed, with outliers in both the ASD and TD groups. Median visual weightings are shown in Figure 3.8. A Kruskal-Wallis test revealed no significant effect of group on visual weighting:  $H(2) = 2.28, p = 0.321$ .

### 3.7.6 Plano conditions: MABC-defined groups

Comparing ASD and DCD highlighted no significant differences, mirroring findings in the previous chapter. As with imitation and cKAT analyses in the previous chapter, children with ASD who failed the MABC-2 (<15%ile) were compared with children with DCD. Where error in these two groups was not significantly different, these children

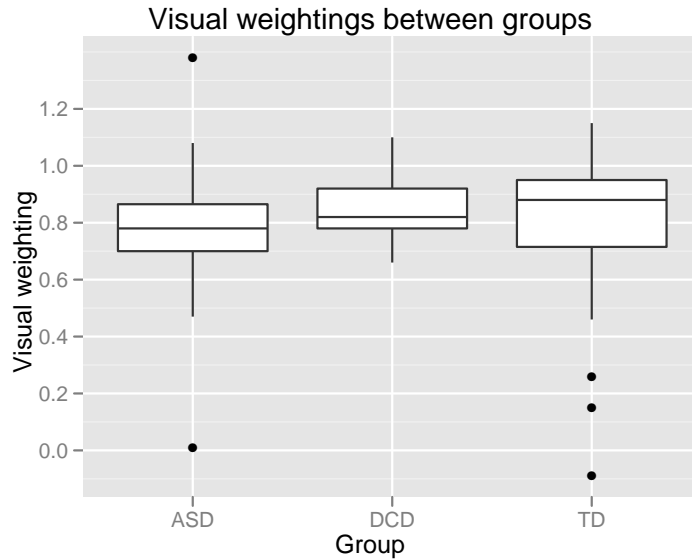


Figure 3.8: Visual weightings for each group. There is no significant effect of group.

were placed into a single ‘clinical motor deficit group’, and compared with the ASD ‘pure’ group (MABC-2 > 15%ile) and TD.

### 3.7.6.1 Absolute error

Nonparametric tests were used as data were not normally distributed. There was no significant difference in absolute error in motor-impaired children with ASD and those with DCD:  $U = 81.5, p = 0.446$  (VPP);  $U = 90, p = 0.695$  (VP);  $U = 61, p = 0.098$  (PP). The analyses reported above were re-run with the ASD pure, clinical motor deficit and TD groups.

Median error for VP, VPP and PP are shown in Table 3.3. Mirroring findings between ASD and TD in the initial analysis, the clinical motor deficit group was significantly less accurate than TD in both VPP and PP conditions:  $U = 239, p = 0.024$ ;  $U = 216.5, p = 0.008$ . The former result is not robust enough to survive a Bonferroni correction. All other comparisons were non-significant (see Table B.2 in Appendix B).

### 3.7.6.2 Proprioceptive and visual benefit

Neither proprioceptive nor visual benefit differed significantly between motor-impaired children with ASD and those with DCD ( $t(29) = -0.65, p = 0.520$ ;  $t(27.968) = 1.84, p = 0.076$ ), therefore analysis with the ‘clinical motor deficit’ was carried out.

Data were normally distributed, therefore groups were compared using two separate one-way ANOVAs. There was a significant effect of group on proprioceptive benefit, with the clinical motor deficit group showing a significantly greater proprioceptive cost than the TD group:  $F(2, 61) = 3.70, p = 0.031, \eta_p^2 = 0.11$ ; mean difference =

0.74,  $SE = 0.27$ ,  $p = 0.009$ . The motor deficit group also showed greater proprioceptive cost than the ASD pure group, however this was not statistically significant: mean difference = 0.43,  $SE = 0.38$ ,  $p = 0.264$ . The difference in proprioceptive cost between ASD pure and TD was also not significantly different: mean difference (ASD pure-TD) = -0.31,  $SE = 0.40$ ,  $p = 0.434$ .

There was no effect of group on visual benefit:  $F(2, 61) = 0.73$ ,  $p = 0.488$ . Again these results mirror the ASD/TD findings in the initial analysis.

Table 3.3: MABC-defined groups' mean absolute error for each plano condition

Condition	Group	Median (range)
VP	ASD 'pure' ( $n=9$ )	1.5 (1.01-2.19)
	Clinical motor deficit ( $n=31$ )	1.56 (0.7-5.97)
	TD ( $n=24$ )	1.49 (0.78-2.54)
VPP	ASD 'pure' ( $n=9$ )	1.69 (1.12-2.88)
	Clinical motor deficit ( $n=31$ )	2.08 (0.66-6.48)
	TD ( $n=24$ )	1.6 (0.61-4.23)
PP	ASD 'pure' ( $n=9$ )	3.54 (1.13-7.44)
	Clinical motor deficit ( $n=31$ )	4.19 (1.2-9.63)
	TD ( $n=24$ )	2.61 (1.17-6.94)

The difference between each condition within groups is statistically significant [all  $p < 0.001$  (uncorrected)]. Errors in the clinical motor deficit group are significantly larger than those in TD for the VPP and PP conditions.

### 3.7.7 Prism condition: MABC-defined groups

Visual weighting did not differ significantly between motor-impaired children with ASD and children with DCD ( $U = 95.5$ ,  $p = 0.881$ ), therefore analysis with the 'clinical motor deficit' group was carried out.

Data were not normally distributed therefore nonparametric tests were performed. There was no significant effect of MABC-defined group on visual weighting:  $H(2) = 3.71$ ,  $p = 0.156$ . Median visual weightings are shown in Figure 3.9. Visual weightings in the ASD 'pure' group appear to be considerably lower than the TD and clinical motor deficit groups on average. While this is in line with the hypothesised double dissociation, the differences are not statistically significant, even without correcting for multiple comparisons ( $U = 62$ ,  $p = 0.065$ ; and  $U = 88.5$ ,  $p = 0.099$  for ASD 'pure' vs. TD, and ASD 'pure' vs. clinical motor deficit respectively). While there may be a trend towards a difference between ASD 'pure' and TD there is insufficient data at present to conclude this with any certainty. This is also true for ASD 'pure' and the clinical motor deficit group: despite a lower weighting in the ASD 'pure' group the difference is not significant. In both cases a relatively small ASD 'pure' group coupled with high variability are likely limiting the power of these analyses.

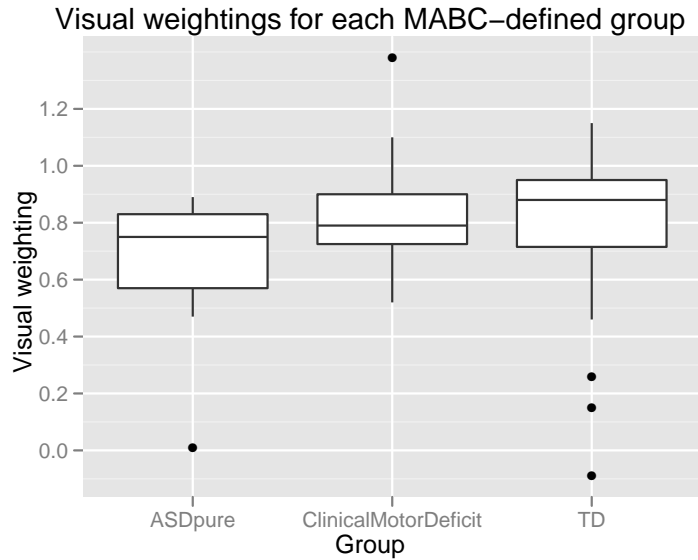


Figure 3.9: Visual weightings for MABC-defined groups

### 3.8 Discussion (Experiment 1)

The aim of this study was to assess the relative weighing of vision and proprioception in children with ASD, DCD and typical development. Previous literature has focused on either ASD or DCD in isolation, and this study aimed to directly compare the two in a controlled manner. Previous literature pointed to a possible double dissociation between ASD and DCD, with ASD favouring proprioceptive cues and DCD favouring visual cues (Rosblad & von Hofsten, 1992; Haswell et al., 2009). Contrary to this apparent double dissociation, the two clinical groups were not differentiated according to proprioceptive benefit, visual benefit or visual weighting. The ASD group did show a significant proprioceptive cost relative to TD [in contrast to a clear proprioceptive benefit in ASD reported by Ranta & Mostofsky (2011), and supporting Weimer et al.'s (2001) much less robust suggestion of a proprioceptive deficit].

It was found in the previous chapter that the two clinical groups were, on average, equivalent in terms of performance on the movement battery (MABC-2), supporting the case for comorbidity, as is now reflected in the revised edition of the DSM (DSM-5: APA, 2013). When considering the motor impaired clinical group (all DCD children, and ASD children who failed the MABC-2) compared to TD, those with motor difficulties had a significantly greater proprioceptive cost than TD, supporting Sigmundsson et al. (1999). The finding that those children with ASD and DCD who failed the MABC-2 weight vision similarly also supports the suggestion that motor impairment more generally is associated with a reliance on visual cues. Additionally, when those with motor difficulties were compared to children with ASD and spared motor skills, a possible difference did emerge in visual weightings, with a lower reliance on visual cues

in the ASD ‘pure’ group. This supports the proposed dissociation in part, however this difference did not reach statistical significance and it is suggested that more extensive investigation of ASD subgroups would be of interest. Unfortunately, only nine children over 8 years of age were able to be classed as ASD ‘pure’, and within this group there was still a large degree of variability. Assuming a proprioceptive bias exists in ASD and can be captured with this kind of task, it might be that a greater sample size would have provided more evidence for this.

These results raise the question of when the vision/proprioception dissociation might arise, and what could have caused the differences in results presented here and those reported previously (particularly reports of an ASD proprioceptive bias). Is it that ASD is associated with an increased reliance on proprioception only when there is not comorbid DCD? This is difficult to ascertain from the small sample of ASD ‘pure’ subjects here, or from previous studies which do not always report motor ability (and when they do the MABC is rarely used). Fuentes et al. (2011) found no difference between ASD and TD in the use of proprioception, despite those in the ASD group having comorbid motor difficulties according to the PANESS. This finding certainly does not support the suggestion that a proprioceptive-reliance in ASD only applies to those cases without comorbid motor deficits. However, the PANESS and MABC have been found not to significantly correlate, making comparisons between the current findings and previous work using the PANESS uninformative (Thompson, Clark, Whitall & Roche, 2008).

Results are difficult to reconcile with the previous literature which prompted the investigation of the proposed dissociation. However a comparison of the results with previous studies using a similar task are more informative. As there was no significant difference between DCD and either ASD or TD on any measure the results do not directly support or contradict any of the previous studies using this task. However when considering the clinical motor deficit group, the results do support Sigmundsson et al. (1999), who found proprioceptive deficits in DCD, and indirectly support Smyth & Mason (1998), who found a visual bias in DCD. However it suggests that findings in these studies may not be specific to DCD, but can be applied to motor impaired children more generally, as reported by Rosblad & von Hofsten (1992).

This study intended to directly examine the double dissociation suggested from reading prior literature. However, it may be the case that the findings from which the double dissociation was identified were from too diverse an array of tasks, age groups, sensory manipulations and subject identification processes. It is possible that if tasks described in previous studies were re-run with both ASD and DCD groups the dissociation would not be apparent, as was the case here. Alternatively, it is possible that the perceptual nature of the task is responsible for the lack of dissociation between groups. The differential use of visual and proprioceptive cues may be more apparent in tasks involving more purposeful movements. This is investigated in the second half of this chapter and Chapter 4.

While the hypothesis was not confirmed, the results of the present study are not thought to have been affected by poor experimental design, as the task has been widely used previously. In addition to the use of a well used task, the study is considered to have a number of other strengths. A major strength is the use of a well-defined and relatively large sample of children with ASD. The majority of similar studies involve groups numbering fewer than fifteen children, sometimes as few as five, which is problematic given the known heterogeneity of the disorder. The DCD group is of a similar size to previous studies and at times considerably larger, although recruitment difficulties meant this group is much smaller than the ASD group. However this is not thought to impact too negatively on results as the question here is one of motor skill, so the DCD group should be less varied in this respect than an ASD group. As discussed previously, the relatively small ( $n=9$ ) ASD ‘pure’ group made it difficult to fully assess the secondary hypothesis regarding MABC-defined groups. Practical restrictions meant it was not possible recruit more children for this subgroup.

In addition to the relatively larger than usual sample sizes and the use of medical records in the identification of subjects, the use of age criteria informed by the typical trajectory of visual and proprioceptive cue weightings throughout childhood, a detail which is often overlooked, is an additional strength of the present study. This negates the concerns of age effects such as those in Smyth & Mason (1998) and Mon-Williams et al. (1999).

The use of a full movement assessment which is often used by clinicians in DCD diagnosis has proved useful in further investigating the alternative dissociation hypothesis. Previous studies tend not to provide detailed information about motor skills so it is difficult to ascertain whether or not proprioceptive bias in ASD in previous studies is true in all cases of ASD, just those with motor deficits, or only those without motor deficits. Results from this study suggest that comparing ASD with and without motor deficits deserves further investigation.

Future research would therefore benefit from targeted recruitment of children with ASD without clinical motor difficulties. However, the practicalities of this are a major problem: firstly, children with ASD without comorbid DCD appear to be a relative rarity (3 in 10 in this case); and secondly, unless a child is presented to a medical team because of poor motor skills a movement assessment is not a routine part of the assessment, even when motor deficits might be apparent. It is possible that the large proportion of children with ASD failing the MABC-2 is partly due to a selection bias on the part of parents choosing whether or not to take part, as the invitation letter made the focus on motor difficulties clear. With no firm statistics on the prevalence of ASD/DCD symptom-overlap (and no published comorbidity rates following the recent changes to diagnostic criteria) it is unclear how representative the ASD group is in this case with respect to movement ability. From discussions with clinicians it appears however that the sample may be largely representative.

Having failed to uncover the hypothesised double dissociation between ASD and DCD in a largely perceptual task, the groups are now compared in a task which requires a more natural pointing action.

### **3.9 Experiment 2: Vision and proprioception in action (mirror reach)**

The evidence for a possible double dissociation between the use of vision and proprioception in ASD and DCD has been reviewed at the start of this chapter. Following on from the null findings in the first experiment, it is possible that a dissociation in sensory processing is better captured in a task involving an action. This is investigated in the second half of this chapter, which reports an investigation of the use of vision and proprioception in a reaching action following visual displacement of the hand via a mirror. If a dissociation exists it is expected that children with ASD will show a relative proprioceptive bias, and DCD a relative visual bias. However given the results of the previous study, it is possible that those ASD with spared motor skills will show evidence of a proprioceptive bias, while children in either clinical group with motor difficulties will again tend to rely on vision.

Mirrors have been used frequently to investigate the use of vision and proprioception in basic reaching movements in typical subjects. When one hand is placed either side of a midsagittally placed mirror, with the reflective side facing one of the two hands, the mirror image can be mistaken for the hidden hand. When the two hands are different distances from the mirror people tend to misjudge where their hand is as they incorrectly make use of the incongruent visual information. This causes them to misreach a target with their unseen hand (Holmes et al., 2004). This effect of the mirror is an example of visual capture, where we are compelled to process and act upon presented visual information (Holmes, Snijders & Spence, 2006; Holmes & Spence, 2005). Such misreaching has been found to be greater when the hands are placed further apart and the visual-proprioceptive discrepancy is larger (Holmes et al., 2004). However this was found in both a mirror and no-vision condition, so the result may be due to poorer proprioceptive acuity when reaching further from the body midline (Wilson, Wong & Gribble, 2010), rather than visual-proprioceptive congruency effects. It should be noted that reaching errors following the mirror illusion tend not to show 100% visual capture: Holmes et al. (2004) found that reaches showed only a 30%-42% bias towards vision. The authors suggest that some other source(s) of information, such as proprioceptive cues or cognitive influences, are also involved. It is these factors that prevent the reach from being programmed solely with respect to the visual information.

The mirror illusion is often elicited in one of two ways: watching the hands in

a passive (still) position, or an active position (tapping). Brief passive exposure has previously failed to elicit significant reaching errors in the mirror condition (Holmes et al., 2004; Holmes & Spence, 2005), however in each case viewing duration was around only 25% that of the active viewing condition. It is likely that this duration was insufficient as reaching errors have been found to increase with exposure duration in both conditions (Holmes & Spence, 2005). After 12 seconds of visual exposure, Holmes et al. (2006) found that reaching errors following passive and active exposure in the mirror were statistically equivalent, although asynchronous active exposure (tapping out-of-phase) did result in significantly smaller errors. This is because the illusion depends on the mirror image appearing to be the hidden hand, so incongruence between visual and proprioceptive representations of the hand hampers the illusion. (This is also true for the rubber hand illusion, described in Chapter 5.)

Few studies have used the mirror illusion with children. Of the small number of examples found, only one used a task similar to the reaching paradigm described above. Bremner, Hill, Pratt, Rigato & Spence (2013) used the task with TD children, who tapped their hands synchronously while looking in the mirror or at an opaque divider. In this case children were allowed an illusion induction period and reach training before beginning experimental trials. The induction period allowed children to look in the mirror until the reflection felt like the hidden hand. They were then trained to reach within 2 cm of the target from a congruent position (a reach straight ahead). Unfortunately there was no analysis of any differences in accuracy from different starting positions or visual condition (mirror/ no mirror) and there are no details of the duration of the tapping phase. The use of repeated practice trials in this study is potentially problematic, as this primes a straight reach, perhaps exaggerating visual capture. Assuming that visual capture was not significantly exaggerated by training, misreaching in the incongruent condition suggests that this task is suitable for young children, although there do not appear to be any studies using this task with clinical groups of children.

The present study uses a similar paradigm to that described above (see also Holmes et al., 2004 and Holmes & Spence, 2005), to assess the effect of incongruent visual and proprioceptive information in a simple reaching task in children with ASD, DCD and TD. The design and procedure is very similar to that reported by Bremner et al. (2013), although in the present study children will not be given practice trials to avoid the possible priming problem highlighted above. The present study does not use a no-vision control condition, due to time restrictions. In this case the control task is the congruent condition, where both hands are equidistant from the mirror. A major difference between the current study and some previous similar studies is the positioning of the target above the mirror, rather than showing the target in the mirror (e.g. Holmes et al., 2006). Having to break visual attention and attend to the target above the mirror is not ideal so this alternative approach was considered, however it was anticipated that

some children would falter when they experienced the sensation of looking in the space their hand apparently occupies, but not being able to see it move as they point to the target in the mirror. As having the target above the mirror has proved successful with TD children (Bremner et al., 2013) it is assumed that this target position should not adversely affect performance.

It is hypothesised that if the proposed ASD/DCD double dissociation regarding the use of visual and proprioceptive cues is correct, the ASD group should produce more accurate reaches following the mirror illusion compared to DCD, who should be more swayed by the visual representation of their arm's positioning. Following results from experiment one and the possibility that the dissociation relates more to 'pure' ASD versus motor-impaired children, an additional hypothesis would expect more inaccurate reaches in any motor-impaired child, and more accurate reaches from those children with ASD who passed the MABC-2.

## **3.10 Methods**

### **3.10.1 Adult pilot study: Methods, results and discussion**

As discussed above, the task tends to use one of two conditions for the non-responding hand: the index finger is tapped synchronously with the index finger of the hidden hand; or both hands are kept still. It is likely that given sufficient time, passive viewing is sufficient to elicit a strong illusion. As this task is being developed for use with children with ASD and DCD, it was noted that performing a tightly coordinated bimanual movement could be problematic for these groups. However, as tapping conditions have in some cases been found to induce a stronger illusion, a simpler alternative was tested in a short pilot study with typical adults. Results from this pilot informed the design of the child study.

#### **3.10.1.1 Design**

This task employed a 2x2 mixed design, with visual input as a between-subjects factor (passive hand or tapping hand), and incongruence condition (near/far) as a within-subjects factor. (The congruent condition is not analysed as it is factored into responses in the incongruent conditions.)

#### **3.10.1.2 Subjects**

Ten adults (7 males) aged 22-43 (mean age 27 years) with normal or corrected-to-normal vision, and right hand dominance participated. Both vision status and hand dominance were self-reported. Half were assigned to the passive condition (4 males) and half to the tapping condition (3 males): age did not differentiate these groups ( $t(8) = 0.66, p = 0.529$ ). Participants were not paid for their participation. Neurotypical adults were used due to the limited availability of child subjects for pilot work.

### 3.10.1.3 Apparatus

Reaches were made in a wooden box with two open faces (front and back), measuring 76.5 x 45 x 31 cm (length x depth x height). A plastic mirror was located in the horizontal midpoint of the box, with the reflective side facing left. On the top of the box to the left of the mirror, the top comprised of two equally sized lids: the right most of these was lifted to allow for vision of the mirror (see Figure 3.10). Marks on the floor of the box (visible only to the experimenter) indicated the 4 starting locations: 7, 12 and 17 cm from the mirror for the right hand and 12 cm from the mirror for the left hand. Each starting position was 17 cm from the front edge of the box. The target was a block of wood on a slider at the top of the box. The target was in a fixed position in line with the 12 cm mark to the right of the mirror. Motion tracking was carried out using a TrakSTAR electromagnetic tracking system (Ascension Technology, Burlington, VT), the magnet for which was located in a fixed position at the back of the box.

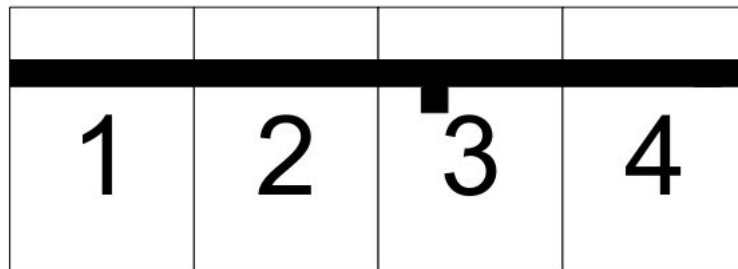


Figure 3.10: Mirror reach apparatus from above. The mirror is between compartments 2 and 3, with the reflective side facing into compartment 2. The left hand is placed to the left of the mirror and lid 2 is removed to allow for a view of the mirror. The right hand is placed in the right compartment and reaches to directly underneath the target bead seen here on the slider.

### 3.10.1.4 Procedure

Subjects sat on a height adjustable chair in front of the apparatus, with the mirror facing the left side. Participants placed one hand on either side of the mirror inside the box and TrakSTAR markers were placed on the nail of each index finger and secured with tape. Subjects were told to hold their hands with the fingers and thumb together as much as possible, and keep their hand still or tap the hands in synchrony, flexing at the wrist. For those subjects in the tapping condition the wires from the markers were attached to the top of the box to avoid noise from the cables hitting the bottom of the box when the hands were tapped. This noise would offer subjects an additional cue to the synchrony of their movements and could either enhance or diminish the illusion, depending on the synchrony of the two hands. While the two hands were covered by the top of the box the experimenter placed the hands in position. The left hand

was always positioned 12 cm from the mirror and the right hand was placed in one of three positions [7, 12, 17 cm to the right of the mirror: incongruent (near), congruent, incongruent (far)] in a fixed random order. The incongruent (near) starting location was far enough from the divider that the thumb did not touch it (and thereby provide additional tactile information of hand location). The left hand position allowed for a clear view of the reflection and also allowed for a peripheral view of the left hand.

Subjects watched the reflection of their hand (passive or tapping) in the mirror for 15 seconds until the experimenter instructed them to “stop, look and reach”. Subjects stopped looking in the mirror, looked up to the target block and then reached to the floor directly below the target with the right index finger. Adherence to the requirement to look in the mirror was monitored visually by the experimenter and trials were repeated if this instruction was not adhered to (this happened very rarely). Subjects completed 15 trials (5x3 unique trials) and were not given feedback during the task.

### **3.10.1.5 Results**

The movement was considered to have begun once the velocity of the reaching hand exceeded 50mm/s, and ended once velocity fell below 50mm/s. In order to control for differing end locations in the depth dimension, the angle of response was measured rather than the response on the x-axis. A response angle was calculated for every trial. The congruent trials were expected to have relatively small response angles as the angular distance between the start position and the target is 0°. Kinematic variables were not considered as the purpose of the pilot was to assess the effectiveness of a passive visual condition in eliciting the mirror illusion as measured by response angle. Additionally, differences in kinematic variables will not be assessed in the child study. Differences in kinematics such as MT and PS in children with motor difficulties may be due to motor deficits in general, or may be due to the uncertainty about hand position due to the incongruence effect in the mirror illusion (Palmer, Paton, Hohwy & Enticott, 2013), and the present study is not designed to assess such differences.

For each subject, the mean signed response angle was calculated for the congruent condition. Mean (SD) signed response in the congruent condition was similar between tapping and passive groups: 0.86° (7.14) and 1.92° (2.92) respectively. Congruent mean angle for each subject was then subtracted from signed response angles on incongruent trials to correct for any baseline directional bias. A signed mean for each incongruence condition was calculated for each subject. In order to assess the effect of starting position it was necessary to multiply the incongruent far responses by -1 so that in both incongruent near and far trials a positive response is in the expected direction. The larger the final response angle, the less visual capture. Only incongruent trials were used in analysis as congruent trials had already been factored into incongruent responses.

Corrected mean responses (factoring in congruent accuracy) were entered into a 2x2 repeated measures ANOVA comparing viewing condition (passive and tapping) and incongruent starting position (near and far). Group means are shown in Table 3.4. There was no significant effect of viewing condition ( $F(1, 8) = 0.80, p = 0.397$ ), suggesting that a passive condition is suitable for use in future studies. Similarly there was no effect of incongruent starting position [ $F(1, 8) = 0.61, p = 0.457$  (GG)], showing that rightward and leftward movements were equivalent. As would be expected, the interaction effect was also not significant:  $F(1, 8) = 0.09, p = 0.771$ .

Table 3.4: Mean (SD) response angle for each congruent condition and corrected mean responses for each incongruent start position and condition

Start position	Condition	Mean (SD) response angle
Congruent	Passive	1.92 (2.92)
	Tapping	0.86 (7.14)
Incongruent near	Passive	6.01 (5.85)
	Tapping	9.82 (11.22)
Incongruent far	Passive	11.25 (2.43)
	Tapping	12.14 (11.69)

The required angle was 20°. Neither the difference between congruency conditions nor the difference between viewing conditions was significant.

### 3.10.1.6 Discussion

The null effect of viewing condition on reach angle suggests that passive viewing of the hand for 15 seconds is sufficient to elicit a visual-proprioceptive conflict strong enough to affect subsequent movements. The small errors in the congruent condition compared to the incongruent conditions suggest that the mirror illusion (visual/proprioceptive incongruence) was successful, although there is no way to be certain that the mirror had an effect as there was no opaque divider condition. However as this paradigm has been used previously it is expected that had an additional no-mirror condition been included it would have verified that the results are due to the sensory incongruence.

Controlling the starting position of the index finger following tapping was problematic in some cases, as unlike finger tapping, only the wrist was anchored in a fixed position. This lack of control is a practical reason to favour passive viewing. Considering the difficulty children with motor deficits may have with synchronous tapping, this also supports the use of passive viewing.

The null effect of incongruent target (near and far) on response angle suggests that all responses for incongruent trials can be combined in future analysis. For this reason the child version of the task will involve an equal number of congruent and incongruent trials, with half of the incongruent trials being near and half far.

### 3.10.2 Child study

#### 3.10.2.1 Subjects

All children had taken part in the studies detailed in Chapter 2 and in the first half of this chapter. This task was completed as part of a follow-up session alongside tasks reported in Chapters 4 and 5. Children participated in the follow-up session between 2 and 18 months after participating in the first round of experiments. All subjects were aged 8 and over in line with the previous study. All but one child (a member of the DCD group) was male. Two children with ASD were not able to complete the task due to difficulties understanding the instruction to reach to the floor of the box or a reluctance to watch the mirror, and there were data recording errors for three children with ASD. Mean age, IQ rank, MABC-2 percentile rank, SRS and DCDQ-07 scores for each group is given in Table 3.5. Groups were age- and IQ-matched:  $F(2, 32) = 2.75, p = 0.079$ ; and  $F(2, 32) = 0.55, p = 0.584$ ). There was a significant difference between groups on SRS scores, MABC-2 percentile rank and DCDQ-07 scores ( $F(2, 30) = 56.62, p < 0.001$ ;  $F(2, 32) = 17.86, p < 0.001$ ; and  $F(2, 35) = 51.90, p < 0.001$  respectively). All groups differed significantly from each other on SRS score, with lowest scores in TD and highest scores in ASD. Both clinical groups had significantly lower MABC-2 and DCDQ-07 scores than TD (all  $p < 0.001$ ).

Table 3.5: Subject demographics (excluding those children who either attempted the task but did not complete it, children whose data were not recorded, and TD children who failed the MABC-2)

	ASD ( $n=18$ )	DCD ( $n=6$ )	Clinical motor deficit ( $n=21$ )	TD ( $n=11$ )
Age	12.11 (1.88)	11.00 (2.53)	11.67 (2.06)	11.27 (1.19)
IQ %ile	45.44 (29.11)	49.17 (42.99)	40.67 (29.83)	60.09 (21.06)
MABC-2 %ile	12.73 (21.60)	4.35 (3.96)	4.87 (3.65)	54.09 (22.44)
SRS	114.50 (23.61)	62.17 (31.86)	97.16 (36.52)	15.82 (18.90)
DCDQ-07	33.47 (10.68)	31.8 (11.17)	32.11 (10.07)	67.91 (9.15)

Values shown are mean (SD). Note that sample sizes differ from those in Experiment 1 in this chapter as not all children were able to return for the follow-up session. There are significant group differences for the MABC-2, DCDQ-07 (ASD and DCD differ from TD) and SRS (all groups differ from each other).

#### 3.10.2.2 Procedure

Children completed a task similar to the pilot described above. Viewing condition was not manipulated in this case, and only viewing of the passive hand was tested. The number of unique trials was also changed from the pilot, with eight congruent trials, and four each for near and far incongruent trials, presented in a fixed random order. The rest of the procedure is as described previously.

### 3.11 Results

Kinematic variables are not included in analysis as per the initial pilot study. The main variable is the angle of response. It is hypothesised that the response angle will be closer to the required angle in the ASD group compared to the DCD group if the proposed double dissociation is true. Alternatively, given the difference in visual weighting in the previous experiment between ASD children with and without motor impairment, it is possible that ASD ‘pure’ children will produce a more accurate response angle than motor impaired children in both clinical groups.

In order to factor in directional bias and accuracy in the congruent condition, baseline-corrected responses were calculated for each subject as per the pilot study. The majority of subjects had a mean response in the expected direction for the illusion. Mean response angles for diagnostic and MABC-defined groups are shown in Table 3.6.

Response angle (and therefore error) is almost identical in ASD, DCD, TD and the clinical motor deficit group (comprised DCD and 15 of the 18 ASD subjects). The target angle from the two incongruent starting positions was  $20^\circ$ , so while subjects are swayed by the mirror image to an extent, they are compensating enough to reach at an angle that is at least 60% that required to reach the target. The ASD ‘pure’ group (which is too small for formal analysis) appears to show greater resistance to the mirror illusion and reach at an angle that is 82% of the required angle.

Mean response angle for the congruent condition was similar between the three main groups: 10.39 (6.67), 15.32 (10.60), and 7.86 (4.74) for ASD, DCD and TD respectively. This supports the exclusion of the congruent condition in analysis.

Mean responses for ASD, DCD and TD (collapsing across targets) were entered into a one-way ANOVA. As expected from the near-identical mean angles there was no significant effect of group on mean response angle:  $F(2, 32) = 0.06, p = 0.943$ . Comparing the DCD group with the motor impaired ASD subgroup, there was no significant difference in mean response angle:  $t(19) = -0.25, p = 0.807$  (2-tailed). This allowed for the formation of the ‘clinical motor deficit group’ as per previous studies. There was no significant difference between the clinical motor deficit group and TD:  $t(30) = 0.03, p = 0.977$  (2-tailed).

Table 3.6: Mean response angle (SD) collapsed across target

	Mean (SD)
ASD ( $n=18$ )	12.75 (4.57)
ASD ‘pure’ ( $n=3$ )	16.47 (1.24)
DCD ( $n=6$ )	12.58 (5.12)
Clinical motor deficit ( $n=21$ )	12.17 (4.66)
TD ( $n=11$ )	12.11 (5.13)

### 3.12 Discussion (Experiment 2)

This study investigated visual capture in children with ASD, DCD and TD during a simple reaching task. Accuracy in the task depends largely on the degree to which the mirror image of the arm influences the perceived starting location of the arm. It was hypothesised that if ASD is associated with a relative bias for proprioceptive over visual cues this group would make more accurate reaches: if DCD is associated with an increased reliance on visual cues at the expense of proprioceptive cues it was expected that this group would show greater visual capture and less accurate reaches. Following results from the spatial location matching task described in the first half of this chapter, an additional hypothesis suggested that children with ASD and motor deficits would behave similarly to those with DCD. Both would be expected to rely on visual cues to a greater extent than ASD children without motor difficulties. As this was a follow-up study not all subjects were able to return for testing and the distribution of MABC-2 scores was too heavily skewed toward the motor deficit range to allow for a complete investigation of this additional hypothesis.

As the average response angle from incongruent starting positions was only 60% of the angle required for an accurate reach it seems that the illusion was effective. The mirror illusion caused equivalent degrees of visual capture in each developmental group and the use of vision and proprioception did not differentiate children with ASD and comorbid motor difficulties from those with DCD. However it is still unclear whether ASD without comorbid DCD is associated with a greater reliance on proprioceptive cues in action. The three subjects in the ASD ‘pure’ group were more accurate than other groups, although it was not possible to perform statistical analyses with such a small sample.

One potential problem with this task is the varying approaches used by different children. While ideally the reach should be made quickly in a single smooth movement, so as not to break the visual capture by looking away from the mirror for too long, this was not always possible, especially in the clinical groups. Despite instructions and demonstrations some children persisted in being relatively slower to initiate their movement (noted through experimenter observations). This may reflect a level of confusion as to the arm’s positioning, or may instead (or also) reflect a general slowness to initiate movements (previously reported in both clinical groups: Donnellan, Hill & Leary, 2013; Johnston et al., 2002, Glazebrook et al., 2006). Individual differences also arose in the style of movement. Some children were able to complete a smooth single pointing movement; others consistently corrected the end point once the finger had landed; and others choose to slide the finger across the floor rather than lifting to point. Some children would slide their hand towards the congruent starting position before moving forward, having realised that there was more than one starting position. When these children asked if there was more than one starting position this was neither confirmed nor denied. Others would move their whole body to the side in order to line-up

with the target. These techniques were discouraged although some children persisted in using them either through a desire to maintain higher accuracy (a recurring theme in the clinical groups) or they were simply unable to modify their approach. Finally, some children did not produce the expected velocity profiles and it was clear that some children considered the task to be self-paced. This supports the decision to consider only response angle in analysis.

Results very clearly show typical accuracy in the clinical groups. The almost identical performance by each group suggests that the null effect of group is not due to a lack of statistical power. However with so much variation in the children's approach to the task it is unclear how suitable this task was to investigate the vision/proprioception dissociation.

### 3.13 General discussion

This chapter has reported two tasks assessing the relative use of visual and proprioceptive cues in perception and action in children with ASD, DCD and typical development. It was hypothesised that children with ASD and DCD would show a double dissociation in their use of visual and proprioceptive cues. Previous research has tended to find a relative bias for proprioceptive cues in ASD (e.g. Masterton & Biederman, 1983; Haswell et al., 2009), while those with DCD tend to show a reliance on visual cues, although the pattern seems less clear for DCD (Wann et al., 1998; Biancotto et al., 2011; cf. Rosblad & von Hofsten, 1994). The present studies are thought to be the first to directly investigate this possible dissociation.

In a test of spatial location matching, it was found that children with poor motor skills (ASD or DCD) tended to have difficulties locating proprioceptively-defined targets. This was also true for the ASD group as a whole, although this finding is likely driven by the large number of children with ASD and comorbid motor difficulties. This suggests that when considering sensory processing in ASD and DCD, diagnostic labels may not be as informative as considering groups based on observed motor ability. Considering task performance when visual cues were prismatically displaced, children with ASD with relatively spared motor skills tended to weight visual feedback less, although this trend was not robust and requires further investigation with a larger sample of children with ASD and typical motor skills. The double dissociation was therefore partly confirmed. However, it appears that the dissociation may relate to ASD with and without motor impairment more than ASD and DCD. This would suggest that previous findings of a proprioceptive bias in ASD is specific to those with typical motor skills, although there are some reports of a proprioceptive bias in ASD despite significant motor deficits (Haswell et al., 2009). It should be noted however that previous literature has not used the MABC-2 to assess motor ability. It is possible that children showing proprioceptive bias alongside significant motor impairment would not be described as motor impaired on the MABC-2, due to low correlations between the MABC

and the frequently used PANESS (Thompson et al., 2008).

In an action task involving reaching after the induction of visual capture of a reflection of the hand, it was expected that if there is a double dissociation, children with ASD would be more accurate than DCD as they would be relatively less swayed by the mirror reflection. Again the double dissociation hypothesis was not supported as there were no group differences, although it was not possible to compare ASD ‘pure’ with the clinical motor deficit group in this case. These findings again suggest that the use of vision and proprioception is not a reliable indicator of clinical group membership (ASD, DCD or TD), but may be more reliable as an indicator of MABC-2 status (pass or fail), however this requires additional research as only possible trends were identified in the data presented here. Changes to diagnostic criteria allowing for dual diagnosis of ASD and DCD will make the question of a possible vision/proprioception dissociation *within ASD* (spared versus impaired motor ability) easier to investigate in the future. It will be possible to gather evidence of motor ability in ASD from clinical observations and assessments, carried out according to strict diagnostic criteria, with less reliance on a single test of motor ability carried out by researchers.

It is concluded that clinical motor deficits, regardless of their status as primary or secondary diagnoses, may be associated with an increased reliance on visual cues at the expense of proprioceptive cues. The null findings in both studies between ASD with motor difficulties and DCD again supports the modified criteria for ASD and DCD in the DSM-5.

Although the reaching task has been described as an action task, it is not a goal-directed action because the reach is made in a different plane to the target. There is evidence to suggest that visuomotor control is different in perceptually-driven and goal-directed actions (Goodale, Jakobson & Keillor, 1994). In order to be a goal-directed and purposeful action the child would need to have touched the target. In this case the movement was used to indicate a perceptual judgement. It could be argued that the two studies presented here have both assessed perceptual abilities, particularly as the movement itself was not considered in either task. The next chapter compares ASD and DCD on a purposeful goal-directed action, with a focus on both end-point accuracy and the execution of the movement as a whole. If this task finds group differences this would point to a double dissociation in sensory processing that is specific to purposeful movements.

## Chapter 4

# Proprioceptive feedback in action

The studies reported in this chapter continue to investigate sensory contributions to perception and action in neurotypical adults, and in children with ASD, DCD and TD. The first study employs neurotypical adults in an investigation of the role of direct and indirect proprioceptive feedback in perception and action. Following on from the investigation of sensory contributions in a perceptual task (spatial location matching) and an indirect action task (reaching to a location below a target) in the previous chapter, the second study assesses the role of vision and proprioception in a goal-directed action in children with ASD, DCD and TD.

### 4.1 The nature of a goal-directed actions

What is the difference between picking up a cup of tea and pretending to pick up a cup of tea? Aside from the latter most often occurring in the presence of a young child, in terms of visuomotor control there seems to be a great deal of difference. There is a large body of evidence to suggest that visuomotor control during target-directed actions differs from visuomotor control when actions are pantomimed or used to indicate a perceptual judgement. Goodale et al. (1994) found that kinematic properties of a reach-to-grasp movement were different in a goal-directed reach (the target is picked up) and a pantomimed reach (subjects pretend to pick up a now absent object). The former uses concurrent visual information to guide the reach while the later must rely on remembered object properties. Movements in pantomimed reaches tended to reach a lower peak velocity, have a longer MT and a less accurate MGA than goal-directed actions. This was also true when the pantomime reach was made to the side of the target while it was still visible (so every property except the exact location of the reach goal was visually-defined at the time of the reach). These results suggest that certain properties of a preprogrammed action are only activated when the action is truly goal-directed and involves a present target.

There is also evidence for different kinematics in actions following a delay compared to immediate goal-directed actions (Hu, Eagleson & Goodale, 1999). The authors

suggest that similar to pantomimed action, visual representations of objects for delayed actions make relative estimates about the object properties, while representations for immediate actions consider targets in absolute terms. This is true even when the delayed action is still goal-directed and the target is actually grasped. Similar findings of a resistance to visual illusion in action but not perception suggest that pantomimed and non-goal-directed actions are reliant on the ventral visual stream, making them more similar to perceptual tasks (Goodale & Milner, 1992), even when they involve a movement (Westwood, Chapman & Roy, 2000).

Neither of the two tasks described in the previous chapter was a true goal-directed action. The spatial location matching task required very little movement, and the movement was not considered a movement per se, but simply the easiest way to indicate the perceptual judgement. It was similar to scrolling a mouse up and down until judging the cursor to be in the centre of the screen. The movement was not a smooth single movement, like pointing underneath the target in the reaching task. However, although the reaching task was described as an action (relative to the almost entirely perceptual spatial location matching task), it shares a number of features with the pantomime action described above. Although a reach was made, the movement was not directed towards the target itself, but was simply a means to indicate the target's position on one axis, while ignoring the height of the target in space. This chapter uses a posting task (a goal-directed action) and a matching task (a perceptual judgement with an incidental movement) to investigate not only the use of vision in these two tasks, as has already been widely researched, but also the effect of proprioceptive feedback.

## 4.2 Reaching to proprioceptively-defined targets

Reaching to and interacting with objects is a typical everyday occurrence, but the sensory conditions these actions are carried out under can vary. We often reach towards visually-defined objects: we see the mug on our desk and we move our hand towards it. We can also use remembered visual information to reach towards the mug without moving our gaze from the computer, if we are confident that it has not moved from where we last put it down. We can also reach to proprioceptively-defined targets both with and without vision; for example, taking a glove off in a light or dark cinema.

Relatively little work has examined motor control when targets are defined proprioceptively. This does not mean that proprioceptive targets are not something we encounter frequently, but perhaps illustrates that proprioceptively-defined targets are more limited in scope. For example, if an outstretched finger is the target, the number of positions the target can be placed in is restricted by the natural postures of the arm. The majority of studies using proprioceptive targets have opted to use remembered targets as opposed to concurrent targets (e.g. Barden, Balyk, Raso, Moreau & Bagnall, 2005; Hocherman, 1993; Gaunet, Ittyerah & Rossetti, 2007). These tasks with remembered targets would not be considered goal-directed. Very few studies have considered

proprioceptive targets in goal-directed actions. If the distinction between goal-directed and non-goal-directed actions is true for proprioceptively-defined targets then it would be expected that the two different tasks would produce a different movement towards them. This chapter focuses on how we approach proprioceptively-defined targets and whether the way in which a target is proprioceptively-defined interacts with the presence or absence of visual cues.

A study with neurotypical adults will investigate the role of vision and proprioception in posting and orientation matching tasks, while also manipulating the spatial location of proprioceptive cues. This spatial manipulation alters the nature of the action: goal-directed or non-goal-directed with respect to the proprioceptively-defined target. This is followed by a report of the posting task with children with ASD, DCD and TD to investigate the use of vision in goal-directed actions towards proprioceptively-defined targets.

### **4.3 Online proprioceptive guidance in the posting and matching task**

Two similar studies conducted by Gosselin-Kessiby, Messier & Kalaska (2008) and Gosselin-Kessiby, Kalaska & Messier (2009) investigated the use of online proprioceptive feedback to guide actions. These studies used a letter posting and orientation matching task originally used with neurological patients to investigate the role of vision in perceptual tasks and goal-directed actions (Goodale, Milner, Jakobson & Carey, 1991). Gosselin-Kessiby et al. (2008, 2009) used a modified version of these posting and matching tasks to explore a gap in the reach-to-grasp literature: previous studies have tended to focus on either the transport of the hand or grasping behaviour, at the expense of the process of rotating the hand during the initial reach towards a target. Gosselin-Kessiby et al. (2008, 2009) investigated how orientation of the wrist during the reaching phase was influenced by whether the target was visually- or proprioceptively-defined. Both studies involved the use of concurrent proprioceptive feedback of a handle which was located 30 cm below a slot or handle. Subjects either rotated the top handle to match the orientation of the lower one, or posted a letter through the top slot, the orientation of which matched the held handle below it.

In the posting task, subjects were instructed not to fine-tune the movement once the hand had left the starting position. However, errors at the end of the movement were smaller than those at the start of the movement, suggesting that small automatic online corrections were made both with and without vision. These corrections were not evident when the posting slot was covered. The authors concluded that goal-directed actions were prone to automatic online corrections in a way that intransitive actions (reaching towards the covered slot) were not. They also found that when

only proprioceptive information about the required orientation was available errors were smaller in a goal directed action, compared to perceptual matching. This sits well with previous findings of kinematic differences between goal-directed actions and pantomimed actions to visually-defined targets, which Goodale et al. (1994) suggest do not make use of the same visuomotor information as goal-directed actions do.

There is a major methodological concern in the proprioceptive posting and matching study however. Having the reference handle at the bottom of the board and then acting on the upper handle or slot means that this is not a true proprioceptively-defined goal-directed action. The orientation information is proprioceptively-defined, but more information is needed to guide the action, e.g. height and size. Conclusions from this study assume that indirect proprioceptive information of the target will guide action in a way comparable to having direct proprioceptive information of target location. It is questioned here whether this spatially removed feedback can actually guide action in the same way as direct continuous proprioceptive feedback would. As the location of feedback alters whether the action is truly goal-directed or not, it is suggested that it cannot.

There do not appear to be any published studies assessing the use of direct proprioceptive orientation information for online guidance of a goal-directed action. (However the visual-proprioceptive matching tasks described in Chapter 3 have done so for perceptual judgements.) The present study investigates whether orientation information is utilised more effectively from direct (spatially congruent) proprioceptive feedback compared to indirect feedback (spatially displaced), in modified versions of the posting and matching tasks described above.

### **4.3.1 Present study**

The main motivation for this study is to determine an appropriate method for the investigation of goal-directed action in ASD and DCD, by resolving prior methodological issues.

With a proprioceptive version of the posting and matching task already reported in the literature it seemed appropriate to use this as a springboard for the present study. However, in situations where continuous proprioceptive feedback of the target has been used (Gosselin-Kessiby et al., 2008, 2009), the feedback is in a location that is not being acted on, that is, the action is still not goal-directed towards a proprioceptively-defined target. To ensure that the action is goal-directed with respect to the proprioceptively-defined target, the proprioceptive information should be at the location being acted on. The present study challenges Gosselin-Kessiby et al.'s assumption that spatially incongruent feedback can be referred to as 'direct feedback', by comparing two kinds of concurrent proprioceptive feedback. Proprioceptively-defined targets will be either directly-defined, that is they are the goal in a goal-directed action; or indirectly-defined, that is they afford the key information necessary for accurate performance (orientation)

however they are spatially displaced, meaning the goal is not in the same location as the proprioceptively-defined orientation information.

### 4.3.2 Hypotheses

It is hypothesised that direct proprioceptive feedback of target orientation will allow for more accurate performance than indirect proprioceptive feedback, due to the goal-directed nature of the action. As vision has previously been found to be weighted more heavily than proprioception in similar tasks (Gosselin-Kessiby et al., 2009), it is expected that vision will be the strongest cue available and so proprioceptive condition may not greatly affect performance when vision is available (i.e. vision will dominate).

## 4.4 Methods

### 4.4.1 Subjects

Twenty four neurotypical adults (8 females, 22-41 years, mean=26 years, SD=4.53) were recruited from an email sent to postgraduates in the school of Philosophy, Psychology and Language Sciences at Edinburgh University. All reported being right handed with normal or corrected-to-normal vision and all indicated that they had no diagnosed motor deficit or ASD. Subjects received £5 for their participation.

### 4.4.2 Apparatus

The apparatus for both posting and matching is shown in Figure 4.1. A wooden frame measuring 62.5 x 28 cm housed one of two boards: one board with two rotatable discs with handles (matching board); the other with two discs in which slots of the same dimensions as the handles were cut out (posting board). On each board the discs were located one above the other, 15 cm apart. In each case the slots or handles were in the centre of a disc. Each handle or slot measured 12.5 x 2 cm. Handles were located in this position at both the front and back of the apparatus. At the back of the posting board, the slot was defined by a 0.3 cm thick border of wood, which could be grasped so that the slot was being held in the hand and therefore proprioceptively-defined. The discs on both boards could be freely rotated through 360° but were lockable in 18 positions at 10 degree intervals. On the matching board, each disc could be rotated independently from the front by the subject and from the back by the experimenter. On the posting board the discs were rotated by the experimenter from the back. This in turn rotated the slots at the front. A wooden 'letter' measuring 10x15 cm was used in the posting task.

A trakSTAR electromagnetic motion tracking system (Ascension Technology, Burlington, VT) was used to record the movement of the handle and letter as they were manipulated by the subject. The electromagnet was housed on a shelf at the back of the wooden frame, 22 cm from the back of the board. In the posting task a tracking sensor

was inserted into the end of the wooden letter, in the centre of the side facing the subject: in the matching task it was inserted into the end of the handle so that as the handle was moved the sensor moved around the circumference of the circle.

The apparatus was made without any metal fixings or materials to avoid interference with the tracking system. The magnet was housed in a consistent position, so that all trials for each subject used a common coordinate frame.

#### 4.4.3 Procedure

Posting and matching task order was counterbalanced across subjects. In each of these tasks, subjects completed five blocks: vision-only; and four experimental conditions which manipulated vision and proprioception in a 2x2 design (vision/no vision and direct/indirect proprioception). The vision-only condition served as a baseline for analysis, firstly to identify any kinematic variables that varied systematically with target orientation, and secondly to investigate the evolution of orientation scaling over the time course of reaches. Experimental conditions were used to investigate the contribution of varying visual and proprioceptive conditions on task performance. After each ‘vision-only’ condition, experimental conditions were given following an ABBA order for proprioception, and ABAB for vision. Once all experimental conditions were completed for the first task (e.g. posting) the ‘vision-only’ condition for the second task (e.g. matching) was completed, followed by experimental conditions.

For each subject their condition order was the same for each task. Each task (posting and matching) used 18 possible target orientations, each presented twice, once in DV or DN and once in IV or IN. Target order was randomised. Each subject was presented with a unique random order for both vision-only and experimental conditions. The order was fixed within each subject for experimental conditions so that trials in posting followed the same order as matching. In vision-only conditions a different random order was used in each task and again each subject received unique random orders.

Each condition consisted of nine unique trials, totalling 36 experimental trials per task and nine randomly assigned trials in the vision-only condition. Procedure and instructions for each task are described below and the procedure for experimental tasks are summarised in Table 4.1.

Participants sat in front of the apparatus with the top disc at eye level. They were asked to reach to the back of the board and hold onto one of the handles, then move their chair so they could sit comfortably in this position. They were then told that the task involved either matching the orientation of a handle or posting a letter through a slot. In all experimental conditions the subject was instructed to reach to the back of the board and hold the handle or slot with their non-dominant (left) hand. In the ‘direct’ condition the top handle or slot was held, and the lower handle or slot was held

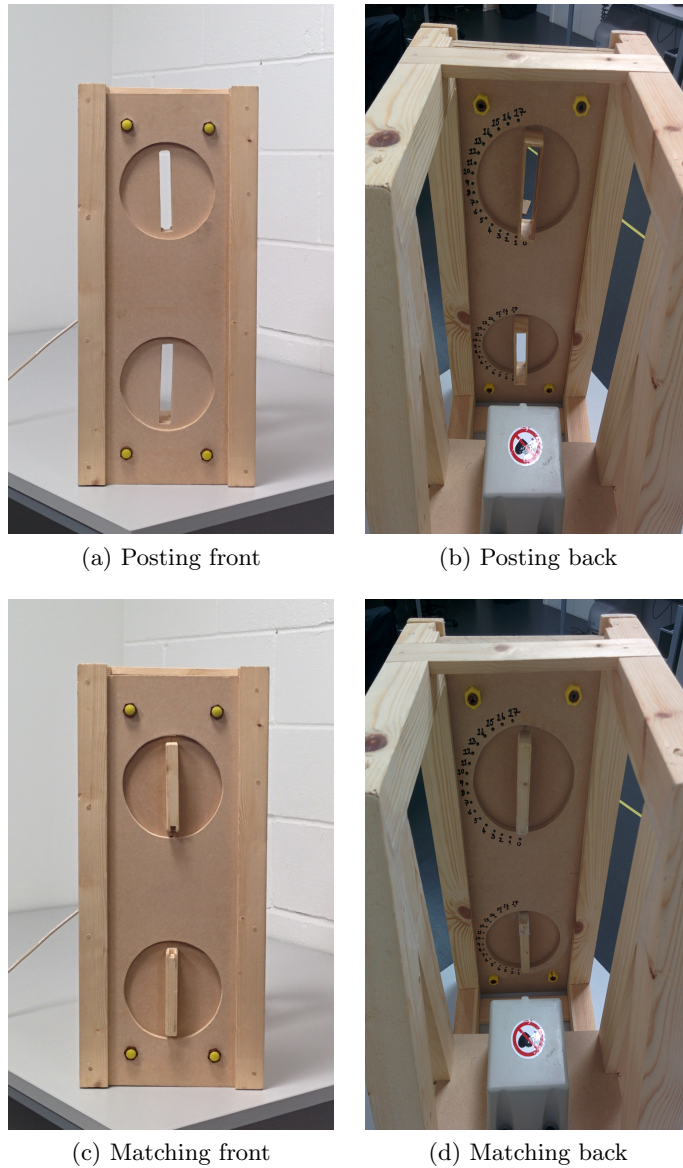


Figure 4.1: Posting and matching apparatus. a) The posting apparatus as viewed by the subject. The letter is posted through the top slot. During testing the lower slot is covered. b) The back of the posting apparatus: The subject holds the back of the slot at the top (direct) or the bottom (indirect). In indirect conditions both slots are set to the same orientation. Orientation is set by inserting a peg into one of 18 holes around the circle. c) The matching apparatus as viewed by the subject. Subjects move the top handle to match the proprioceptively-defined handle at the back of the board (either at the top or bottom), or visually match the front lower handle. During testing the lower handle is covered in proprioception conditions. d) The back of the matching apparatus. The top is held for direct trials, the bottom for indirect. Orientation is set as per posting.

in ‘indirect’ conditions. In all cases the top handle or top slot at the front was the target (i.e. the handle to be turned or the slot to be posted through). The ‘vision-only’ condition included matching or posting with vision but no proprioceptive information about the target. The matching task was to move the top handle until it matched the lower handle; the posting task was to post the letter through the top slot (the lower slot was covered). The posting no-vision condition closely replicates Gosselin-Kessiby et al. (2008, 2009) and Milner, Perretti, Johnston, Benson, Jordan, Heeley, Bettucci, Mortara, Mutani, Terazzi & Davidson (1991). The matching no-vision condition differs from these previous studies. All direct conditions are novel. Indirect conditions are similar to Gosselin-Kessiby et al. (2008, 2009), except in this case the back of the board is held, not the front.

Subjects were asked to close their eyes before each trial in both tasks. This was to ensure that in conditions where vision was allowed during the action, the subject had not seen the target move from the previous orientation, thereby giving them additional information about the target, relative to its previous orientation. Subjects were not blindfolded at any point during the experiment and compliance with instructions to close eyes was monitored by the experimenter.

Table 4.1: Instructions for experimental conditions in the posting and matching tasks

Task	Common instructions	Condition	Task-specific instructions
Posting	Hold the letter with the right hand, starting with it flat on the table, marked side up, far edge against the starting block. The palm should be down, covering the top of the letter. Movements should be made at a quick but comfortable speed using one smooth, single movement. If the letter hits the board it should not be moved from this position. The letter is always posted through the <i>top</i> slot with the right hand. Eyes should be closed while the experimenter moves the target to its new position at the start of each trial.	DV	Reach to the back of the board with the left hand and grasp the back of the top slot and hold it throughout the action.
		DN	As above, with eyes closed throughout.
		IV	Reach to the back of the board with the left hand and grasp the back of the bottom slot and hold it throughout the action.
		IN	As above, with eyes closed throughout.
Matching	The top handle should be moved with the right hand until the subject is satisfied that the angle matches the angle defined proprioceptively at the back of the board. Eyes should be closed while the experimenter moves the target to its new position at the start of each trial.	DV	Reach to the back of the board with the left hand and grasp the back of the top handle and hold it throughout the action.
		DN	As above, with eyes closed throughout.
		IV	Reach to the back of the board with the left hand and grasp the back of the bottom handle and hold it throughout the action.
		IN	As above, with eyes closed throughout.

## 4.5 Results

Results from the vision-only conditions in posting and matching are reported first and these include both orientation and kinematic measures. This is followed by results from the experimental conditions in the posting task, and then by results from the experimental conditions in the matching task.

### 4.5.1 Measures

#### 4.5.1.1 Terminal orientation

Terminal orientation (TO) was analysed using three measures: mean absolute error (the average unsigned difference in degrees between terminal orientation and target orientation), mean constant error (the average signed error, used to assess clockwise or anticlockwise error bias) and variable error (SD of constant error) to give an indication of precision. Each mean value or variable error was calculated per condition for each subject in posting and matching.

#### 4.5.1.2 Speed of movement measures

Speed of movement measures were obtained for posting trials, including movement time (MT), peak orientation speed (POS), time to peak orientation speed (TPOS), peak speed (PS), time to peak speed (TPS), and reaction time (RT). These measures are described in Table 4.2. These measures are only available for posting trials as matching was self-paced. A preliminary analysis of the vision-only condition provides a way to identify the most useful speed measures to include in the analysis of experimental conditions.

Table 4.2: Posting measures

Measure	Definition
Movement Time (MT) (s)	Duration of the movement.
Peak Orientation Speed (POS)	Maximum rate of change of orientation.
Time to Peak Orientation Speed (TPOS) (s)	Time taken to reach peak orientation speed.
Peak speed (PS) (mm/s)	Top speed reached at the end of the acceleration phase.
Time to Peak Speed (TPS) (s)	Time taken to reach peak speed.
Reaction Time (RT) (s)	Time taken to initiate movement.

### 4.5.1.3 Planned analyses

The experiment consisted of four sub-experiments, each analysed separately. First, the vision-only matching task was analysed using correlational analysis to ensure that subjects were able to accurately match the two handles. The vision-only posting data were then analysed to assess how wrist rotation is scaled in the classic version of the posting task (Goodale et al., 1991). This will inform analysis of the experimental posting and matching conditions.

The experimental conditions in the matching task will be analysed using a 2x2 (vision: present/absent; proprioception: direct/indirect) factorial analysis. The posting task proper will follow a similar factorial analysis. Where the sphericity assumption is violated, degrees of freedom will be adjusted using the Greenhouse-Geisser adjustment (GG).

The two tasks (matching and posting) are not compared directly, as the availability of vision of the target in experimental conditions is different in each task: in posting the visual feedback is of the target and the responding hand approaching the slot: in matching it was not possible to create this situation and the visual feedback is of the responding hand, not the target.

### 4.5.2 Vision-only matching

As expected, terminal orientation strongly positively correlated with target orientation:  $r > 0.99$ . Figure 4.2 shows mean constant, absolute and variable errors for each of the 18 target orientations in the vision-only condition. Error associated with each target is broadly similar.

### 4.5.3 Vision-only posting

As with the matching task, terminal orientation correlated almost perfectly with target orientation ( $r > 0.99$ ). Errors were broadly similar across targets (see Figure 4.3).

#### 4.5.3.1 Choosing clockwise or anticlockwise rotations

Subjects were allowed to post the letter following a clockwise or anticlockwise rotation of the wrist. There was no instruction specifying that one was to be preferred over the other. Some targets were easy to reach using either a clockwise or anticlockwise rotation, for example a vertical slot, which is  $90^\circ$  from the starting position in either direction; others were easier to reach using one rotation over the other. The percentage of subjects using clockwise and anticlockwise rotations for each target is shown in Table 4.3.

The majority of subjects chose the shortest rotation for all targets, with the exception of one of the targets  $10^\circ$  from vertical. When the longest route was taken for targets with a large discrepancy between shortest and longest route (e.g. the horizontal

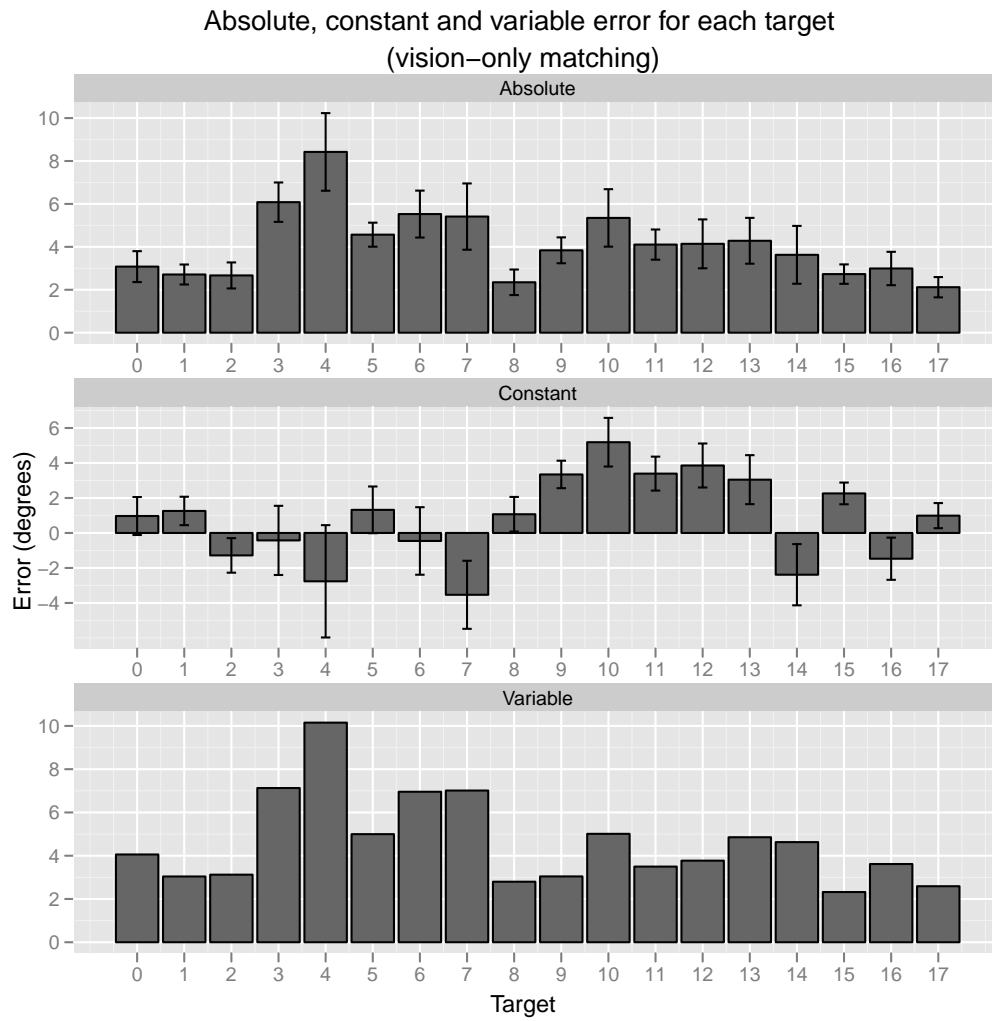


Figure 4.2: Absolute, constant and variable error for each target in the vision-only matching condition. Error bars show SE. Target 0 is vertical, and 9 is horizontal.

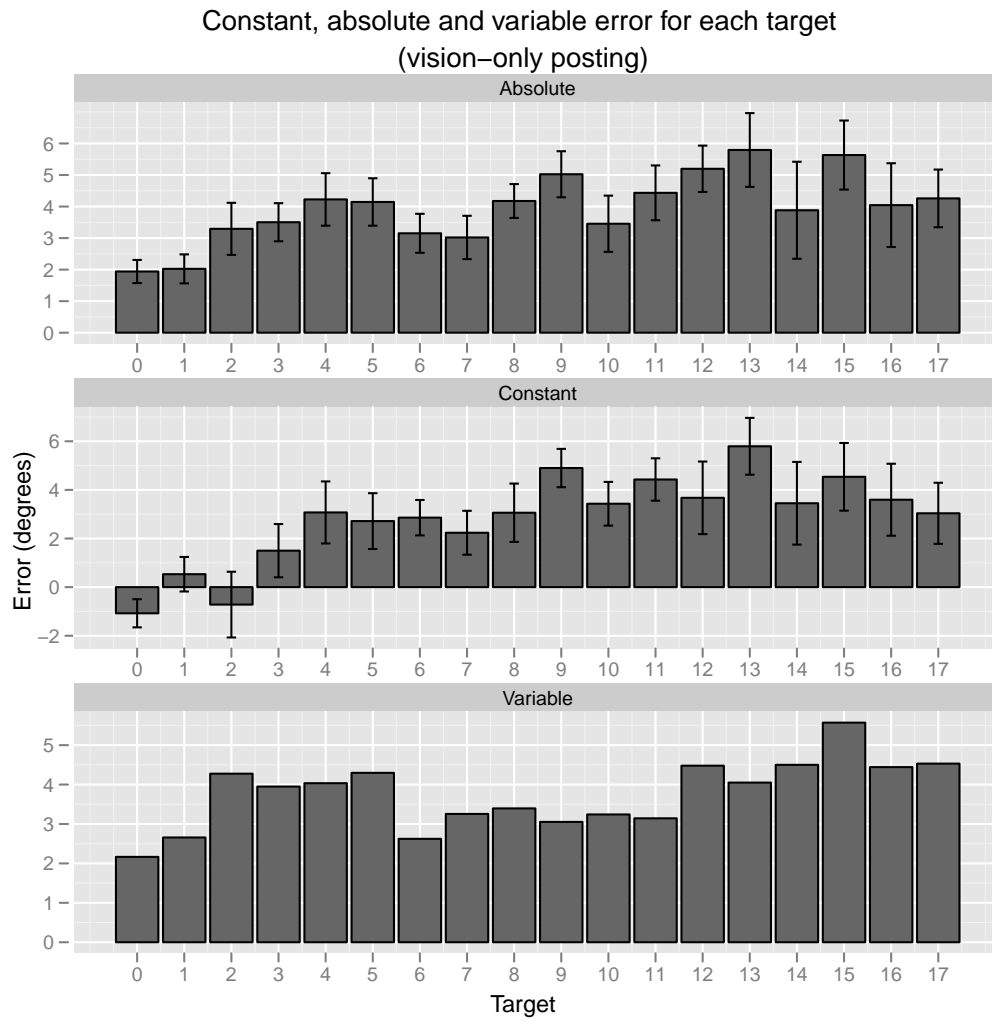


Figure 4.3: Absolute, constant and variable error for terminal orientation for each target in the vision-only posting condition. Error bars show SE.

slot which required either no wrist rotation or 180° rotation), those subjects following the longest route approached the final orientation much later in the movement than subjects who chose the shortest route (see Figure 4.4). This likely explains why the majority chose the most efficient rotation.

Table 4.3: Percentage of clockwise and anticlockwise rotations for each target for vision-only posting

Target angle	No. subjects	% Anticlockwise rotations	% Clockwise rotations
0	14	50	50
10	14	71	29
20	10	70	30
30	13	77	23
40	10	80	20
50	14	79	21
60	13	77	23
70	13	92	8
80	8	100	0
90	15	87	13
100	13	8	92
110	13	0	100
120	9	0	100
130	12	0	100
140	7	0	100
150	16	13	87
160	9	22	78
170	13	54	46

The line shows the point at which the most common rotation direction changes. (There are unequal numbers of subjects between angles because angle was randomised without counterbalancing.)

#### 4.5.3.2 The time-course of visually-guided movements

Due to the clear bias for the shortest rotation it was not possible to consider mean orientation error separately for those trials in which the less favoured rotation was used. For this reason the overall mean absolute error for each target was calculated from all trials. Each subject's movement<sup>1</sup> on each trial was normalised so that the movement was framed in terms of percentage movement time, rather than seconds. Absolute error for each target was then calculated for each 1% of normalised time. This preliminary analysis of the vision-only condition allows for an investigation of orientation scaling during normal visually-guided action. This analysis is used to identify the point in time during a movement when orientation becomes relatively stable in order to identify

<sup>1</sup>Movements were deemed to have started once velocity exceeded 50mm/s and ended once velocity fell below 50mm/s

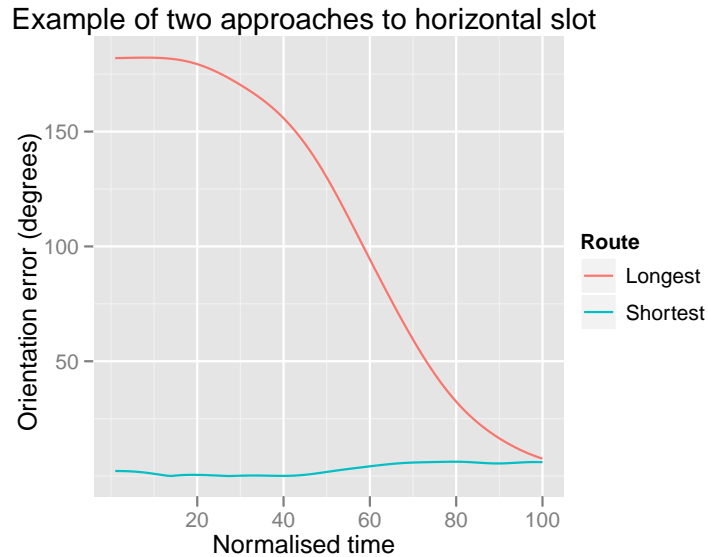


Figure 4.4: Orientation error over normalised time for two subjects approaching the horizontal slot

an additional measure of orientation other than TO (i.e. orientation part of the way through a complete movement). Figure 4.5 shows the mean orientation error through the time course of the movement. The graph shows that by 60% MT the orientation error is close to the error present at the end of the movement (100% normalised time). Therefore by 60% MT large wrist rotations have largely been completed and this point seems to mark the beginning of the fine-tuning phase of the movement.

The key question relates to the effect of different kinds of orientation information on task accuracy, therefore final orientation is obviously most informative. In addition to this, using orientation at 60% MT will allow for an analysis of orientation matching before fine-tuning in the later stages of the deceleration phase of the movement.

#### 4.5.3.3 Other measures

Each output measure from the vision-only condition was correlated with target orientation to assess which measures are sensitive to varying orientation. Figure 4.6 shows the correlations for each subject for each output measure against target orientation. As expected it is clear that TO is highly dependent on target orientation, with an average correlation of  $r = 0.997$ . The other kinematic measures show either consistently weak correlations or a high degree of variability between subjects. For this reason orientation (at 100% and 60% MT) will be the only measures included in analysis of the experimental conditions. Mean error (absolute, constant and variable) will be used to measure the effect of condition on orientation at each time point.

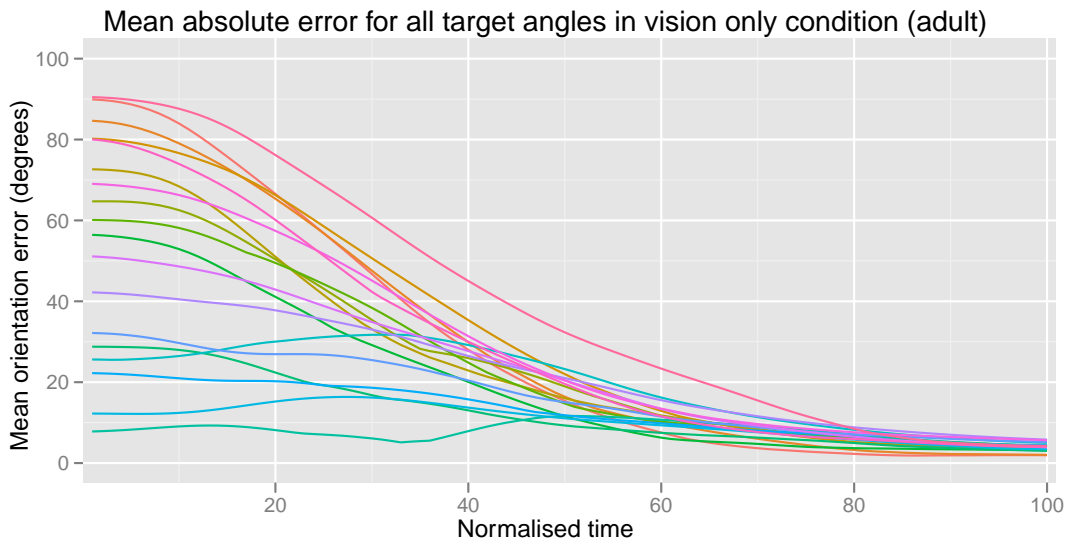


Figure 4.5: Mean absolute orientation error across normalised time. By 60% MT large wrist rotations have been completed and the rest of the movement involves smaller adjustments.

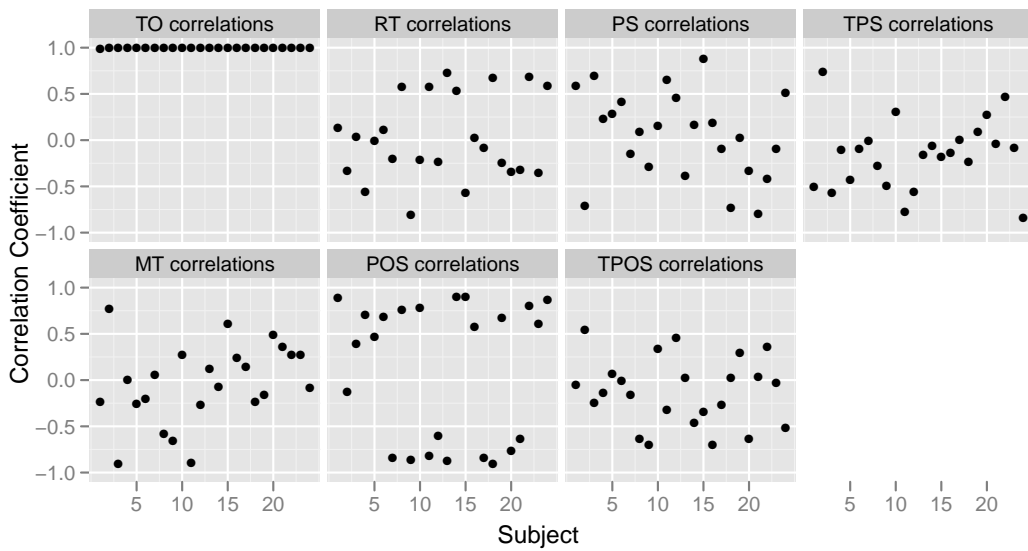


Figure 4.6: Pearson correlation between target orientation and each measure for each subject in the vision-only condition

## 4.5.4 Posting main analysis

### 4.5.4.1 Orientation absolute error

Mean absolute error was calculated for each subject and entered into a separate 2x2 ANOVA each for 60% MT and 100% MT. Means for each condition are shown in Figure 4.7.

At 60% MT there was no significant vision\*proprioception interaction ( $F(1, 23) = 0.39, p = 0.537$ ) however the effect of visual condition was significant, with smaller errors when visual feedback was provided ( $F(1, 23) = 4.97, p = 0.036, \eta_p^2 = 0.18$ , mean difference (SE)=1.80 (0.81)). There was no significant effect of proprioceptive condition on error:  $F(1, 23) = 2.68, p = 0.116, \eta_p^2 = 0.10$ .

By 100% MT sensory cues significantly interacted:  $F(1, 23) = 10.40, p = 0.004, \eta_p^2 = 0.31$  (GG). Proprioceptive condition only had a significant effect on orientation in the absence of visual feedback. With no visual feedback, indirect proprioceptive feedback led to greater errors compared to direct proprioceptive feedback. Both the main effect of vision and proprioception on error were significant, with smaller errors in vision compared to no vision conditions, and direct compared to indirect conditions, however the interaction effect better explains these effects:  $F(1, 23) = 136.54, p < 0.001, \eta_p^2 = 0.86$ , mean difference (SE) =7.02 (0.60); and  $F(1, 23) = 9.78, p = 0.005, \eta_p^2 = 0.30$ , mean difference (SE) =1.53 (0.49).

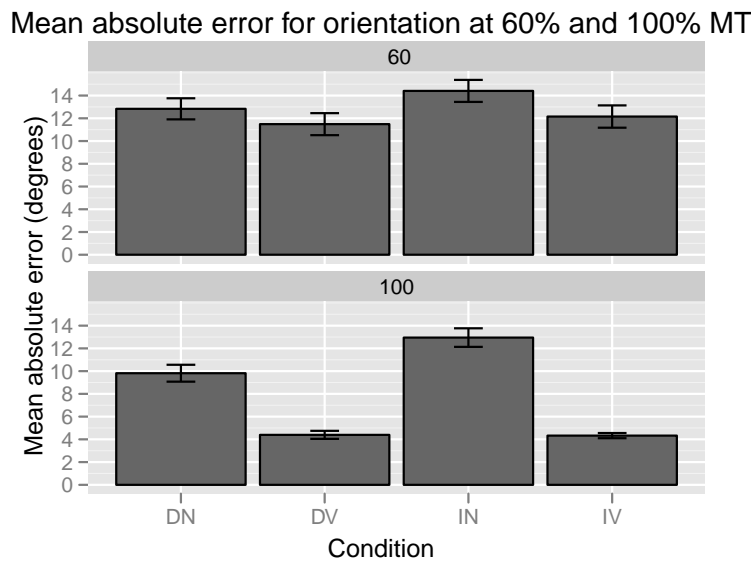


Figure 4.7: Orientation error in each condition at 60% and 100% MT. Error bars show SE. There is a significant vision\*proprioception interaction at 100% MT.

#### 4.5.4.2 Orientation constant error

Mean constant errors are shown in Figure 4.8. At 60% MT, there was no significant interaction between sensory cues ( $F(1, 23) = 0.05, p = 0.828$ ) and no significant effect of proprioceptive condition ( $F(1, 23) = 0.03, p = 0.859$ ). However there was a significant effect of visual condition, with no-vision conditions producing smaller constant errors  $F(1, 23) = 25.52, p < 0.001, \eta_p^2 = 0.53$ , mean difference (SE) = 7.87 (1.56).

At 100% MT there was still no significant interaction between sensory cues (vision and proprioception), nor a significant effect of proprioceptive condition on mean constant errors:  $F(1, 23) = 0.19, p = 0.666, \eta_p^2 = 0.01$  (GG);  $F(1, 23) = 0.29, p = 0.594, \eta_p^2 = 0.01$  (GG). At 100% MT there was a significantly smaller mean error in conditions with vision compared to those without:  $F(1, 23) = 17.99, p < 0.001, \eta_p^2 = 0.44$  (GG), mean difference (SE) = 6.58 (1.55).

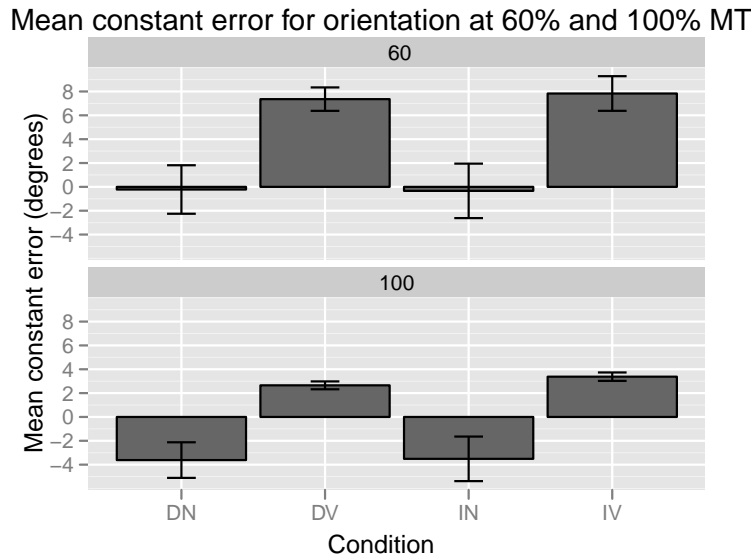


Figure 4.8: Mean constant error in each condition at 60% and 100% MT. Error bars show SE. At both time points there is a significant effect of vision: at 60% MT error is lower when vision is removed, although the removal of vision significantly adversely affects accuracy at 100% MT.

#### 4.5.4.3 Orientation variable error

Mean variable error in each condition is shown in Figure 4.9. At 60% MT there was no significant interaction between sensory cues ( $F(1, 23) = 1.64, p = 0.213$ ), however response variability was significantly reduced in conditions with visual feedback:  $F(1, 23) = 7.19, p = 0.013, \eta_p^2 = 0.24$ , mean difference (SE) = 3.06 (1.14). Proprioceptive condition did not significantly affect precision ( $F(1, 23) = 1.65, p = 0.212$ ).

By 100% MT the cues did significantly interact, with direct proprioception allow-

ing for more precise movements than indirect in the absence of vision:  $F(1, 23) = 12.73, p = 0.002, \eta_p^2 = 0.36$  (GG). Both main effects of vision and proprioception were significant, with greater variability in no vision compared to vision conditions and indirect compared to direct conditions, however the interaction better explains these effects:  $F(1, 23) = 251.44, p < 0.001, \eta_p^2 = 0.92$  (GG), mean difference (SE) = 7.50 (0.47); and  $F(1, 23) = 5.36, p = 0.03, \eta_p^2 = 0.19$  (GG), mean difference (SE) = 1.13 (0.49).

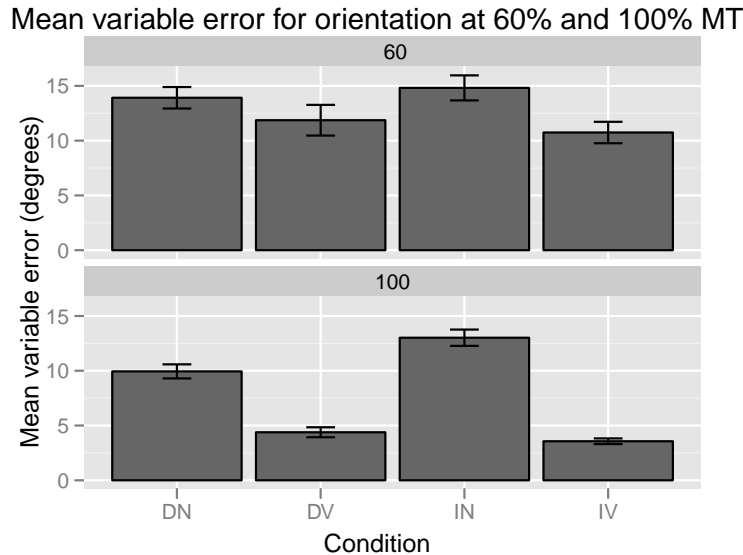


Figure 4.9: Mean variable error in each condition. Error bars show SE. At 60% MT there is a significant main effect of vision, and at 100% MT there is a significant vision\*proprioception interaction.

#### 4.5.4.4 Posting summary

At 100% MT mean absolute orientation error and variable error were affected differentially by proprioceptive condition only in the absence of vision. With no visual feedback accuracy and precision were better when direct proprioceptive feedback was provided, when compared with indirect proprioceptive feedback, confirming the initial hypothesis. This differential effect was not apparent at 60% MT. It is clear in both cases that at 60% MT the pattern noted at 100% MT is beginning to emerge, although it fails to reach statistical significance.

#### 4.5.5 Matching

Only terminal orientation was used as an output measure in matching as data were not collected when the handle was moving. As above, three measures were extracted for each subject: mean absolute error, mean constant error, and variable error. These are shown in Figure 4.10.

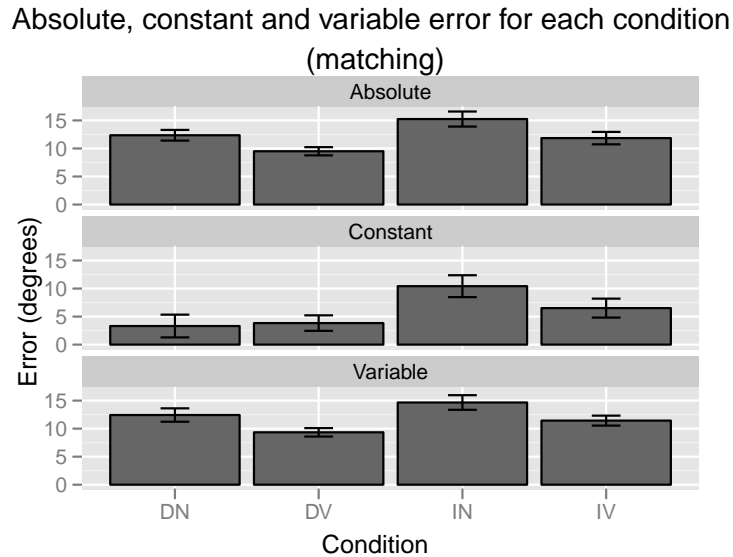


Figure 4.10: Mean error between conditions for absolute, constant and variable error. Error bars show SE. There is a significant effect of vision and proprioception on absolute error and variable error. There is a significant vision\*proprioception interaction for constant error.

#### 4.5.5.1 Orientation absolute error

Data were entered into a 2x2 ANOVA, comparing orientation errors within subjects according to visual condition (vision/ no vision) and proprioceptive condition (direct/indirect feedback). Visual condition had a significant effect on absolute error, with greater errors in the vision absent condition:  $F(1, 23) = 23.79, p < 0.001, \eta_p^2 = 0.51$  (GG), mean difference (SE)=3.13 (0.64). Similarly, proprioceptive condition had a significant effect, with greater errors in the indirect condition:  $F(1, 23) = 6.59, p = 0.017, \eta_p^2 = 0.22$  (GG), mean difference (SE)=2.62 (1.02). Unlike posting, there was no significant vision\*proprioception interaction:  $F(1, 23) = 0.19, p = 0.669$  (GG).

#### 4.5.5.2 Orientation constant error

A different pattern of effects emerged for constant error, with no significant effect of visual condition:  $F(1, 23) = 1.89, p = 0.182$  (GG). Again, responses in direct proprioception conditions were significantly less prone to error than in indirect conditions:  $F(1, 23) = 13.20, p < 0.001, \eta_p^2 = 0.37$  (GG), mean difference (SE)=4.89 (1.35). There was also a significant vision\*proprioception interaction [ $F(1, 23) = 5.95, p = 0.023, \eta_p^2 = 0.21$  (GG): errors are reduced in the no-vision condition by defining the target directly].

### 4.5.5.3 Orientation variable error

As was the case with absolute error, there were significant effects of visual and proprioceptive conditions:  $F(1, 23) = 15.48, p < 0.001, \eta_p^2 = 0.40$  (GG) and  $F(1, 23) = 6.19, p = 0.021, \eta_p^2 = 0.21$  (GG). Responses in conditions with vision were more consistent than responses in the no vision conditions [mean difference (SE)=3.16 (0.80)], and responses following direct proprioceptive feedback were more consistent than those following indirect feedback [mean difference (SE)=2.16 (0.87)]. There was no significant vision\*proprioception interaction effect:  $F(1, 23) = 0.008, p = 0.931$  (GG).

### 4.5.5.4 Matching summary

Vision of the responding hand led to significantly smaller unsigned errors compared to matching under no-vision conditions. Vision of the responding hand also led to more consistent responses (variable error). Similarly, proprioceptive feedback had a significant effect on the magnitude of unsigned errors and response variability: direct proprioceptive feedback of the target led to smaller errors and less variability. The vision\*proprioception interaction was only apparent when considering constant error.

## 4.6 Discussion

The study aimed to assess the use of direct proprioceptive feedback of target orientation versus indirect proprioceptive feedback, using a modified version of the posting and matching task described by Gosselin-Kessiby et al. (2008, 2009). There was a consistent effect in both posting and matching: in the absence of vision, estimates of orientation were more accurate when subjects had direct proprioceptive feedback of the target compared to indirect proprioceptive feedback. This suggests that Gosselin-Kessiby et al.'s reference to indirect proprioceptive cues as 'direct' target information is incorrect, as in this case orientation information was better utilised when it was located at the position of the target. It should be noted that spatial accuracy was not considered in analysis, so inaccuracy in judging the height of the target did not affect the measures used in analysis. It is expected that had the Gosselin-Kessiby et al. studies involved a truly direct source of ongoing proprioceptive feedback, a similar effect on accuracy would have been found.

Results from the present study provide evidence for automatic proprioceptive guidance in the absence of vision, as reported by Gosselin-Kessiby et al., although in this case the effect was specific to the direct-proprioception condition. As the significant interaction between sensory cues was most evident in the posting task, this suggests that proprioceptive cues to target orientation are better utilised in goal-directed actions in the same way that visual cues are processed differently for goal-directed and non-goal-directed actions (Goodale et al., 1994).

The results of this study highlight a difference between goal-directed and non-goal-directed actions similar to that previously reported in vision (Goodale et al., 1994). Just as visual cues are processed differently depending on the action being performed, so too are proprioceptive cues. The effect of proprioceptive condition on performance suggests that direct proprioceptive feedback of this nature is suitable for investigating how we approach proprioceptively-defined targets. The direct proprioceptive condition will be used in the next study to further investigate the roles of visual and proprioceptive cues in guiding actions in ASD and DCD.

## **4.7 Posting using vision and proprioception in children with ASD, DCD and TD**

Children completed a shortened version of the posting task detailed above. By altering the availability of visual information, this task aims to investigate the potential differential use of visual cues in ASD, DCD, TD, and the efficacy with which proprioceptive information is used in the absence of vision. These results will complement those detailed in Chapter 3. It is expected that if DCD is associated with an increased reliance on visual information, children with DCD should show marked impairment in conditions without visual feedback. If ASD is associated with a relative reliance on proprioceptive feedback, they should not show such marked impairment. Alternatively, if sensory weighting in ASD is related to motor ability, as suggested by results in Chapter 3, those children with ASD who failed the MABC-2 should show patterns similar to DCD. Those children with ASD with spared motor skills should show a relative bias towards proprioceptive cues.

## **4.8 Methods**

### **4.8.1 Subjects**

All subjects had completed all child studies detailed in Chapters 2, 3 and 5. Of 28 TD children, permission was granted for 14 and the majority took part in their schools. Of the children with ASD ( $n=33$ ), 23 returned after the initial testing period and were tested either at school or in the university. Of those 10 children in the original DCD group, 6 returned for further study and again were tested at school or at the university. All children completed this task 2-18 months after initial testing, in a follow-up session which also included the reaching task reported in Chapter 3 and the RHI task reported in Chapter 5. Data were not used for 4 children with ASD (all analyses) and one TD child (all but the vision-only analysis) due to TrakSTAR recording errors. Three children in the TD group had previously failed the MABC-2 so their data were not included, in line with previous chapters.

Data were analysed for 21 ASD, 6 DCD and 10 TD children. All but one child (a

Table 4.4: Subject demographics (values shown are mean (SD))

	ASD ( $n=21$ )	DCD ( $n=6$ )	Clinical motor deficit ( $n=23$ )	TD ( $n=10$ )
Age	12.29 (1.95)	11 (2.53)	11.83 (2.17)	11.30 (1.25)
IQ %ile	44 (27.94)	49.17 (42.99)	39.35 (29.06)	61.40 (21.72)
MABC-2 %ile	12.74 (20.98)	4.35 (3.96)	4.51 (3.68)	54.50 (23.61)
SRS	111.89 (28.71)	62.17 (36.52)	100.10 (36.14)	12.20 (15.40)
DCDQ-07	34.10 (10.91)	31.80 (11.17)	32.10 (10.06)	70.00 (6.29)

Groups were age- and IQ-matched (group difference  $p > 0.05$ ). As with the reaching task in Chapter 3, SRS, DCDQ-07 and MABC-2 scores differentiated clinical groups from TD ( $p < 0.05$ ) and the SRS differentiated ASD and DCD from each other and from the TD group ( $p < 0.05$ ).

member of the DCD group) was male. Demographics are broadly similar to those in the reaching task in the previous chapter (see Table 4.4).

## 4.8.2 Apparatus

The apparatus described previously for the posting task was used.

## 4.8.3 Procedure

The following changes were made to the procedure used in the adult study: the board was positioned slightly side-on, to minimise the reach required to hold on to the back of the board, while still allowing for full vision of the front of the board. While each subject in the adult study completed trials in their own unique random order, all children in the study completed trials in the same fixed random order. The vision-only condition included nine trials, with four positions from the proprioception+vision condition and four positions from the proprioception condition, with a vertical slot as the last trial. Trials in the proprioception+vision and proprioception conditions were counterbalanced, with the first condition always including half the unique trials in a fixed order and the second condition always comprising the remaining half in a fixed random order.

Importantly, only direct proprioception conditions were included. The previous study found significantly greater accuracy and precision when proprioceptive information was direct, making the action a true goal-directed action. This study focuses on the role of vision in such goal-directed actions. As with the adult study, the vision-only condition always came first and the remaining conditions were counterbalanced.

## 4.9 Results

Data were processed in the same way as described above in the adult study. Again, the correlation between target orientation and each output measure was calculated for each subject for the vision-only condition. As with the adult study, those measures that vary systematically with target orientation are analysed further. The vision-only condition is also used to identify an additional time point at which to measure error. Again, the vision-only condition is used to identify measures for use in final analysis, and is not directly compared to either proprioceptive condition.

### 4.9.1 Vision-only

Only TO varied systematically with target orientation, as was the case in the adult study (see Figure 4.11). There was no strong correlation between any other measure and target orientation, and this is true both considering the sample as a whole and considering each group separately. For this reason only TO will be considered in final analyses of experimental conditions.

To determine whether a secondary time point for orientation could be considered, normalised time for the vision-only condition was plotted for each target to see if the lines converged at a certain point before the end of the movement (see Figure 4.12). Orientation tended to be relatively stable from 60% MT in the adult sample, and this was also true for the child sample. There does not appear to be any difference between groups in their wrist orientation scaling throughout the movement (see Figure 4.13), and kinematic variables did not differ significantly between groups (see Table 4.5).

Table 4.5: Main effect of group for kinematic variables

RT	$F(2, 34) = 2.21, p = 0.125$
MT	$F(2, 34) = 1.84, p = 0.174$
PS	$F(2, 34) = 1.73, p = 0.193$
TPS	$F(2, 34) = 0.89, p = 0.422$
POS	$F(2, 34) = 2.86, p = 0.071$
TPOS	$F(2, 34) = 0.67, p = 0.519$

### 4.9.2 Experimental conditions

Where data were not normally distributed, nonparametric analyses (Kruskal-Wallis and Wilcoxon) were conducted, otherwise an ANOVA was conducted.

#### 4.9.2.1 Absolute error

Mean absolute error at 60% and 100% MT were calculated for each subject for each condition (DV, DN). Data were not normally distributed therefore nonparametric tests

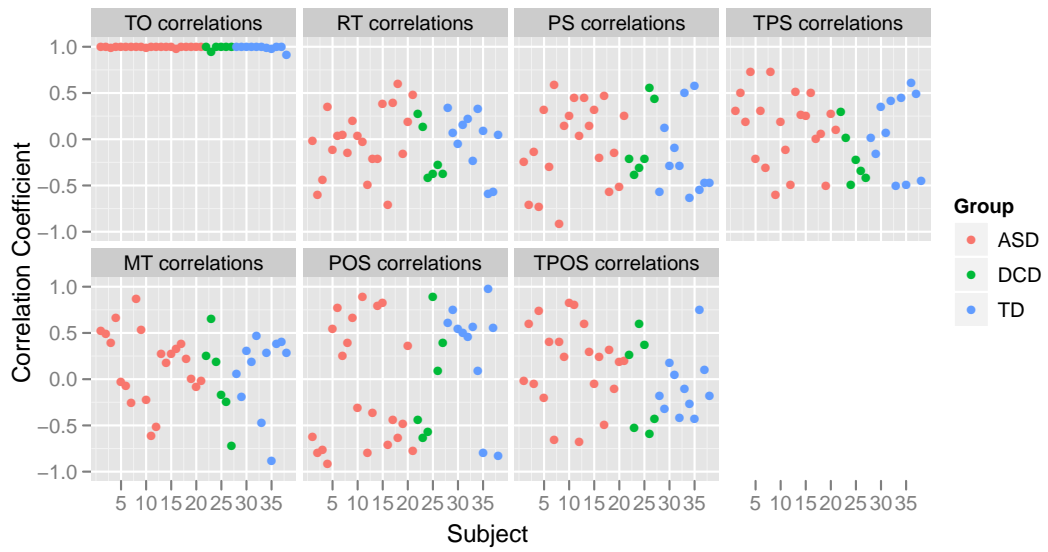


Figure 4.11: Pearson correlation between target orientation and each measure for each subject in the vision-only condition. Red, green and blue represent ASD, DCD and TD respectively.

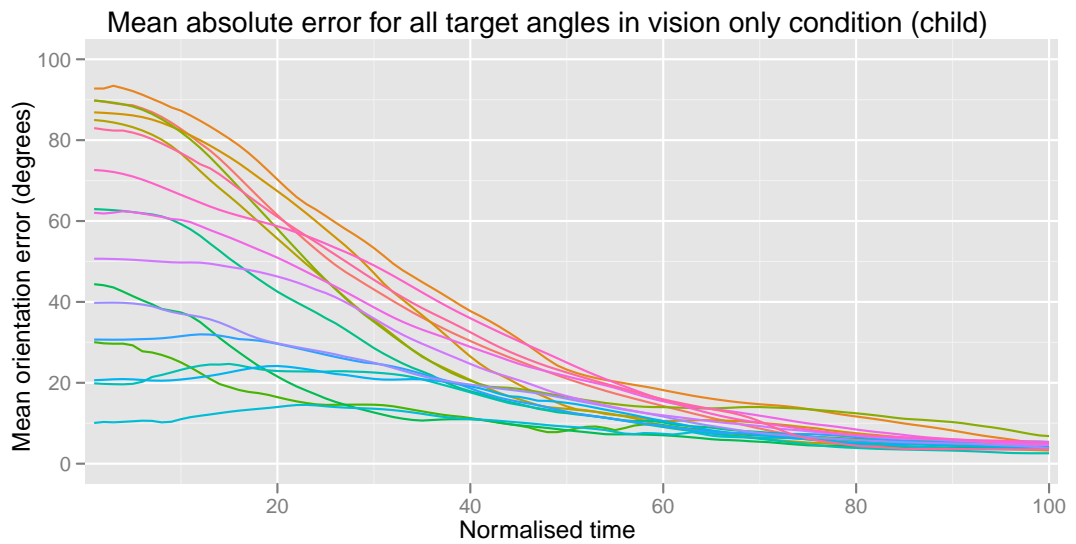


Figure 4.12: Mean absolute orientation error across normalised time. Data from all groups have been combined for each target due to insufficient numbers in the DCD and TD groups. As with the adult study, by 60% MT large wrist rotations have been completed and the rest of the movement involves smaller adjustments.

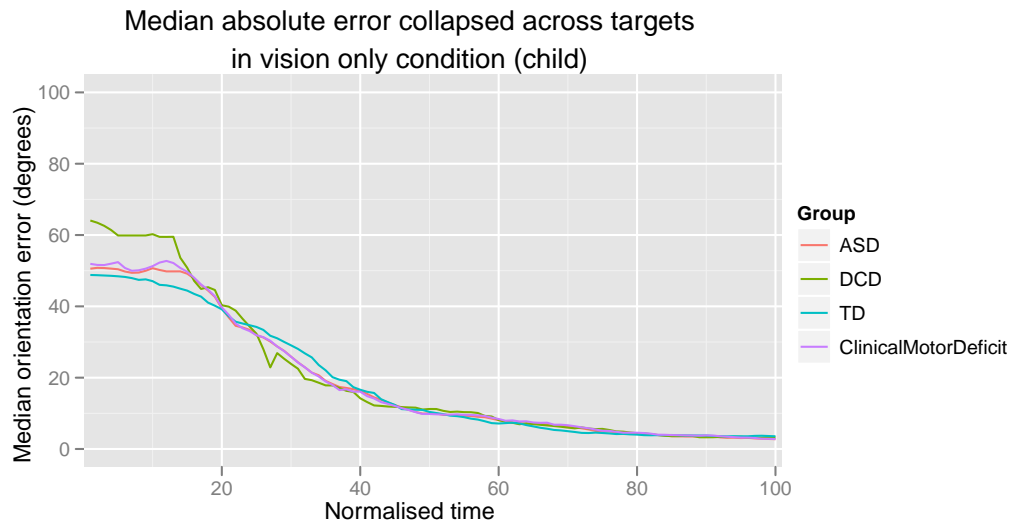


Figure 4.13: Median absolute orientation errors collapsed across targets for each group. Errors are shown across normalised time (from 0-100% movement time.)

were used. The spread of absolute errors in each group across conditions is shown in Figure 4.14. With vision, there was no significant effect of group on accuracy at 60% MT ( $H(2) = 1.11, p = 0.575$ ) or 100% MT ( $H(2) = 0.75, p = 0.687$ ). This was also true when vision was removed ( $H(2) = 2.28, p = 0.32$  and  $H(2) = 1.17, p = 0.557$  for 60% and 100% MT respectively). The null effect of group at both 60% and 100% MT suggests that groups completed the action in a similar way, with even those children with motor difficulties approaching their final orientation by the same time point as those with intact motor skills, suggesting similar motor planning strategies and capabilities.

As hypothesised and supported by findings in the adult study, there was a significant effect of visual feedback on accuracy at 100% MT, with a significant decrease in accuracy when vision was removed ( $Z = -4.20, p < 0.001$ ). There was no such effect at 60% MT ( $Z = -1.80, p = 0.071$ ).

Contrary to the hypothesis of significantly poorer performance in DCD when vision was removed, there did not appear to be any significant group\*visual condition interaction.

#### 4.9.2.2 Constant error

Mean constant error is shown in Figure 4.15. Mean constant error at 60% and 100% MT was calculated for each subject for each condition (DV, DN) and entered into a 3x2 ANOVA comparing groups on the two visual conditions. At 60% MT there was no effect of group ( $F(2, 34) = 1.97, p = 0.155$ ) and no effect of visual condition on constant error ( $F(1, 34) = 3.15, p = 0.085$  (GG)). There was no significant group\*visual condition

Absolute error in DN and DV between groups  
at 60% and 100% MT

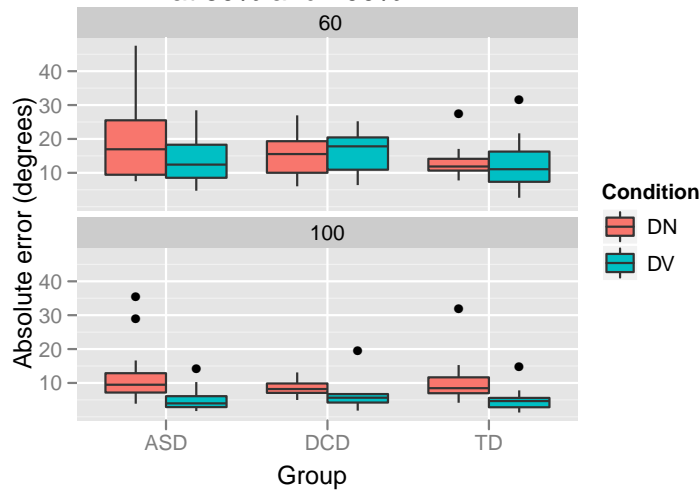


Figure 4.14: Mean absolute orientation error at 60% and 100% MT for each condition in each group. Accuracy significantly decreases when vision is removed. Group and group\*vision effects are not significant.

interaction:  $F(2, 34) = 0.19, p = 0.831$  (GG).

By 100% MT visual condition significantly affected constant error, with significantly greater error in the absence of vision:  $F(1, 34) = 15.09, p < 0.001, \eta_p^2 = 0.97$  (GG), mean difference (SE)=5.43 (1.40). Groups were still not significantly different ( $F(2, 34) = 0.9, p = 0.416$ ). The groups appear to be differentially affected by the removal of vision, with less effect on error in the ASD group, however the interaction effect fails to reach statistical significance ( $F(2, 34) = 3.01, p = 0.063$ ).

#### 4.9.2.3 Variable error

Standard deviations for each subject in each condition were calculated as a measure of variable error, and average error in each group is shown in Figure 4.16. Again data were not normally distributed therefore nonparametric analyses were conducted. There was no significant effect of group on precision with vision at 60% MT ( $H(2) = 1.69, p = 0.429$ ) or 100% MT ( $H(2) = 0.99, p = 0.609$ ). This was also true when vision was removed ( $H(2) = 2.28, p = 0.319$  and  $H(2) = 0.62, p = 0.734$  for 60% and 100% MT respectively). There was a significant main effect of vision at both 60% MT and 100% MT, with more precise actions when vision was provided ( $Z = -2.59, p = 0.01$  and  $Z = -3.92, p < 0.001$ ).

Constant error in DN and DV between groups  
at 60% and 100% MT

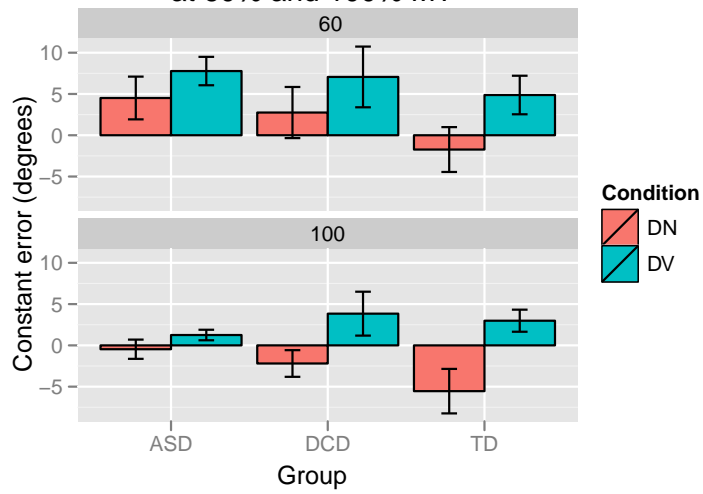


Figure 4.15: Mean constant orientation error between groups and condition at 60% and 100% MT. Error bars show SE. There is a significant effect of vision at 100% MT.

Variable error in DN and DV between groups  
at 60% and 100% MT

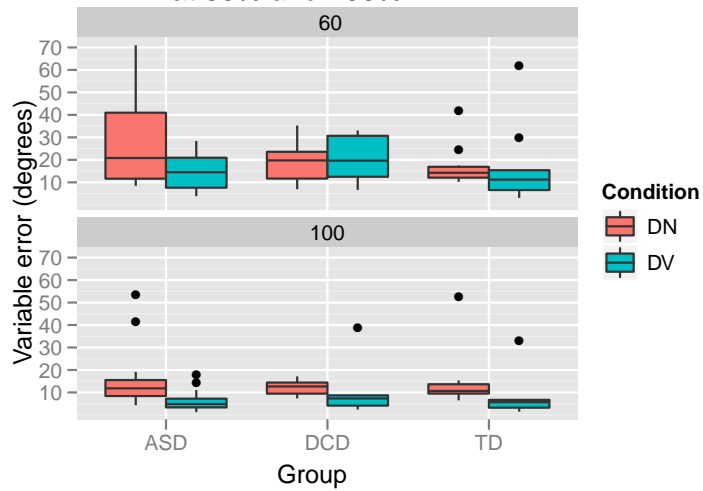


Figure 4.16: Mean variable orientation error at 60% and 100% MT for each condition in each group. Precision is significantly lower when vision is removed.

#### 4.9.2.4 Comparing MABC-defined groups

As with studies in previous chapters, performance by motor-impaired children with ASD and children with DCD was compared to ensure they were statistically equivalent, before entering these children into a single motor-impaired group. There was no significant difference between the two subgroups of motor-impaired children on any measure (see Tables C.1 and C.2 in Appendix C), supporting the use of the ‘clinical motor deficit’ group in further analysis.

Mean constant error between MABC-defined groups is shown in Figure 4.17 and the range of absolute and variable errors is shown in Figure 4.18. The ASD ‘pure’ group was too small to include in formal analyses therefore the following analyses compare the clinical motor deficit group and TD.

For constant error the group difference at 60% MT approached significance ( $F(1, 31) = 4.13, p = 0.051, \eta_p^2 = 0.58$ ), with greater error in the clinical motor deficit group [mean difference (SE)=4.50 (2.21)]. By the end of the movement there was no significant effect of group on constant error ( $F(1, 31) = 1.99, p = 0.168$ ). Absolute and variable error did not differ between groups at either 60% or 100% MT (see Table C.3 in Appendix C).

The effect of vision mirrors that of the previous analyses with diagnostic groups. There was no significant effect of visual condition on constant errors at 60% MT ( $F(1, 31) = 3.027, p = 0.092$  (GG)) but by 100% MT the difference was significant, with lower errors when vision was provided:  $F(1, 31) = 14.99, p = 0.001, \eta_p^2 = 0.96$ , mean difference=5.42, SE=1.40. There was also a significant group\*condition interaction, with greater sensitivity to the removal of vision in the TD group:  $F(1, 31) = 4.95, p = 0.033, \eta_p^2 = 0.58$  (GG). Absolute error was significantly lower with vision at 60% MT ( $Z = -1.99, p = 0.046$ ) and 100% MT ( $Z = -3.90, p < 0.001$ ). The same was true for variable error ( $Z = -2.42, p = 0.015$  and  $Z = -3.57, p < 0.001$  for 60% and 100% MT respectively).

## 4.10 Discussion

This study continued the investigation of visual and proprioceptive cue processing in children with ASD, DCD, and TD, after identifying a possible double dissociation (discussed and tested in Chapter 3). The study intended to assess the effect of the removal of vision on the ability to use proprioceptive target information in a goal-directed action.

As with findings in the previous chapter, there was no significant difference between ASD and DCD (and TD) in either the effect of the removal of vision, or their ability to use proprioceptive orientation information to guide movements. All groups were better able to perform the posting task when vision was allowed, and there was no significant

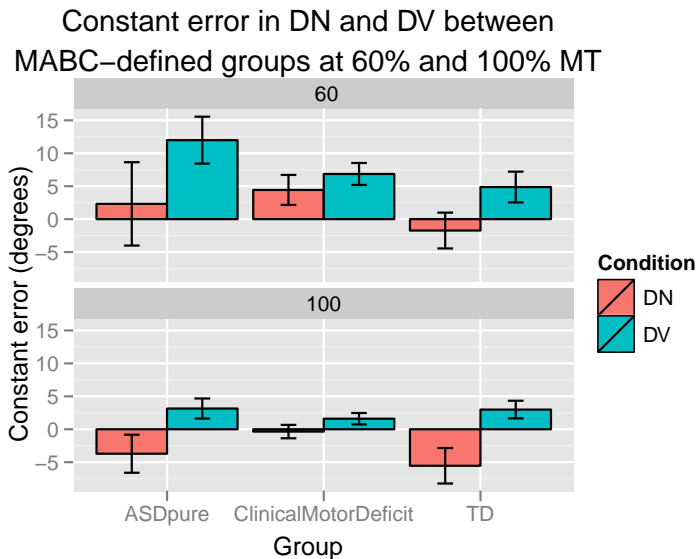
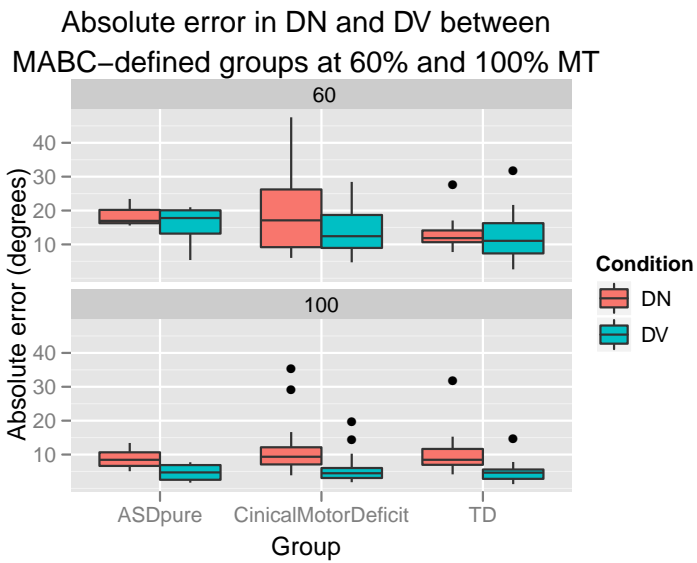


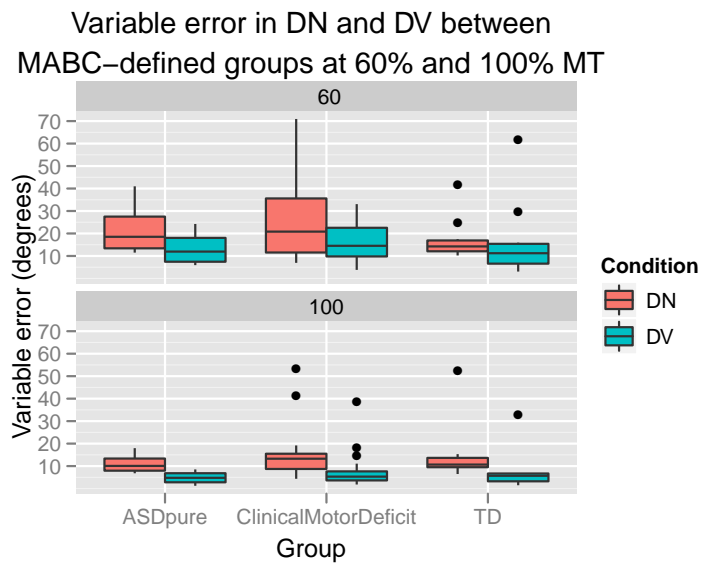
Figure 4.17: Constant error at 60% and 100% MT for MABC-defined groups. Note that the ASD pure group was not included in analysis due to small sample size. There is a significant effect of vision at 100% MT.

group difference in the use of proprioceptive information in the absence of vision. This was true for all measures when comparing diagnostic groups and also for the majority of measures when comparing groups based on observed motor ability. At 60% MT the motor impaired group showed larger constant errors than TD, however this difference disappeared by the end of the movement, and throughout the movement variability and absolute error did not differentiate motor-impaired and TD children. These null results are not thought to be due to low statistical power, particularly the comparison of the clinical motor deficit group and TD, as sample sizes were within the typical range for similar studies, and an inspection of the average error associated with each group shows that they are often strikingly similar. Counter to the initial hypothesis, there is therefore still no evidence to suggest that ASD and DCD are associated with a proprioceptive and visual bias respectively. It is still possible that ASD with spared motor skills is associated with a proprioceptive bias, as found in previous research (see Chapter 3), however as with the follow-up study detailed in the second half of Chapter 3, there were too few of these children to investigate this additional hypothesis. These findings again support recent changes in the DSM-5 comorbidity criteria for ASD and DCD. The findings do however raise an important question regarding the nature of motor difficulties in motor-impaired children more generally.

Why was there no greater negative effect of the removal of vision in the DCD or clinical motor deficit group compared to TD, and little evidence of overall poorer performance in the motor-impaired children? These findings are puzzling given the demanding nature of the task without vision. The children with motor deficits had



(a) Absolute error. Note data were not normally distributed. There is a significant effect of vision at both time points.



(b) Variable error. Note data were not normally distributed. There is a significant effect of vision at both time points.

Figure 4.18: Posting errors in MABC-defined groups. Note that the ASD pure group was not included in analysis due to small sample size.

clear impairments in general motor ability, as measured by the MABC-2 administered during initial testing, and often noted in medical records. It is therefore unlikely that the unexpected and counter-intuitive results are due to poorly defined groups. However there is no obvious explanation for these findings. It was ensured that children always kept their eyes closed when necessary, and if they did not they were blindfolded. In trials where the letter hit the board and was then moved around until it could be forced through the slot, the data from this last portion of the action were not included (as by this point the velocity would have fallen below the 50mm/s threshold). It can be assumed therefore that data were not affected by either inadvertent vision in the no-vision condition, or corrective movements once the letter had made contact with the board.

It is unlikely that children were well-practised in tasks similar to the blind posting task. However, children with motor impairment have been found to persist in using ineffective motor plans (Astill & Utley, 2006), so perhaps the novelty in the present study encouraged the use of an ‘off-the-cuff’ approach, as there was no preexisting motor plan for this task. This may explain their preserved ability. All children will have been familiar with the MABC-2 tasks to some extent so their familiarity with these tasks perhaps exacerbated their motor problems as they persisted in using inefficient strategies. However evidence for motor deficits in these groups comes from studies using a variety of tasks, a number of which are novel (e.g. Ranta & Mostofsky, 2011; Haswell et al., 2009).

So how could children who had difficulty catching a ball, assembling Meccano and placing pegs in a board complete a task which is arguably much more demanding? The jarring difference in performance on a blind posting task and the MABC-2 tasks perhaps highlights the limited scope of the MABC-2, which focuses on the end product of an action, rather than the sensory and kinematic components it is built from. If motor-impaired children fail the MABC-2 but pass this task, what else are they able to do, and why? Understanding what motor impaired children *can* do may be more instructive to understanding ASD and DCD than focusing on what they cannot do, particularly given the extraneous variables that could contribute to high-level difficulties, such as motivational or attentional problems.

This task was thought to be the most appropriate to test the effect of sensory cues on action, as it is a true goal-directed action (cf. the reaching task in the previous chapter), and the adult study provided evidence that the proprioceptive manipulation was effective. However children with motor difficulties were not significantly impaired. As well as comparable orientation scaling, kinematic variables did not significantly differentiate groups, contrasting with a number of previous studies finding extended MT and RT in motor impaired children (e.g. Johnston et al., 2002; Glazebrook et al., 2006; Donnellan et al., 2013). Results from this study provide no evidence to support either the initial double dissociation proposed, or the alternative dissociation concerning ASD ‘pure’ and children with motor impairment. These findings do however sit well

alongside the null effect of group in the reaching task reported in the previous chapter.

## 4.11 General Discussion

This chapter has reported two studies assessing the use of visual and proprioceptive cues to target orientation in perception and action tasks. The adult study found that the spatial location of proprioceptive orientation information only differentially affected the control of wrist orientation in the absence of vision. With visual information of target orientation, proprioceptive information was of little benefit. When vision was removed, errors were significantly reduced when proprioceptive information of the target orientation coincided spatially with the end-point of the action, compared to spatially indirect proprioceptive information. These findings extend those from Gosselin-Kessiby et al. (2008, 2009), suggesting that proprioceptive information can inform online correction of transitive actions, and this use of proprioceptive information is enhanced when spatially congruent with the end-point of the required movement. Gosselin-Kessiby et al. (2008, 2009) found greater errors for proprioceptively-defined targets in matching compared to posting, and this was also found in the present study, although in this case both tasks showed an effect of the directness of proprioceptive information. The effect of proprioceptive condition in the present matching task suggests that intransitive actions are also sensitive to proprioceptive cues in the absence of vision. The clear differences between direct and indirect conditions in the present study and the small magnitude of errors made in no-vision conditions supports the suggestion that proprioception can be used to effectively guide actions, both automatically (Gosselin-Kessiby et al. 2008, 2009) and intentionally.

The second study further investigated the role of visual and proprioceptive cues on task performance in children with ASD, DCD and TD. As with findings detailed in the previous chapter, there is still no evidence for a dissociation in the use of vision and proprioception in ASD and DCD, and no significant deficit in the ability of children with motor deficits to use proprioceptive information to inform goal-directed actions.

Results from both studies support and extend previous evidence for the dissociation between goal-directed and intransitive or pantomimed actions. Previous studies have focused on actions which are only visually-defined, and the present study extends these findings to actions towards proprioceptively-defined targets.

## Chapter 5

# Proprioception and susceptibility to the Rubber Hand Illusion

This chapter continues the investigation of sensory contribution to perception and action. Two studies are reported here, each investigating the role of proprioceptive acuity in resolving cue conflict. The first study employed neurotypical adults in two tests of proprioceptive acuity and a visuo-tactile/proprioceptive conflict task (the rubber hand illusion: RHI). The second study assesses susceptibility to the RHI in children with ASD, DCD and typical development. Work in this chapter compliments work detailed in Chapters 3 and 4, involving more basic tests of visual and proprioceptive cue weighting in children with ASD and DCD.

### 5.1 The rubber hand illusion

When there is a conflict between our senses, the brain must weight the sensory information according to what is deemed most reliable. This conflict has already been used in Chapter 3, which reported one experiment using a prism and another using a mirror to create a conflict between seen and felt location. Another example of cue conflict is the RHI, which uses a conflict between visual and tactile input to create visual capture of touch prior to a judgement of proprioceptive positioning. If a rubber hand placed in front of a subject is stroked in a manner identical to tactile stimulation given simultaneously to their real (hidden) hand, the rubber hand may be perceived as belonging to the subject: as the sensory cues coincide, they are experienced together so the felt touch is misattributed to the seen touch (Botvinick & Cohen, 1998; Tsakiris & Haggard, 2005).

When asked to estimate where their hand is following synchronous tactile and visual stimulation, subjects tend to estimate that it is closer to the rubber hand: this is termed proprioceptive *drift*. Drift is commonly used as an objective indication of illusion strength, with larger drift towards the rubber hand following synchronous stimulation indicative of a stronger sense of ownership of it (although evidence of a

reported illusion without accompanying drift prompted Rohde, DiLuca & Ernst (2011) to challenge this assumption of ownership). Proprioceptive *shift* (the difference in drift following synchronous and asynchronous visuo-tactile stimulation) is also commonly used. Asynchronous stimulation means the stroking on the real and rubber hand is out-of-time. The subjective illusion and/or proprioceptive drift tends to occur only when tactile stimulation delivered to the (real and rubber) hands is temporally and spatially congruent. This effect of congruency is the main finding of the majority of studies, including those assessing whole body illusions such as Ehrsson (2007), however various studies have also investigated other properties of the rubber hand (or other foreign object) that affect the illusion. For example, drift tends not to occur when the rubber hand is at a biologically implausible angle (Tsakiris & Haggard, 2005; Ehrsson, Spence & Passingham, 2004). The resemblance of the foreign object to a real hand also affects proprioceptive drift, with a wooden stick proving less successful than a rubber hand (Tsakiris & Haggard, 2005), although the handedness of the rubber hand and real hand must be congruent (Tsakiris & Haggard, 2005; Petkova & Ehrsson, 2009).

### **5.1.1 What can explain individual differences in susceptibility to the RHI?**

It has been noted that not everyone, even in typical groups, experiences the RHI and similar illusions of body representation and ownership. While the majority of studies do not give figures for failure rate, Ehrsson and colleagues report that approximately 3 in 10 typical adults do not experience the illusion (7 out of 25 (28%) in Ehrsson et al., 2004, and 9 out of 28 (32%) in Ehrsson, Wiech, Weiskopf, Dolan & Passingham, 2007). In both studies subjects experienced a 60 second stimulation duration and illusion susceptibility was measured with questionnaires. The distance between the two hands has been found to affect illusion rates, with 50% of subjects failing to consistently experience the illusion across a range of separation distances between the real and rubber hand (15-60 cm) (Davies, White & Davies, 2013). At each separation (15, 30, 45 and 60 cm) around a third of 28 subjects failed to experience the illusion, in line with Ehrsson et al. (2004) and Ehrsson et al.'s (2007) failure rates.

As the RHI is a kind of body-distortion illusion, a number of studies have investigated the relationship between the RHI and our everyday awareness of our bodies. David, Fiori & Aglioti (2013) found no significant correlation between self-reported 'bodily awareness' and subjective RHI strength in either typical adults or those with high self-reported 'bodily awareness'. It was anticipated that the latter would have better proprioceptive acuity and show reduced RHI susceptibility. The null result may be due to poorly defined groups however, with the 'bodily awareness' measure failing to capture actual proprioceptive acuity. (It was measured with questions about people's awareness of experiencing goosebumps etc.) A significant positive correlation *was* found between RHI susceptibility in the typical group and visual bias in a rod-and-frame test,

which measures the effect an angled framing square has on the perceived angle of a rod inside it (visual-context effects). There was also a slight trend for a similar effect using the Stroop task assessing word-colour conflict, with higher RHI susceptibility associated with greater errors in the Stroop task. The authors suggest that an inability to disengage attention from certain aspects of the stimulation may increase illusion susceptibility. Another correlational study found that RHI strength (again measured by questionnaires) was positively correlated with body plasticity and suggestibility (measured using the Trinity Assessment of Body Plasticity and Creative Imagination Scale respectively) (MacLachlan, Desmond & Horgan, 2003). While these results are informative it is important to note that the RHI only involved synchronous stimulation, and in some cases only associations between subjective measures are reported.

Although some work has considered self-reported body awareness, there do not appear to be any studies investigating the RHI in neurotypical adults in relation to an objective measure of proprioceptive acuity. The role of proprioceptive acuity has however been investigated with regards to eating disorders. Those with eating disorders such as Anorexia Nervosa have been found to have poor proprioceptive acuity (Grunwald, Ettrich, Busse, Assmann, Dahne & Gertz, 2002) and tactile sensitivity (Keizer, Smeets, Dijkerman, van den Hout, Klugkist, van Elburg & Postma, 2011; Keizer, Smeets, Dijkerman, van Elburg & Postma, 2012). Additionally, Eshkeviri, Rieger, Longo, Haggard & Treasure (2012) report that they show proprioceptive deficits and/or an increased sensitivity to visual representations of the body (focusing on the body in observable, objectified terms). These features were associated with increased sensitivity to the RHI. There was also a significant positive correlation ( $r=0.3$ ) between drift and embodiment, and interoceptive deficits (a disturbance of internal body representation) were found to significantly predict RHI susceptibility (Eshkeviri et al., 2012). However, it is not clear whether the correlation with drift (the difference between pre- and post-stimulation estimates) is specific to drift in the synchronous condition.

The relationship between interoceptive sensitivity and proprioceptive acuity is unclear. Tsakiris, Tajadura-Jimenez & Costantini (2011) found that highly interoceptive individuals (those able to count their heartbeat without taking their pulse) were less susceptible to the RHI, despite proprioceptive acuity being statistically equivalent between high and low interoceptive subjects. It should be noted however, that the baseline proprioceptive acuity measure used in Tsakiris et al. (2011) seems problematic, as it requires only a verbal estimate of hand positioning and benefits from the use of visual landmarks. Having perhaps hastily ruled out the effect of proprioceptive acuity, the authors suggest that findings might be explained by an increased allocation of attention to multi-sensory processing in those with lower interoceptive sensitivity; or alternatively, those with high interoceptive sensitivity may be able to make use of both interoceptive and exteroceptive cues when perceiving the body, compared to a bias towards exteroceptive cues in those with lower interoceptive sensitivity.

In summary, the association between proprioceptive acuity and RHI susceptibility is interesting both in its own right and also with reference to recent studies involving people with ASD, given the suggestion of a proprioceptive bias in ASD. From the studies detailed above, it appears that proprioceptive acuity may affect susceptibility to the RHI, although to date the most compelling evidence comes from studies involving those with eating disorders. The effect of individual differences has been investigated in part, but has not been directly related to standard measures of proprioceptive acuity. The first study in this chapter aims to investigate whether the acuity/susceptibility association can be generalised to a typical adult population using standard tests of proprioceptive acuity. It is hypothesised that people with higher proprioceptive acuity, who are better able to locate their body using proprioceptive information, will show relative resistance to the rubber hand illusion, compared to people with lower proprioceptive acuity. The second study reported in this chapter compares ASD, DCD and TD children on the RHI, using proprioceptive acuity measures detailed in previous chapters to investigate whether diagnosis or proprioceptive acuity is significantly associated with varying illusion susceptibility. This continues the investigation of the proposed vision/proprioception double dissociation in ASD and DCD.

Before conducting a study using the RHI it is important to consider that the illusion can be carried out in a number of ways. The RHI procedure used in the adult study here was intended to be identical to that used in the child study, so design variations had to allow for effective testing of both populations, and be suitable for the specific questions of the two studies. Commonly used variations in procedure and variables are described in the sections below.

## **5.1.2 Variations in stimulation duration and type**

### **5.1.2.1 Stimulation duration**

It is not clear how long the hand needs to be stroked for before the subject begins to experience the illusion. Reports of the time required vary from less than 30 seconds [11 seconds (Ehrsson et al., 2004); 23-25 seconds (Davies et al., 2013)] to as long as 2.5 minutes (Armel & Ramachandran, 2003). The length of time may be heavily affected by individual differences, or alternatively may be fairly arbitrary, with studies giving longer durations as a fail-safe precaution. It is important not to give too short a stimulation period as there is evidence of brief periods of asynchronous stroking eliciting proprioceptive drift, even though drift should be specific to synchronous conditions (Rohde et al., 2011). Additionally, Rohde et al. (2011) found that very frequent interruptions to synchronous stimulation disrupted the illusion, and a large number of short bursts of stimulation did not lead to the illusion through a cumulative effect.

Stimulation durations used in previous studies tend to range from 90 seconds to 4 minutes of continuous stimulation (e.g. Kammers, deVignemont, Verhagen & Dijker-

man, 2009; Costantini & Haggard, 2007; Ocklenburg, Peterburs, Pinnow & Gunturkun, 2011; Tsakiris & Haggard, 2005), although stimulation for as long as 30 minutes has also been reported (Botvinick & Cohen, 1998). Some studies break up longer stimulation durations with periodic proprioceptive estimates (e.g. Tsakiris & Haggard, 2005). Such interruptions to stimulation have been found to have an adverse effect on illusion strength when given very frequently. Rohde et al. (2011) found no robust illusion (no significant effect of synchrony) with interruptions every 10 seconds, however with interruptions at 40 second intervals there was a clear difference between synchronous and asynchronous conditions (in the expected direction) by the end of the 2 minute stimulation period. With the exception of the 10 second condition described in Rohde et al. (2011), all of the above studies replicated the expected illusion using slightly different stimulation durations.

There are obvious negative effects to using either too long or too short a stimulation period. If insufficient time is given then the illusion will not work optimally as the sensory congruency has not had sufficient time to be processed: if too much time is given this may affect attention and concentration (particularly in children and clinical groups) which in turn could affect the strength of the illusion. Some studies allow subjects as long as they need to experience similar illusions (e.g. Bremner et al., 2013). While this ensures that every child experiences the illusion before any measurements are taken, it also introduces exposure time as an extraneous variable which could impact on the results of these studies. This would be a particular concern if testing clinical groups or young children: due to their varied approach to tasks it is expected that such procedures could result in a large degree of variance in exposure time.

#### **5.1.2.2 Type of stimulation**

Just as stimulation duration varies, so too does the type of stimulation. Some procedures focus stimulation on just one finger (e.g. Kammers et al., 2009) while others focus on various locations on the hand (e.g. Rohde et al., 2011). Both approaches can elicit the illusion, and similarly both manual and mechanical delivery of stimulation can result in a successful illusion (e.g. Tsakiris & Haggard, 2005). If stimulation is delivered manually this reduces the precision with which a complex pattern can be reproduced, as spanning multiple hand locations is both difficult for the experimenter to remember and difficult to coordinate with both hands. It has been shown that stroking stimulation leads to a stronger illusion than tapping (Haans, Kaiser, Bouwhuis & Ijsselstein, 2012), which is less commonly used (e.g. Paton, Hohwy & Enticott, 2012). It is suggested that the stroking advantage is due to the greater amount of information available through stroking compared to tapping: the increased stimulation allows for more visual and tactile/proprioceptive information to be correlated and integrated to form the illusion (Haans et al., 2012). It is also possible that asynchronies are simply harder to detect when given minimal tapping stimulation.

### 5.1.3 Methods of measurement

As well as differing stimulation procedures, there is also variation in the method of measurement of the illusion. Often the strength of the illusion is measured using a subjective measure and/or objective measure, with the three most common being questionnaires [often from Botvinick & Cohen (1998)], skin conductance and proprioceptive drift (Ocklenburg et al., 2011). Using subjective measures alone is insufficient and tends to focus on the illusion as one of agency and self awareness. For example, self reported ‘eeriness’ in a whole body mirror illusion used by Altschuler & Ramachandran (2007) tells us little about the underlying sensory processing behind the illusion. Subjective measures are also highly variable between subjects and may be sensitive to perceived expectations. More objective measures, such as skin conductance and proprioceptive drift or shift tend to allow for an investigation of the sensory basis of the illusion, without the need to address the phenomenological experience of ownership. Skin temperature and conductivity of the real hand(s) seems to be a less reliable objective measure compared to proprioceptive drift. For example, Moseley, Olthof, Venema, Don, Wijers, Gallace & Spence (2008) found significant cooling in the stimulated hand following a synchronous RHI condition, while Paton et al. (2012) failed to find an effect in either hand. Similarly, Ocklenburg et al. (2011) and Armel & Ramachandran (2003) found an effect of the illusion on skin conductance in the non-stimulated hand only when the rubber hand was threatened, but not in the standard illusion.

Drift is usually defined as either the difference between the post-stimulation response and the veridical position of the hand (e.g. Tsakiris & Haggard, 2005); or the difference between the pre- and post-stimulation response (e.g. Cowie, Makin & Bremner, 2013). The use of the veridical position as a comparison is arguably the most appropriate, as pre- and post-stimulation conditions differ as the pre- estimate often does not follow tactile stimulation or viewing of the rubber hand. However proprioceptive *shift* is arguably the most sensitive measure, as only the synchrony of stroking differentiates experimental (synchronous) and baseline (asynchronous) trials. When proprioceptive drift/shift is used there are a number of methods used to indicate the felt location of the hand. Some studies use a matching technique, whereby the non-stimulated hand is moved above or below the table to the position felt to be adjacent to the hand. This is done either in the absence of vision (e.g. Cascio, Foss-Feig, Burnette, Heacock & Cosby, 2012; Cowie et al., 2013; Haans et al., 2012; Botvinick & Cohen, 1998), or with vision of the apparatus, but neither real nor rubber hands (e.g. Kammers et al., 2009). Others have used a similar pointing method but do not use an active movement: the experimenter will take the blindfolded subject’s hand and move it until they say ‘stop’ (e.g. Paton et al., 2012). Others do not require any kind of proprioceptive matching and instead require the subject to verbally report the number on a ruler that corresponds to the felt location, or to verbally guide the experimenter’s finger along a ruler (e.g. Costantini & Haggard, 2007; Germine, Benson, Cohen & Hooker, 2013;

David et al., 2013; Palmer et al., 2013; Davies et al., 2013; Tsakiris et al., 2011). A potential problem with both this and the visually-guided proprioceptive response is the possibility for subjects to be guided visually to repeat the same responses. It may also inhibit responses that do not cross the midline, as visually it is very apparent that these are inaccurate as the subject is aware that the hand is roughly in line with the shoulder, which is clearly beyond the body midline.

While drift is often taken as a proxy for subjective experience of the illusion, Rohde et al. (2011) challenge this assumption, as they found drift following no tactile stimulation and brief asynchronous stimulation (which rarely elicits feelings of ownership). For this reason they suggest that drift is not a reliable measure of illusion susceptibility. There is some support for this: Davies et al. (2013) also report drift in asynchronous conditions (relative to baseline) without accompanying subjective experience of the illusion. Conversely they also found that when the real and rubber hand were placed 60 cm apart subjects reported a subjective illusion (in 68% of cases), but showed no significant drift. It is important to note however that very few studies separate the hands to this extent, and this finding was specific to this separation distance. This study highlighted that the objective-subjective measure dissociation (apparent illusion in one measure but not another) was only true for proprioceptive drift in the synchronous condition (compared to a pre-stimulation baseline). There was no dissociation when considering proprioceptive *shift*, which describes synchronous drift relative to performance in the asynchronous condition. This discrepancy supports the idea that an asynchronous baseline is most appropriate, and therefore proprioceptive shift is more appropriate than drift measures.

While the observations of drift following brief asynchronous or vision-only conditions, and a slight discrepancy between subjective and objective measures in some circumstances is interesting, it does not affect the present study as subjective feelings of ownership are not of interest. Additionally, the unexpected drift in asynchronous and vision only conditions could merely highlight the known tendency for us to estimate towards our midline under uncertainty (Wann & Ibrahim, 1992; Gross & Melzack, 1978; Gross, Webb & Melzack, 1978).

## 5.2 Measuring proprioceptive acuity

The focus of this chapter is the role that proprioceptive acuity plays in the RHI. Proprioceptive acuity is often measured using some form of postural matching task, or a spatial location matching task. Postural matching requires subjects to move one limb so that it feels that it is in the same posture as the other, resulting in a mirror image down the midsagittal plane. This is usually done without vision (see Goble (2010) for an overview of variations of the basic joint position matching paradigm). Spatial location matching uses the PP condition of the first task described in Chapter 3. A minority of studies use other tasks to assess proprioceptive acuity such as weight dis-

crimination (Tremblay, Estephan, Legendre & Sulphur, 2001) and detection thresholds for passive movements (Cammarata, Schnitzer & Dhaher, 2011).

The effect of arm used to match has been investigated, with a general consensus that matching is easier when using the non-dominant arm to match the dominant one (Goble, 2010). The present study requires right handed subjects to use their left hand as a target and move the right until it matches, in order to increase task difficulty, and also so that it matches the procedure used in the spatial location matching task in Chapter 3, which is adapted for the present study.

The RHI cannot be solved through postural matching, and instead requires subjects to identify a tactile/proprioceptively-defined spatial location (similar to the PP condition in the matching task in Chapter 3). The present study will use both a postural matching task and a spatial location matching task, in which postural information is uninformative. Both are included in order to investigate the relationship between different types of proprioceptive acuity, and whether high acuity in one will always result in high acuity in the other.

### **5.3 Study 1: The relationship between proprioceptive acuity and RHI susceptibility in neurotypical adults**

This study investigates the effect proprioceptive acuity has on cue conflict resolution in the RHI in typical adults.

## **5.4 Methods**

### **5.4.1 Subjects**

Twenty self-reported right-handed adults (6 males) aged 21-33 (mean=26 years, SD=3.44) participated, following recruitment via an email advertisement or an advert on a university website. All reported having no diagnosis of ASD, DCD, CP, Multiple Sclerosis or other neurological condition affecting movement, and all reported having normal or corrected-to-normal vision. One subject was removed and replaced as he reported having scoliosis (curvature of the spine to the side) and it was unclear whether this would affect postural matching ability as the trunk of the body was noticeably asymmetric. No subject reported having experienced the RHI before. All subjects received £5 for their participation.

### **5.4.2 Procedure**

All participants completed three tasks in a fixed order: RHI; postural matching; and proprioceptive spatial location matching. This order was chosen so that both tasks involving motion tracking were completed successively, and so that the two most similar

tasks (RHI and spatial location matching) were not completed one after the other. Furthermore, breaks were given between tasks to minimise any residual effect of earlier tasks on later ones. Instructions were given at the start of each task. There was no feedback given throughout any of the tasks, however subjects were debriefed after testing.

### 5.4.3 Rubber hand illusion

#### 5.4.3.1 Apparatus

A wooden box with two open faces (front and back), measuring 76.5 x 45 x 31 cm (length x depth x height) was used for the RHI (this is the same apparatus used in the reaching task detailed in Chapter 3). A diagram of the apparatus is shown in Figure 5.1. A central right/left wooden divider separated the real hand (on the left) and the rubber hand (on the right). The top of the box comprised four equally sized lids, two on either side of the divider. The lid closest to the divider on the right side was lifted to allow for vision of the rubber hand and was replaced when estimates of hand location were made. Estimates were made using a bead on a slider at the top of the box. The slider was 17 cm from the front of the box, in easy reach of all subjects. A TrakSTAR electromagnetic motion tracker (Ascension Technology, Burlington, VT) was used for recording the position of the real left hand (via a marker on the index finger) and the response coordinates (via a marker attached to the top of the bead) once the estimate had been made. The stationary left hand was monitored to ensure that it was in approximately the correct position when the estimate was made. The TrakSTAR magnet was located in a fixed position at the back of the box.

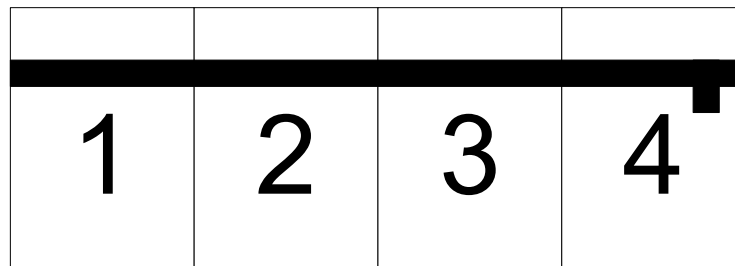


Figure 5.1: RHI apparatus showing the four lids, slider and response bead. The real hand is placed under lid 2 and the rubber hand is placed under lid 3. The areas under lids 2 and 3 are separated by a wooden divider.

#### 5.4.3.2 Procedure

A rubber model of a left arm was placed inside the apparatus to the right of the central divider. The participant placed their own left hand on the other side of the central divider (left compartment), placing it flat on the bottom of the box. The participant

adjusted the seat so that they were able to look inside the box from above, whilst being able to comfortably keep the left hand in position. The participant's left hand was positioned 7 cm from the divider and they were instructed to maintain the same posture as the rubber hand. The participant was told that the experimenter would stroke their hand with a brush, and they should keep it still throughout. They should also ensure that they look at the rubber hand. Subjects completed three practice trials, which allowed them to experience brushing on the real hand and gain practice using the response bead. For practice trials neither real nor rubber hand were visible and the index finger was brushed for 10 seconds. Subjects then closed their eyes and moved their right hand (previously on their knee or by their side) up to the response bead and moved it until it was felt to be above the real index finger. The subject opened their eyes once the response was recorded and the bead had been reset to the start of the runner. Vision was not allowed during responding to prevent the use of visual landmarks to guide responses.

Experimental trials were delivered in two blocks of four, with synchronous and asynchronous blocks counterbalanced across subjects. The switch from one condition to the other was not signalled to the subject. In the first trial of each block, 60 seconds of stimulation was delivered to the (hidden) real and (seen) rubber hand. In subsequent trials stimulation was delivered for 30 seconds<sup>1</sup>. After each stimulation period the rubber hand was covered, subjects closed their eyes and moved the bead to indicate finger position, as per practice trials. During stimulation periods the finger was stroked approximately once per second<sup>2</sup>.

#### **5.4.4 Proprioceptive postural matching**

##### **5.4.4.1 Apparatus**

A wooden board with two handles, spaced 15.4 cm apart was used (see Figure 5.2). Each handle could be rotated through 360° or secured in one of 18 positions. Again, a TrakSTAR system was used to collect data. One marker was attached to the end of each handle and the magnet was located in a fixed position at the side of the board.

##### **5.4.4.2 Procedure**

The participants sat at a table and were blindfolded throughout the experiment. The board was placed flat on the table straight in front of the subject and their left hand/forearm was placed on the left handle which was in one of three possible ori-

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<sup>1</sup>A short pilot study tested subjects for 5 minutes in each experimental condition with 5 trials each lasting 60 seconds. Subjects were unable to sustain attention throughout the 5 minute trial. It was decided to allow 60 seconds for the first trial and 30 seconds for each of 3 subsequent trials per condition.

<sup>2</sup>The experimenter could have used a pacing metronome through headphones to deliver temporally controlled stimulation, however this would have been impractical in the child version of the task (where being able to carefully monitor the child's visual attention was deemed more important than having tightly controlled stimulation).

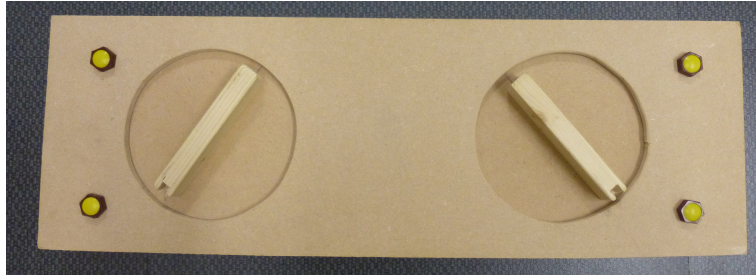


Figure 5.2: Postural matching board

entations. The right hand/forearm was placed on the other handle. The right arm always started at the same orientation ( $20^\circ$ ). The participant was instructed to move their lower right arm until the right side of the body felt the same as the left side. If subjects matched the hands so that they both faced the same way they were reminded of the instruction and if necessary given a demonstration (the experimenter moved the blindfolded subject's arms, using a target angle not used in the experiment). This happened very rarely. Subjects completed fifteen trials, five per three unique targets. The targets given were restricted by the maximum flex of the wrist. Targets were vertical and  $30^\circ$  clockwise and anticlockwise from vertical. The shortest and longest required distance to move the right arm from the starting position was therefore  $40^\circ$  and  $100^\circ$ .

#### 5.4.5 Proprioceptive location matching

##### 5.4.5.1 Apparatus

The apparatus described in the first experiment in Chapter 3 was used and set up as per the PP condition described in that chapter.

##### 5.4.5.2 Procedure

Participants sat at a table in a height adjustable chair, which was adjusted so that the eyepiece was at eye level. They were instructed to look through the aperture at all times, to ensure that their trunk remained relatively stable across trials. The aperture was covered from the inside so the target could not be seen. They were instructed to put their non-dominant hand (left) through the top left hole in the apparatus and hold the bottom of the target (stick) with a pencil grip. They then reached their right hand through the lower right hole and found the slider. Once it was established that the participant could comfortably move the bead along the whole length of the slider the experiment began. Participants were instructed to move the bead to the location of the target. The target positions used in the plano conditions described in Chapter 3 were used (three unique targets), with each presented five times in a fixed random order.

## 5.5 Results

Results for each task will be detailed separately, followed by correlational analyses investigating relationships between the three tasks. Where the sphericity assumption is violated in parametric analyses the degrees of freedom will be adjusted using a Greenhouse-Geisser (GG) adjustment.

### 5.5.1 RHI

Note that responses in the no-vision practice trials are not used in any analysis as they were only intended to familiarise subjects with the tactile stimulation and the use of the response bead.

#### 5.5.1.1 Effect of estimate number

The effect of estimate number was explored as a within subject variable in a 2x4 ANOVA (stimulation condition x estimate number). This analysis should inform of the best measure to use in final analysis of overall susceptibility to the illusion.

Mean constant error for each trial is shown in Figure 5.3. There appears to be no discernible trend with regard to change in drift over time for either condition. Figure 5.4 shows constant errors for each trial for each subject. It is clear that subjects did not all show a similar pattern in drift, reflected in the large standard errors associated with the group mean errors across trials.

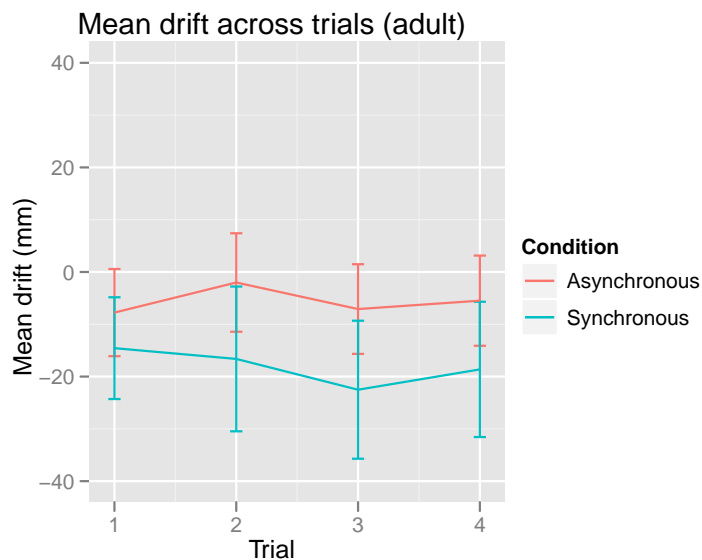


Figure 5.3: Mean constant error across trials for synchronous and asynchronous conditions. Negative drift is towards the rubber hand and zero corresponds to the veridical location of the real hand. (Error bars show SE.)

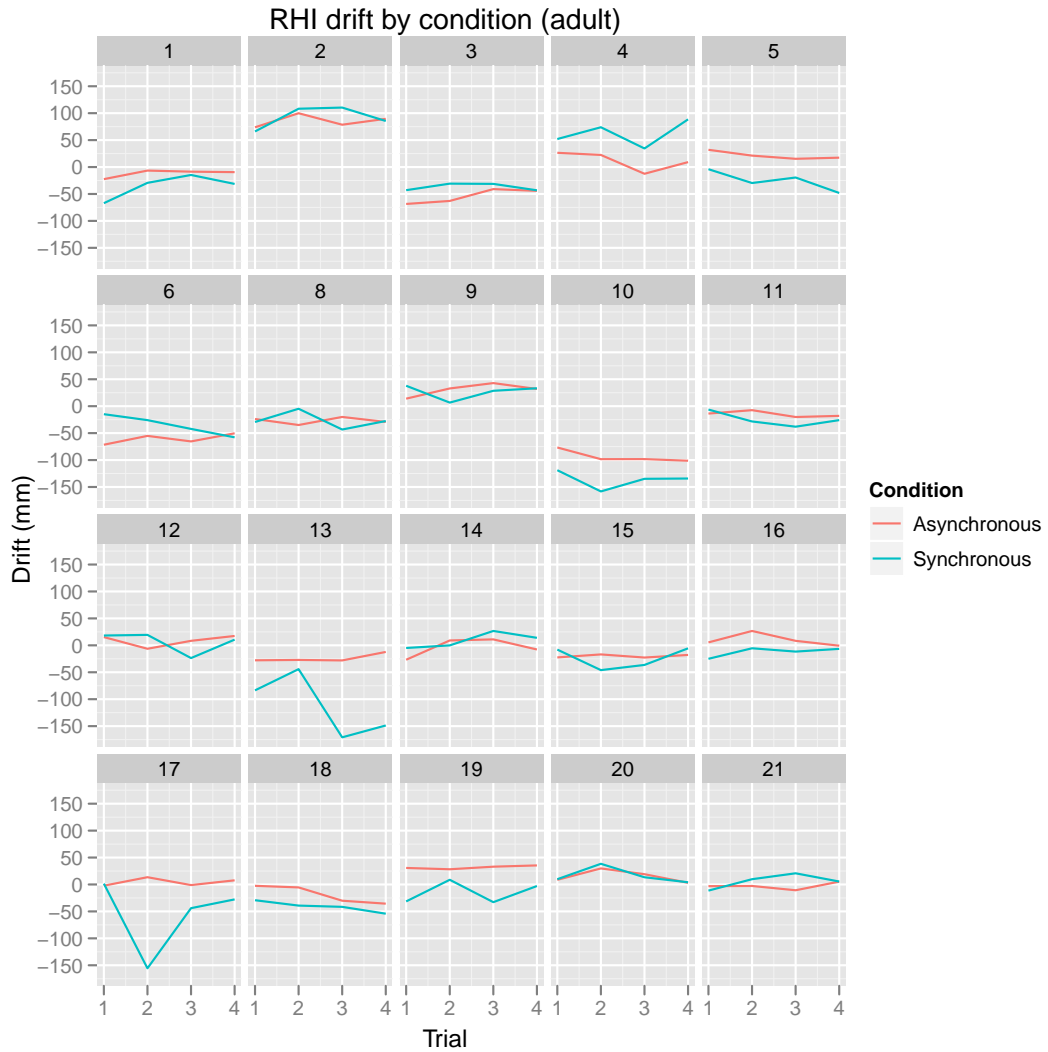


Figure 5.4: Each subplot shows constant error for each subject across trials for synchronous and asynchronous conditions. Negative drift is towards the rubber hand.

Estimate did not have a significant effect on drift:  $F(3, 57) = 0.50, p = 0.682$ . Additionally, as suggested by the visible overlap in SE between conditions across trials, there was no significant effect of condition (synchronous and asynchronous) on drift:  $F(1, 19) = 2.96, p = 0.102$  (GG). This suggests that overall there was no robust illusion, at least when measured using proprioceptive drift. Finally, there was no significant interaction between estimate number and condition ( $F(2.02, 38.38) = 0.39, p = 0.68$  (GG), so there is no clear pattern of drift in either condition.

As the first estimate in each condition was after 60 seconds compared to only 30 for the remaining estimates, an additional analysis compared responses according to condition and stimulation duration. There was no significant effect of duration using either the mean or median error for the 30 second duration ( $F(1, 19) = 0.05, p = 0.82$  and

$F(1, 19) = 0.001, p = 0.98$ , both GG).

Non-significant effects of estimate and duration support the use of an average error for each condition in further analyses. Due to the small number of observations for each subject, median error will be used.

### 5.5.2 Proprioceptive shift

Constant errors (proprioceptive drift) were calculated for each trial by subtracting the response from the veridical location of the real hand. Proprioceptive shift was then calculated by subtracting the median drift in synchronous trials from median drift in asynchronous trials. A negative shift is in the expected direction for the illusion<sup>3</sup>. Mean drift in synchronous and asynchronous conditions is shown in Figure 5.5 and the difference in drift between conditions for each individual is shown in Figure 5.6.

Overall shift towards the rubber hand occurred in 65% of the group, with shifts ranging from -88.74 mm to -1.51 mm (mean=-25.84, SD=25.39). The remaining 35% of the group had an overall shift away from the rubber hand, ranging from 2.57 to 47.16 (mean=17.44, SD=15.14). Supporting the null effect of condition in the previous analysis, there was no overall rubber hand illusion, as shift was not significantly from zero  $t(19) = 1.57, p = 0.067, d = 0.720$  (1-tailed), although the difference was in the expected direction and did tend towards significance.

As illustrated in Figure 5.6, those who received asynchronous stimulation first tended to show more proprioceptive shift than those who received synchronous stimulation first (mean difference 20.50 mm). To explore this, constant errors (drift) for synchronous and asynchronous conditions were compared separately for these two subgroups. There was significantly greater drift in the synchronous condition for those with asynchronous first:  $t(9) = 2.33, p = 0.045, d = 0.736$  [1-tailed, Bonferroni corrected, mean difference (SE)= 20.94mm (28.45)]. Although drift was also slightly higher following synchronous stimulation in the synchronous first group, the difference is not statistically significant:  $t(9) = 0.05, p = 0.964$ , 1-tailed, Bonferroni corrected, mean difference (SE)=0.44mm (9.57). Note that these results hold even when the outlier subject is removed. There was no significant difference in acuity between the two subgroups for either spatial acuity or postural acuity:  $t(18) = -0.29, p = 0.778$  and  $t(18) = -0.17, p = 0.864$ . This suggests the difference between subjects from either side of the condition counterbalancing is likely due to an effect of condition order rather than chance differences in proprioceptive acuity.

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<sup>3</sup>One subject's shift was slightly outwith  $\pm 2SD$  of the group mean, however as this was in the expected direction for the illusion the subject was not removed from analysis.

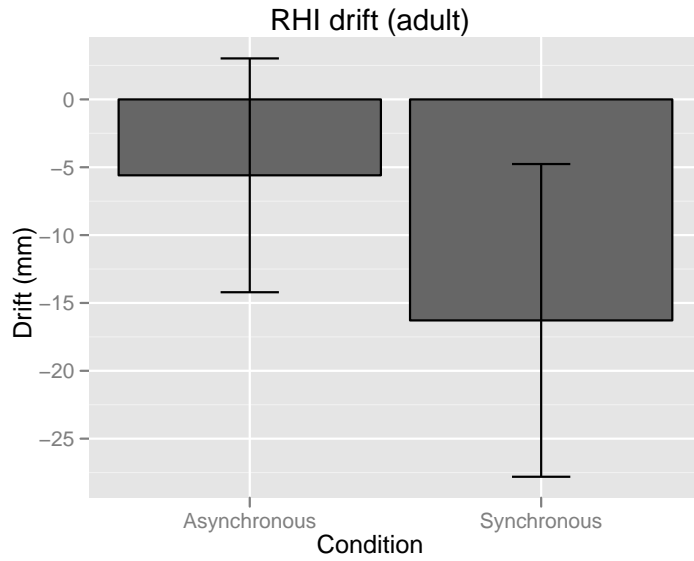


Figure 5.5: Effect of condition on subjects' proprioceptive drift (median constant error). Drift towards the rubber hand is coded as negative. Error bars show SE.

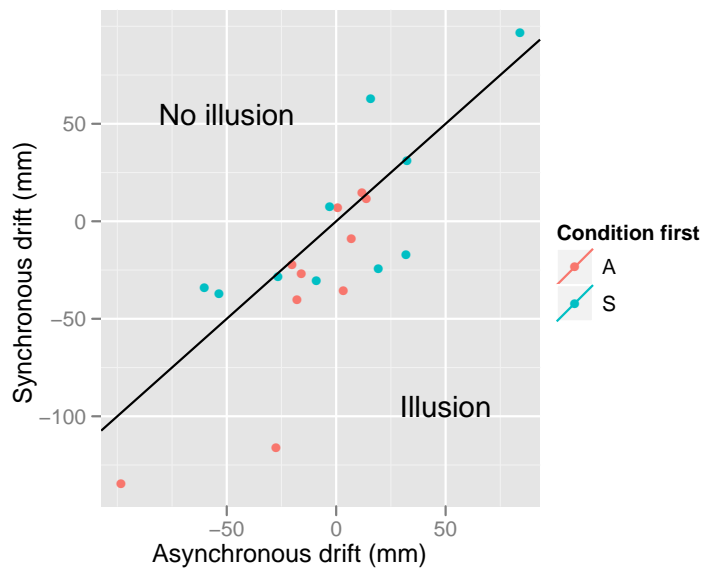


Figure 5.6: Median constant error for synchronous versus asynchronous conditions for each subject. The line shows slope 1, intercept 0 (no illusion). Subjects who completed asynchronous trials first (red) tended to show greater drift in synchronous than asynchronous trials, compared to subjects who completed synchronous trials first (blue).

### 5.5.3 Postural matching

Median absolute, constant and variable errors for each target angle were entered into three separate within subjects ANOVAs (all post hocs use Bonferroni correction for multiple comparisons). Mean absolute, constant and variable errors are shown in Table 5.1, and the mean absolute error for each target is shown in Figure 5.7. There was a significant effect of target angle on absolute error:  $F(2, 38) = 8.12, p = 0.001, \eta_p^2 = 0.30$ . This effect was driven by significant differences between 120° and both 60° and 90°, with greatest errors for 120° ( $p=0.018$  and  $p=0.015$  respectively).

There was also a significant effect of target on constant error ( $F(2, 38) = 44.07, p < 0.001, \eta_p^2 = 0.70$ ). All targets were significantly different from each other (all  $p < 0.001$ ). Variable error (SD) was also significantly affected by target angle ( $F(2, 38) = 3.86, p = 0.03$ ). This was driven by a significant difference between 120° and 60° ( $p = 0.037$ ).

Table 5.1: Mean constant, absolute and variable error (degrees) for each of the three target angles

Angle	Mean (SD) absolute error	Mean (SD) constant error	Mean (SD) variable error
60	10.71 (5.24)	-6.10 (10.29)	5.45 (2.37)
90	9.00 (6.17)	0.86 (10.60)	6.49 (2.61)
120	16.57 (7.63)	-15.92 (8.97)	7.86 (3.26)

All target angles are relative to horizontal: a 90° target is vertical. There are significant differences between targets for each measure.

### 5.5.4 Spatial location matching

Median error (absolute, constant and variable error) was calculated for each target for each subject. These were then compared in within-subjects ANOVAs (one per measure for constant and variable error) and a Friedman nonparametric test for absolute error as data were not normally distributed. Average absolute, constant and variable errors are shown in Table 5.2. There was a significant effect of target on absolute and constant error, but no effect on variable error.

Absolute error:  $X^2(2) = 10.33, p = 0.006$ ; constant error:  $F(2, 38) = 8.89, p = 0.001, \eta_p^2 = 0.32$ ; and variable error:  $F(1.49, 28.35) = 3.45, p = 0.058, \eta_p^2 = 0.15$  (GG). Post hoc comparisons highlight a significant difference between the centre and rightmost target for absolute error:  $Z = -2.69, p = 0.021$  and between the outermost targets for constant error: mean difference=1.83,  $SE = 0.51, p = 0.006$  (both Bonferroni corrected).

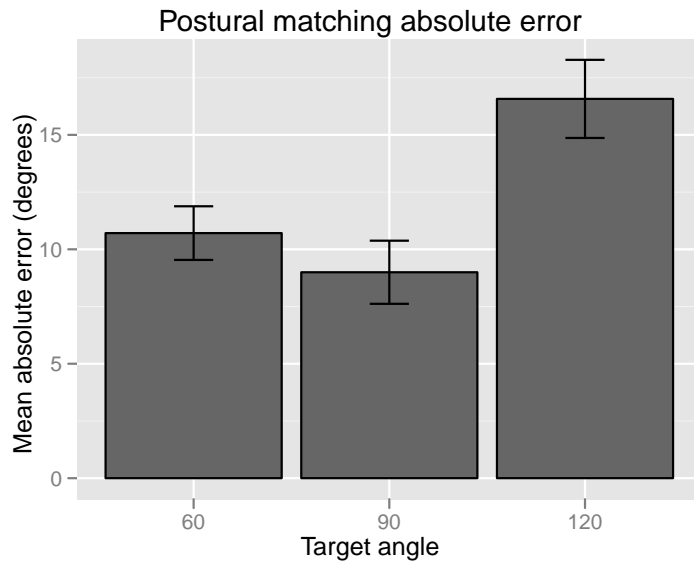


Figure 5.7: Effect of target on median absolute error (significant difference between 120 and 60, and 120 and 90). All target angles are relative to horizontal: a 90° target is vertical.

Table 5.2: Mean (SD) constant and variable error, and *median (range)* absolute error for each target in the spatial location matching task

	Mean (SD) constant error	Mean (SD) variable error	Median (range) absolute error
Right	3.50 (2.88)	0.74 (0.33)	4.00 (8.30)
Centre	2.45 (2.71)	0.55 (0.24)	2.60 (7.60)
Left	1.68 (3.16)	0.58 (0.22)	2.30 (7.20)

All errors are in cm and right and left is relative to the subject. There are significant differences between targets for absolute and constant error.

### 5.5.5 Correlational analysis

Data for the spatial location matching task were not normally distributed, therefore nonparametric Spearman's correlation was used for spatial versus postural matching and spatial matching versus RHI. Postural matching and RHI data were normally distributed, therefore Pearson correlation was used for RHI versus postural matching.

There was a significant negative correlation between RHI shift and spatial location matching absolute error:  $r_s = -0.42, p = 0.032$  (one-tailed). As hypothesised, greater shift was associated with greater matching error.

No other correlations were significant: the two proprioceptive acuity tasks (using median absolute error) were only very weakly correlated [ $r_s = 0.04, p = 0.44$  (1-tailed)]; and RHI shift was weakly negatively correlated with postural matching acuity (using median absolute error):  $r = -0.27, p = 0.122$  (1-tailed).

## 5.6 Discussion

This study investigated the relationship between proprioceptive acuity and susceptibility to the RHI, as measured by proprioceptive drift and shift. The tasks will first be discussed individually (RHI, postural matching, spatial location matching), followed by a discussion of the findings from the correlational analyses.

### 5.6.1 RHI

The rubber hand illusion can be operationally defined by greater proprioceptive drift towards the rubber hand following synchronous stimulation compared to asynchronous. Despite the majority of subjects (65%) in the present study following this pattern, there was no significant effect of stimulation condition on drift and therefore no robust illusion. This was verified by an analysis of proprioceptive shift. The reasons for this are unknown as the procedure was almost identical to the majority of previous RHI studies, and statistical power is not thought to be compromised as sample size was within the usual range for similar studies. The most obvious differences in the methodology used here are the stimulation duration, blind responding and the lack of any pause for questionnaires. Stimulation duration can be ruled out as there was no evidence for either growth or decay of the illusion over time and brief interruptions to stimulation did not adversely affect the illusion [compare with the effect of interruptions every 10 seconds (Rohde et al., 2011)]. If we assume that drift and shift are adequate measures of experiencing the illusion from a purely sensory perspective, it would appear that the percentage of the group with positive shift (away from the rubber hand) is in line with the failure rates reported by Ehrsson et al. (2007) and Ehrsson et al. (2004) using questionnaire ratings (32% and 28% respectively). In order to ascertain that minimal or positive shift is associated with a lack of subjective illusion it may have been instructive to use questionnaires. Despite their frequent use in previous studies,

questionnaires were not used for a number of reasons. Firstly, subjective measures are very difficult to administer properly with children with autism (this is discussed fully in the following section). As this study was always intended to be coupled with clinical work, it was important that the RHI procedure be identical in both. It would have been possible to give adults the questionnaire after the entire RHI procedure and simply not do so for children, leaving the illusion procedure identical, however this option presents its own difficulties. Half of the subjects will have experienced the illusion condition most recently, so condition-specific responses would not be immediate in every case. By giving the questionnaire after each condition (common in previous studies) this could have a confounding effect on behaviour and subjective reporting after the second condition due to a priming effect. It would also violate the need for the adult and child procedure to be the same. As well as practical difficulties in using questionnaires, from a theoretical perspective, the experience of ownership per se is not of interest here, again supporting the decision to use only objective measures.

The difference in drift in each condition depending on condition order, (with less drift in the synchronous condition when it was completed first), suggests that the RHI may be driven in part by visual capture which is not specific to stimulation condition (Rohde et al., 2011), and this may increase with time, thereby artificially increasing drift in the second condition. This would then subsequently affect proprioceptive shift measures.

### **5.6.2 Postural matching and Spatial location matching**

These tasks assessed postural acuity and spatial acuity respectively. The significant effect of target angle in postural matching, with more oblique angles more difficult to accurately match, has been reported previously (Goble, 2010; Adamo, Martin & Brown, 2007). The task being sensitive to these differences suggests that it is a sensitive measure of postural proprioceptive acuity. Similarly the effect of target location in the spatial location matching task suggests that this acuity task is also sufficiently sensitive to accurately estimate proprioceptive acuity.

### **5.6.3 Correlational analyses**

Shift in the RHI was found to significantly correlate with spatial acuity, but not postural acuity. The latter two measures did not intercorrelate. This dissociation can be explained by considering the requirements of each task. Pointing directly above the index finger in the RHI is essentially the same task as pointing underneath the hand holding the target in the spatial location matching task. Postural matching makes no use of this localising technique as though the arm was an external object, but instead must consider the arm as a part of the body (i.e. uses intrinsic instead of extrinsic coordinates). Although the RHI and location matching task use the same kind of proprioceptive acuity, it is unlikely that there was any residual practice effect from the

RHI on location matching, as they were not completed successively.

## **5.7 Study 2: RHI susceptibility in ASD, DCD and typical development**

The RHI has already been discussed with reference to previous work with neurotypical adults, and the above study which found a significant negative correlation between spatial proprioceptive acuity and RHI susceptibility in a group of neurotypical adults. The second half of this chapter introduces previous findings using the RHI in children with typical development and ASD. Compared to the wealth of study devoted to the RHI in adults, it is a little-used paradigm in either developmental psychology or autism research. The present study, which compares susceptibility in ASD, DCD and TD and relates this to the proposed vision/proprioception dissociation will then be reported and discussed. A general discussion will follow, discussing findings from the adult and child studies together.

### **5.7.1 RHI in autism**

It has been found that neurotypical adults with more autistic traits (high AQ scores) show decreased RHI susceptibility compared to low AQ scorers (Palmer et al., 2013). Verbal estimates of hand position were collected before and after each three minute block and questionnaire data were collected after each trial. Groups differed only on drift measure. Questionnaire items were not sensitive to group differences but did differentiate synchrony conditions. There was also a significant effect of rubber hand position in the synchronous condition, which was specific to the low AQ group. Those with fewer autistic traits drifted towards the rubber hand more when it was in a less biologically plausible position. The angle of the rubber hand was changed by rotating it at the elbow, resulting in more biologically plausible positions than studies in which the arm is not anchored at the elbow before being rotated (e.g. Ide, 2013). It may be that the effect of hand angle in the low AQ group in Palmer et al.'s (2013) study is due more to the confound of the hand being moved closer to the midline than the angle per se: we tend to drift towards the midline when we are unsure of our positioning (Wann & Ibrahim, 1992). Palmer et al. (2013) also found differences in normalised jerk and acceleration in a reaching task completed after the RHI. The low AQ group produced less smooth movements after synchronous compared to asynchronous stimulation. There was no such difference in the high AQ group. Overall the high AQ group produced significantly smoother movements following synchronous stimulation, despite both groups subjectively experiencing the illusion. It is suggested that smoother movements reflect a lack of expected uncertainty: the low AQ group (fewer ASD symptoms) realised that following the illusion their expectations of their current hand positioning was uncertain and this is reflected in a less smooth

movement. This is interesting in a typical sample, however it is unlikely that the same could be said in an ASD group, as high jerk has often been found when observing movements in ASD (Nobile, Perego, Piccinini, Mani, Rossi, Bellina & Molteni, 2011). Kinematic data from children, especially those with ASD and DCD, tends to be noisy, so data necessary for an investigation of jerk were not collected in the present study. For kinematic data to be informative every child must perform the same kind of action (e.g. a smooth single movement), which tends not to be the case (discussed in Chapter 3).

Few studies include true ASD samples, although one recent study has provided an argument for a reduced RHI in ASD, lending some support to the AQ study detailed above. Cascio et al. (2012) report a delay in experiencing significant drift towards the rubber hand in ASD. Children and adolescents ( $n=21$ , aged 8-17) with ASD or PDD-NOS experienced two three-minute trials each for synchronous and asynchronous RHI stimulation. Each trial was followed by three consecutive blind proprioceptive estimates of finger location. Additional questionnaire measures were gathered after each two-trial block. Those with ASD showed significant drift in the synchronous condition after two three-minute trials, but not after the first three-minute trial. Those in the TD group showed significant drift after both synchronous trials. The authors suggest that the ASD group is more resistant to the illusion and need more exposure to it to show the expected pattern of drift. (It is not clear whether this finding is due to timing per se, or whether the ASD group simply needed to familiarise themselves with the task, as there were no practice trials.)

A closer inspection of the results however suggests that these conclusions may be unsound. There was no robust RHI, with no effect of condition on drift. Similarly there was no effect of trial on drift (the first three-minute trial produced similar drift to the last three minutes). Finally there was no main effect of group. The only significant effect was a three-way group\*condition\*trial interaction. Post hoc analyses used to investigate this interaction did not use any correction for multiple comparisons, and if  $p$ -values are corrected using a Bonferroni correction then only one result remains significant: the TD group showed significant drift in the first three minutes of synchronous stimulation. The findings of significant drift in the second synchronous trial for both ASD and TD do not survive corrections for multiple comparisons. These corrected results suggest that the study has in fact found that children with ASD do not reliably experience the RHI after either 3 or 6 minutes, and TD children's responses to the RHI may not be as robust as initially claimed. The TD children may be exhibiting the previously observed plateau in drift following a period of initially rapid drift towards the rubber hand (Tsakiris & Haggard, 2005).

As well as peculiarities in the analyses detailed, the methods are not without question either. The rubber hand was positioned very far to the subject's right hand side<sup>4</sup>

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<sup>4</sup>No details of the exact position are given, although it appears to be in line with the shoulder of the responding arm.

[compared to the more common positioning closer to the midline (e.g. Davies et al., 2013)], making it appear to be in a biologically implausible position. As discussed above, biological implausibility appears to differentially affect high- and low-AQ groups (Palmer et al., 2013), and some studies report a complete eradication of the illusion with biologically implausible stimuli (Tsakiris & Haggard, 2005). Finally, four children with ASD were removed from analysis as the experimenter felt they paid insufficient attention to the stimulus. Considering the frequency of attentional difficulties in ASD it is surprising that the remaining 21 children with ASD, with some as young as 8 years old, had the attentional capacity to attend to stimuli for six minutes with only a brief pause after three minutes to give estimates.

Testing adults with ASD, Paton et al. (2012) report a subjective illusion (using questionnaire measures) comparable to TD. However drift measures showed greater drift in TD, although this drift was not specific to the synchronous condition. The authors suggest that this reflects either better proprioceptive acuity or poorer sensory integration in ASD. The former seems unlikely, as a sizeable proportion of the ASD group had substantial negative drifts in the opposite direction of the rubber hand (beyond both the real and rubber hand, towards the far right of the body), suggesting poor proprioceptive acuity. It may be that this arguably poor proprioceptive acuity is related to motor ability in the ASD group, however no indication of motor ability or sensory acuity is provided.

### 5.7.2 RHI in typical development

While a large amount of research has been conducted with TD adults, the use of the RHI in typically developing children is rare. Indeed, of the two published studies found that use the paradigm with children, only one explicitly examined the illusion in TD (Cowie et al., 2013), while the other used TD as a control for a clinical group (Cascio et al., 2012). Using a between-subjects design, in which children received *either* synchronous or asynchronous visuo-tactile stimulation, Cowie et al. (2013) found that children aged 4-9 years ( $n=90$ ) showed a greater response to the RHI than adults ( $n=30$ ) using drift and questionnaire measures, although drift was not specific to the synchronous condition in the child groups. Response to the illusion appears to be stable between 4-9 years and differences between children and adults suggest that the underlying processes involved in the RHI have not fully matured until some time beyond 9 years of age. While these results are interesting, it is important to note that this study used a between-subjects design, meaning that condition-comparisons are being made between different children, rather than the more conventional within-subjects design.

### 5.7.3 Using the RHI with children and clinical groups

There are a number of factors to consider when applying the RHI paradigm to children, particularly those with ASD. With high rates of comorbid ADHD or sub-clinical ADHD

symptoms it is likely that long stimulation periods could be problematic, as attention may wander and any illusion is likely to be broken if attention is not sustained. With some children with ASD experiencing tactile hypersensitivity, it is also possible that some may be averse to the stimulation and this is another reason to favour shorter stimulation periods. Problems with questionnaire responses have already been discussed and they seem unsuitable for use with children, particularly those with ASD, as they might have difficulty understanding that a rubber hand can feel like it belongs to them.

#### **5.7.4 Hypotheses**

Hypothesis 1: As RHI shift was found to correlate negatively with performance on a proprioceptive acuity test in adults, it is hypothesised that the ASD group will show less shift towards the rubber hand compared to DCD and TD, if ASD is associated with a proprioceptive bias, as discussed previously. Following the proposed vision-proprioception double dissociation it is expected that with a greater reliance on vision in DCD, this group might show the most extreme shift towards the rubber hand.

Hypothesis 2: As 70% of the original sample of children with ASD failed the MABC-2 and were considered no different from DCD in a spatial location matching task (Chapter 3) which is similar to the RHI, it is possible that if sensory cues are used in the same way in these two tasks there will be no significant difference between ASD subjects who failed the MABC-2 and children with DCD. It is expected that these children will show greater susceptibility to the illusion than children with ASD who passed the MABC-2.

### **5.8 Methods**

#### **5.8.1 Subjects**

All children had previously completed the tasks detailed in Chapters 2, 3 and 4. Twenty three ASD, 6 DCD and 14 TD children participated. All but one participant (a member of the DCD group) was male. The majority of the ASD group failed the MABC-2 (17), all DCD failed and three children in the TD group failed. Data from the three TD children who failed the MABC-2 were not included as per previous chapters and responses were not recorded for three children in the ASD group due to equipment failure. Subject demographics are detailed in Table 5.3.

#### **5.8.2 Procedure**

The procedure for the RHI matches that given previously for adults. In this case the RHI was completed in a testing session alongside the posting task reported in Chapter 4 and the reaching task reported in Chapter 3. It is not anticipated that other tasks conducted in the testing session will have carry-over effects, especially given the break between tasks.

Table 5.3: Subject demographics (values shown are mean (SD))

	ASD ( $n = 20$ )	DCD ( $n = 6$ )	Clinical motor deficit ( $n=23$ )	TD ( $n = 11$ )
Age	12.25 (2.00)	11.00 (2.53)	11.83 (2.17)	11.27 (1.19)
IQ %ile	43.45 (28.55)	49.17 (42.99)	39.35 (29.06)	60.09 (21.06)
MABC-2 %ile	11.53 (20.76)	4.35 (3.96)	4.51 (3.68)	54.09 (22.44)
SRS	116 (23.11)	62.17 (36.52)	100.10 (36.14)	15.82 (18.90)
DCDQ-07	33.34 (10.61)	31.80 (11.17)	32.10 (10.06)	67.91 (9.15)

Note, TD and DCD are the same as the reaching task in Chapter 3. The ASD group includes two children who did not complete the reaching task. Groups were age- and IQ-matched (group difference  $p > 0.05$ ). As with the reaching task, SRS, DCDQ-07 and MABC-2 scores differentiated clinical groups from TD ( $p < 0.05$ ) and the SRS differentiated ASD and DCD from each other and from the TD group ( $p < 0.05$ ).

## 5.9 Results

Data were processed and analysed in the same way as the RHI for adults, detailed previously. In addition to analysis of the three diagnostic groups (ASD, DCD, TD), as with previous chapters an additional hypothesis using group comparisons based on MABC-defined ASD subjects was also considered.

The distributions of average shift (median synchronous error - median asynchronous error) for each group appear to be reasonably normal (see Figure 5.8), therefore parametric analyses are performed. The ASD group show a more varied response, with more subjects averaging large positive shifts (shifts away from the rubber hand) or very small negative shifts. Some children with ASD did show proprioceptive shift towards the rubber hand as would be expected, and some did so to the same extent as some TD children. The DCD group has only one deviant response, of a seemingly very large magnitude. This suggests that this subject was affected by the sensory incongruence, but for some reason did not respond in the typical way expected from the illusion. With only six subjects however, the DCD group is difficult to draw conclusions from. The range of proprioceptive shift in ASD and TD groups is fairly similar.

### 5.9.1 Effect of estimate number

Figures 5.9, 5.10 and 5.11 show each subject's errors across trials in synchronous and asynchronous conditions. (Note that the vertical scale is dictated by individual group extreme scores therefore the scale differs between figures.) It is clear that there is no obvious pattern across trials. The effect of trial is not significant, as was the case in the adult study:  $F(2.37, 80.63) = 1.81, p = 0.163$  (GG). There was no interaction with trial and either group or condition:  $F(4.74, 80.63) = 1.11, p = 0.344$  (GG); and  $F(3, 102) = 1.53, p = 0.213$ .

Due to the nonsignificant effect of trial on drift, an average (median) for the four

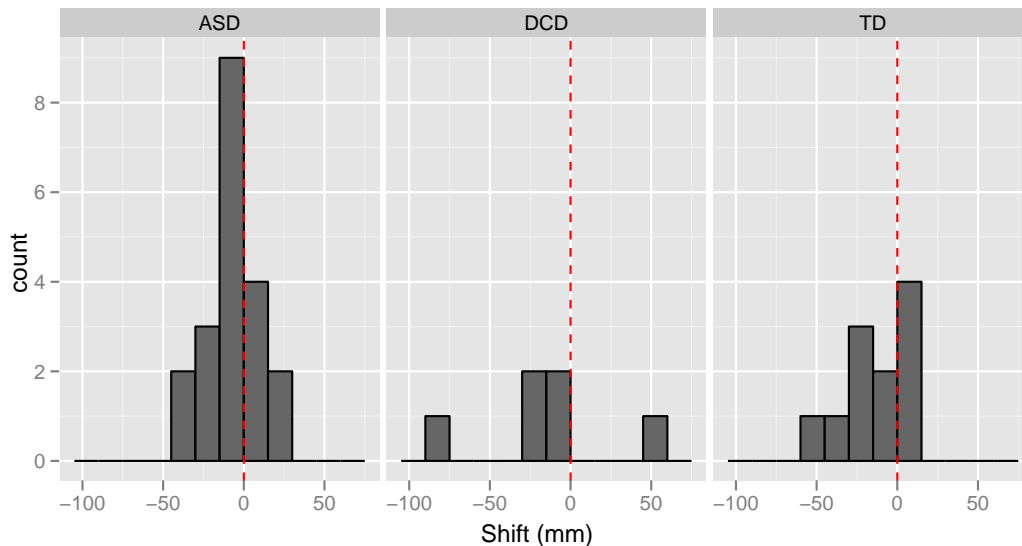


Figure 5.8: Distribution of average shift for diagnosis-defined groups. Data to the left of the red line is in the expected direction for the illusion.

trials was calculated per subject, as per the adult study. Median constant errors for synchronous and asynchronous conditions were calculated for each subject as drift measures, and these were then used to obtain shift measures as per the adult study. In each of the following analyses the TD group does not include those children who failed the MABC-2, however all results remain the same if these three children are included.

### 5.9.2 Hypothesis 1 analysis (diagnostic groups)

Figure 5.12 shows the median error for synchronous and asynchronous trials for each child in each of the three diagnostic groups. If we define successfully eliciting the illusion as the average drift in the direction of the rubber hand being greater in synchronous than asynchronous trials, it is clear that the majority of subjects experienced the illusion, although some to negligible degrees. Those points closer to the line have a smaller discrepancy between synchronous and asynchronous drift, while those further from the line show a larger difference.

Drift for each condition was entered into a 3x2 ANOVA (group x condition). In contrast to the adult study, in this case there *was* a robust illusion, as drift was significantly larger in the synchronous condition:  $F(1, 34) = 7.07, p = 0.012, \eta_p^2 = 0.17$  (GG), mean difference (SE)=11.28 (4.24). Contrary to the hypothesis, there was no group difference in proprioceptive drift:  $F(2, 34) = 1.12, p = 0.338$ . Although there was considerably less drift in the ASD group (see Figure 5.13) it is clear that responses are very varied within the DCD and TD group. There was also no group\*condition interaction, suggesting a similar rubber hand effect in each developmental group:  $F(2, 34) = 0.60, p = 0.552$  (GG). Confirming the null effect of group, proprioceptive shift also failed to differentiate

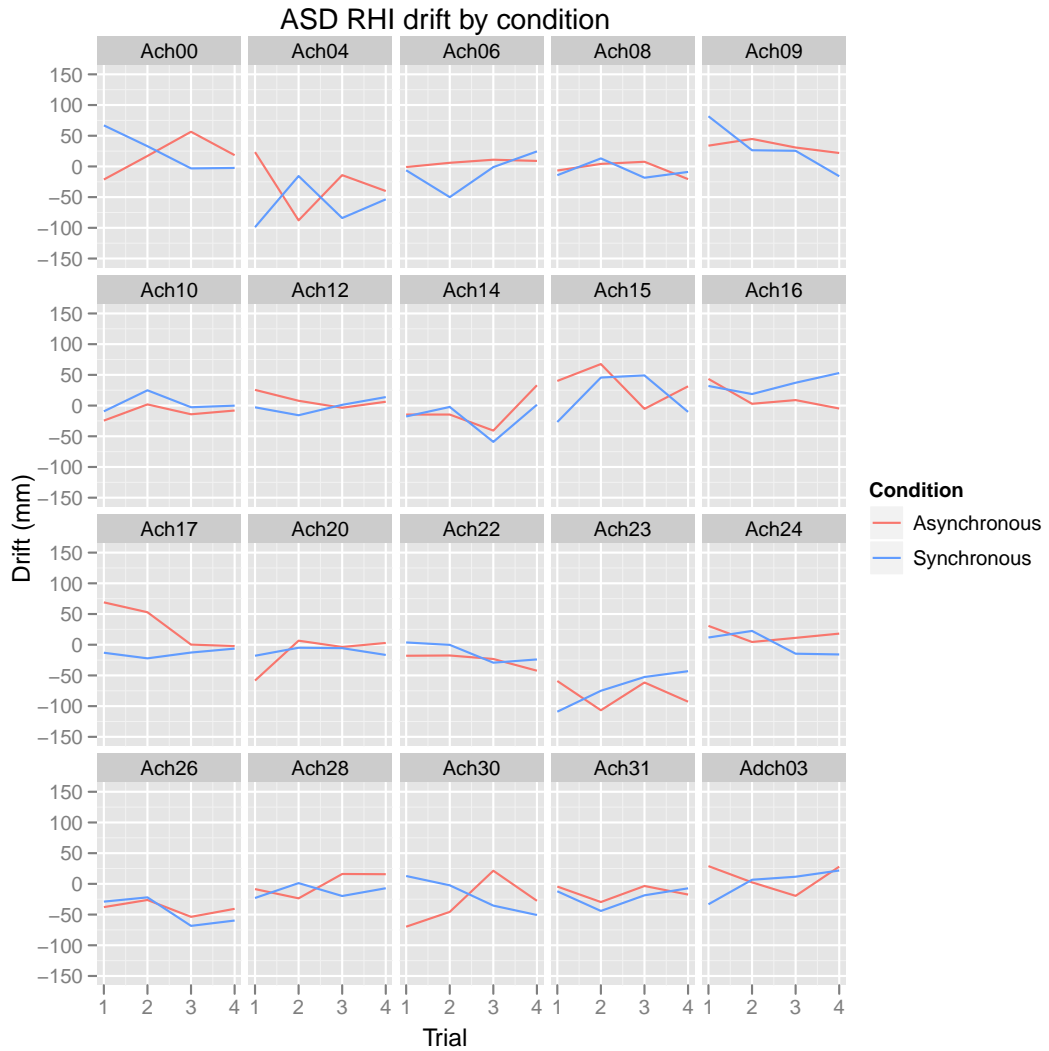


Figure 5.9: Proprioceptive drift across trials (ASD). There is no clear pattern over time and there does not appear to be a strong illusion as synchronous and asynchronous drift overlap to a large extent.

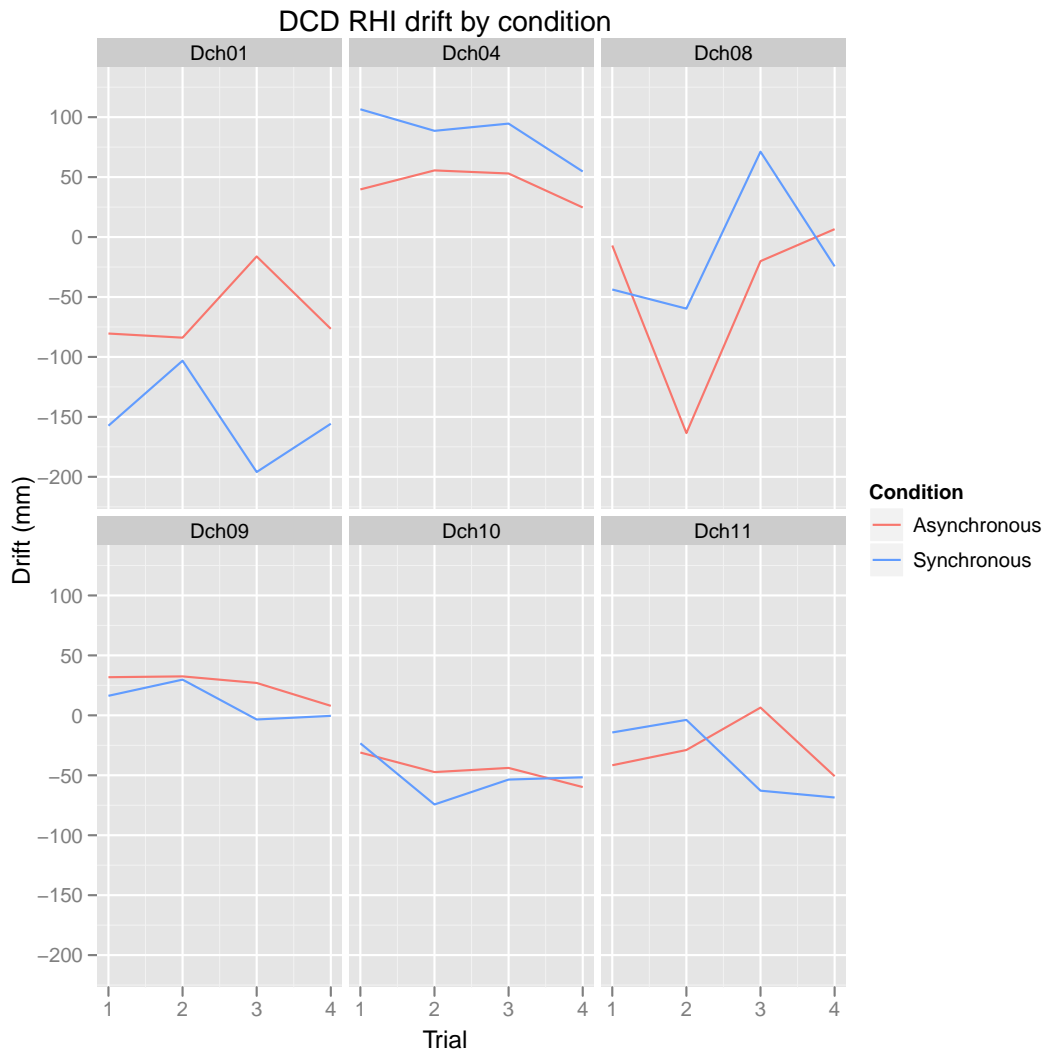


Figure 5.10: Drift across trials (DCD). Again there is no clear pattern over time and there does not appear to be a strong illusion as only one child shows consistently greater drift in the synchronous condition.

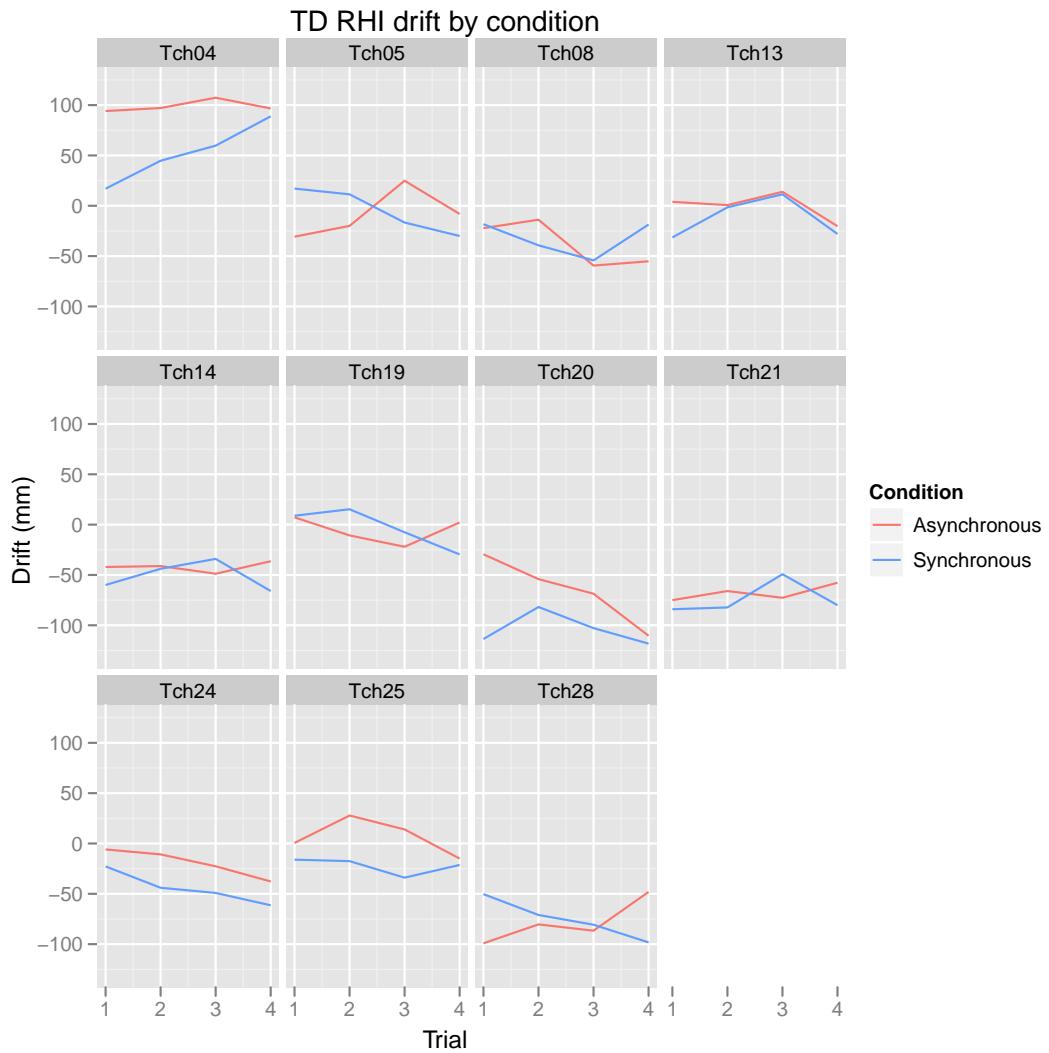


Figure 5.11: Drift across trials (TD). As with ASD and DCD there is no clear pattern over time. There is still some overlap between synchronous and asynchronous, however some children show consistently greater drift following synchronous stimulation.

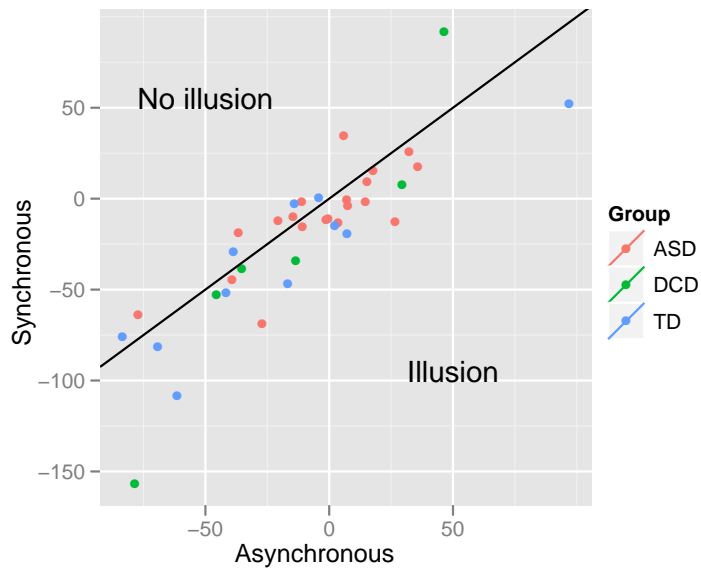


Figure 5.12: Average drift for asynchronous against synchronous conditions. Line shows slope 1, intercept 0 (no illusion).

diagnostic groups:  $F(2, 34) = 0.60, p = 0.552$  (see Table 5.4).

Table 5.4: Mean shift (mm), grouping subjects by clinical diagnosis

	Mean (SD) proprioceptive shift
ASD ( $n = 20$ )	-5.71 (17.10)
DCD ( $n = 6$ )	-14.19 (39.67)
TD ( $n = 11$ )	-13.95 (21.19)

Negative drift is towards the rubber hand.

### 5.9.3 Hypothesis 2 analysis (MABC-defined groups)

Before comparing the ‘clinical motor deficit’ group with TD children, the DCD group was compared with the ASD children who failed the MABC-2. There was no significant difference between motor impaired ASD and DCD children for drift or shift measures ( $F(1, 21) = 1.33, p = 0.252$  and  $t(21) = 0.66, p = 0.518$  respectively), supporting the use of the ‘clinical motor deficit’ group.

Mean drift and shift for clinical motor deficit, TD and ASD ‘pure’ groups are shown in Table 5.5. There were too few children from the original redefined ASD ‘pure’ group to include this subgroup in formal analysis, so the following analysis only includes the motor deficit group and TD. Median drift for each condition was entered into a 2x2 ANOVA (group x condition). Contrary to the hypothesis, there was no significant difference in proprioceptive drift in the motor deficit group and

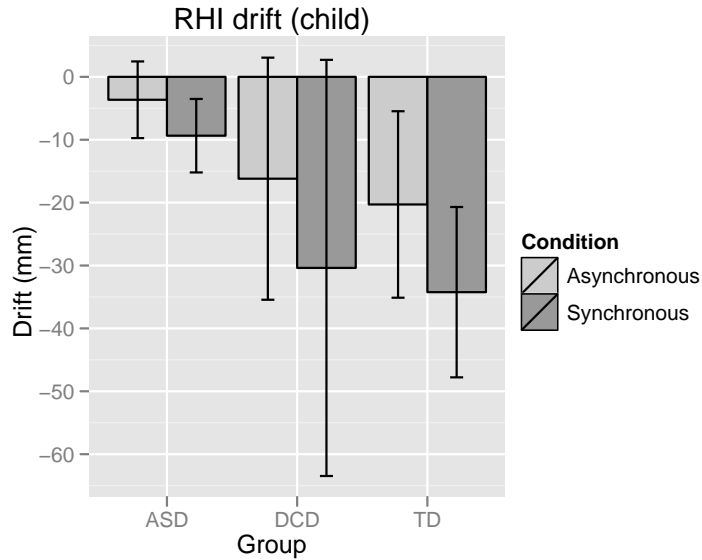


Figure 5.13: Mean drift in each condition between developmental groups. Error bars show SE. There is a significant effect of condition but no group effect or group\*condition interaction.

TD:  $F(1, 32) = 1.65, p = 0.209$ . As expected, the significant effect of condition remained, with greater drift towards the rubber hand in the synchronous condition:  $F(1, 32) = 6.71, p = 0.014, \eta_p^2 = 0.17$  (GG), mean difference (SE)=11.20(4.32). Again, there was no significant group\*condition interaction:  $F(1, 32) = 0.41, p = 0.529$  (GG). The difference in proprioceptive shift in each group was also not significant ( $t(32)=0.64, p=0.529$ ), confirming the null effect of group on drift.

Table 5.5: Mean drift (synchronous and asynchronous) and mean shift (all mm) for MABC-defined groups

	Synchronous Mean (SD)	Asynchronous Mean (SD)	Shift Mean (SD)
Clinical motor deficit ( $n = 23$ )	-12.77 (45.31)	-4.33 (30.63)	-8.44 (24.58)
TD ( $n = 11$ )	-34.24 (44.93)	-20.29 (49.18)	-13.95 (21.19)
ASD 'pure' ( $n = 3$ )	-25.23 (33.80)	-23.50 (46.60)	-1.73 (13.13)

Note the ASD 'pure' group is not included in analysis as there are too few children.

#### 5.9.4 The effect of proprioceptive acuity in RHI shift

In order to assess the effect of proprioceptive acuity in the child study, data from the PP condition in the spatial location matching task reported in Chapter 3 were correlated with RHI shift. Visual weighting from the spatial location matching prism condition was also correlated with RHI shift.

There was no significant correlation between shift and mean visual weighting:  $r(35) = -0.02, p = 0.918$ , however there was a significant correlation between acuity (PP mean error) and shift:  $r(35) = 0.42, p = 0.005$  (one-tailed). Unexpectedly, greater proprioceptive acuity tended to correlate with more pronounced proprioceptive shift towards the rubber hand. When diagnostic groups were analysed separately only the ASD group's correlation was significant:  $r(18) = 0.52, p = 0.018$ . The clinical motor deficit group also showed a trend towards a significant positive correlation (although this would not survive correction for multiple comparisons):  $r(21) = 0.41, p = 0.041$ . Proprioceptive acuity was not related to proprioceptive shift in the RHI in the TD group:  $r(9) = 0.27, p = 0.417$ .

## 5.10 Discussion

This study investigated the RHI in children with ASD, DCD and TD, and considered how susceptibility related to diagnosis, motor ability and proprioceptive acuity. The finding of a significant effect of stimulation condition on drift suggests the illusion was successful, however an examination of each child's drift suggests that the illusion may have been weaker than in previous studies. There was no evidence of a differential response to the RHI in children with ASD and TD with regards to time to illusion onset (Cascio et al., 2012), however it should be noted that 85% of the ASD group failed the MABC-2 and were considered to be more similar to the DCD group in a vision-proprioception perceptual task (see Chapter 3).

Contrary to the initial hypothesis of a double dissociation between vision and proprioception manifesting in group differences in RHI susceptibility, there was no significant effect of group. However, the DCD group was very small, and MABC-2 scores in the ASD group were skewed towards the motor-deficit range. The second hypothesis, comparing ASD 'pure' and the clinical motor deficit group was not able to be investigated fully, as the ASD 'pure' group was too small. In line with visual weighting results from the spatial location matching task in Chapter 3, there was no difference between the clinical motor deficit group and TD in RHI drift or shift. The three children in the ASD 'pure' group appear to have been relatively resistant to the illusion, similar to the lower visual weighting reported in Chapter 3, however further study with such children is required.

The finding that proprioceptive acuity positively correlated with proprioceptive shift seems counterintuitive, and cannot easily be explained, particularly when contrasted with a (weaker) significant *negative* correlation in adults. It is acknowledged that the acuity measure is based on slightly fewer trials in the child groups, and the tasks were completed sometimes months apart, however it is not expected that this would cause such large discrepancies between child and adult results. This counterintuitive finding in children (driven primarily by the clinical groups) perhaps serves to highlight the difficulty in using the RHI in children with attentional and behavioural difficulties. It

is assumed that the finding is likely due to the unreliable nature of behaviours captured in atypical groups, as there is no theoretical explanation for the finding.

Some children found it difficult to maintain attention during the task, with some very reluctant to watch the rubber hand. These children were prompted throughout, although their data may be less reliable than would be expected from TD children. While previous studies have reported removing subjects who did not appear to maintain close attention to the rubber hand (e.g. Cascio et al., 2012), this has not been done here as the experimenter was not blind to either hypothesis or diagnoses. Attentional difficulties do raise some concerns about previous studies involving children with ASD, particularly when the stimulation duration is longer than that used in the present study. The high incidence of comorbid ADHD-type symptoms in the clinical groups used here, and in these clinical populations in general, presents an attentional challenge in these types of tasks which is difficult to overcome. However, as there was no group difference in this case it is expected that poor attention in some members of the groups (primarily the clinical groups) was not detrimental and has not impacted negatively on the results. Another problem encountered during testing was some children becoming aware of the expected illusion and trying to avoid the illusion by looking away from the rubber hand. Some children also tended to look away more during the synchronous than asynchronous condition, possibly because the illusion was uncomfortable to experience, however this suggestion remains speculative. The suggestion does perhaps find support in accounts of sensory sensitivities in ASD and the effects of sensory overload (Harrison & Hare, 2004).

While subjective measures could have helped address some of these questions, it is felt that only using objective measures was justified. Reasons for this have previously been outlined. After testing, children were informally asked about the illusion, and a number of younger children and those with ASD were unable to coherently verbalise the experience and gave inconsistent answers when asked more direct questions. While these observations have not been recorded in any standardised way, they illustrate the potential problems faced when using both open-ended and direct subjective measures.

This study has failed to provide support for the proposed sensory dissociation in ASD and DCD. It is possible that a task with additional cognitive and attentional components such as this was not appropriate for investigating sensory processing in these groups. However the null effect of group is in line with findings reported in previous chapters.

## 5.11 General discussion

This chapter assessed the relationship between proprioceptive acuity and the RHI in both typical adults and typical and atypical developmental groups. The finding of no robust RHI in an adult sample of comparable size to previous studies is unexpected,

although evidence of a stronger illusion in TD children compared to adults supports findings from Cowie et al. (2013), with the benefit of a more appropriate within-subjects design in the child study. The main difference between this study and previous RHI studies (which tended to produce larger effects) is the use of proprioceptive drift and shift as a main measure, without questionnaire measures. It might be that previous studies with adults find strong effects in part due to priming from questionnaire measures. Given the significant association with spatial acuity in adults, the lack of a robust illusion in adults is not seen as a failure of procedure, as drift varies systematically with acuity in the expected direction.

## Chapter 6

# Investigating the related nature of motor and social skills in typical development

The role of motor deficits in ASD and DCD has been discussed in previous chapters. So far the focus has been on how motor aspects of these two disorders might be similar or dissimilar in kind and origin. In the studies reported in earlier chapters, motor skills in ASD and DCD have not been significantly different on a range of tasks, ranging from high-level motor skills and visuomotor imitation to visual and proprioceptive processing necessary for movement. This chapter reports a study conducted with parents of typically developing children using the SRS and DCDQ-07 questionnaires to assess the relationship between ASD and DCD-like symptoms in typical development.

### 6.1 Motor deficits in ASD and social deficits in DCD: What separates these two disorders?

A number of childhood neurodevelopmental disorders can co-occur, resulting in comorbid social, motor, attentional and behavioural difficulties. Co-occurring social and motor deficits are most apparent in the high (unofficial) comorbidity<sup>1</sup> rate of ASD and DCD (Gillberg & Kadesjo, 2003). Results from studies reported in previous chapters also provide support for this large overlap.

In the ICD-10, motor deficits are listed as a possible symptom of AS: individuals within the rest of the spectrum, according to its diagnostic criteria, do not suffer from motor difficulties. As discussed in the introductory chapter however, a number of studies have suggested that in practice there is no discernible difference between AS and other autistic disorders when considering motor skills (e.g. Seal & Bonvillian, 1997; Page & Boucher, 1998). With a recent major overhaul in the definition of autistic

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<sup>1</sup>The change in DSM criteria allowing dual diagnosis is too recent for any published prevalence rates of official comorbidity.

disorders, and a new relationship between ASD and DCD (APA, 2013), the question of the specificity of motor deficits in ASD becomes increasingly relevant.

Social deficits tend to vary across the spectrum, with classic autism often representing those with the most profound social and communication difficulties, and AS and HFA often including those with clear social difficulties but with much better social functioning and some of these children are able to attend mainstream education. It has been suggested that fine motor skills in ASD can predict adaptive social skills (MacDonald, Lord & Ulrich, 2013). Couple this with the observation that social deficits are over-represented amongst individuals with DCD (Jarus et al., 2011) and we are left with an interesting question: would it be appropriate to consider ASD and DCD as two intersecting spectra, with the majority of ASD and DCD children likely to have symptoms from both the social dysfunction and motor dysfunction spectrum? This chapter addresses this question by considering the related nature of social and motor skills in typical development, and whether the trajectory of development in one domain is indicative of the trajectory of the other.

## **6.2 The interrelated nature of social, motor, attentional and educational aspects of typical development**

There has been much investigation of the link between social skills, attention and academic attainment in childhood. Motor skills have been considered in a small number of studies, most often with regards to academic attainment: the link between motor and social skills is often confined to the study of children with ASD and in some cases DCD. Most studies report clear links between skills in different domains of development and ability in one is often predictive of skills in another.

### **6.2.1 The relationship between social and academic skills**

A number of studies have reported a significant association between social competence and academic achievement. It is possible that this is simply an artefact of the ways children with different temperaments respond in testing situations. Alternatively, there may be a real difference between shy children, and socially-comfortable children, with the former managing to elicit less attention and fewer interactions, thereby reducing their opportunities for informal learning before starting school (Lamb, Garn & Keating, 1982).

A number of areas of social development have been found to correlate with academic success. For instance, social skills have been found to relate to both a child's ability to foster positive relationships with their teacher and their academic achievement (Newcomb, Bukowski & Pattee, 1993). Prosocial behaviours (e.g. following instructions, helping others) have also been found to positively correlate significantly with pre-literacy, language and maths skills in a group of 467 four-year-old American

preschoolers<sup>2</sup> (Arnold, Kupersmidt, Voegler-Lee & Marshall, 2012). Looking specifically at maths ability in a group of fifty 3-5 year olds, Dobbs, Doctoroff, Fisher & Arnold (2006) found that initiative and self control correlated positively with academic ability, whilst behavioural concerns correlated negatively with academic achievement ( $r=0.57$ ;  $0.33$ ; and  $-0.42$  respectively).

Social skills have also been found to predict future academic achievement (e.g. Malecki & Elliott, 2002). In a longitudinal study of 1756 5-6 year old children in the US, Duncan, Dowsett, Claessens, Magnuson, Huston, Klebanov, Pagani, Feinstein, Engel, Brooks-Gunn, Sexton, Duckworth & Japel (2007) failed to find an effect of socio-emotional development on future academic success, although they did find an effect of attention (measured using the attention problems subscale of the IOWA Conners Teacher Rating Scale), which Arnold et al. (2012) include as a distinct component of social development.

It should be noted that in a number of these studies correlations are of a relatively small magnitude ( $r=0.07-0.19$  in Duncan et al., 2007 and Arnold et al., 2012) and these are perhaps only marginally significant after necessary corrections for multiple comparisons.

It has been suggested that the common finding of related social and academic skills demonstrates a bidirectional influence of one skill on the other in at least some stages of development. Welsh, Parke, Widaman & O'Neil (2001) found a bidirectional effect in a longitudinal study of American 2nd-3rd graders ( $n=163$ ), with a reciprocal relationship between social and academic skills from one school year to the other, although this was not found in slightly younger children. It seems clear that various aspects of social and emotional development are related to academic achievement in a number of ways and it appears that an initial set-back in one will likely lead to delay in the other.

## **6.2.2 The relationship between motor development and academic achievement**

Just as better social skills have been linked to better academic outcomes, so too have early motor skills. For example, Grissmer, Grimm, Aiyer, Murrah & Steele (2010) found that fine motor skills in American children in kindergarten were a significant predictor of future maths and reading attainment at age 10. This finding was replicated in a group of French-speaking Canadian children tested in kindergarten and 2nd grade (Pagani, Fitzpatrick, Archambault & Janosz, 2010). Testing children in kindergarten and then the first year of primary school, Son & Meisels (2006) also found that both gross and visuo-motor skills predicted future maths and reading ability. The relationship between fine motor (specifically visuo-spatial integration skills such as hand-eye coordination) and academic skills has been found throughout childhood and adolescence (Carlson, Rowe & Curby, 2013). It should be noted however that this study only tested 57

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<sup>2</sup>It should be noted that the majority of studies described have been conducted in North America, so it is important to bear in mind the possible effect of culture on findings.

children between 5-18 years of age. Despite their intention to use such a wide age range to investigate the persistence of the association between motor and academic skills with increasing age, there are perhaps insufficient numbers representing each age to suggest with any certainty that the association is robust across much of childhood and adolescent development. Additionally, there was no analysis to rule out an effect of age, and it is not clear that all tests administered were age-normed.

### **6.2.3 The relationship between social and motor development**

The link between social and motor skills in ASD and DCD has been discussed in previous chapters, and the consensus seems to be that these two disorders share a number of social, motor, behavioural and emotional symptoms. The link between social and motor skills in typical development has been less extensively researched, particularly in school-aged children (Leonard & Hill, 2014), although available evidence does suggest that a similar pattern of coexisting social and motor strengths or weaknesses is apparent in typical development.

A link between social and motor ability has been identified in children as young as 8 months old (Lamb et al., 1982), and this [small ( $r=0.18$ )] correlation was of comparable strength to previous findings comparing social and cognitive skills (Lamb, Garn & Keating, 1981). Lamb et al. (1982) conclude that the effect is one driven by competence rather than an artefact of performance differences brought on by the testing situation. The relationship between social and motor skills in older children has received little attention outwith the neurodevelopmental disorder literature. A number of studies have however investigated the relationship between social skills, academic attainment and a child's participation in organised sports and other clubs. It has generally been found that those children who attend after school activities have better social development than children who do not attend organised clubs, with sports activities promoting conflict resolution skills, social competence and social maturity (Howie, Lukacs, Pastor, Reuben & Mendola, 2010; Fletcher, Nickerson & Wright, 2003). It should be noted that children attending after school activities of any kind were likely to show better social skills, although sports and non-sports activities seem to be related to distinct aspects of social behaviour. Fletcher et al. (2003) suggest that the relationship might be due to those children with better social skills being more inclined to participate in sports activities as they are at ease working with their peers, compared to children with less well-developed social skills.

As well as evidence of direct links between social and motor skills, Wilson, Piek & Kane (2013) have suggested that social skills in a sample of 4-6 year olds ( $n=475$ ) may play a mediating role in the development of their motor skills and internalising behaviours (anxiety, depression, worry etc.). Supporting previous findings considering the relationship between academic achievement and social and motor development, Wilson et al. (2013) also found significant correlations between IQ and both motor and

social skills, although both were of a relatively small magnitude ( $r=0.19$  and  $0.27$ ). A partial correlation controlling for age, gender and verbal IQ found a significant positive correlation between observed motor skill (using the BOTMP second edition: BOTMP-2, Bruininks & Bruininks, 2005) and teacher-rated social skills ( $r=0.27$ ). The authors suggest that motor deficits reduce the amount of social play these children participate in, and this may affect emotional factors such as internalising behaviours. This is supported by findings of social and emotional difficulties in children with DCD (Gillberg & Gillberg, 1983; Schoemaker, Lingam, Jongmans, van Heuvelen & Emond, 2013). Finally, considering emotion recognition as a key skill in social development, Cummins, Piek & Dyck (2005) found that children aged 6-12 ( $n=39$ ) with mild to moderate motor difficulties (although no official diagnosis of DCD) were less able than TD children to recognise static and dynamic emotions from faces. When visuospatial skills and emotion recognition were controlled for, motor ability predicted children's level of social competence, with those children with observed motor difficulties also displaying difficulties in social functioning (as measured by a parent questionnaire).

#### **6.2.4 Summary**

It is clear that many aspects of child development work in parallel, with progress in one domain being held back if there are significant delays in the other. It is not clear how these relationships work and what the key directions of causation are, however it is clear that children will experience more global difficulties if for example motor skills are delayed (and vice versa), than would be expected if developmental domains were truly independent. To further assess the link between social and motor development, the present study uses the SRS and DCDQ-07, both used in clinical settings, to measure social and motor development in a group of 5-11 year old TD children.

### **6.3 Methods**

#### **6.3.1 Subjects**

With ethical approval from the City of Edinburgh Council, every City of Edinburgh Council mainstream primary school, and all East Lothian and West Lothian Council mainstream primary schools were phoned, written to or emailed individually with a request to take part. A number of schools received follow-up phone calls or letters with a note from a consultant paediatrician at the Edinburgh Royal Hospital for Sick Children, explaining the clinical relevance of the study and encouraging schools to take part. All schools were invited to take part to provide as wide a range of respondent backgrounds as possible, although ultimately subjects and participating schools were self-selected.

Seven local primary schools agreed to take part. Participating schools included one independent school and 6 City of Edinburgh Council schools. Participating schools were from different areas of Edinburgh, each drawing pupils from different socioeconomic backgrounds. The percentage of pupils receiving free school meals is a good indicator of socioeconomic status and has been used here to describe the schools involved. The total for Edinburgh City primary schools for the academic year 2012/2013 was 19.9%. Scores on this index for participating schools for this period are detailed in Table 6.1 alongside school roll and response figures. (Note that data were collected in the academic year 2012/2013 and 2013/2014, however free school meal data for the most recent year is not available at the time of writing.) One of the schools that chose to take part is classed as a Positive Action School: these are schools in which at least 40% of pupils are eligible for free school meals and/or a clothing grant from the local government. Positive Action schools receive additional funding for extra classroom staff and resources.

In each school, parents (or guardians, referred to henceforth as ‘parents’) of all children in P1-P7 (all primary school classes except nursery) were sent a letter outlining the study and a consent form. Of the 2758 parents who were sent the initial request to participate, 378 gave written consent. Of those consenting, 256 returned completed questionnaires. Additional responses were gathered from 6 parents (of 100, of whom 8 gave initial consent) whose children attend a local after school club for primary-aged children. (Note that this a fee-paying after school club with children typically coming from schools with a low free-school-meals index, all located within broadly the same catchment area.) Of those parents approached via their child’s school or the after school club, 9.17% completed a questionnaire pack. Details of parents completing the questionnaires were not collected.

Table 6.1: School roll, percentage of pupils receiving free school meals, number of parents giving consent, and number of completed questionnaire packs returned

School	School roll	Free school meal (%)	Initial consent	Returned
Abbeyhill	145	27.5	18	10
ASC	100	NA	8	6
Balgreen	367	31.9	35	21
Castleview	204	60.7	0	NA
Currie	354	7	40	31
Gylemuir	411	13.6	67	45
Murrayburn	365	32.9	37	20
Watson’s	912	NA	181	129

Note, ASC is an after school club and Watson’s is a private school so free school meal uptake is not available.

### 6.3.2 Materials

Parents received four questionnaires, outlined below and given in Appendices D-F (note that the SRS is not provided due to copyright restrictions). All parents completed paper copies of the questionnaires. Parents also completed a consent form identifying their child if they gave permission for their child to be considered for future behavioural studies. Parents who did not want their child to be considered for future studies were not required to identify their child<sup>3</sup>.

- Social Responsiveness Scale (SRS)

This is a standardised questionnaire. The SRS assesses five domains of social functioning, with questions from each randomly distributed throughout the questionnaire. Parents are not aware of the five subcomponents. The five subcomponents include: social awareness (8 statements); social cognition (12 statements); social communication (22 statements); social motivation (11 statements); and mannerisms (12 statements). Each subcomponent is given a score and the sum of these gives a total score. There are a total of 65 statements regarding various aspects of social behaviour and typically autistic symptoms. These all use a 1-4 rating system, ranging from ‘not true’ to ‘almost always true’. Parents are asked to consider their child’s behaviour in the last six months, and no reference is made to comparison with the child’s peer group.

- Developmental Coordination Disorder Questionnaire-2007 (DCDQ-07)

This is also a standardised questionnaire. The DCDQ-07 is split into three subcomponents (again, this is not made clear to parents although questions from each subcomponent are given together in this case): control during movement (6 statements); fine motor/ handwriting (4 statements); general coordination (5 statements). A score is calculated for each subcomponent and the sum of these gives a total score. All 15 statements use the same 1-5 response scale, ranging from ‘not at all like your child’ to ‘extremely like your child’. Parents are asked to consider their child’s ability relative to others of the same age.

- Baseline development questions

This questionnaire was designed specifically for the present study. This is a brief questionnaire about general academic attainment and aspects of behaviour not assessed in the SRS or DCDQ-07. Parents are asked to consider their child compared to other children of the same age. Questions include ratings of perceived academic attainment in English and maths, and competitiveness when playing with other children and when playing single player computer games. They are also asked if their child takes part in any extra curricular sports and how long they spend playing outside or taking part in sports (less than 2 hours/week, 2-4 hours, 5-7 hours, 8+ hours).

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<sup>3</sup>To maintain anonymity, consent forms have not been stored with completed questionnaires.

- Child's familial risk of neurodevelopmental disorders

This questionnaire was also designed specifically for the present study. It is a brief questionnaire asking about the presence of any neurodevelopmental disorders in the child's family. Diagnoses of interest included ASD, HFA, AS, other autism diagnosis (which should be specified), DCD, dyspraxia, speech dyspraxia, dyslexia, dyscalculia, dysgraphia, ADHD, CP and other motor difficulty (which should be specified).

### 6.3.3 Procedure

Parents completed the above questionnaires and returned them to their child's teacher. All parents received questionnaire packs with questionnaires in the order above, however there were no instructions as to the order these should be completed in. Questionnaires were completed anonymously, using only a subject number, and details of related disorders in families did not require disclosure of the exact diagnosis, nor details of the exact relationship of the affected person to the child. This extra level of anonymity was added to ensure that confidentiality was not seriously compromised for parents who opted to identify their child on the consent form. (However the majority of parents who did indicate that there was a relevant diagnosis in the family went on to detail at least the diagnosis, with many also detailing the relationship of the family member to the child.)

## 6.4 Results

262 questionnaire packs were returned (68% of those expected from initial consent form returns) and in over 99% of cases the respondent was a parent, most often the child's mother. Of the 262 returned packs 73 (28%) of these were not able to be used in analysis. Data were excluded for a number of reasons. Data for children with any relevant diagnosis were excluded from analysis (this included children currently undergoing assessment for relevant diagnoses). This group included children with ASD, DCD, dyspraxia, ADHD, dyslexia, dysgraphia and CP. These diagnoses were reported by respondents and were not able to be verified via medical records. Data for those children whose parents did not complete the diagnosis questionnaire were also not included in analysis. There were too few cases for each diagnosis to warrant separate diagnosis-specific analysis, or an adequate analysis considering all diagnoses together. Those children with a family history of any neurodevelopmental or motor deficit but no diagnosis themselves *were* included in analyses ( $n=57$ ). Children scoring above clinical cut-offs on the SRS or DCDQ-07 ( $n=22$  and  $n=5$  respectively) were included in analyses as scores on these questionnaires alone are not adequate for a diagnosis of either ASD or DCD. Data were not included if a subject did not have a full compliment of completed questionnaires, so that every subject could be included in each correlational

analysis. If one question was not answered in the DCDQ-07 or SRS then the mean of the relevant subcomponent was used for the missing question. For questionnaires that omitted the child's age, if the child's class was known then the modal age for that class was assigned to the subject (so five years for P1, six for P2 etc.), otherwise the data were not used. Details of the demographics for the final data set are shown in Table 6.2 and details of the main measures are shown in Table 6.3. Figure 6.1 shows the distribution of DCDQ-07 and SRS scores, showing skews towards the lower end of the typical range in both cases.

#### 6.4.1 Preliminary analyses

A preliminary analysis (Kruskal-Wallis H test as data were not normally distributed) was carried out to ensure that ratings did not depend on school (i.e. parents from one school were not more likely to consistently select the most desirable ratings than parents from another school). Median ratings for each measure split by school is shown in Table 6.4. There was no significant effect of school on any measure:  $H(6) = 12.03, p = 0.061$  (SRS);  $H(6) = 7.50, p = 0.277$  (DCDQ-07);  $H(6) = 3.24, p = 0.779$  (school attainment);  $H(6) = 3.78, p = 0.706$  (sports participation).

A second preliminary analysis concerned the SRS and DCDQ-07. Factor analysis was carried out to ensure that the SRS and DCDQ-07 were loading on separate factors, and this was verified (see Table 6.5). The inclusion of school attainment and sport time resulted in a third factor being identified, which these two questions loaded on.

Data from the questions relating to competitiveness were not included in analysis. As previous literature has focused on the relationship between academic attainment, sports participation and either social or motor skills it was decided that these would be the most informative baseline questions to compare against the social-motor correlation. Academic attainment in English and maths were summed to give a single attainment score. By excluding children with dyslexia, dyscalculia etc. this summing should not disadvantage children who may have poor English or maths attainment due to a diagnosed disorder. While it is possible that some children may not have English as a first language and this would impact on their English score, there was no formal question of the child's first language so it is not possible to filter out this small subgroup. This is acknowledged as a slight limitation although with relatively small numbers of foreign-language pupils in the primary schools involved any adverse effect of this oversight will be of a very small magnitude. Additionally, completing the questionnaires requires a good level of English, therefore participating parents whose first language is not English are assumed to have a good standard of English and it is assumed the same will be true of their children.

Table 6.2: Age, gender and school demographics for the final data set

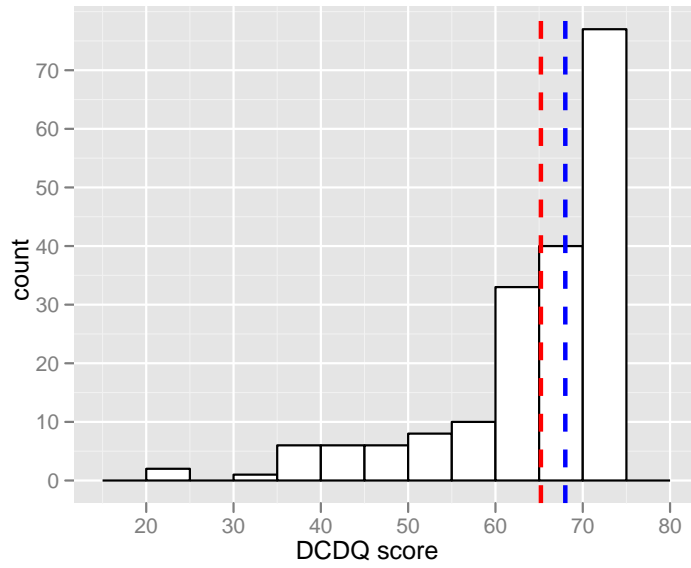
School	Age 5 (M:F)	Age 6 (M:F)	Age 7 (M:F)	Age 8 (M:F)	Age 9 (M:F)	Age 10 (M:F)	Age 11 (M:F)	Total (M:F)
A	1 (1:0)	1 (0:1)	2 (2:0)	0 (NA)	0 (NA)	0 (NA)	0 (NA)	4 (3:1)
ASC	0 (NA)	3 (1:1)	0 (NA)	1 (1:0)	0 (NA)	0 (NA)	0 (NA)	4 (2:1)
B	2 (0:2)	3 (2:1)	0 (NA)	0 (NA)	1 (1:0)	0 (NA)	0 (NA)	6 (3:3)
Cu	2 (1:1)	7 (6:1)	4 (3:1)	1 (0:1)	4 (0:4)	2 (1:0)	1 (0:1)	21 (11:9)
G	4 (3:1)	6 (4:1)	9 (6:3)	3 (0:2)	6 (3:3)	7 (1:5)	3 (0:3)	38 (17:18)
M	4 (3:1)	3 (0:3)	2 (2:0)	3 (0:3)	1 (1:0)	1 (0:1)	0 (NA)	14 (6:8)
W	28 (11:16)	13 (8:4)	11 (4:6)	15 (8:6)	9 (5:2)	10 (7:2)	16 (9:7)	102 (52:43)
<b>Total</b>	<b>41 (19:21)</b>	<b>36 (21:12)</b>	<b>28 (17:10)</b>	<b>23 (9:12)</b>	<b>21 (10:9)</b>	<b>20 (9:8)</b>	<b>20 (9:11)</b>	<b>189 (94:83)</b>

Note that some parents did not indicate their child's gender ( $n=12$ ) although their data were included as gender was not part of any planned analyses: where the sum of male and female totals differ from the overall total the extra data did not specify the child's gender.

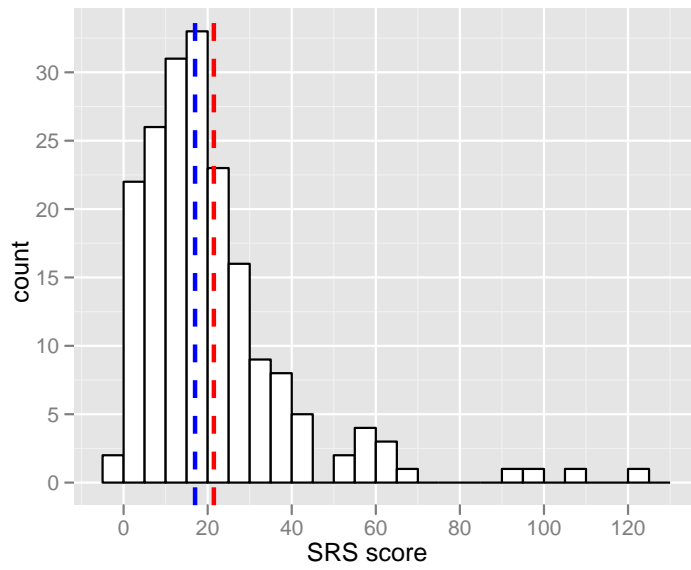
Table 6.3: Details of the four main measures split by age

Measure (possible range)	Age (years)	Median	Range	Number in clinical range
DCDQ-07 (0-75)	5	67	38-75	5
	6	68	43-75	1
	7	67	37-75	4
	8	68	25-75	3
	9	71	24-75	3
	10	71.5	47-75	3
	11	70.5	36-75	3
SRS (0-195)	5	16	0-63	0
	6	17	1-56	0
	7	17	4-124	1
	8	20	2-108	2
	9	19	0-92	2
	10	16.5	1-34	0
	11	13.5	2-45	0
School attainment (0-8)	5	5	2-8	NA
	6	6	2-8	NA
	7	6	3-8	NA
	8	5	3-8	NA
	9	7	2-8	NA
	10	6	2-8	NA
	11	7	2-8	NA
Sports participation (0-3)	5	2	0-3	NA
	6	2	0-3	NA
	7	2	0-3	NA
	8	2	0-3	NA
	9	2	1-3	NA
	10	2	1-3	NA
	11	2	1-3	NA

Clinical range for DCDQ-07 is: 15-46 (5-7 years); 15-55 (8-9 years); 15-57 (10-15 years).  
 Clinical range for SRS raw scores is 70+ for males and 65+ for females.



(a) Distribution of DCDQ-07 total scores.



(b) Distribution of SRS total scores.

Figure 6.1: Distribution of DCDQ-07 and SRS total scores. The red line shows the mean score, and the blue line shows the median score. The average score is within the typical range for both measures.

Table 6.4: Median scores for each measure according to school

	School	Median
DCDQ-07	Abbeyhill	64
	ASC	68
	Balgreen	64
	Currie	72
	Gylemuir	65.5
	Murrayburn	66
	Watson's	69
School attainment	Abbeyhill	7
	ASC	6
	Balgreen	6.5
	Currie	6
	Gylemuir	5.5
	Murrayburn	4.5
	Watson's	6
SRS	Abbeyhill	22
	ASC	20.5
	Balgreen	17
	Currie	16
	Gylemuir	19.5
	Murrayburn	28
	Watson's	15
Sport time	Abbeyhill	2
	ASC	2.5
	Balgreen	2
	Currie	2
	Gylemuir	2
	Murrayburn	1.5
	Watson's	2

Table 6.5: Rotated component loadings for SRS and DCDQ-07 components

Component	1	2
SRS Awareness	0.77	
SRS Cognition	0.88	
SRS Communication	0.91	
SRS Motivation	0.81	
SRS Mannerisms	0.81	
DCDQ-07 Control during movement		0.84
DCDQ-07 Fine motor and handwriting		0.87
DCDQ-07 General coordination		0.88

Note that loadings less than 0.4 are excluded.

### 6.4.2 Correlational analysis

Spearman correlation was used as data were not normally distributed. In each case  $n=189$  and Bonferroni corrections were made for multiple comparisons. There was no significant correlation between age and scores on the SRS, DCDQ-07, academic attainment or sports participation (see Table 6.6). To ensure there was no non-linear relationship between the two main variables (SRS and DCDQ-07), with age as a possible moderating factor, SRS and DCDQ-07 scores were correlated separately for each age group (see Table 6.7). The youngest age groups do appear to have stronger correlations than the oldest age groups, however the pattern is inconsistent. Differences between the youngest and oldest groups are likely due to the difference in sample size, with more than twice as many five year olds as there were eleven year olds. There is no compelling evidence that age has an effect on the relationship between SRS and DCDQ-07 scores, therefore age was not statistically controlled for in final correlational analysis.

Correlations between the main measures were significant for each comparison with the exception of sports participation and academic attainment ( $r_s = 0.03, p = 0.734$ ). Scores on the DCDQ-07 correlated significantly with SRS score ( $r_s = -0.49$ ), academic attainment ( $r_s = 0.26$ ) and sports participation ( $r_s = 0.27$ ), all at  $p < 0.001$ . The moderate correlation between the DCDQ-07 and SRS is of a considerably larger magnitude than the correlation between DCDQ-07 and either academic attainment or sports participation. As hypothesised, more DCD symptoms is associated with more ASD symptoms (see Figure 6.2). SRS score also correlated significantly with academic attainment and sports participation, although again this was to a lesser extent than the SRS correlated with the DCDQ-07 ( $r_s = -0.27, p < 0.001$  for SRS/academic attainment and  $r_s = -0.18, p = 0.007$  for SRS/sports participation).

As expected from previous research, all aspects of development assessed appear to be interrelated, although the relationship is noticeably stronger between social and motor development.

Table 6.6: Age correlated with each main measure

	Spearman's <i>rho</i> statistic
SRS	$r_s(187) = -0.08, p = 0.29$
DCDQ-07	$r_s(187) = 0.13, p = 0.07$
Academic attainment	$r_s(187) = 0.11, p = 0.15$
Sports participation	$r_s(187) = 0.09, p = 0.21$

## 6.5 Discussion

This chapter has reported a study examining the related nature of parents' perceptions of their typically developing child's social, motor and academic development. This question was driven primarily by the clinical work detailed in previous chapters, in

Table 6.7: SRS correlated with DCDQ-7 scores for each age group

Age	DCDQ-07/SRS correlation
5 years	$r_s(39) = -0.52, p < 0.001$
6 years	$r_s(34) = -0.58, p < 0.001$
7 years	$r_s(26) = -0.55, p = 0.002$
8 years	$r_s(21) = -0.24, p = 0.263$
9 years	$r_s(19) = -0.63, p = 0.002$
10 years	$r_s(18) = -0.18, p = 0.346$
11 years	$r_s(18) = -0.38, p = 0.099$

Note  $p$ -values are uncorrected.

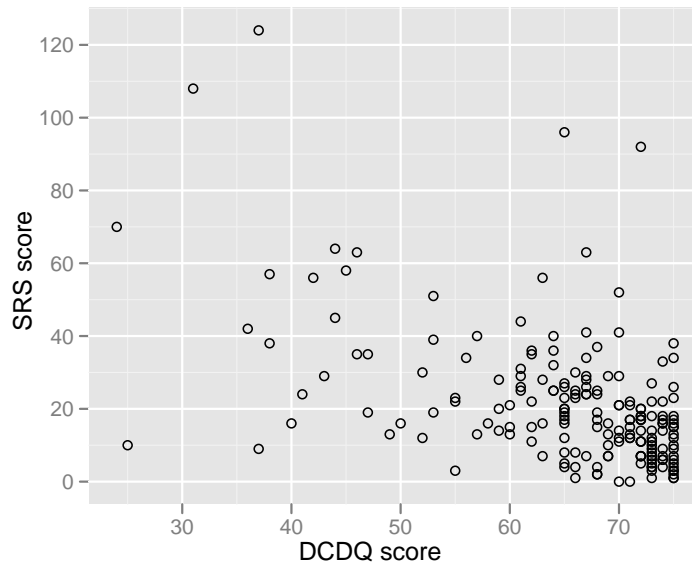


Figure 6.2: SRS/DCDQ-07 correlation. Higher scores on the SRS are indicative of more ASD symptoms, and lower scores on the DCDQ-07 are indicative of more DCD symptoms.

which the social and motor development of children and adults with ASD and DCD was found to overlap to a sizeable degree.

Findings support previous research which has highlighted the interrelated nature of various aspects of development, with clear links between social skills, motor skills and academic attainment (Lamb et al., 1982; Newcomb et al., 1993; Dobbs et al., 2006; Arnold et al., 2012; Malecki & Elliott, 2002; Welsh et al., 2001; Grissmer et al., 2010; Pagani et al., 2010; Carlson et al., 2013; Lamb et al., 1982, 1981; Howie et al., 2010; Fletcher et al., 2003; Wilson et al., 2013). It is important to note that in this case the strongest relationship was found between social and motor skills. This relationship did not depend significantly on age, and was seen throughout early childhood (5-11 years), extending previous findings of a significant correlation between motor skills and academic attainment across the 5-18 year age range (Carlson et al., 2013). The present study differs from previous studies however in the magnitude of the correlation, with previous studies reporting relatively small correlations. The majority of previous studies focus on pre-school children and this may partly explain the discrepancies between studies: it is possible that the effect is to some degree age-dependent, although with no effect of age in the present study any age-dependent factors would have to be apparent only in the first few years of development. Further ruling out an effect of age, a similar association has also been found previously between older children's participation in sports and their social competence, and effects still tend to be relatively small ( $r=0.21-0.27$  in Fletcher et al., 2003). However, studies of sports participation do not directly assess motor competence and it is unclear whether the associations are brought about by better motor skills related to their sports participation or whether the structure of extra-curricular sports activities lends itself to improving social skills.

It should be noted that all questionnaires except the SRS asked about aspects of development relative to the child's peer group, which perhaps nullified any age effect, although it seems likely from previous research that progress in different developmental domains is interrelated throughout childhood. If a wider age range is not responsible for the larger than average correlations reported here it may be that different measures of ability can in part explain these differences. Previous studies have used a wide variety of measures of social competence, with each focusing on different social skills, and some tending to be quite narrow in focus: a strength of the present study is the use of a measure that covers a range of social and communication skills. It is also possible that skewing toward the lower ends of the normal range in both the SRS and DCDQ-07 artificially inflated the correlations.

It is assumed that the greater correlations for social and motor skills compared to associations with academic attainment or sports participation reflects a greater interdependence of social and motor skills in development, however it is possible that the stronger correlation may be an artefact of the questionnaires used, with the two standardised questionnaires correlating most strongly. It may be that the baseline questionnaire which included items about academic attainment and sports participa-

tion were not sufficiently sensitive. Standardised questionnaires with proven construct validity addressing these areas might highlight relatively larger correlations with social and motor skills. However, the majority of academic attainment questionnaires are designed for teachers, (e.g. Mercugliano, Power & Blum, 1999; DuPaul, Rapport & Perriello, 1991) and it was not possible to involve teachers in the present study.

The study has two main limitations: first is the generalisability of the results. Despite extensive recruitment efforts, it was not possible to collect data from a representative sample of children across the wide range of socioeconomic backgrounds. There was both a poor response from schools in allowing parents to be contacted, and also a poor response from parents in schools that chose to take part. Those that did take part tended to be from areas of less social deprivation: although a Positive Action school sent letters to parents ( $n=204$ ), no consent forms were returned. It should be noted however that three of the schools from which data were collected had a higher percentage of pupils receiving free school meals than the total for all Edinburgh city council schools. While it was not possible to obtain data for those children in schools in the poorest areas, a variety of backgrounds were captured with the exception of this extreme end of the scale. A second limitation concerns the use of parent ratings for all measures. It is acknowledged that parent ratings do lead to a large degree of variance as the number of respondents is so large, and each respondent will interpret questions differently and use different peer groups as a comparison for their child. For this reason teacher ratings or objective ratings would be preferable. Teacher ratings would reduce the number of respondents, and therefore reduce variance in respondent's interpretation of questions, however it was not possible to gather teacher ratings. Similarly, objective measures would address the problem associated with multiple respondents, however objective measures of a wide range of social behaviours are not readily available and would be impractical with such a large sample.

While parent ratings may be a potential drawback, the use of questionnaires widely used in ASD and DCD diagnosis is considered a clear strength. These questionnaires are well suited to investigate a range of social and motor behaviours and are well standardised and have demonstrable construct validity. Additionally, the DCDQ-07 has been found to correlate moderately with performance-based measures used in DCD diagnosis (Wilson et al., 2000) and the SRS has been found to adequately address a number of subtle atypical social behaviours associated with ASD (Constantino, Davis, Todd, Schindler, Gross, Brophy, Metzger, Shoushtari, Splinter & Reich, 2003). Other studies tend to use brief questionnaires or subscales of questionnaires to assess these constructs and this reduces the scope of these studies to the single aspect of social or motor development being assessed.

Although the SRS is primarily used for ASD screening, and is not strictly speaking a test of social skills per se, the SRS has been found to correlate well with other measures of social functioning, such as the Vineland Adaptive Behavior Scale (Pine, Luby, Abbacchi & Constantino, 2006). The SRS has also been used to assess the effect

of social skills training in ASD and there were significant improvements in items on the SRS specifically addressing social competence, but no improvement in language or repetitive interests items (Tse, Strulovitch, Tagalakakis, Meng & Fombonne, 2007). This suggests that the SRS is useful for assessing social skills in their own right, and is useful in investigations of social functioning in TD groups (Dixon, Tarbox & Nejdowski (2009), p122). Also, the questionnaire has been designed so that non-social domains of autism symptomatology such as repetitive and stereotyped interests are framed in the context of the impact they have on reciprocal social skills (Constantino et al., 2003). Finally, previous studies assessing social skills in TD have used questionnaires often used in ASD research (e.g. the Social Skills Rating System in Wilson et al., 2013), again supporting the use of the SRS. The SRS therefore seems ideal for use in a study investigating social and motor skills in typical development, particularly as the study was prompted by study of symptom overlap in ASD and DCD.

This study has shown a clear relationship between perceived social and motor development across pre-adolescent childhood. Using a large group of UK children, this work complements a large body of research previously conducted in North America. The interrelated nature of developmental domains in typical development seems to mirror the frequent comorbidity between ASD and DCD described in Chapter 2 and reported throughout this thesis. Given the social-motor relationship in TD, the frequent occurrence of social and motor difficulties in clinical groups is unsurprising.

# Chapter 7

## Conclusions

### 7.1 Research question

The aim of this thesis was to assess the nature of motor deficits in ASD and DCD. This was motivated by a conflict between empirical findings of frequent comorbid social and motor deficits in ASD and DCD, and both a lack of any real mention of motor deficits in the DSM-IV's ASD criteria, and exclusion criteria making a dual ASD/DCD diagnosis impossible. The bulk of the work reported in this thesis was completed before the introduction of the DSM-5, which changed the exclusion criteria and allowed for dual diagnosis, in line with the frequently high comorbidity rates found in empirical studies. The broader topic of visual and proprioceptive contributions to perception and action was also investigated in neurotypical adults, with results informing follow-up work with clinical groups.

#### 7.1.1 Why do motor skills matter?

Children with ASD and DCD have a number of motor difficulties, which affect their school life and academic attainment, their home life, their choice of hobbies, and their self confidence as they begin to recognise the difference in ability between them and their peers. Difficulties can be simple and relatively hidden, such as having messy handwriting or wearing Velcro or slip-on shoes to avoid having to tie laces. However some difficulties are more obvious to children's peers, such as not being able to ride a bike when everyone else can and struggling in P.E. at school. Children with DCD involved in the present studies had all received some form of occupational therapy, physical therapy or physiotherapy to try to address problems they were experiencing. While a number of children with ASD had similar motor difficulties, these were not always treated, although often children received some form of therapy. This thesis investigated the differences between motor difficulties in these two groups, primarily as it is an intriguing area of research, particularly given the recent diagnostic overhaul, but also because it has real world application: having 'two left feet' and being 'all thumbs' is not just a minor inconvenience for these children, but has a knock-on effect

on other areas of their development. If these disorders are better understood, it will be possible to keep improving therapies for different children with motor deficits, to improve the motor skills themselves and also to limit the damage of this knock-on effect.

An account of the experience of collecting data for the studies in this thesis is given below. This will focus on the difficulties of conducting this kind of work, from difficulties recruiting subjects, to practical difficulties in the lab, and how these difficulties affect the development of the area as a whole. Strengths and limitations of the present studies and scope for future research will also be discussed. This is followed by brief summaries of the findings from each chapter and general conclusions.

## **7.2 Working with children and clinical groups**

Working with children, particularly in clinical groups, presents a number of challenges. An obvious difficulty is recruitment. Even with the support of health professionals, hospitals, and schools, convincing parents and children that participation in research is worthwhile can be difficult, more so when testing cannot routinely take place at school. Particularly with ASD recruitment, it is not uncommon for children to need to be persuaded by parents to take part. When assessing motor difficulties, it is understandable that some children with motor difficulties will be less keen to take part as they are aware of their limitations and difficulties. For children like this who do take part, keeping them motivated when they know they are performing well below average is a constant concern, and one that must be addressed properly, particularly if continued participation in further tasks is required.

Recruitment problems have an obvious negative impact on a study's sample size, and the time taken to build even modest sample sizes is prolonged, particularly when recruitment requires other agencies to make initial contact with parents and children. The use of clinical samples recruited through the NHS is something of a double-edged sword. On the one hand it has clear benefits: well-defined groups and an entire diagnostic history for subjects. On the other hand, the length of time needed to complete tortuously long forms, procedures, background checks and finally the ethics committee meeting and subsequent corrections and amendments, puts great time pressures on any work using this recruitment method. In this case the whole process took 15 months before recruitment could even begin.

Once children have been recruited, the next challenge is gathering data. Motivation and engagement is difficult in any study involving children, particularly those in clinical groups who perhaps require simplified instructions and fairly brief tasks due to difficulties maintaining attention. A consequence of working with participants with relatively short attention spans is the need to reduce the number of trials and conditions compared to what would be possible with neurotypical adults. If more trials were possible it would be easier to capture a representative snapshot of performance in different

conditions. In developmental and atypical development studies it is not always possible to be certain that the measure of average performance in each condition is anything but a numeric average, rather than being truly representative of the child's typical behaviour. This problem of small data sets is seen in the majority of developmental studies, particularly in experimental designs involving multiple manipulated variables.

Once the child is motivated and engaged, the next challenge is to maintain that engagement. Particularly with children with ASD, it is often necessary to steer conversation away from the child's current topic of interest. This topic is often something you either know nothing about, or your reference point is 20 years out of date (e.g. being told about the new 3D Sonic game on the 3DS and only being able to comment on Sonic's early years on the humble Mega Drive). It is sometimes difficult to strike a balance between being friendly, and putting the child at ease by allowing them to go off-topic, and ensuring that testing is not prolonged unreasonably due to an extended discussion about video games, Pokemon, or other irrelevant topic.

Another important consideration is the need for flexibility in the experimental procedure. In studies involving neurotypical adults, the procedure outlined in the report is usually the procedure that has been carried out by every subject, with very little variation. In developmental studies procedures need to have a certain degree of flexibility to allow for individual children's needs. This holds especially true for studies involving clinical groups: some children will not tolerate a blindfold, therefore it must be possible for the experimenter to monitor the child to ensure the eyes are kept closed; some children will resist being placed in a particular position and will consistently move to a slightly different position. While this is not ideal, there are few alternatives to letting these children make minor adjustments to testing conditions. Excluding every child that is unable to follow the outlined procedure to the letter would result in the loss of the majority of the sample, particularly in clinical groups with various sensitivities and attentional and behavioural issues such as those in ASD.

With so many difficulties introduced by working with children and clinical groups, the biggest challenge arises when designing tasks for such multifaceted disorders as ASD and DCD. Tasks need to assess the research question, be suitable for the children taking part, must be engaging, and ideally have some ecological validity so that findings can be taken from the artificial lab setting and inform our understanding of these children's difficulties in daily living. The need for ecologically valid tasks is highlighted by the puzzling finding that children with known motor difficulties were able to complete some tasks as well as TD children with normal motor skills. (This was evident in an indirect action in the reaching task, and the more ecologically valid direct action in the posting task.) If some tasks are not problematic for these children, why are they failing the MABC-2, and why are they noted to have difficulties with every day tasks such as tying shoe laces and walking around an environment without bumping into things? This raises the question of how we decide what skills should be (and can be) tested in motor batteries, which form a large part of the diagnostic procedure for DCD. It

is true that assessment involves both objective measures from batteries and subjective observations of the way in which children complete tasks, however when motor batteries are used in research settings the subjective component is largely ignored. This is because many psychology researchers are not in a position to make such subjective observations, as they lack the training a qualified OT has. A problem with using only the objective component is that important details can be lost or can obscure the child's ability. For example, two of the three manual dexterity tasks in the MABC-2 are timed. Often less conscientious children were much more concerned with the speed element, resulting in a speed accuracy trade-off, with even typical children faltering because precision was deemed less important than speed. The ecological validity of these timed tasks is low, however an untimed task would need to be scored based on the style of movement execution and this is much more difficult to assess relative to normative data. Concerning the ecological validity of motor batteries, it is curious that navigating around an environment is not assessed as part of the gross motor subscale of the MABC-2: bumping into things is a common problem in DCD, and obstacle avoidance would be a simple task to assess in a controlled manner in the lab or clinic.

### **7.3 Working in schools**

While there is always a sigh of relief when a school agrees to take part, it is inevitably a logistical nightmare. First you must find a suitable place to work and set up the apparatus. Designing or choosing necessary apparatus must be considered early in task development. For instance, when carrying out motion tracking it was not possible to use the large, expensive and not-at-all-portable Optotrak system in schools, therefore the smaller magnetic tracking system was the only option. This meant that in each school the janitor was asked to look out a non-metal table, which is surprisingly hard to come by in modern schools. It is also necessary to consider how you will be travelling to the school, and if you are not driving door-to-door, how much you can physically carry or drag behind you. Once everything you need is in the school, the next hurdle is finding somewhere to set up. Testing environments in schools are often more cramped than the lab and when using the MABC-2 it was sometimes difficult to find a room large enough (and not in constant use) to complete the heel-toe walking task. The task was always completed, but sometimes required a quick dash to the gym hall in between P.E. lessons. With unused space hard to come by in schools, there is also the inevitable uninvited visitor (staff or pupil) walking through the room or knocking on the door during testing. It is therefore important to have an experimental procedure that allows for these disruptions, for example by being able to redo trials if there is a disruption.

Once the make-shift lab is set up, the real challenge begins: testing. Although it is time efficient in the long run to have the entire control group under one roof, it is sometimes difficult to pin children down, and day-to-day the process seems much less time efficient. The child you had arranged to test at 11:00 could be taken out

of class for a number of reasons, from helping to weed the school's nature garden, to being reprimanded in the headteacher's office. This results in either waiting until the child is back in class, or trying to find another child who is willing to complete testing now, and a teacher who is willing to let one of their pupils out of class at short notice. Timetabling also runs into problems when considering the three highlights of the day: break-time, lunch-time, and home-time, none of which a child is willing to be late for. By testing just before any of these daily highlights you run the risk of attention wandering. Time of the year is also a consideration: the start of the year is often ruled out as teachers want to get used to their new class; the run up to Christmas, with song practice and play rehearsals throughout December, is always a difficult time to (a) find the child you were expecting to test (b) keep over-excited children on track and (c) find a place to work that is not so close to carol practice that you can sing along, and has not been transformed into a Christmas craft area; then there is sports day, transition days for P7 pupils and finally the lead up to the summer holidays.

With so much outwith your control, and so many constraints when testing in schools it is inevitable that data will be less-than-perfect. However it is a convenient and effective way to collect control data relatively quickly, and is standard procedure in the majority of work involving school-aged children.

## **7.4 Strengths and weaknesses of the present studies**

A clear strength in the present studies is the inclusion of children identified by relevant health care professionals. Working with paediatricians and OTs during the recruitment phase has resulted in well-defined clinical groups, and the availability of medical records has been useful in ensuring eligibility. Access to medical records has also been very interesting. As a researcher, the world of diagnosis and assessment is typically someone else's domain. It has been interesting to see how these children can have very different diagnostic histories, how clinicians deal with such heterogeneous disorders, and the process by which other disorders (such as ADHD) are investigated and later eliminated. Reading through medical records also highlighted the difficulty in diagnosing ASD using DSM-IV criteria, particularly when severe motor deficits are apparent. This was particularly well illustrated by one child who was diagnosed with DCD 6 years before being assessed for and diagnosed with ASD, even though previous assessments for ASD placed him in the normal range. The ability to use these records and case histories to give the DSM-IV criteria context has been important when reflecting on the key question of this thesis and considering its clinical application.

The use of the same subjects throughout the series of studies allowed for performance in follow-up studies to be considered according to both group membership (ASD, DCD and TD) and also by observed motor ability (via MABC-2 scores gathered in initial testing). This is considered a clear strength when investigating the proposed vision-proprioception dissociation in ASD and DCD as there appeared to be some pos-

sible differences between children with spared and deficient motor skills. An associated weakness however was the difficulty in recruiting sufficient numbers, and this is a common problem in work with clinical groups. The initial study (Chapter 2 and the first half of Chapter 3) had a good number of TD and ASD subjects (the latter considerably more than similar previous studies). However attrition between studies meant that follow-up studies were less powerful. Additionally, it meant that it was not possible to fully investigate the alternative hypothesis that motor ability rather than ASD/DCD diagnosis affects sensory processing. However, in this case the problem was confounded by the high prevalence of motor deficits in the original ASD sample, which in itself is of great interest, as it provides support for the revised relationship between ASD and DCD in the DSM-5. It should be noted that recruitment efforts were extensive, with a number of paediatricians and OTs working within the NHS approaching suitable families, and a number of inquiries made to various schools and voluntary organisations. The high proportion of children with ASD with significantly impaired motor skills may in part be due to selection bias, as parents were aware of the nature of the studies. However with such strict guidelines about informed consent, particularly when working with vulnerable groups and recruiting through the NHS, the large amount of detail given to parents before testing was unavoidable.

The majority of the paradigms used here are well-established in the relevant literature (spatial location matching, mirror reach, posting/matching, RHI and postural matching). It can therefore be assumed that the tasks are effective. When working with highly variable groups such as children and clinical groups it is important to use tasks which have proven successful in the past. This reduces the chance of results being confounded by both inherent variability *and* poorly designed tasks. That being said, it is important to make any necessary changes to tailor common tasks to the needs of different subject groups, e.g. housing the prism inside the apparatus in the spatial location matching task rather than asking children to wear glasses. In some cases the task was also altered to better fit the specific research question (e.g. designing the posting/matching task so that targets could be proprioceptively-defined). However in each case the tasks stemmed from well-established tasks, which have produced consistent results, as reported in prior literature.

It is acknowledged that the majority of tasks used lack ecological validity, and for the most part do not resemble tasks that children with ASD and DCD are reported to find difficult. Practical restrictions made the study of ecologically valid tasks difficult. For example, basic tasks were favoured when investigating vision and proprioception in ASD and DCD, as high-level ecologically valid tasks are made up of a number of constituent parts. For instance, tasks such as dressing involve motor planning (putting a shirt on before a jumper and making sure the jumper is the right way round), visual and proprioceptive guidance (guiding a jumper cleanly over the head), and motor execution (pulling a jumper over the body). High-level tasks such as these are also more open to individual differences in how much children have practised them. Some children

with motor difficulties are helped with such tasks, and others are simply woken up 20 minutes earlier so they can get dressed on their own without time pressure. For these reasons using less ecologically valid tasks is a much cleaner way to investigate basic sensory processing. Once the fundamental details of sensory processing in these groups are understood, it should then be easier to extend research into more ecologically valid tasks to address this limitation.

## 7.5 Future research

Given the new possibility of a dual ASD/DCD diagnosis, it is suggested that future research focus on the best way to deliver OT and other therapies for motor difficulties to children with ASD. As social and motor deficits do appear to correlate significantly it is important to consider perhaps combined social and motor therapies both for children with ASD and DCD so that these children will be better able to adapt and function in school and at home.

With evidence of motor deficits persisting into adulthood, it may be beneficial to focus efforts on better understanding motor difficulties in adults with ASD and DCD. Currently therapies for young adults with motor difficulties like DCD are not readily available, perhaps as it is only relatively recently that DCD has been given as a routine childhood diagnosis. Diagnostic criteria also need to be reconsidered to include adult symptoms if it is assumed that children's problems will not resolve over time and the focus of difficulties may change. However, as early diagnosis and intervention is vital if children are to develop as well as they can, it is by no means suggested that the focus of research should be shifted from children to adults, but that the ongoing needs of children into adulthood should be considered.

Working with adult clinical groups would also avoid some of the problems associated with working with children in these groups, such as limited attention. Subjects with ASD and DCD in the adult study presented in Chapter 2 did not fatigue in the way that children did and would have been able to complete more trials, and perhaps less engaging and more thorough tasks, such as threshold detection tasks. A more rigorous approach with adults may help in confirming that motor difficulties in ASD are not condition-specific and are in fact symptoms of an additional DCD diagnosis.

If future research is to inform clinical services for ASD and DCD, research may benefit from more ecologically valid tasks, to ensure that results tap into real-life problems encountered by children with motor difficulties. This research would be best suited to an interdisciplinary approach, involving OTs, physiotherapists, paediatricians and research psychologists, to ensure that current assessments, interventions, and neuropsychological evidence is considered together. Assessing sensory contributions to tasks such as hand writing, safely navigating through the environment and basic self-care tasks may be beneficial in informing therapies and interventions for children receiving treatment for motor difficulties. Given the comparable skills of motor impaired ASD and

DCD children and non-impaired TD children on some of the tasks reported in this thesis, it may also be beneficial for future research to consider what motor impaired children *can* do, and try to understand their impairment in other tasks with respect to spared abilities.

## 7.6 Chapter summaries

### 7.6.1 Chapter 1

The first chapter gave an overview of the DSM-IV criteria for ASD and DCD and described the common symptoms shared by these conditions. Although ASD is primarily thought of as a social deficit, the study of motor difficulties has received increased attention in recent years. The question was raised as to whether motor symptoms apparent in ASD are characteristically different from those in DCD, supporting the DSM-IV's exclusion criteria making a formal ASD/DCD comorbid diagnosis impossible. The recent changes regarding comorbidity in the DSM-5 makes this a timely question. The conclusion drawn from the previous literature was that there do not appear to be any high-level differences in motor symptoms in ASD and DCD, although the lack of direct comparisons of these two groups, and the wide range of approaches taken to investigate motor difficulties made it difficult to draw firm conclusions. This provided the motivation for this thesis, which was designed to provide a direct comparison of ASD and DCD using both standard diagnostic tools, and well-established tests of sensorimotor ability.

### 7.6.2 Chapter 2

The first study presented in this thesis was designed to profile high-level motor skills in adults and children with ASD, DCD and TD using three different tests: a standardised and commonly used motor battery (MABC-2), a new computerised battery (cKAT), and a computerised imitation task. These tests allowed for measures of fine and gross motor skills in standard tasks used in therapy and clinical assessment; basic visuomotor skills; and ability to imitate a visuomotor action defined by a person or computer.

Results from the MABC-2 showed that 70% of children with ASD had motor skills at or below the 15th percentile and were therefore deemed to have either definite DCD or probable DCD (although in a clinical setting more tests and procedures would be carried out before giving a diagnosis).

Results from the cKAT battery suggested that ASD and DCD did seem to have poorer visuomotor skills on some tasks compared to TD, although a host of practical problems with this battery which seemed to selectively affect children in the clinical groups to a much greater extent than TD, suggests that these results should be viewed with caution. The results certainly do not suggest that this is a viable tool for assessing motor skills in young children or those with preexisting attentional, behavioural and

movement difficulties until practical limitations are addressed.

The imitation task found no selective impairment in the ASD group for actions performed by a person and those with no motor impairment seemed to have no tangible imitation deficit in any condition relative to TD. The DCD group was no different from the ASD subgroup with motor difficulties, suggesting that imitation ‘deficits’ previously reported in ASD may be rooted in motor difficulties rather than more classic autistic difficulties. These tasks were also administered to adults and major findings were broadly similar.

### **7.6.3 Chapter 3**

Chapter 3 reported a study assessing the relative contribution of visual and proprioceptive cues in perception and action in ASD and DCD. This was prompted by the lack of any clear differential pattern in high-level motor tasks as measured by the MABC-2 in Chapter 2. There is a growing body of research suggesting that ASD may be related to an over-reliance on proprioceptive cues, while children with DCD may execute movements based more strongly on visual cues. This hypothesis was tested in a perception task which calculated a visual weighting during a prismatic displacement task, in which a visual-proprioreceptive cue conflict was created. Results indicated that there was no robust effect of group on weighting, although the ASD group without motor difficulties did show a slight trend towards lower visual weightings, however this was driven by one child with very accurate proprioceptive acuity when faced with a cue conflict.

The hypothesis was tested a second time using a reaching task which again created a cue conflict, this time with a mirror. Again, the hypothesis was not supported and there was no difference between ASD, DCD and TD with regards their relative use of vision and proprioception during cue conflict. In the latter case however there were fewer children with ASD and spared motor skills so it was not possible to fully investigate whether a bias towards proprioception is only evident in those children with ASD without comorbid DCD.

### **7.6.4 Chapter 4**

Chapter 4 reported two studies investigating the role of proprioceptive cues in a goal-directed action and an orientation matching task. The first study used neurotypical adults to assess the role of proprioceptive feedback of targets in perception and action, specifically, whether the location of this feedback is important. The target was either direct: the hand is the target; or indirect: the proprioceptive feedback of the target is spatially displaced from the target location. It was found that direct proprioceptive feedback of target orientation, in which a letter is posted through a slot which is being held, produces a more accurate action than indirect feedback, in which the action is not directed to the location of the proprioceptive feedback. This was also true for orientation matching (a perceptual task).

The second study used the posting task with children with ASD, DCD and TD and assessed differences between groups when targets were defined proprioceptively (direct only), visually, or visually and proprioceptively. It was expected that if there is a visual-proprioceptive double dissociation in ASD and DCD that the groups would be differentially affected by the removal of vision. This hypothesis was not supported, and children with ASD and DCD appeared to make use of visual and proprioceptive feedback in similar ways, and were not significantly different from TD children. However as with the reaching task in chapter 3 the ASD group was largely made up of children with significant motor difficulties.

### 7.6.5 Chapter 5

Chapter 5 reported two tasks using the rubber hand illusion to assess the role of proprioceptive acuity and proprioceptive reliance on eliciting the illusion. The first involved neurotypical adults in two tests of proprioceptive acuity and the standard rubber hand illusion, with proprioceptive shift as the measure of illusion strength. It was hypothesised that those with better proprioceptive acuity would be less susceptible to the illusion that tends to arise following the cue conflict between visual and tactile stimulation. It was found that proprioceptive acuity in a spatial location matching task was significantly correlated with illusion susceptibility, with better acuity reducing the strength of the illusion. The same was not true for postural acuity. This is likely due to the very similar task requirements in spatial location matching and the illusion, and the lack of any need for postural acuity in the illusion. Interestingly however, there was no robust illusion, with drift following synchronous and asynchronous stimulation not differing significantly. This was unexpected and highlights a possible confound in previous research, whereby the use of questionnaire responses artificially inflates differences, suggesting that differences in previous studies may not be driven solely by the sensory effect of the illusion.

The task was also used with children with ASD, DCD and TD to further explore the proposed vision/proprioception double dissociation and also to explore claims that it takes longer to elicit the illusion in ASD than TD. It was hypothesised that if ASD and DCD are dissociated by the relative weightings of vision and proprioception then the ASD group should experience the illusion to a lesser extent than DCD, who should show greatest proprioceptive shift towards the visual stimulation (the rubber hand). In this case there *was* a robust illusion, however the hypothesis was not supported, with groups almost identical on the proprioceptive shift measures. Additionally, there was no evidence for a delayed illusion in ASD. Again it should be noted that too few children with ASD and spared motor skills participated to allow for a full investigation of ASD subgroups.

### 7.6.6 Chapter 6

The final study investigated parents' ratings of their typically developing children's social skills, motor skills and academic attainment. Previous research has found that social skills tend to be good predictors of academic achievement, and a small number of studies have found links between social and motor skills. With social and motor deficits apparent in ASD and DCD, it is of interest to understand the relationship between these basic skills in typical development. There was a significant positive correlation between social and motor skills across the primary school age range (5-11 years) in a group of 189 children. This correlation was of a greater magnitude than either social or motor skills correlated with perceived maths and English attainment relative to peers. These findings suggest that the related nature of social, motor and other cognitive abilities in neurodevelopmental disorders is unsurprising given the similar pattern in typically developing children. This study supported and extended previous findings, with a greater focus on social and motor skills specifically, and in a large group of children from UK schools, complementing previous work carried out in North America.

## 7.7 Conclusions

Neurodevelopmental disorders tend to cluster, and the overlap between ASD and DCD has been demonstrated here, with specific focus on motor deficits. This overlap suggests some underlying susceptibility in these children, which could well point to a shared causal origin of motor deficits in ASD and DCD. This sits opposite the idea of comorbidity reflecting two *distinct* diagnoses (Feinstein, 1970), however the term is often used to simply mean 'co-occurring' (Maj, 2005), and it is unclear which definition is used in new DSM-5 criteria which allow for comorbid diagnosis. Results from the novel direct comparisons of ASD and DCD presented in this thesis suggest that motor deficits in ASD are not ASD-specific, as there were no qualitative or quantitative differences in motor proficiency in children with ASD and DCD. As other direct comparisons are currently lacking in the literature, until further research is conducted to explore the aetiology of motor deficits in these two disorders, the term 'co-occurring' may be a more appropriate term than the currently poorly defined term 'comorbid'.

Having directly compared ASD and DCD, there is no evidence for characteristic motor deficits specific to ASD, and no evidence for the proposed double dissociation between groups in the use of visual and proprioceptive cues. An investigation of both high level motor ability, and the role of relevant sensory cues for movement and perception support these conclusions. Motor impaired children (with either diagnosis) showed a tendency to rely on visual information, pointing to a dampened sense of proprioceptive associated with motor difficulties. This is supported by previous research involving DCD and other motor impairments. Previous reports of a relative reliance on proprioception in ASD were not supported here, although 70% of the original ASD group (up

to 86% in follow-up studies) had motor difficulties in the clinical range. In some cases there did appear to be some differences between ASD with and without movement difficulties, however the relatively small subsample without motor difficulties makes it difficult to identify this possible trend with any certainty.

The exact nature of motor difficulties in ASD and DCD is still not fully understood. Given the clear relationship between social and motor development in children without neurodevelopmental disorders (as discussed in Chapter 6), it seems appropriate to consider the relationship between ASD and DCD as an extension of the interwoven nature of different developmental domains in typical development. It is not clear why some children with ASD have spared motor skills, and some children with DCD are less socially impaired, however it appears that in the majority of cases symptoms of ASD and DCD tend to overlap quite considerably. For this reason research into this area would be of great benefit to children with both ASD and DCD, and other neurodevelopmental disorders with shared symptoms.

Work from this thesis has highlighted the extent of motor difficulties in ASD and has consistently reported results that support the new possibility of an ASD/DCD dual diagnosis. As diagnostic criteria and procedure is constantly changing it is vital that we continue to look at this under-investigated area of ASD so that diagnostic criteria can continue to reflect the most recent findings from research.

## Appendix A

# Additional non-significant main and interaction effects: Chapter 2

Table A.1: Analysis of constant error for ASD, DCD and TD (adult imitation)

Condition	$F(1, 43) = 0.20, p = 0.658$
Condition*Group	$F(2, 43) = 0.06, p = 0.943$
Measure	$F(1, 43) = 2.37, p = 0.131$
Measure*Group	$F(2, 43) = 1.59, p = 0.215$
Condition*Measure	$F(1, 43) = 0.79, p = 0.38$
Condition*Measure*Group	$F(2, 43) = 1.20, p = 0.311$

Table A.2: Analysis of variable error for ASD, DCD and TD (adult imitation)

Condition	$F(1, 43) = 0.04, p = 0.852$
Condition*Group	$F(2, 43) = 0.27, p = 0.763$
Measure	$F(1, 43) = 0.29, p = 0.591$
Measure*Group	$F(2, 43) = 1.16, p = 0.325$
Condition*Measure*Group	$F(2, 43) = 2.88, p = 0.067$

Table A.3: Non-significant group effect for cKAT measures (child cKAT)

Pent RT	$F(2, 66) = 1.03, p = 0.363$
Pent MT	$F(2, 66) < 0.001, p = 1$
Pent DT	$F(2, 66) = 0.28, p = 0.760$
Pent PS	$F(2, 66) = 1.38, p = 0.259$
Pent PL	$F(2, 66) = 2.08, p = 0.133$
Tracing PA	$F(2, 63) = 2.65, p = 0.078$

Table A.4: Non-significant group effect for cKAT measures using MABC-2-defined groups (child cKAT)

Pent MT	$F(2, 62) = 1.93, p = 0.154$
Pent DT	$F(2, 62) = 0.66, p = 0.523$
Pent PS	$F(2, 62) = 0.61, p = 0.547$
Pent PL	$F(2, 62) = 2.86, p = 0.065$

Table A.5: Analysis of subject/model correlation for ASD, DCD and TD (child imitation)

Condition	$F(1, 62) = 2.62, p = 0.110$
Condition*group	$F(2, 62) = 1.75, p = 0.183$
Measure*group	$F(2, 62) = 1.43, p = 0.248$

Table A.6: Analysis of subject/model correlation for DCD and motor impaired ASD (child imitation)

Group	$F(1, 29) = 0.001, p = 0.946$
Group x Condition	$F(1, 29) = 2.40, p = 0.133$
Group x Measure	$F(1, 29) = 0.55, p = 0.466$
Condition x Measure x Group	$F(1, 29) = 0.06, p = 0.815$

Table A.7: Analysis of constant error for ASD, DCD and TD (child imitation)

Condition	$F(1, 62) = 2.67, p = 0.108$
Condition*Group	$F(2, 62) = 0.42, p = 0.662$
Measure	$F(1, 62) = 0.18, p = 0.670$
Condition*Measure	$F(1, 62) = 0.13, p = 0.725$
Condition*Measure*Group	$F(2, 62) = 0.34, p = 0.717$

Table A.8: Analysis of constant error for DCD and motor impaired ASD (child imitation)

Group	$F(1, 29) = 1.11, p = 0.301$
Group x Condition	$F(1, 29) = 0.18, p = 0.678$
Group x Measure	$F(1, 29) = 2.94, p = 0.097$
Condition x Measure x Group	$F(1, 29) = 1.21, p = 0.281$

Table A.9: Analysis of constant error for ASD pure, clinical motor deficit and TD (child imitation)

Condition	$F(1, 62) = 2.07, p = 0.155$
Condition*Group	$F(2, 62) = 0.30, p = 0.739$
Measure	$F(1, 62) = 0.06, p = 0.801$
Condition*Measure	$F(1, 62) = 0.31, p = 0.580$
Condition*Measure*Group	$F(2, 62) = 0.81, p = 0.449$

Table A.10: Analysis of variable error for ASD, DCD and TD (child imitation)

Condition	$F(1, 62) = 0.01, p = 0.915$
Condition*Group	$F(2, 62) = 0.44, p = 0.646$
Measure	$F(1, 62) = 0.57, p = 0.452$
Measure*Group	$F(2, 62) = 1.03, p = 0.364$
Condition*Measure*Group	$F(2, 62) = 2.30, p = 0.109$

Table A.11: Analysis of variable error for DCD and motor impaired ASD (child imitation)

Group	$F(1, 29) = 0.10, p = 0.752$
Group x Condition	$F(1, 29) = 0.36, p = 0.552$
Group x Measure	$F(1, 29) = 0.13, p = 0.721$
Condition x Measure x Group	$F(1, 29) = 3.81, p = 0.061$

Table A.12: Analysis of variable error for ASD pure, clinical motor deficit and TD (child imitation)

Condition	$F(1, 62) = 0.33, p = 0.571$
Condition*Group	$F(2, 62) = 0.18, p = 0.840$
Measure	$F(1, 62) = 0.22, p = 0.640$
Measure*Group	$F(2, 62) = 1.34, p = 0.268$
Condition*Measure*Group	$F(2, 62) = 1.74, p = 0.184$

## Appendix B

# Additional non-significant main and interaction effects: Chapter 3

Table B.1: Non-significant group comparisons for each plano condition (spatial location matching)

Condition	Comparison groups	Mann-Whitney U
VP	ASD/TD	U=368, p=0.946
	DCD/TD	U=99, p=0.716
	ASD/DCD	U=128, p=0.709
VPP	DCD/TD	U=72.5, p=0.151
	ASD/DCD	U=126, p=0.662
PP	DCD/TD	U=81, p=0.275
	ASD/DCD	U=102, p=0.225

VP=Target defined by vision only; VPP=Target defined by vision and proprioception; PP=Target defined by proprioception only

Table B.2: Comparison of ASD pure, clinical motor deficit and TD for each plano condition (spatial location matching)

Condition	Comparison groups	Mann-Whitney U
VP	Clinical motor deficit/TD	$U = 370.5, p = 0.98$
	Clinical motor deficit/ASD pure	$U = 139, p = 0.987$
	ASD pure/TD	$U = 104.5, p = 0.887$
VPP	Clinical motor deficit/ASD pure	$U = 113.5, p = 0.4$
	ASD pure/TD	$U = 76.5, p = 0.203$
PP	Clinical motor deficit/ASD pure	$U = 110, p = 0.339$
	ASD pure/TD	$U = 92, p = 0.518$

VP=Target defined by vision only; VPP=Target defined by vision and proprioception; PP=Target defined by proprioception only.

## Appendix C

# Additional non-significant main and interaction effects: Chapter 4

Table C.1: Analysis of constant error for DCD and motor impaired ASD (child posting)

	60% MT	100% MT
Group	$F(1, 21) = 0.31, p = 0.585$	$F(1, 21) = 0.02, p = 0.899$
Group x condition	$F(1, 21) = 0.17, p = 0.685$	$F(1, 21) = 2.39, p = 0.137$

Table C.2: Analysis of absolute and variable error for DCD and motor impaired ASD (child posting)

		DN	DV
Absolute error	60% MT	$U = 41, p = 0.516$	$U = 34, p = 0.256$
	100% MT	$U = 38, p = 0.392$	$U = 39, p = 0.431$
Variable error	60% MT	$U = 35, p = 0.286$	$U = 38, p = 0.392$
	100% MT	$U = 48, p = 0.865$	$U = 42, p = 0.562$

DN=Direct proprioception, no vision; DV=Direction proprioception, vision.

Table C.3: Analysis of absolute and variable error for clinical motor deficit and TD at 60% and 100% MT (child posting)

Time point	Condition	Absolute error	Variable error
60% MT	DN	$U = 81, p = 0.193$	$U = 85, p = 0.253$
	DV	$U = 105, p = 0.714$	$U = 91, p = 0.363$
100% MT	DN	$U = 95, p = 0.451$	$U = 91, p = 0.363$
	DV	$U = 107, p = 0.773$	$U = 97, p = 0.499$

DN=Direct proprioception, no vision; DV=Direction proprioception, vision.

# Appendix D

## DCDQ-07

**COORDINATION QUESTIONNAIRE** (REVISED 2007)

<b>Name of Child:</b> _____	<b>Today's Date:</b>
<b>Person completing Questionnaire:</b> _____	<b>Child's Birth:</b>
<b>Relationship to child:</b> _____	<b>Child's Age:</b>

Year	Mon	Day

Most of the motor skills that this questionnaire asks about are things that your child does with his or her hands, or when moving. A child's coordination may improve each year as they grow and develop. For this reason, it will be easier for you to answer the questions if you think about other children that you know who are the same age as your child. Please compare the degree of coordination your child has with other children of the same age when answering the questions. Circle the one number that best describes your child. If you change your answer and want to circle another number, please circle the correct response twice. If you are unclear about the meaning of a question, or about how you would answer a question to best describe your child, please call \_\_\_\_\_ at \_\_\_\_\_ for assistance.

	Not at all like your child 1	A bit like your child 2	Moderately like your child 3	Quite a bit like your child 4	Extremely like your child 5
1. Your child <i>throws a ball</i> in a controlled and accurate fashion.	1	2	3	4	5
2. Your child <i>catches</i> a small <i>ball</i> (e.g., tennis ball size) thrown from a distance of 6 to 8 feet (1.8 to 2.4 meters).	1	2	3	4	5
3. Your child <i>hits</i> an approaching <i>ball</i> or <i>birdie</i> with a bat or racquet accurately.	1	2	3	4	5
4. Your child <i>jumps</i> easily <i>over</i> obstacles found in garden or play environment.	1	2	3	4	5
5. Your child <i>runs</i> as fast and in a <i>similar</i> way to other children of the same gender and age.	1	2	3	4	5
6. If your child has a <i>plan</i> to do a motor <i>activity</i> , he/she can organize his/her body to follow the plan and effectively complete the task (e.g., building a cardboard or cushion "fort," moving on playground equipment, building a house or a structure with blocks, or using craft materials).	1	2	3	4	5 (OVER)

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	Not at all like your child 1	A bit like your child 2	Moderately like your child 3	Quite a bit like your child 4	Extremely like your child 5
7.	Your child's printing or <i>writing</i> or drawing in class is <i>fast</i> enough to keep up with the rest of the children in the class.				
	1	2	3	4	5
8.	Your child's printing or <i>writing</i> letters, numbers and words is <i>legible</i> , precise and accurate or, if your child is not yet printing, he or she <i>colors and draws</i> in a coordinated way and makes pictures that you can recognize.				
	1	2	3	4	5
9.	Your child uses appropriate <i>effort</i> or tension when printing or writing or drawing (no excessive <i>pressure</i> or tightness of grasp on the pencil, writing is not too heavy or dark, or too light).				
	1	2	3	4	5
10.	Your child <i>cuts</i> out pictures and <i>shapes</i> accurately and easily.				
	1	2	3	4	5
11.	Your child is interested in and <i>likes</i> participating in <i>sports or active</i> games requiring good motor skills.				
	1	2	3	4	5
12.	Your child learns <i>new motor tasks</i> (e.g., swimming, rollerblading) easily and does not require more practice or time than other children to achieve the same level of skill.				
	1	2	3	4	5
13.	Your child is <i>quick and competent</i> in tidying up, putting on shoes, tying shoes, dressing, etc.				
	1	2	3	4	5
14.	Your child would <i>never</i> be described as a " <i>bull in a china shop</i> " (that is, appears so clumsy that he or she might break fragile things in a small room).				
	1	2	3	4	5
15.	Your child does <i>not fatigue easily</i> or appear to slouch and "fall out" of the chair if required to sit for long periods.				
	1	2	3	4	5

*Thank you.*

# Appendix E

## Baseline questionnaire

Subject number:

Circle your response to the following questions. If you make a mistake score through the original response and circle a second response.

**Compared to other children of your child's age:**

**How do you perceive your child's academic attainment in English (reading comprehension, writing skills etc.)**

Very poor    Below average    Average    Above average    Very good

**How do you perceive your child's attainment in mathematics**

Very poor    Below average    Average    Above average    Very good

**When playing games with other children (sports, board games, computer games), how competitive is your child?**

Not at all (shows little interest in winning)    Moderately    Very competitive (a 'sore loser')

**When playing computer games alone, how important are 'high scores' and beating AI (virtual) players to your child?**

NA (does not play)    Not at all    Moderately    Very competitive

**Does your child take part in extra-curricular sports (football, swimming etc)**

YES/NO

**How long does your child spend participating in physical activities outside of school, such as extra curricular sports, playing in the park/garden? (This should NOT include PE and break times at school.)**

□    Less than 2 hours a week    2-4 hours a week    5-7 hours a week    8+ hours a week

# Appendix F

## Familial risk questionnaire

Subject number:

All answers you give are strictly confidential.

Does your child have any of the following diagnoses (please tick any appropriate):

ADHD

Autism Spectrum disorder (ASD)

Asperger Syndrome

High Functioning Autism

Other autism diagnosis (please specify) \_\_\_\_\_

Dyspraxia

Speech dyspraxia

Developmental coordination Disorder (DCD)

Dyslexia

Dysgraphia

Dyscalculia

Cerebral Palsy

Other motor difficulty (please specify) \_\_\_\_\_

Do any members of your child's family have any of the above diagnoses? YES\*/NO

\*If you would be happy to give more information please state the diagnoses below (you are not required to give details of the relation of this person/people to your child).

□

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