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Subtypes in the autism spectrum: relating cognition to behaviour

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Thesis submitted for the degree of PhD in Psychology,

University of London, October 2006

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...those who hope in the LORD will renew their strength. They will soar on wings like eagles; they will run and not grow weary, they will walk and not be faint. Isaiah 40v31

Abstract

Three main theories have attempted to characterise autism at the cognitive level: theory of mind, executive function and central coherence; but none are able to account for all the behavioural manifestations seen across different children. This thesis is concerned with heterogeneity within the autism spectrum that might exist at the cognitive level and how this relates to behaviour. 57 high-functioning 7-12 year olds with autism spectrum disorder (ASD) and 28 normally-developing children participated in tasks tapping into these three cognitive domains, as well as intelligence and head size. Their parents completed interviews concerning their behavioural symptoms. Support for the relevance of all three theories to ASD was found, with significant group differences between the ASD and control groups. Approximately 50% of the children with ASD displayed a detectable theory of mind impairment, 33% executive dysfunction and 20% weak central coherence, and all possible combinations of impairment were found. A further puzzling 40% of children displayed no detectable impairment, indicating either a misdiagnosis, that the tests were not sensitive enough, or that there was an additional cause not investigated here. Theory of mind and executive function abilities were found to be closely related, whilst central coherence was independent of these skills, indicating that at least two cognitive subtypes were present in the current sample. The emerging hypothesis was that theory of mind impairment adversely affected performance in unstructured executive function tests through a lack of understanding of implicit task demands. Only theory of mind and verbal ability were found to predict specific aspects of the behavioural triad. In addition, the presence of weak central coherence was related to the increased head size found in 20% of ASD cases, providing a possible endophenotype for this cognitive skill.

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Chapter I: Cognitive theories of autism

Autism is a disorder that was first recognised as a clinical entity in the 1940s by two independent clinicians. Both Kanner (1943) and Asperger (1944) described groups of children who had various symptoms or behaviours in common; notably from Kanner ‘an autistic aloneness, a desire for sameness and islets of ability’, and from Asperger ‘difficulties with social integration, eye gaze and voice tone’. Since then, interest in autism has greatly increased, particularly over the last 25 years, resulting in a better knowledge of the core features (Wing & Gould, 1979), the course (Schonauer, Klar, Kehrer & Arolt, 2001) and the prevalence (Baird et al., 2006) of the disorder, as well as earlier detection (Baird et al., 2000) and standardised diagnostic tools (Lord et al., 2000). Autism is now widely acknowledged to be a neurodevelopmental disorder with a genetic basis (Rutter, 2000), resulting in atypical development of the brain (Bauman & Kemper, 1985). Despite this assertion, very little is known about the exact genetic or biological abnormalities underlying the disorder and so diagnosis still relies upon a defined set of behavioural criteria. These are often referred to as the typical triad of impairments: problems with social interaction; problems with communication; and restricted, repetitive and stereotyped patterns of behaviour, interests and activities (DSM-IV, American Psychiatric Association, 2000; ICD-10, World Health Organisation, 1993).

While the stereotypical image of an individual with autism is of a child lost in their own world, unaware of even the presence of others and strongly reactive against any changes perceived to affect their world, higher-functioning individuals, who are now known to make up a sizeable proportion of the autistic population (45% with IQs greater than 70; Baird et al., 2000), reveal that social behaviours are not absent, but rather abnormal. Poor comprehension and poor use of facial expressions, gestures and vocal prosody are a common giveaway (Tantam, Holmes & Cordess, 1993), and language and conversation,

rather than absent, is commonly repetitive and egocentric (Tager-Flusberg, 1999), one-sided and non-reciprocal (Fine, Bartolucci, Szatmari & Ginsberg, 1994). Whilst the defining social and communication impairments appear to overlap, this may simply be a result of the way in which they are defined (Tager-Flusberg, 1999), with some behaviours such as social aspects of communication being used to define both areas. Similarly, repetitive behaviours can be expressed in a number of different ways, including basic behaviours such as motor mannerisms and sensory preoccupations, but more complex behaviours such as circumscribed interests and repetitive language tend to be more commonly seen in higher-functioning individuals (Bodfish, Symons, Parker & Lewis, 2000).

Although necessary, a behavioural diagnosis has the disadvantage of heterogeneity; there can be many different reasons for or causes of the same behaviour. Equally, different behaviours in different individuals can result from the same underlying cause due to interaction with other factors, such as general ability, education, temperament and family situation (Rutter, 2000). Indeed, parents and clinicians often comment that every autistic child is different and has their own individual needs, yet seem happy to acknowledge that autism is a single unified disorder (Jackson, 2003). In order to accommodate such behavioural heterogeneity, the notion of an autism spectrum has been introduced (Wing & Gould, 1979), varying in multiple directions including severity of symptoms and the pattern of symptoms present at different developmental stages and general ability levels. (For ease, the term 'autism' is used to refer to the whole autism spectrum for the remainder of this chapter, unless reference to specific diagnoses are of importance.)

Such heterogeneity at the behavioural level of course makes the identification of the biological or genetic underpinnings of autism extremely difficult (Folstein & Rosen-Sheidley, 2001). At the biological level, an independent marker of autism is lacking, leaving diagnosis at the mercy of unrefined behavioural measures. An intermediary level of explanation between biology and behaviour is therefore helpful, describing the functions of

the brain in terms of the cognitive systems they may mediate. This can be termed the cognitive level of description and allows a range of behaviours to be linked to a unitary cause. A number of theories have attempted to characterise the key behavioural symptoms of autism at the cognitive level; most notably the mentalising theory, the executive function theory and the central coherence theory. This thesis will be concerned with these three theories only; whilst other theories do exist at the cognitive level, these either have not stood the test of time or attempt to explain only very specific behaviours. These three key theories equally do not attempt to explain all aspects of autistic behaviour but do attempt to draw a variety of different behaviours together through a common cognitive cause. The remainder of this chapter will therefore evaluate these three theories briefly; more detailed discussion of particular paradigms tapping into these cognitive domains can be found in the experimental chapters 3-5.

1.1 Mentalising

A widely tested and prominent cognitive theory of autism is the mentalising or 'Theory of Mind' (ToM) theory (Baron-Cohen, 1995; Frith, 2003b; Leslie & Frith, 1990). Individuals with autism are thought to show delay in or lack the ability to represent other people's mental states (such as desire, knowledge and belief) as distinct from their own, an ability which allows us to intuitively understand and predict other's behaviour on the basis of their thoughts. In normal development, the explicit ability to represent mental states appears to emerge at a distinct point in time between 3 to 5 years of age (Wimmer & Perner, 1983). The universality of this achievement across different cultures is impressive (Callaghan et al., 2005). However, there is consensus that this achievement is preceded by the implicit ability to take account of others' mental states (Astington, 1993; Flavell, 1999) although a number of developmental milestones in the understanding of others' minds are typically seen. Thus from 9 months of age, infants are able to implicitly attribute goals and intentions to actions (Csibra, 2003). At about the same time, infants engage in joint attention, sharing their

attention both with another person and an object in the environment, indicative of a desire to communicate and share their own visual experience (Bakeman & Adamson, 1984).

Children as young as 18 months are able to perform and therefore to understand the concept of pretend play, where reality is suspended and mental representations of both reality and pretence are necessary (Leslie, 1987). An understanding of other people's desires and emotions as different from one's own is displayed explicitly by about 3 years and children of this age can talk about these concepts in mental state terms (Wellman & Liu, 2004). By 4 years of age children explicitly display an understanding of more complex terms, such as belief, and particularly belief that goes against reality, commonly known as false belief; by this age, children appear to understand that people's minds are not copies of reality but are representations that can change and may be false (Wimmer & Perner, 1983).

It is not clear at this stage whether one or more underlying cognitive components come on line for this gradual development to proceed. Leslie (1987) suggested a cognitive component known as an expression raiser to be necessary, though not sufficient, for the development of a ToM. This expression raiser allows multiple mental representations to exist at the same time, each representing a different mental view of the world, without being in competition with each other. Whilst normally developing children possess this component, they may not display behaviour consistent with its use at an early age, due to other factors. For example, a child must be able to form such representations at all before they can be dissociated, must have a certain level of experience of how humans interact with the environment, must be able to inhibit their own representation of reality or reality itself in order to acknowledge another's mental state distinct from this, and, at least for false belief tasks, must have the verbal ability to understand and express such mental state terms in language. Other theoretical accounts of ToM also exist, including the 'theory' theory (Perner, 1991; Wellman & Gelman, 1998) and simulation theory (Harris, 1992), although the debate surrounding these different theories is outside the scope of this thesis.

1.1.1 Mentalising in autism

The mentalising theory proposes that in autism either an implicit ToM never emerges as the necessary expression raiser is absent or faulty (Leslie & Frith, 1990) or it appears only after a gross delay (Baron-Cohen, 1989b, 1991). Thus in its strongest form, children with autism should not be able to represent other people's minds as representations distinct from both reality and their own mental states. While this theoretical stance is only a hypothesis, its validity can be tested as the cognitive phenotype can be defined in terms of behavioural predictions independent of an autism diagnosis, thus avoiding circularity. Support for the theory therefore came initially from behavioural experiments using false belief tasks (Baron-Cohen, Leslie & Frith, 1985) on which individuals with autism were predicted to and did perform more poorly than comparison groups matched for age, mental age or language ability, despite their mental age being above 4 years. On the other hand, children with autism had no problem solving false photograph (Leekam & Perner, 1991) or false drawing tasks (Charman & Baron-Cohen, 1992), indicating that their problem lay in the realm of mental rather than pictorial representation.

This robust result has been reproduced by different researchers over numerous paradigms and mental states, providing converging evidence of a ToM impairment in autism. For example, Perner, Frith, Leslie & Leekham (1989) found children with autism to perform poorly on a seeing-leads-to-knowing task; Baron-Cohen (1989a; 1992) noticed a failure to distinguish mental and physical entities, an unawareness of the mental function of the brain, a lack of understanding of the appearance-reality distinction, and problems with deception through a simple penny hiding game in children with autism; and Tager-Flusberg & Sullivan (1994) showed that children with autism were worse at giving mental state explanations for actions. Similarly, whilst children with autism are capable of labelling emotions and explaining actions, they find it much more difficult to identify the causes of internal states (Capps, Losh & Thurber, 2000). Furthermore, some of the earlier

manifestations of mental state understanding, such as joint attention and pretend play, are absent or deviant in autism (Charman, 1998; Wing & Gould, 1979).

1.1.2 Brain basis of mentalising in autism

More recently, functional neuroimaging studies have begun to define a network of brain regions involved in mental state attribution and reasoning with remarkable consistency, including medial prefrontal cortex, superior temporal sulcus & the temporal pole (Baron-Cohen et al., 1999b; Berthoz, Armony, Blair & Dolan, 2002; Brunet, Sarfati, Hardy-Bayle & Decety, 2000; Castelli, Happé, Frith & Frith, 2000; den Ouden, Frith, Frith & Blakemore, 2005; Fletcher et al., 1995; Gallagher & Frith, 2004; Gallagher et al., 2000; Gallagher, Jack, Roepstorff & Frith, 2002; German, Niehaus, Roarty, Giesbrecht & Miller, 2004; Goel, Grafman, Sadato & Hallett, 1995; Mason, Banfield & Macrae, 2004; McCabe, Houser, Ryan, Smith & Trouard, 2001; Rilling, Sanfey, Aronson, Nystrom & Cohen, 2004; Spiers & Maguire, 2006; Takahashi et al., 2004; Vogeley et al., 2001). Activity in these regions has been shown to be reduced in individuals with autism whilst performing mentalising tasks (Castelli, Frith, Happé & Frith, 2002; Happé et al., 1996; Nieminen-von Wendt et al., 2002), as has connectivity between visual areas and this network (Castelli et al., 2002). Structural imaging also supports these findings, with increased grey matter volume localised to this network of brain regions in autism (Abell et al., 1999; Waiter et al., 2004). Two recent experiments have measured blood flow in the brain as an indicator of cortical activity and have found it to be reduced in autism in those brain regions associated with the ToM network. The first study indicated reduced blood flow in the superior temporal sulcus (Gendry Meresse et al., 2005) and the second showed reduced blood flow in medial prefrontal cortex, anterior cingulate gyrus and right medial temporal lobe (Ohnishi et al., 2000).

Several studies involving patients whose lesions lie in brain regions involved in this mentalising network have been studied. A number of authors have found that patients with frontal lesions show impairments on ToM tasks (Happé, Malhi & Checkley, 2001; Rowe, Bullock, Polkey & Morris, 2001; Stuss, Gallup & Alexander, 2001) and similar findings have emerged for patients with damage to the temporoparietal junction (Apperly, Samson, Chiavarino & Humphreys, 2004; Samson, Apperly, Chiavarino & Humphreys, 2004). In another study, those with orbitofrontal but not dorsolateral prefrontal cortex lesions showed similar patterns of performance on mentalising tasks to adults with Asperger Syndrome (Stone, Baron-Cohen & Knight, 1998). However, one study involving a patient with medial frontal lobe damage found no evidence of mentalising problems despite extensive testing (Bird, Castelli, Malik, Frith & Husain, 2004). In general, these studies seem to suggest that damage to either medial prefrontal cortex or the superior temporal sulcus may be sufficient but not necessary to cause ToM impairment.

1.1.3 Broader phenotype

A new direction that has helped to inform the search for the cognitive basis of autism has been the study of the broader phenotype: the presence of similar cognitive patterns of performance in unaffected relatives of individuals on the autism spectrum, indicative of a genetic basis for the cognitive impairments seen. Ozonoff, Rogers, Farnham & Pennington (1993) were the first to attempt this kind of study in siblings of children with autism and found no differences in performance on ToM tasks; however, power analyses indicated the measures used were not sensitive enough to detect group differences. More recently, Dorris, Espie, Knott & Salt (2004) have found siblings of children with Asperger Syndrome to have mild mentalising problems on the reputedly more sensitive Eyes test (Baron-Cohen, Jolliffe, Mortimore & Robertson, 1997b). Using this same test, Baron-Cohen et al. (2006) found mothers and fathers of children with autism to show decreased activity in the network of brain areas associated with mentalising whilst showing good

behavioural performance on the task. Shaked, Gamliel & Yirmiya (2006) found no difference between the younger siblings of children with and without autism on ToM measures however but this may simply reflect poor sensitivity given the findings of Baron-Cohen et al. (2006).

1.1.4 Relation to symptoms

The mentalising theory of autism has the advantage of being able to explain plausibly many of the social and communication problems seen in individuals with autism spectrum disorders and therefore a large range of symptoms. If ToM provides a basis on which to understand others thoughts, intentions and behaviour, a ToM impairment would make it difficult to interact in a flexible and responsive way to people and to communicate reciprocally. Indeed, the first study to compare ToM task performance to everyday behaviour in individuals with autism found those who passed false belief tasks to show more insightful social behaviour and better verbal and communication skills than those who failed, although even the passers were behaving below the level expected for their age and ability (Frith, Happé & Siddons, 1994; see also Hughes, Leboyer & Bouvard, 1997a). Capps, Kehres & Sigman (1998) also found that false belief task performance in children with autism was related to the ability to contribute novel information to a conversation and Hale & Tager-Flusberg (2005) found a similar relationship between performance on a battery of ToM tasks and the ability to maintain the topic of conversation initiated by their conversational partner and therefore to make relevant utterances. Ability on ToM tests in children with autism has also been linked to the understanding of non-literal language (Martin & McDonald, 2004) and the ability to understand other people's embarrassment (Hillier & Allinson, 2002).

Surprisingly, it has also been suggested that the ToM impairment could provide an explanation for some of the routine and ritualistic behaviours that are one of the diagnostic

criteria, as a secondary consequence (Baron-Cohen, 1989c); if a ToM impairment results in social impairment, this in turn may produce distress in social interactions due to difficulties in understanding others' intentions and predicting why they act in a particular manner, hence producing predictable but routine & ritualistic behaviours in compensation. These sorts of arguments may suffer from circularity though, making it difficult to establish external validity. In any case, Joseph & Tager-Flusberg (2004) recently found that ToM deficits were linked only to communication difficulties in autism, but not to the social or repetitive behaviours seen and Turner (1996) found the degree of repetitive behaviour present in autism to be unrelated to mentalising ability or IQ.

The two experiments reporting reduced blood flow in autism in brain regions associated with the ToM network (see section 1.1.3), also related this measure to certain symptoms, making a link from brain through cognition to behaviour. Reduced blood flow in the superior temporal sulcus was found to be correlated with the overall extent of symptoms (Gendry Meresse et al., 2005) as measured by a standard parental interview (ADI-R; Lord, Rutter & Le Couteur, 1994), while reduced blood flow in medial prefrontal cortex & anterior cingulate gyrus was associated with the level of social and communication impairments, and reduced blood flow in right medial temporal lobe was associated with the degree of desire for sameness (Ohnishi et al., 2000) as measured by a short parental interview (CARS; Schopler, Reichler, DeVellis & Daly, 1980).

1.1.5 Negative findings

Despite the mentalising theory being able to account for much of the variety of social symptoms, heterogeneity can still be observed at the cognitive level. The mentalising impairment is generally observed to be less severe in children with Asperger syndrome and Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS) than in children with autism (Ozonoff, Rogers & Pennington, 1991b; Sicotte & Stemberger, 1999; Ziatas,

Durkin & Pratt, 1998, 2003), although some researchers do not find such differences (Buitelaar, van der Wees, Swaab-Barneveld & van der Gaag, 1999; Dahlgren & Trillingsgaard, 1996), and in some cases of adults with Asperger Syndrome none or very little impairment may be found (Bowler, 1992; Hill, Sally & Frith, 2004). Indeed, even in the first study supporting the mentalising theory with low-functioning children (Baron-Cohen et al., 1985), 20% of the autistic children passed the false belief task. Furthermore, a small number of negative findings exist which are difficult to reconcile. Russell & Hill (2001) found no evidence of impairment in reporting own intentions that differed from the outcome of a task in autistic children, and Yirmiya & Shulman (1996) found no differences between children with autism and those with mental retardation on a standard false belief task. A minority of autistic individuals also show evidence of mentalising even in everyday life (Frith et al., 1994) and those with Asperger Syndrome are thought to have better social insight compared to those with high-functioning autism (Ozonoff et al., 1991b).

Negative findings are difficult to interpret as they could result for a number of different reasons: the tasks may be insensitive to the underlying impairment, the clinical group may be heterogeneous, improvement and compensation may occur, or the theory may simply be wrong. It is important to remember that in many of these experiments, mentalising ability is measured through a simple task that will also depend on a range of other abilities and which may possibly be passed in a number of different ways. Task difficulty and sensitivity are therefore important factors when interpreting such results, particularly when floor and ceiling effects are involved. Indeed, ToM task performance does undergo developmental change over time in people with autism (Steele, Joseph & Tager-Flusberg, 2003) but, though less severe, mentalising impairments have still been documented in high-functioning individuals including adults when performing more complex tests, for example, advanced mentalising stories (Happé, 1994; Jolliffe & Baron-Cohen, 1999a; Kaland et al., 2002), judging mental state from the eyes or from the voice (Baron-Cohen et al., 1997b; Golan & Baron-Cohen, 2006; Kleinman, Marciano & Ault, 2001; Rutherford, Baron-

Cohen & Wheelwright, 2002), understanding *faux-pas* (Baron-Cohen, O'Riordan, Stone, Jones & Plaisted, 1999a), video-clips of situations that are close to real life encounters (Channon, Charman, Heap, Crawford & Rios, 2001; Heavey, Phillips, Baron-Cohen & Rutter, 2000) and silent animations (Abell, Happé & Frith, 2000; Castelli et al., 2002; Klin, 2000).

1.1.6 Improvement and compensation

One important observation indicating that factors other than mental state understanding may play a role in ToM task performance is the relationship between verbal ability and mentalising ability (Happé, 1995; Yirmiya, Erel, Shaked & Solomonica-Levi, 1998).

Children with autism were found to have a 80% probability of passing the standard false belief task when they reached a verbal mental age of more than 13 years, more than 7 years later than the normally developing child. This indicates that although their ToM impairment delays the age at which they can pass this task, at a certain level of verbal skill they are capable of producing the correct solution. Fisher, Happé & Dunn (2005) also recently showed that the relationship between ToM and verbal ability was stronger in children with autism than those with moderate learning difficulties or normal development, indicating a greater reliance on verbal ability to perform these tasks.

Exactly what this apparent improvement with age and verbal ability means is still unresolved; it may be that verbal skill causes this insight into ToM understanding (Astington & Jenkins, 1999) or that a third factor causes both verbal ability to increase and social insight to develop. Alternatively, improved ToM task performance may not be indicative of the development or acquisition of an implicit ToM; rather it may indicate some, possibly verbal, method of compensation (Happé, 1995; Tager-Flusberg, 2000) or a conscious method of 'hacking out' the solution (Frith, Morton & Leslie, 1991). Indeed, it has been suggested that ToM development is both delayed and deviant in autism (Baron-

Cohen, 1989b, 1991) and there is some evidence for the existence of a qualitatively different pattern of ToM development (Serra, Loth, van Geert, Hurkens & Minderaa, 2002). For example, children with autism are more likely to use desire terms in conversation than children with Specific Language Impairment (SLI) or controls, who are more likely to use thought and belief terms (Ziatas et al., 2003). Similarly, children with autism are as likely as children with Downs Syndrome to talk about desire, perception & emotion but are less likely to use mental states and to ask for another's attention (Tager-Flusberg, 1992) and have a better understanding of intentions than beliefs (Carpenter, Pennington & Rogers, 2001).

Still further suggestions for the relationship between verbal ability and language have been made: poor mentalising skill may actually limit language learning, as this may require the child to understand a speaker's intentions (Bloom, 2000). Then again, the relationship may be spurious and a product of the task design, as performance on a non-verbal mentalising task (the Eyes test, Baron-Cohen et al., 1997b) was found to be unrelated to language ability in autism, indicating that the relationship may be a result of the language component of the majority of ToM tasks (Senju, Tojo, Konno, Dairoku & Hasegawa, 2002).

A number of task manipulations have been shown to aid children in their performance on false belief tasks. Wellman et al. (2002) used thought bubbles as a physical representation of the mind and found this helped autistic children to pass false belief tasks. Swettenham (1996) taught children with autism to understand the mind as a camera and that, as photos can be out of date, so can the mind. He found that, similar to Wellman et al., the children could use this analogy to successfully predict the subsequent behaviour of agents; however, they still could not predict the agents' mental states accompanying the behaviour.

Unfortunately, although the belief term use of children with autism develops in line with their ToM task performance (Ziatas et al., 1998) and explicit teaching about mental states improves false belief task performance, it does not appear to affect social competence in

the same way (Ozonoff & Miller, 1995). Similarly, the translation of learnt social knowledge into novel environments and real life situations appears to be extremely poor (Parsons & Mitchell, 2002). It seems therefore that improvement may occur in autism in either the domain of social competence or the domain of mental states through explicit teaching but that this knowledge is not transferable either between or within domains, lending support to the idea that improvement may indeed be through compensation and that the skills acquired are not the same as the skills of normally developing children, who have an implicit understanding of mental states as well as an explicit one (Frith, 2004).

1.1.7 Heterogeneity

For the most part, therefore, research suggests that ToM deficits are wide ranging in autism, particularly in children. However, there is some recent evidence that ToM problems are not present in all children with autism even when using a battery of tasks (Pellicano, Maybery, Durkin & Maley, 2006; 68% of children performed more than 1 SD below the control mean). Indeed, Murphy (2006) found highly variable performance within a group of autistic adults, despite significant differences between this group and other clinical patients. The issue still remains therefore of whether these individuals who pass even advanced mentalising tasks truly have a mentalising problem. If mentalising is really intact in these cases, it would be extremely important to explain their social impairments in everyday life by some other concept.

1.1.8 Other explanations of social failure in autism

While often seen as part and parcel of mentalising ability, poor joint attention (Sigman & Mundy, 1989), lack of imitation & pretend play (Meltzoff & Gopnik, 1993; Rogers & Pennington, 1991) and poor affective contact (Hobson, 1990) have all been proposed as possible causes of autism themselves with mentalising failure as a secondary consequence. Whilst it is undeniable that these more basic problems exist in individuals with autism, it

can also be argued that they result from a ToM deficit. Furthermore, while many of these abilities appear in normal development prior to explicit mental state understanding, it has already been argued that signs of an implicit ToM appear at a much earlier age (see introduction to section 1.1). The most notable of these more basic problems is the striking lack of pretend play (Wing & Gould, 1979) which would normally be seen in early childhood; pretence involves the decoupling of one's own representation of reality from the make-believe representation (Leslie, 1987), exactly the problem proposed by the mentalising theory. Similarly, joint attention is known to be impaired in young children with autism (Charman, 1998); this also requires mentalising skill as the child must understand another's direction of eye gaze to be meaningful and therefore to consider their thoughts or intentions in order to look to the same place to gain the same information (Baron-Cohen, 1989d). The imitation deficits seen in autism (Dawson, Meltzoff, Osterling & Rinaldi, 1998) may occur due to a lack of understanding of the other person's intentions and a poor representation of that person in relation to themselves.

1.1.9 Specificity of mentalising impairment to autism

In a meta-analysis of ToM performance across children with autism, those with mental retardation and normally developing children, Yirmiya et al. (1998) found no differences between the former two groups of children and thus suggest that autism does not differ from certain other disorders involving mental retardation when careful group matching is employed. A number of studies have also indicated that children with specific language impairment (SLI) may perform poorly on false belief tasks compared to IQ-matched controls (Bishop, 1997; Farmer, 2000; Shields, Varley, Broks & Simpson, 1996), and it has been suggested that mentalising ability may rely on language development and communicative competence. However, one study has indicated that in everyday life children with SLI use belief and thought terms in speech in the same way as normally developing children do, whilst those with autism are more likely to refer to desires (Ziatas

et al., 2003), indicating that their poor performance on the false belief task may not result from mentalising failure. Additionally, another recent study found good false belief task performance in children with SLI when the linguistic demands of the task were reduced (Miller, 2004), supporting the idea that these children fail such tasks due to their linguistic complexity rather than their mentalising demands.

Furthermore, ToM task performance has been found to be impaired in children who are congenitally deaf (Peterson & Siegal, 1995) or blind (Minter, Hobson & Bishop, 1998). However, the ToM development of deaf children has also been shown to be delayed but not deviant in its progression (Peterson, Wellman & Liu, 2005) and deaf children with deaf parents who therefore have access to fluent sign-language from birth have normal ToM task performance (Courtin, 2000). It seems likely that both language learning and experience are necessary for good ToM task performance, as well as an implicit ToM, and it is these former factors that are deficient in deaf and blind children and the latter in children with autism.

There does however appear to be robust evidence of ToM deficits in schizophrenia (Frith, 1992), with a number of studies reporting impaired performance (Harrington, Langdon, Siegert & McClure, 2005; Langdon, Coltheart & Ward, 2006; Mazza, De Risio, Surian, Roncone & Casacchia, 2001; Murphy, 2006) even below own control task performance. It appears that only a subpopulation of individuals with this diagnosis have such difficulties however, namely those with negative or disorganised symptoms, and that the impairment in these patients is not as severe as in autism (Pickup & Frith, 2001). As in autism, mentalising ability seems to predict social functioning (Roncone et al., 2002). However, the differences between autism and schizophrenia in terms of onset of the disorder may be key in differentiating them and retaining their specificity.

1.2 Executive function

The second major cognitive theory of autism, executive function, was initially suggested due to similarities in behaviour that were noted between patients with acquired frontal lobe damage and individuals with autism, particularly repetitive and socially inappropriate behaviour (Damasio & Maurer, 1978; Ozonoff et al., 1991b; Prior & Hoffmann, 1990; Rumsey, 1985). Frontal lobe patients commonly have problems with tasks tapping executive functions, including planning, flexibility, inhibition, working memory, generativity and self-monitoring, all thought to be involved in maintaining a problem-solving set for attainment of a future goal (Duncan, 1986).

1.2.1 Executive function in autism

A number of experiments have now reported similar deficits in autistic populations across the different executive functions. Problems with planning have been seen on the Tower of Hanoi task (or its variants the Tower of London, the Nepsy Tower and the Stockings of Cambridge), revealed in less efficient strategy use particularly for more complex problems (Bennetto, Pennington & Rogers, 1996; Hughes, Russell & Robbins, 1994; Joseph, McGrath & Tager-Flusberg, 2005a; Landa & Goldberg, 2005; Ozonoff et al., 2004; Ozonoff & Jensen, 1999; Ozonoff, Pennington & Rogers, 1991a), as well as on Milner mazes (Prior & Hoffmann, 1990) and the Luria bar task (Hughes, 1996). Ozonoff et al. (2004) have recently found this to be the case across a wide range of ages and ability levels.

The most commonly used test of mental flexibility is the Wisconsin Card Sorting Task (WCST). Individuals with autism typically have problems with unprompted shifting between sets, tending to perseverate with the previously used strategy despite feedback after each trial (Bennetto et al., 1996; Ozonoff & McEvoy, 1994; Ozonoff et al., 1991a; Prior & Hoffmann, 1990; Rumsey, 1985; Shu, Lung, Tien & Chen, 2001; Steel, Gorman & Flexman, 1984; Szatmari, Tuff, Finlayson & Bartolucci, 1990). The Trail-making test, in

which individuals must shift flexibly between consecutive numbers and letters, has also revealed flexibility impairments in autism (Rumsey & Hamburger, 1988), as has the Intra-/Extra-Dimensional Shift Task (ID/ED); in this task, problems were limited to shifting between sets (ED) rather than general response perseveration (Hughes et al., 1994; Ozonoff et al., 2004). Again, similar to the planning domain, it seems that these problems occur at all ages and ability levels (Ozonoff et al., 2004). Courchesne et al. (1994) have suggested that such problems with mental flexibility may reflect a difficulty in switching attention between stimuli or tasks. This is also supported by evidence that individuals with autism are slower to orient to peripheral stimuli (Wainwright & Bryson, 1996) and slower at responding to cues compared to controls (Townsend, Harris & Courchesne, 1996; Wainwright-Sharp & Bryson, 1993).

Tests of inhibition and working memory have less often been used with individuals with autism. A few inhibition tasks have highlighted problems in children with autism; on the Windows task requiring the participant to point away from a reward in order to receive it (Hughes & Russell, 1993; Russell, Hala & Hill, 2003; Russell, Mauthner, Sharpe & Tidswell, 1991), on a Go/No-Go task requiring inhibition of a prepotent response (Ozonoff, Strayer, McMahon & Filloux, 1994), on the similar Knock-Tap task requiring the child to knock in response to the experimenter's tap and vice versa (Joseph et al., 2005a) and on antisaccade and delayed saccade tasks requiring the individual to either look in the opposite direction from a peripheral stimulus or to look at the stimulus after its offset (Luna, Doll, Hegedus, Minshew & Sweeney, 2006). Indeed, Luna et al. found this to be true over a range of ages, but for development to occur at the same rate as in normal development despite this delay. Working memory problems have been located in both adults and children with autism through spatial working memory tasks, such as the block span task and the CANTAB spatial working memory task (Bennetto et al., 1996; Goldberg et al., 2005; Joseph et al., 2005a; Landa & Goldberg, 2005; Morris et al., 1999). Verbal working memory deficits have also been observed (Digit Span backwards: Kenworthy et al., 2005).

Generativity is a further area of executive function that has been investigated in autism. This is normally assessed through fluency tasks, in which the participant must produce responses according to a particular category given to them. This has been found with verbal fluency tasks (Boucher, 1988; Kleinmans, Akshoomoff & Delis, 2005; Minshew, Goldstein, Muenz & Payton, 1992; Rumsey & Hamburger, 1988; Turner, 1999), as well as in ideational fluency (Turner, 1999) and design fluency (Lewis & Boucher, 1995). Furthermore, the strategies used to produce responses tend to be more repetitive; Williams, Moss, Bradshaw & Rinehart (2002) found that individuals with autism were more likely to repeat the previous digit than controls in a random number generation task and Rinehart, Bradshaw, Moss, Brereton & Tonge (2006) observed repetitive strategies such as alternating between two responses.

1.2.2 Brain basis of executive function in autism

As already mentioned in section 1.1.2, abnormalities in prefrontal cortex are known to exist in autism and executive dysfunction is naturally associated with frontal lobe dysfunction in patients with acquired brain lesions. Whether any of these prefrontal abnormalities are associated with executive functions has recently begun to be investigated through functional imaging studies. Muller, Pierce, Ambrose, Allen & Courchesne (2001) required their adults to plan and perform a simple finger movement task and found identical but increased activation in the autism group compared to controls in areas of prefrontal cortex. More recently, Schmitz et al. (2006) also found increased activity in areas of the parietal & frontal lobes when adults with autism performed tasks of inhibition (Go/No-Go, Stroop) and flexibility. These authors suggested that this increase in activity may be due to compensation as no behavioural differences were observed between the groups. One further study has shown reduced activation in dorsolateral prefrontal cortex and the posterior cingulate in adults with autism whilst performing a spatial working memory task (Luna et al., 2002). Furthermore, Just, Cherkassky, Keller, Kana & Minshew (2006)

compared brain activity on a planning task (Tower of London) and, whilst finding very similar brain activity in adults with autism and controls, found lower synchronization between frontal and parietal areas in autism, indicative of underconnectivity and therefore a lack of integration of information. However, despite these positive results, it is still a matter of debate whether executive dysfunction and prefrontal abnormalities are primary in autism, are the result of abnormalities in other related brain areas or are purely comorbid due to proximity of brain areas responsible for executive functions and for other functions primary to autism.

1.2.3 Broader phenotype

Evidence for the existence of the broader phenotype in autism in the domain of executive function comes from a number of studies looking at different aspects of executive dysfunction. Ozonoff et al. (1993) were the first to study the siblings of children with autism, finding impaired performance compared to the siblings of learning disabled children on a planning task (Tower of Hanoi) and a flexibility task (WCST; borderline significance). Similarly, Hughes et al. (1997a; 1999) tested parents and siblings of children with autism and found poor planning (Tower of London), flexibility (ID/ED) and generativity (verbal fluency; siblings only) but good or even superior working memory (verbal and spatial memory span) and Piven & Palmer (1997) showed that two parents of autistic children performed worse than the parents of children with Downs Syndrome on both performance IQ tests, thought to tap into executive dysfunction, and a planning task (Tower of Hanoi). However, more recently Bolte & Poustka (2006) found no differences between parents of children with autism, early onset schizophrenia or learning difficulties on tests of planning (Tower of Hanoi and Trail-making) or flexibility (WCST) but this may have been due to issues of task sensitivity.

1.2.4 Relation to mentalising

Russell (1997) argues that an inability to inhibit prepotent responses may not only be able to explain the pattern of executive results but also those supporting the mentalising theory. Specifically, an inability to stop oneself from responding to the prepotent stimulus could explain the failure of many autistic children on false-belief tasks. More generally, this would suggest that individuals with autism are unable to think about others' mental states as their own mental state is too potent. Although the false photograph task (Charman & Baron-Cohen, 1992; Leekam & Perner, 1991) was designed to rule out exactly this problem, Russell, Saltmarsh & Hill (1999b) claim that a novel version of this task is just as difficult for children with autism as the false belief task, indicating a general inability to inhibit prepotent responses.

From this hypothesis, correlations between performance on mentalising and prepotent inhibition tests would be expected, or at least co-occurrence of deficits. Very few studies have measured abilities on both types of task in the same individuals with autism and those that do, do not always report such relationships (eg. Pellicano et al., 2006). The first study to compare these abilities examined the proportion of individuals passing both mentalising and executive function tasks and found fewer children to be impaired on mentalising than executive function tasks (Ozonoff et al., 1991a), possibly indicative of primacy of executive function in autism. However, this may say more about the levels of task difficulty than about underlying impairments. In addition though, these abilities were also correlated in Ozonoff et al.'s study. More recently, Joseph & Tager-Flusberg (2004) found that ToM performance only related to performance on the Knock-Tap test of prepotent inhibition, not to other executive measures, after accounting for general ability and language level.

However, two problems also immediately arise with this hypothesis. Firstly, individuals with autism have difficulty with some mentalising tasks that do not involve prepotent responses, for example, the Eyes Task (Baron-Cohen et al., 1997b) and silent geometric

animations (Abell et al., 2000; Castelli et al., 2002; Klin, 2000). Secondly, executive function may not be the limiting factor in mentalising task performance in children with autism. From the literature on normal development, there is some indication that both mentalising skills and the ability to inhibit prepotent responses are necessary to pass false belief tasks (Roth & Leslie, 1998). These authors also suggest that the pattern of performance in 3 year olds and children with autism on false belief tasks is different; whilst 3 year olds may fail due to poor executive control needed to allow false beliefs to conceptually stand out, they suggest that autistic children may fail due to an impairment in the mentalising mechanism.

1.2.5 Relation to symptoms

The executive function theory attempts primarily to account for many of the non-social features of autism, particularly the obsessive interests, rituals, insistence on sameness, and repetitive & stereotyped thoughts, words & behaviour, although some accounts go so far as to explain the social features as well (eg. Russell, 1997). However, many of these behaviours are also found in individuals with other developmental disorders and more generally in learning disability, so they may not be specific to autism or a defining feature (Hagerman, 1999; Mazzocco, Baumgardner, Freund & Reiss, 1998). This opens the question of specificity, hence only those studies accounting for IQ will be considered in this section and other developmental disorders will be addressed in section 1.2.8.

Furthermore, there is evidence that repetitive behaviours may emerge after other autistic features (Cox et al., 1999; Stone et al., 1999), possibly indicating that they are secondary features; alternatively, this late emergence may be due to brain maturation affects and genetic influences coming online later in development.

The first study to consider the relationship between executive function and the symptoms of autism found strong relationships between each of inhibition and generation with

specific classes of repetitive behaviours (Turner, 1996). More recently, a number of studies have also begun to investigate this relationship in other aspects of executive function and the behavioural triad. Verte, Guerts, Roeyers, Oosterlaan & Sergeant (2006) found that performance on tasks of working memory, planning and generativity by children with autism was related to the overall severity of symptoms seen. Liss et al. (2001) found that performance on a number of different tests of executive dysfunction in autism was related to the severity of social impairments and repetitive behaviours, although these relationships were no longer significant after accounting for verbal ability. More specifically, spatial working memory has been shown to be weakly related to the impaired social behaviours seen in autism (Landa & Goldberg, 2005) and perseveration to joint attention and social interaction skills, independent of verbal ability (although the correlation was calculated across both the autism and control groups; McEvoy, Rogers & Pennington, 1993). Furthermore, flexibility has been found to predict later social functioning (Berger, Aerts, van Spaendonck, Cools & Teunisse, 2003). Gilotty, Kenworthy, Sirian, Black & Wagner (2002) reported correlations between a parental report of executive problems and both social and communication abilities in children with autism and Happé, Booth, Charlton & Hughes (2006) report similar findings from tests of executive function. Bishop & Norbury (2005a) also found correlations between both verbal and non-verbal fluency and communication abilities in children with autism as well as children with pragmatic language impairment and specific language impairment. In the domain of restrictive and repetitive behaviours, Lopez, Lincoln, Ozonoff & Lai (2005) found relationships between the presence of these behaviours and flexibility, working memory and inhibition but not planning and generativity tasks.

However, Joseph & Tager-Flusberg (2004) found that executive functions could only explain significant variance in communication ability after accounting for language, but not in social or repetitive behaviours. Furthermore, Stahl & Pry (2005) found no relationship between flexibility and joint attention in young children with autism, Bishop & Norbury

(2005b) found no relationship between verbal and non-verbal inhibition and any aspect of autistic symptomatology, Ozonoff et al. (2004) found no relationship between either a planning task (stockings of Cambridge) or a flexibility task (ID/ED) and any aspect of autistic symptoms or adaptive behaviour, and Teunisse, Cools, van Spaendonck, Aerts & Berger (2001) found no relationship between executive dysfunction and symptomatology. Such mixed results are likely to reflect the range of measures used, both in the cognitive and behavioural domains, making it difficult to draw any firm conclusions.

1.2.6 Negative findings

The executive dysfunction theory of autism has proved more difficult to replicate from experiment to experiment than the mentalising theory and this lack of group differences across a number of studies should be taken especially seriously for a deficit theory as impaired performance is generally expected in a clinical sample. This is the case across all aspects of executive function, although more so for some than others, implying that some aspects of executive function may be impaired whilst others may not be. It is also common to find double dissociations within the literature, with some authors, for example, finding intact planning but impaired flexibility and vice versa. Again, it is important to remember that there can be a number of different explanations for negative results: task sensitivity, heterogeneity, improvement and compensation, or an incorrect theory.

Relatively few negative results appear to be present in the domains of planning and flexibility. Goldberg et al. (2005) found good planning (Stockings of Cambridge) and flexibility (ID/ED) performance compared to normally developing children and those with Attention Deficit Hyperactivity Disorder (ADHD). Flexibility was also intact in a number of studies showing good performance on the WCST (Minshew et al., 1992; Nyden, Gillberg, Hjelmquist & Heiman, 1999; Ozonoff & Jensen, 1999) and on the ID/ED task (Edgin & Pennington, 2005). Indeed, on the ID/ED task, Landa & Goldberg (2005)

found problems with the ID component of the task, where perseveration of the same rule despite non-critical stimuli changes is favoured, and enhanced ED ability, which is thought to reflect cognitive flexibility. Similarly, Kleinmans et al. (2005) found enhanced performance in adults and adolescents with autism on the Trail-making task.

In the areas of working memory and generativity, a similar proportion of negative findings exist. A number of studies have reported no problems with spatial working memory in autism compared to controls (Edgin & Pennington, 2005; Ozonoff & Strayer, 2001). Both Minschew, Goldstein & Siegal (1995) and Scott & Baron-Cohen (1996) found little evidence of generativity problems on verbal fluency tasks, as did Turner (1999) on a design fluency task, and Kleinmans et al. (2005) actually found superior performance on a design fluency task.

A much larger number of negative results exist in the domain of inhibition, which therefore may be relatively intact in autism. Intact performance has been found on the Stroop (Eskes, Bryson & McCormick, 1990; Goldberg et al., 2005; Ozonoff & Jensen, 1999) and one study even found superior performance on this task (Kleinmans et al., 2005), indicating a lack of the normal interference effect seen; this was not due to poor reading automaticity as reading ability was within the normal range. On a Go/No-Go task, Ozonoff et al. (1994) found intact inhibition of neutral responses; both Ozonoff & Strayer (1997) and Brian, Tipper, Weaver & Bryson (2003) have found normal levels of inhibition on a negative priming task requiring suppression of a response to irrelevant distractors; and Ozonoff & Strayer also reported normal performance on a stop-signal task requiring the inhibition of a motor response to neutral and prepotent stimuli. Russell, Jarrold & Hood (1999a) similarly found intact performance on a day-night task, where the child must first respond correctly to day and night pictures and later give the opposite response, and on the tubes task, where the child must predict where an object dropped down a bent tube will land (directly below the drop point or directly below the base of the bent tube).

Russell (1997) provides a possible explanation for the rather mixed inhibition findings. He suggests that individuals with autism have an inability only to inhibit prepotent responses, those with an existing affordance attached to them. Russell goes further in explaining why this specific problem occurs, suggesting that children with autism have problems following arbitrary rules, which is particularly the case when inhibiting a prepotent response. This begins to address some of the difficulties individuals with autism have with other executive tasks that require the child to perform tasks with arbitrary rules that would not normally occur in everyday life. This appears to explain some but not all of the discrepant results. However, more recently Biro & Russell (2001) have suggested that individuals with autism also have problems with self-monitoring through inner speech. A common strategy for solving executive tasks with arbitrary rules is to use verbal mediation or inner speech to verbally encode the rules in order to prompt and monitor oneself throughout the task. This strategy can only be used when a verbal response does not need to be made; when a verbal response is involved, interference between the response and inner speech makes this strategy counterproductive. Russell therefore suggests that problems will occur for individuals with autism only on non-verbal tasks when arbitrary rules are present.

In addition to those studies showing intact inhibition of neutral responses and impaired inhibition of prepotent responses, a couple of studies specifically provide support for Russell's hypothesis. Hughes' (1996) study using the Luria hand game found that inhibition of a prepotent response was impaired and was related to self-awareness and self-control of thoughts, as well as to verbal ability, indicating an inability to use language in the control of thoughts in this task. Joseph, Steele, Meyer & Tager-Flusberg (2005b) found that high-functioning children with autism had problems with visual working memory tasks only when verbal encoding was possible from meaningful as opposed to non-meaningful pictures. Furthermore, their verbal memory span was not impaired, indicating a problem with using verbal mediation to monitor goals rather than with remembering the stimuli. However, it was unclear from this study whether the children with autism were not

processing the stimuli as meaningful objects, rather than not verbally labelling them. In addition, a study by Kleinhans et al. (2005) revealed the opposite pattern with problems on verbal but not non-verbal tasks.

It seems that careful group matching is particularly critical for studies of executive function as IQ has a huge effect on executive function performance (Duncan, Burgess & Emslie, 1995); indeed this may go some way towards explaining some of the discrepant findings. In addition, it may be that executive problems are related to the presence of neuropathology rather than autism *per se* and so would be undetectable in lower functioning individuals with learning disabled comparison groups. Furthermore, a number of studies using very young children with autism have found a lack of group differences across a wide range of executive tests (Dawson et al., 2002; Griffith, Pennington, Wehner & Rogers, 1999; Rutherford & Rogers, 2003), suggesting either that these problems are secondary to autism and develop later or that all young children also show executive problems, albeit for different reasons. At the other end of the age range, Happé et al. (2006) recently found no differences in executive abilities between older high-functioning children with autism (13 years) and IQ-matched controls, whilst seeing stark differences in the younger but equally high-functioning sample (9 years), indicating improvement in function with age. Only a narrow age window may be available in which to locate executive dysfunction. This may indicate that executive control does not develop immediately in young normally-developing children and that it can be overcome or compensated for with age and ability. Alternatively, this may simply indicate that the tasks used are not sensitive to the underlying impairments at different ages due to ceiling or floor effects.

1.2.7 Heterogeneity

As will now be obvious, the literature on executive function deficits in autism is highly inconsistent (Hill, 2004b), despite its explanatory power particularly in terms of the non-

social symptoms. In such a case, there is merit in investigating individual results; these are rarely reported, making it hard to assess whether executive functions are impaired in a subgroup of the autistic population. A small number of studies do provide such information on individual performance however. Whereas Ozonoff et al. (1991a) reported executive dysfunction, as assessed by the WCST and Tower of Hanoi task, in 96% of their autism sample, few replications of this level of universality have since been reported. Pellicano et al. (2006) report executive impairment in at most 50% of individuals across a range of executive tasks, as do Kenworthy et al. (2005) on tasks of inhibition, flexibility, planning and working memory, Berger et al. (2003) on a range of flexibility tasks, Teunisse et al. (2001) on a flexibility task, Liss et al. (2001) on the WCST and Ozonoff & Jensen (1999) on the WCST and Tower of Hanoi task. Although this may say more about test sensitivity than universality, the estimate of less than 50% of the autism population having a detectable executive function deficit seems fairly robust across studies (Hill, 2004a).

1.2.8 Specificity of executive dysfunction to autism

Probably the most challenging problem for the executive function theory of autism is that of discriminant validity: whether the executive deficits that are seen are specific to autism or also seen in other disorders. In particular, many other developmental disorders have been linked to executive dysfunction, including but not limited to attention deficit disorders, phenylketonuria, Tourette's Syndrome, oppositional defiant disorder and conduct disorder, indicating that such specificity is not present (Diamond, Prevor, Callender & Druin, 1997; Sergeant, Geurts & Oosterlaan, 2002; Toupin, Dery, Pauze, Mercier & Fortin, 2000; Watkins et al., 2005). It is currently unknown whether different aspects of executive function can discriminate between these different disorders and therefore which aspect is impaired in autism, or whether all of these disorders have the same underlying problem that therefore cannot discriminate between disorders, explain disorder specific-behaviour or be causal to them.

An explosion of experiments comparing different clinical groups has occurred over the last five years as the merits of this methodology have come to light. Two studies have indicated planning problems in autism in comparison to ADHD (Geurts, Verte, Oosterlaan, Roeyers & Sergeant, 2004; Ozonoff & Jensen, 1999) and Tourette's Syndrome (Ozonoff & Jensen, 1999), a number have indicated flexibility problems in comparison to ADHD (Geurts et al., 2004; Gioia, Isquith, Kenworthy & Barton, 2002; Ozonoff & Jensen, 1999) and Tourette's Syndrome (Ozonoff & Jensen, 1999; Ozonoff et al., 1994) and inhibition of a prepotent response has also been suggested as a problem in comparison to Tourette's Syndrome in one study (Verte, Geurts, Roeyers, Oosterlaan & Sergeant, 2005). Generativity has also been identified as a problem in autism in comparison to both ADHD (Happé et al., 2006) and Tourette's Syndrome (Verte et al., 2005). Children with ADHD on the other hand tend to show problems with inhibition (Gioia et al., 2002; Happé et al., 2006; Ozonoff & Jensen, 1999). However, a handful of studies find no differences between children with autism and those with ADHD on tasks tapping planning (Booth, Charlton, Hughes & Happé, 2003; Goldberg et al., 2005), flexibility (Goldberg et al., 2005; Tsuchiya, Oki, Yahara & Fujieda, 2005), or inhibition and spatial working memory (Goldberg et al., 2005), or with those with Tourette's on a range of working memory tasks (Ozonoff & Strayer, 2001). Again, mixed results dominate the literature.

A recent interest in the role of language in performance of executive function tasks has utilised the same methodology with language-impaired groups in comparison to autism. Liss et al. (2001) compared children with autism to those with specific language impairment and found more perseverative responses on the WCST in children with autism but comparable performance on a planning task. However, these differences disappeared when differences between the groups in verbal ability levels were controlled. Similarly, Bishop & Norbury (2005b) compared children with autism to those with either specific or pragmatic language impairment on tasks of verbal and non-verbal inhibition and found

worse performance compared to controls across all three groups. These results indicate that language may play an important role in performance on executive function tasks.

1.3 Central coherence

The central coherence theory is quite different in nature to the mentalising and executive function theories as it proposes a different information processing style rather than a specific deficit (Happé & Frith, 2006). Individuals with autism are thought to be good at processing 'local' details of information rather than applying the context and extracting the 'global' meaning. This enhanced ability to perceive the elements rather than the whole and the subsequent lack of drive for gist or gestalt has therefore been termed 'weak central coherence' (Frith, 1989).

1.3.1 Central coherence in autism

Initial experiments supporting this theory involved mainly visuo-spatial tasks requiring attention to detail, such as the Embedded Figures Test (Shah & Frith, 1983) & Block Design subtest of the WISC (Shah & Frith, 1993). Specifically, these tasks required the participant to disengage from the overall image and focus on smaller components within the image. Individuals with autism typically perform *better* than normally developing individuals on such tasks, a surprising finding especially given superiority relative to a typically developing group is difficult to find in any disordered population. Indeed, the Block Design result was a particularly interesting finding as performance on this task was compared to performance when the designs were pre-segmented, a condition that greatly aided the normally developing children but not those with autism. This was a strong indicator that the children with autism were naturally perceiving the designs to be composed of their constituent parts. These results have been replicated by a variety of authors with participants of differing ages and abilities (Embedded Figures: Edgin & Pennington, 2005; Jarrold, Gilchrist & Bender, 2005; Jolliffe & Baron-Cohen, 1997;

Morgan, Maybery & Durkin, 2003; Ropar & Mitchell, 1999; van Lang, Bouma, Sytma, Kraijer & Minderaa, 2006b; although see section 5.1 for a more detailed analysis of these results) (Block Design: Caron, Mottron, Berthiaume & Dawson, 2006).

Another popular method utilised to investigate central coherence involves the use of Navon letters; these are large letters made up of multiple smaller letter components, hence introducing a single global element made up of multiple local elements (see section 5.2 for examples and a more detailed review). Although the exact paradigms used vary, individuals with autism generally show enhanced detection of local over global targets compared to controls, particularly when their attention is not directed to a particular level within the stimuli (Plaisted, Swettenham & Rees, 1999). Similarly, children with autism often show interference from local information when performing global processing whilst controls tend to show the opposite pattern (Behrmann et al., 2006; Plaisted et al., 1999; Rinehart, Bradshaw, Moss, Brereton & Tonge, 2000).

In addition to these commonly used techniques, a range of different paradigms across different modalities have been employed at both high and low levels of processing to support the existence of weak central coherence in autism. In the visual domain, Happé (1996) found evidence of a lack of integration of features in autism through a range of illusion tasks tapping into low level visual perceptual processes; individuals with autism were less likely to succumb to these illusions. Jolliffe & Baron-Cohen (1997) found a trend towards superior performance in adults with autism on a novel version of the Rey Figure test, replicated by Ropar & Mitchell (2001). Mottron, Belleville & Menard (1999a) noted that children with autism were more likely to first produce local elements when attempting a copying task whilst controls tended to start with the global outline. Additionally, they were less affected by figure impossibility than controls, indicating a lack of effect of perception of the global object on their drawing. Similar results were also reported by Booth, Charlton, Hughes & Happé (2003) in comparison to a clinical ADHD group.

Jarrold & Russell (1997) showed that normally developing children were quicker at counting dots when they were arranged canonically (as on dice) rather than randomly, whereas children with autism showed no advantage from canonical arrangement. This finding has since been replicated by Gagnon, Mottron, Bherer & Joannette (2004). However, the individual results in Jarrold & Russell's study did not indicate that this pattern of performance was present in more individuals in the autism group than the control group, instead possibly indicating a stronger effect in those children with autism who did show it. Jolliffe & Baron-Cohen (2001) showed that adults with autism were worse than controls at mentally integrating separate puzzle pieces to recognise an object, whilst performing as well as controls when recognising an object from a single puzzle piece. Interestingly, as in Jarrold & Russell's study, these authors comment on individual performance and indicate that this pattern of abnormal performance is present in the majority of the participants diagnosed with autism but fewer of those with Asperger Syndrome.

More recently, studies have involved other domains such as language and audition, indicating that this is indeed a general processing style rather than restricted to a single domain. In the auditory domain, Mottron, Peretz & Menard (2000) found that individuals with autism were better at detecting local changes in melody such as pitch changes of single notes that did not affect the overall pitch contour, whilst being matched to controls in global changes such as contour change or transpositions. Using a more rigorous method of defining local and global changes in pitch, Foxton et al. (2003) also showed that adults with autism do not show the normal interference from global structure in an auditory pitch change direction-matching task.

In the verbal domain, the most commonly used paradigm involves homographs: two words that are spelt the same but have different pronunciations and meanings. Individuals with autism have been shown a number of times to make more errors with the less frequent

word pronunciation, whilst controls modulate their pronunciation dependent on the context in which the word is presented (Burnette et al., 2005; Frith & Snowling, 1983; Happé, 1997; Jolliffe & Baron-Cohen, 1999b). This may indicate that, instead of reading a sentence for the overall global meaning, individuals with autism may be more inclined to read word by word.

The other main area of language that has been investigated in autism and related to weak central coherence is text comprehension. In order to comprehend language and create a global representation of meaning, information must not only be integrated across sentences (as in the homographs above) but also between sentences across whole passages of text. In addition, language is commonly used in such a way that it is assumed the listener will make inferences that are implied in the text, not explicitly supplying such information as it would be redundant; this also requires the listener to have a global understanding of the implied meaning of the text. Jolliffe & Baron-Cohen (1999b) have shown that individuals with autism had problems with exactly this sort of language processing as they had difficulty making simple inferences across sentences. The same authors (2000) also found that adults with autism were worse at arranging sentences coherently and at making inferences and that these abilities were related to each other. Similarly, Norbury & Bishop (2002) found that high-functioning individuals with autism were poor at making inferences in story comprehension whilst showing good performance on literal understanding of a text. Interestingly, whilst previous research has shown comprehension problems in specific language impairment (SLI) and pragmatic language impairment (PLI) as well as autism (Bishop & Adams, 1992), these authors found that individuals with SLI and PLI who have problems with inferences also have problems with literal understanding, indicating that their problems are more general than in autism.

1.3.2 Brain basis of central coherence in autism

Unlike the mentalising domain, there has been very little research using imaging studies to investigate central coherence. Ring et al. (1999) scanned adults with autism whilst performing the Embedded Figures Task and found greater activation in the visual system in comparison to control adults, suggesting that the adults with autism were analysing object features more whilst the controls were performing the same task through top down control and working memory. Belmonte & Yurgelun-Todd (2003) conducted a functional brain imaging study with adults with autism that indicated that low level problems with selective attention arose from overconnectivity and hyperarousal but reduced selectivity, resulting in a WCC processing style. However, these experiments need to be extended to include proper control tasks and the area of central coherence generally needs further investigation before conclusions about the brain systems involved can be formed. One recent scanning study of the Embedded Figures task in normal adults has used a more strict control task and found activation in left inferior and superior parietal cortex (Manjaly et al., 2003). Further replication and application to autism, as well as more rigorous comparison conditions, are therefore required to make sense of these disparate findings.

At the biological level, a number of suggestions have been made concerning the cause of weak central coherence. In their recent review, Dakin and Frith (2005) discuss the possibility that the magnocellular pathway, an early section of the dorsal visual stream processing primarily motion information, is deficient in autism. Children with autism show impaired detection of motion coherence using random dot kinematograms (Milne et al., 2002; Spencer et al., 2000) and performance on this task is related to performance on the Embedded Figures Test (Pellicano, Gibson, Maybery, Durkin & Badcock, 2005).

However, children with autism also show retained low level dorsal stream functioning through a flicker contrast sensitivity paradigm (Pellicano et al., 2005) and a Gabor patch detection paradigm (Del Viva, Iglizzi, Tancredi & Brizzolara, 2006), indicating that

abnormalities in this pathway do not occur until a later stage of processing and are therefore unlikely to indicate magnocellular abnormalities. On the other hand, Mottron, Dawson, Soulières, Hubert & Burack (2006) claim that, consistent with these results, differences should only be seen at later stages of visual processing as these later stages are likely to tap into global as well as local processing.

A further suggestion considers the possibility that weak central coherence results from neuronal under-connectivity. This under-connectivity may result from either structural or functional differences, due to a lack of synchronisation in activation (Brock, Brown, Boucher & Rippon, 2002) or abnormal physical connectivity (Just, Cherkassky, Keller & Minshew, 2004) between relevant brain areas resulting in a lack of binding of parts into wholes, or numerous and inefficient feedback connections resulting in a lack of top-down modulation of early sensory processing through a lack of integration of sensory processing with cognitive monitoring (Frith, 2003a). All of these ideas predict that the under-connectivity would give rise to a preserved or enhanced ability for exemplar-based information processing, in addition to a reduced ability to generalise across examples or process information in context, reminiscent of the style of processing proposed by the central coherence theory.

One of the most consistent neurobiological findings, which was also noticed by Kanner (1943), is of increased head and brain size and weight in autism. Approximately 20% of the autistic population are thought to have macrocephaly, when defined as having a head circumference greater than the 97th percentile of the normal population (Bailey et al., 1995; Lainhart et al., 1997; Stevenson, Schroer, Skinner, Fender & Simensen, 1997) and two postmortem studies of increased brain weight have supported this finding (Bailey et al., 1993; Bauman & Kemper, 1985). This enlargement appears to be general across the whole of the cerebral cortex (Hazlett et al., 2005). Very recently, a possible genetic mutation in

the PTEN gene has been suggested as the cause of macrocephaly in autism (Butler et al., 2005).

However, macrocephaly cannot normally be detected until approximately 2 years of age (Courchesne et al., 2001; Lainhart et al., 1997; Stevenson et al., 1997) although a recent brain imaging study indicated that the increased rate of head growth starts at about 12 months of age (Hazlett et al., 2005). These findings suggest a decrease in the neuronal elimination processes of pruning and apoptosis, which take place early on in postnatal development, leading to an excess number of neurones. After an initial proliferation of synapses early in development, pruning occurs in normal development to eliminate faulty feedback connections and optimise co-ordinated neural functioning. Furthermore, such elimination is dependent on the information entering the system so that feedback connections particularly can be refined. Additionally, there is evidence that feed forward connections are established very early in development whereas feedback connections are continually refined (Price et al., 2006); this coincides with the increased rate of head growth from about 12 months of age. This suggestion is therefore consistent with the under-connectivity hypotheses mentioned above and therefore also with weak central coherence. Furthermore, a computational model of autism has been constructed in which a lack of generalisation results from an increase in units (Cohen, 1994; Gustafsson, 1997).

1.3.3 Broader phenotype

It has been suggested that there may be a continuum in processing style from weak to strong central coherence and therefore that weak central coherence may also be present in the normal population albeit less commonly (Happé, 1999), although there is limited evidence that this processing style is qualitatively different in autism (Jarrold et al., 2005). The presence of weak central coherence in the unaffected parents of individuals with autism has been investigated and indicates that fathers show a piecemeal processing style

across a number of tasks (Happé, Briskman & Frith, 2001), as well as showing similar levels on an individual basis of preference for non-social activities and detail-focussed processing on self-reports (Briskman, Happé & Frith, 2001). This finding was replicated by Bolte & Poustka (2006) using the Embedded Figures Test, although they found no effects for the Block Design task. Furthermore, Baron-Cohen et al. (2006) recently found that parents of children with autism show reduced activity in extrastriate visual brain areas whilst performing the Embedded Figures Test, and engineers and other scientists have been found to be over-represented among the unaffected parents of children with autism (Wheelwright & Baron-Cohen, 2001), indicating that this processing style may be advantageous for these professions.

1.3.4 Relation to mentalising and executive function

Weak central coherence may therefore be a beneficial skill to possess in some cases. How then might this processing style be related to the problems seen in autism? It was originally proposed that weak central coherence might be the primary abnormality responsible for all key features of autism (Frith, 1989), causing the mentalising impairment as well as savant skills. However, Frith later abandoned this notion (Frith & Happé, 1994) as this would not fit with the idea of a continuum across the normal population or correlational evidence that performance in these domains is unrelated (Happé, 1994; Pellicano et al., 2006) (although see Jarrold, Butler, Cottington & Jimenez, 2000 for evidence in favour of the existence of such a relationship). Weak central coherence is therefore now seen as an independent factor from ToM in terms of its causal effect in autism. One alternative suggestion has been that, in combination with the mentalising deficit, this processing style allows less compensation and therefore more severe symptomatology (Happé, 2000), although this has yet to be investigated.

The central coherence account of autism is also similar to the executive function theory in that both attempt to explain non-social features of autism. It has been suggested that executive dysfunction could be the result of a lack of top-down control leading to reduced inhibition (Frith, 2003a), a hypothesis reminiscent of the poor feedback connections suggested in the central coherence theory. One study has shown these two cognitive abilities to be related in autism (Pellicano et al., 2006), although this may have been due to overlap in requirements of the tasks involved, whilst a second study found no support for a relationship (Teunisse et al., 2001), with weak central coherence being present equally in autistic individuals with good and poor executive function.

1.3.5 Relation to symptoms

As weak central coherence is seen as an independent causal factor in autism, it does not try to explain many of the defining diagnostic features of autism and indeed symptom profiles do not appear to be related to central coherence test performance. Morgan et al. (2003) found central coherence to be unrelated to joint attention, pretend play or verbal ability and central coherence does not seem to predict later social functioning in adults (Berger et al., 2003). Teunisse et al. (2001) also found no relationship between weak central coherence and any aspect of autistic symptomatology. Rather, weak central coherence attempts to account for some of the non-social features, particularly obsessive interests, attention to detail, poor comprehension, literal understanding of language, hypersensitivity, savant skills, indiscriminate approach to people and the uneven spikey IQ profile that is typically seen. To mention one of these, savant skills are present in a higher proportion of individuals with autism than in the general population and it is thought that piecemeal processing may be advantageous in developing such skills, for example, segmenting ability for artistic talent (Pring & Hermelin, 2002; Pring, Hermelin & Heavey, 1995). Mottron & Belleville (1993) reported the case of an artistic savant who showed a local bias in Navon and other central coherence tasks. Furthermore, it has been suggested that central

coherence may explain some of the face processing peculiarities seen in autism (Davies, Bishop, Manstead & Tantam, 1994), such as the use of high spatial frequencies to match faces (Deruelle, Rondan, Gepner & Tardif, 2004) and the reduced activity in face specific brain areas and increased activity in areas normally recruited to process objects or perform visual search when processing faces (Hubl et al., 2003). Indeed, face processing ability has been correlated to performance on the Navon task (Behrmann et al., 2006).

1.3.6 Negative findings & heterogeneity

The central coherence theory has met with even greater opposition than the mentalising theory, not in terms of plausibility but, similar to the executive function theory, in terms of negative findings. Again, it should be remembered that negative results may occur for a number of reasons: task sensitivity, heterogeneity, changes over time and strategy use, or that the theory is incorrect. A lack of group differences between controls and individuals with autism have been found on the Embedded Figures Test and other disembedding tasks (Brian & Bryson, 1996), the Block Design subtest (Ozonoff et al., 1991a) and in susceptibility to visual illusions (Hoy, Hatton & Hare, 2004; Ropar & Mitchell, 1999). Motttron et al. (1993; 1999a; 2003), Ozonoff et al. (1994) and Edgin & Pennington (2005) found no group differences on a Navon task and suggested particularly a lack of evidence for impaired global processing. Furthermore, whilst finding impairments on the homographs test, Burnette et al. (2005) also found *impaired* performance on the block design test, Embedded Figures Test and a pattern construction task. Whilst some of these findings appear to result from methodological issues, such as group matching, task design and instructions (see Chapter 5 for a discussion of the issues surrounding the Embedded Figures Test and Navon tasks), the quantity of negative results is notable.

A further problem for the theory is encountered when examining experiments testing weak central coherence across tasks and domains. Ropar & Mitchell (2001) found no

relationship between performance of individuals with autism on visual illusions and the Embedded Figures Test, Block Design or the Rey Figure test. Lopez & Leekham (2003) found evidence in support of weak central coherence in an autism group on verbal but not visual tasks involving processing complex information in context, although this may have been due to task difficulty. Moreover, the global impairment in using context mainly comes from evidence in the verbal domain and it is possible that these problems are purely linguistic rather than general, given the frequent presence of language impairment in autism (Norbury, 2005). Indeed, the group differences found by Hoy et al. (2004) on the homographs task appeared to be mediated by verbal ability; these vanished when verbal ability was accounted for. Burnette et al. (2005) also found performance on the homographs task to be related to performance on ToM tests, while other central coherence measures were not. It is as yet unknown how these domains within central coherence relate to each other and whether it is possible to have weak central coherence manifested in one domain only, with different domains affected in different individuals.

The original version of the theory has therefore developed and transformed over the years to accommodate many of these findings. A more recent and detailed version of the central coherence theory states that, rather than being poor at global processing, autistic individuals are simply either superior at, have a bias towards or have a preference for local processing when both are in competition but are quite capable of global processing if required under the right conditions (Happé, 1999; Happé & Frith, 2006). Indeed, there is now general consensus in the field that global processing is intact (Motttron et al., 2006). This goes some way towards incorporating those studies finding a lack of group differences or finding intact global processing, particularly those when global and local processing are not in competition. Equally, the idea of heterogeneity of this processing bias in the autistic population has rarely been studied so this may be a further reason. One recent study showing individual results however, reported that some aspect of weak central coherence was present in almost all of the young children with autism tested on a battery of

visuospatial tasks (Pellicano et al., 2006), although some of these tasks may have produced deviant performance for other reasons, such as their executive demands.

1.3.7 Other explanations of intact and superior abilities in autism

The symptoms of autism that are addressed by the theory of weak central coherence have also been addressed by other theories. Rather than a higher-level cognitive phenomenon, both Mottron et al. (2006) and Plaisted, Saksida, Alcantara & Weisblatt (2003) have suggested that the local bias may result instead from a difference in low level sensory processing, termed 'Enhanced Perceptual Functioning' by Mottron et al.. This alternative theory emphasises strengths in cognitive processes even more than the central coherence theory. The main difference between these theories is whether the local bias results from superior bottom-up sensory processing or impaired top-down cognitive control. Evidence supporting superior low-level processes comes from findings that children with autism do not show the normal advantage for familiar over novel stimuli in a discrimination task (Plaisted, O'Riordan & Baron-Cohen, 1998a), indicating enhanced discrimination of stimuli features. Similarly, a group of children with autism had faster reaction times on a conjunctive visual search paradigm, indicating enhanced item detection but a lack of collation of information (Plaisted, O'Riordan & Baron-Cohen, 1998b). Caron et al. (2006) also found that individuals with autism, unlike controls, were unaffected by increasing the perceptual cohesiveness of the Block Design task, whilst performing well on a number of task manipulations requiring global processing, indicating that whilst being capable of processing global information, this did not aid their already superior performance any further. All these findings have been attributed to low-level perceptual abnormalities.

These results could be incorporated by either theory and there is currently no evidence to enable these theoretical positions to be differentiated. In fact, one recent experiment (Pellicano et al., 2006) supports neither position, instead finding normal performance on a

Navon task aimed to differentiate the theories. In the discussion, Plaisted suggests a modified theoretical position, that the intact and superior abilities seen in autism could be explained rather by a difficulty in moving between local and global processing, specifically in the direction local-to-global. While this is reminiscent of executive function theories of attention switching in autism (Courchesne et al., 1994), the directionality of the switch would be problematic for a general executive theory. Interestingly, this reintroduces the idea of a deficit as opposed to a processing style and still leaves open the question of whether the origins are in bottom-up or top-down processes. Mann & Walker (2003) found that children with autism found it harder to attend to stimuli outside their current focus of attention and Rinehart, Bradshaw, Moss, Brereton & Tonge (2001) found worse performance on global stimuli in a Navon task when a shift in attention was demanded by the proceeding trial requiring local processing. Iarocci, Burack, Shore, Mottron & Enns (2006) found that, despite normal performance in global and local visual search tasks, children with autism were more sensitive than controls to an implicit local task bias. These results appear to be consistent with the idea of a bias towards local processing, resulting in performance that is only sometimes deficient or enhanced, rather than an absolute deficit or enhancement and that the origin of this bias may conceivably be in attentional processes.

1.3.8 Specificity of weak central coherence to autism

Weak central coherence has not as yet been thoroughly investigated in other clinical disorders. William's Syndrome is thought to involve some element of featural processing, such as when drawing (Bellugi, Lichtenberger, Jones, Lai & St George, 2000), but individuals with this disorder show normal Embedded Figures Test performance and instead show weak mental imagery (Farran, Jarrold & Gathercole, 2001). Patients with schizophrenia have been found to show impaired performance when processing motion stimuli requiring global visual processing (Chen, Nakayama, Levy, Matthyse & Holzman,

2003; Johnson, Lowery, Kohler & Turetsky, 2005), although they conversely appear to show a local deficit on a Navon task (Bellgrove, Vance & Bradshaw, 2003) and even an abnormal global advantage (Granholm, Cadenhead, Shafer & Filoteo, 2002). Patients with depression or anxiety with negative mood tend to show a local bias (Hesse & Spies, 1996); however, depressed patients also appear to perform more poorly than controls on the Embedded Figures Test (Calamari, Pini & Puleggio, 2000). It seems that research on central coherence within each of these disorders is inconsistent and positive results would require further replication before a lack of specificity of weak central coherence to autism can be assumed.

1.4 Conclusion

It can thus be seen that none of the three major cognitive theories appear to be able to account for all the behavioural manifestations of autism. Particularly, none can account for the diversity of experimental findings and behaviours seen in different children. Indeed, no autism theory or theorist really claims to be able to explain all the features of autism; it is generally accepted that autism is likely to include a number of different deficits, and indeed the evidence so far supports this concept (Frith, 1996; Frith & Happé, 1994; Hill & Frith, 2003; Ronald et al., 2006; Tager-Flusberg, Joseph & Folstein, 2001). However, it is also generally accepted that each theory is true for all individuals on the autism spectrum; as can be seen, this has been less well investigated.

The idea that the autism spectrum varies quantitatively, including different severities as well as different combinations of symptoms, can possibly be extended to also include qualitative differences at the cognitive level, with different children having different underlying aetiologies. Indeed, even at the behavioural level, my own anecdotal observations of autistic children lead me to believe that certain children diagnosed with autism, particularly some of those who are high-functioning, are socially impaired in a different way to the

majority of autistic children. Some of these children appear to have a sense or knowledge of what is socially appropriate but do not act accordingly, whilst more typical autistic children appear unaware of appropriate social behaviour. This leads to the hypothesis that there may be subtypes within the autism spectrum with different underlying aetiologies producing some similar behaviours. This calls for the investigation of cognitive phenotypes, which may be a more pertinent way of classifying children than through behaviour.

Indeed, the introduction of different cognitive phenotypes within autism makes sense of the heterogeneity within the disorder, as only those individuals who are affected by the particular cognitive impairment are included in the explanation. Furthermore, the problems of lack of specificity and heterogeneity may also result from studying children grouped by diagnosis, rather than by symptoms, given that diagnostic categories are composed of 'pick and mix'-style criteria. Relating cognition to behaviour is therefore essential to reduce heterogeneity, increase specificity and provide validity for the cognitive theories. Ideally, the search for biological markers would also occur in order to search for consistency across levels, but this is mostly beyond the scope of this thesis.

A further dilemma is that these theories are currently being investigated through tests with unproven sensitivity and unknown psychometric properties. The assumption made in this thesis is therefore that, despite holes and gaps in knowledge, rigorous methodology can be applied in order to tie together performance across tests guided by psychological theory and behavioural symptoms.

1.5 Aim

The aim of this PhD is therefore to assess each of the three main cognitive theories of autism in a selected sample of children in order to study their relationship to each other with selected tests. This enables cognitive subtypes to be located within the autism

spectrum, at least for a selected sample, and for these to be related to the symptoms seen at the behavioural level. In order to do this, I have collected data from tasks tapping mentalising, executive functions and central coherence, as well as verbal and performance IQ, head-size and information on behavioural symptoms, from a large number of high-functioning children on the autism spectrum, as well as normally developing children, providing data on possible cognitive (and one biological) markers. This is a multiple case study design where individual performance can be assessed within each domain and related across domains (see White et al., 2006). Rather than attempting to create new definitions of autism or within autism, this study rather attempts to investigate the adequacy of current behavioural tests in their ability to tap into the hypothesised cognitive impairments and their ability to explain everyday symptoms.

Only one other study has addressed all three cognitive domains in the same children and related these to symptoms. Pellicano et al. (2006) studied the relationship of weak central coherence with mentalising and executive function in high-functioning 4-7 year old children with autism compared to controls. Whilst revealing distinct heterogeneity at least within the mentalising and executive function domains, this study needs to be extended using different measures to evaluate whether any effects were test dependent, and whether they were tied to particular children and methods of diagnosis. Furthermore, it has great potential to be extended by also assessing the critical relationship between mentalising and executive function and more extensively comparing the cognitive theories to behavioural manifestations of the disorder.

Chapter 2: Population

2.1 Sample characterisation

The population I have chosen to study is high-functioning junior-school children with a range of autism spectrum disorders, as well as normally developing control children.

(From here on I use the term 'autism spectrum disorder' or 'ASD' to refer to diagnoses anywhere on the spectrum, including autism and Asperger Syndrome, not just Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS).) The majority of recent research into the cognitive impairments found in ASD has been conducted in higher-functioning populations as they have a broader behavioural repertoire and are able to cope with higher task demands. Given that the demands of my battery of tests was high, involving 3-4 hours of testing, it was considered essential that the children should be relatively able and have good concentration. Additionally, as general ability would be within the normal range, any effect that general mental retardation might have on performance could be eliminated. Furthermore, the use of a sample with intelligence in the normal range circumvents the problem of comparison to younger normally developing children as well as age and intelligence matched controls. The limited age group here also makes the group more homogeneous, making task performance easier to interpret. Even more crucially, many high-functioning children, particularly those with Asperger Syndrome, are not diagnosed until about 7 years so a junior age but not a younger sample is likely to pick up these later-diagnosed children. On the other hand, older children are more likely to have received some form of remediation or be compensating for their problems so this possibility is minimised in a junior age sample. Thus the choice of this young but high-functioning group aims to maximise the presence of abnormal behaviours whilst allowing a large battery of tasks to be utilised.

Of course the disadvantage of using such a high-functioning population is that some abnormal behaviours or performance may have been overlooked or compensated for and the results therefore have a more limited application and generalisation to the whole autism spectrum. Any conclusions in this thesis therefore must be viewed within these limits. However, whilst this type of sample could not be described as the classic 'text book' description of ASD, these high-functioning children still have significant problems in everyday life. Furthermore, such children are numerous and a burden on the educational system, thus deserving investigation and recognition.

2.1.1 Recruitment

In order to recruit these children, the head Educational Psychologist at both Surrey and Sutton Local Education Authorities was contacted and, through this contact, all mainstream schools with statemented children on the autism spectrum were approached. Approximately 50% of these schools agreed to send consent forms to parents and approximately half of the parents approached gave consent. Each school that provided a positive consent form for a child on the autism spectrum was also asked to recruit a control child of approximately the same age, gender, ethnicity and general ability as the child with ASD. Not every school was able to provide a control child for the study however. In addition, specialist schools within Greater London and Surrey with provision for children at the high-functioning end of the autism spectrum were approached and a small number of these schools were able to recruit children to take part. The Surrey branch of the National Autistic Society also included an advertisement in its newsletter and a number of parents subsequently contacted me directly in order to become involved in the study.

In total, 84 children on the autism spectrum and 31 control children were recruited for the study. This imbalance in numbers between the groups was intentional, partly because a

greater drop-out was expected in the ASD group but also to allow for sufficient power once subgroups had been created within this group. Of these 115 children, 2 control children and 3 children with ASD dropped out after the first session due to a change of school, 1 control was excluded after the first session due to an additional diagnosis of Oppositional Defiant Disorder (ODD) and possible Obsessive Compulsive Disorder (OCD), and 22 children with ASD were excluded due to an inability to understand and comply with the task demands. The remaining 28 control children and 59 with ASD therefore comprise the data set. Of the children with ASD, the majority came from mainstream primary schools, 8 were recruited through autism units attached to mainstream schools (although all these children spent the majority of time in mainstream education) and 10 attended schools specialising in autism and related disorders (but these children can be considered high functioning given they were able to complete an intensive battery of tests).

2.1.2 Prior diagnoses

All 59 children in the ASD group had previously received a diagnosis somewhere on the autism spectrum from a qualified clinician, most commonly from a Child and Adolescent Psychiatrist at the local Child and Adolescent Mental Health Service. 10 children had received diagnoses of autism, 31 of Asperger Syndrome, 16 of autism spectrum disorder or PDD-NOS and two of semantic pragmatic disorder (SPD) related to the autism spectrum; these latter two children were not excluded as it is thought that pragmatic language impairment is related to the autism spectrum (Bishop, 1989; Bishop & Norbury, 2002). Furthermore, both children were dealt with by the schools in the same way as other children on the spectrum; they had been identified as having similar needs and given the same help, support and provision. Of all the children involved, 3 also had diagnoses of ADHD, one of dyspraxia and one of mild Tourette's Syndrome. 45 of these children had statements of special educational needs.

2.1.3 Verification of diagnoses

Interviews are commonly used with parents at diagnosis, in conjunction with behavioural observation, in order to obtain a detailed history of the child's development as well as information on the child's present behaviours. A number of standardised interviews have been developed (Autism Diagnostic Interview-Revised (ADI-R); Lord, Rutter & Le Couteur, 1994; Diagnostic Interview for Social and Communication Disorders (DISCO); Wing, Leekam, Libby, Gould & Larcombe, 2002) although many of these are lengthy and impractical in a research setting. One such interview, the Developmental, Dimensional and Diagnostic Interview (3Di; Skuse et al., 2004) has recently been developed to be shorter and more suitable for research purposes, as well as being particularly suitable for detecting mild and high-functioning cases. As the children in the present sample would have been diagnosed by different clinicians, possibly using different criteria, diagnoses were verified using the 3Di. Interviews were conducted with one or both parents of each child involved in the study, both those with ASD and control children, after all testing had been completed. In addition to verifying diagnoses, the data collected from the parental interview was also useful as a measure of the severity of the behavioural symptoms of ASD present in each child.

The 3Di provides information on the triad of symptoms used for diagnosis: the social and communication impairments as well as the presence of repetitive behaviours. Each area of the triad is composed of a number of categories that correspond to those given in ICD-10 (World Health Organisation, 1993), each category of a number of items and each item of a number of questions. In the social domain, for example, one of the categories is peer and sibling relationships, one of the items within this category concerns imaginative play with others and one of the questions within this item asks whether the child ever plays imaginative games with other children. This structure creates quantitative measures of impairment in each of the three areas critical for diagnosis. Along these three continua,

cut-offs for clinically significant impairment are provided in order to provide a categorical diagnosis. Children above the cut-offs for all three areas are then considered eligible for an autism or Asperger Syndrome diagnosis, dependent on the presence or absence of language delay (see below), while those meeting criteria for one or two of the areas only would meet criteria for a diagnosis of atypical autism or Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS). Additionally, those children with significant impairment in only the communication area may be classed as showing a pragmatic language impairment.

On the basis of the cut-offs advised by the 3Di, 1 control was excluded due to high scores on two of the three diagnostic areas as well as a referral to the Child and Adolescent Mental Health Service for suspected ASD and ADHD after testing had been completed, and 2 children in the ASD group were excluded due to not reaching criteria for an ASD on any of the three areas of the triad. Interestingly, both of these children had very early childhood diagnoses (one of autism, one of ASD), were relatively low-functioning and attended special schools. The remaining sample therefore consisted of 57 children with ASD as well as 27 control children. 3Di data was unavailable or incomplete for 4 children with ASD due to parental non-compliance; whilst these children therefore cannot be included in analyses involving behavioural symptoms, they were not excluded from other analyses as there was no reason to believe their original diagnoses were not valid.

Data from the 3Di are shown in Table 2.1 and, as would be expected, indicate severe impairment in all three domains in the ASD group (social $t(77.1)=13.6, p<.001$; communication $t(78)=19.5, p<.001$; repetitive behaviours $t(60.7)=13.1, p<.001$). The communication domain appears to be the best discriminator between the groups with no overlap in the ranges, although the other two domains only show minimal overlap and a clear shift in their distributions. Within the communication domain, all children with ASD fell above the cut-off whereas this was not the case for the other two domains. While this

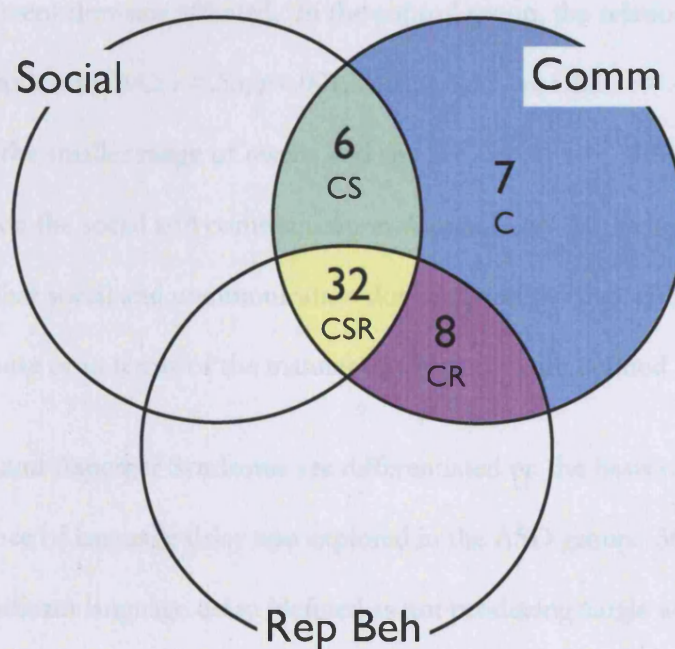
Table 2.1 Means (and standard deviations) with *range* for the three diagnostic domains of the 3DI: social interaction, verbal and non-verbal communication, and repetitive and stereotyped behaviours and mannerisms; high scores indicate impairment.

	Control group (27)	ASD group (53)
Social *** (30 max; 10 cut-off)	3.3 (2.0) .3–8.9	13.2 (4.5) 5.9–22.5
Communication *** (26 max; 8 cut-off)	3.7 (1.7) .7–6.7	14.8 (3.4) 8.2–22.9
Repetitive behaviours *** (12 max; 3 cut-off)	.3 (.5) 0–2	5.1 (2.5) .6–10

*** $p < .001$

may be dependent on the characteristics of the particular sample used here, it may be that the cut-offs provided by the 3DI are not consistent across the different areas of the triad, with the communication cut-off being relatively low compared to the other two areas. The pattern of above cut-off symptoms present in different children is shown in Figure 2.1.

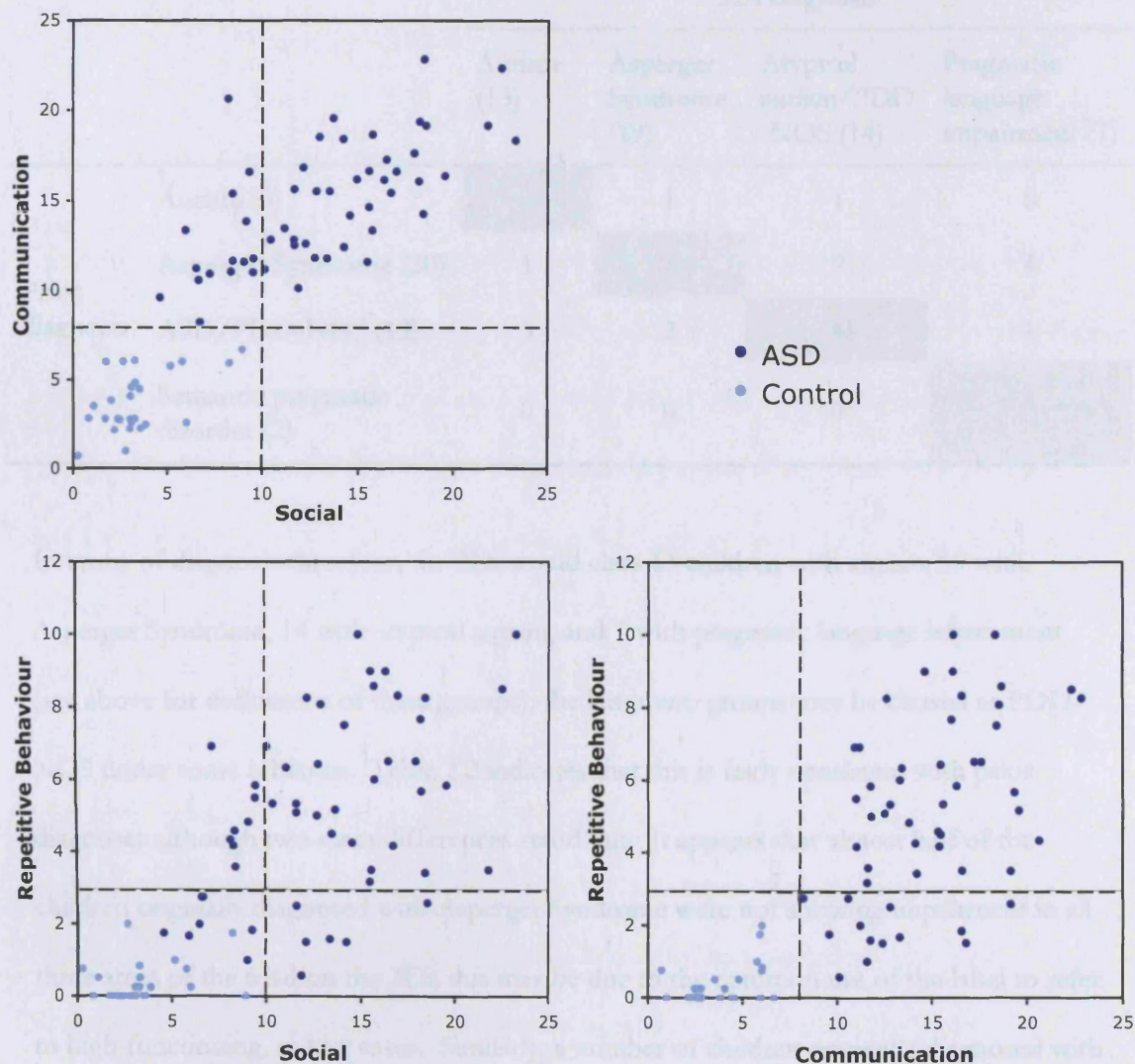
Figure 2.1 Venn diagram showing the presence of above cut-off symptoms on the different 3DI domains for each child in the ASD group; S=social, C/Comm= communication, R/Rep Beh=repetitive behaviour.



The 7 ASD children presenting with only a communication impairment (C) on the 3Di showed a lesser degree of communication problems than the remaining 46 children with ASD, who all had additional domains of impairment ($t(51)=2.9, p=.006$), indicating that they generally had milder symptoms. Similarly, the 32 children with all three areas of impairment (CSR) showed more severe communication problems ($t(38)=2.3, p=.024$) and more repetitive behaviours ($t(20.9)=2.8, p=.010$) than the 8 children with just communication impairment and repetitive behaviours (CR), indicating that they generally had more severe symptoms. However, the 6 children with only social and communication problems (CS) were similarly impaired to those children with all 3 impairments (CSR) in both the social ($t(36)=1.4$) and communication domains ($t(36)=1.7$), indicating that severity might be more highly related between the social and communication impairments than with the presence of repetitive behaviours. All three areas of the triad were highly correlated to each other within the ASD group (S&C: $r=.67, p<.001$; S&R: $r=.46, p<.001$; C&R: $r=.45, p<.001$; see Figure 2.2) although the relationship between the social and communication domains was strongest. This supports the idea that the autism spectrum is a continuum of severity across the three domains rather than a collection of similar disorders with different domains affected. In the control group, the relationships were weaker or non-significant (S&C: $r=.53, p=.001$; S&R: $r=.15, ns$; C&R: $r=.49, p=.010$), most likely due to the smaller range of results and smaller sample size, although again the relationship between the social and communication domains was the strongest. This supports the idea that social and communication domains may overlap, either in terms of their underlying cause or in terms of the manner in which they are defined.

Given that autism and Asperger Syndrome are differentiated on the basis of early language abilities, the presence of language delay was explored in the ASD group. 36% of these children had a significant language delay (defined as not producing single words by 2 years or meaningful phrases by 3 years); this was present in 40% of the children displaying all 3 symptom impairments (CSR), 38% of the children with communication impairment and

Figure 2.2 Summary of correlations between the 3Di data on the three areas of the triad: social, communication and repetitive behaviour; the dotted lines indicate the cut-offs from the 3Di and scores above this indicate abnormal behaviour.



repetitive behaviours (CR), 33% of the children with social and communication impairment (CS), and 14% in the children with only communication impairment (C). Again, this possibly indicates a general lower severity in the children with only communication impairment, although this difference was not significant ($\chi^2(1)=1.6$). However, when comparing those children with language delay to those without, no differences on the social, communication or repetitive behaviour domains were found.

Table 2.2 Classification of children according to prior diagnoses and 3DI diagnoses; shaded cells show the equivalent diagnosis under the two classification schemes.

		3Di diagnosis			
		Autism (13)	Asperger Syndrome (19)	Atypical autism/PDD-NOS (14)	Pragmatic language impairment (7)
Prior diagnosis	Autism (9)	7	1	1	0
	Asperger Syndrome (30)	1	16	9	4
	ASD/PDD-NOS (12)	5	2	4	1
	Semantic pragmatic disorder (2)	0	0	0	2

In terms of diagnosis therefore, the 3Di would class 13 children with autism, 19 with Asperger Syndrome, 14 with atypical autism, and 7 with pragmatic language impairment (see above for definitions of these groups); the latter two groups may be classed as PDD-NOS under some schemes. Table 2.2 indicates that this is fairly consistent with prior diagnoses although two main differences stand out. It appears that almost half of the children originally diagnosed with Asperger Syndrome were not showing impairment in all three areas of the triad on the 3Di; this may be due to the common use of the label to refer to high-functioning, milder cases. Similarly, a number of children originally diagnosed with ASD were classified as having autism or Asperger Syndrome by the 3Di; this may be due to the use of ASD to refer to the whole of the spectrum when the clinician does not want to make a more specific diagnosis. However, the above analyses suggest that the severity of symptoms (quantitative differences) across all 3 domains may be a better characterisation of these children than using a cut-off approach (qualitative differences) as those children with more areas of the triad affected also show a greater severity.

Indeed, many authors assume that the autism spectrum is a continuum rather than a group of related but distinct disorders (de Bruin, Verheij & Ferdinand, 2006; Frith, 2004; South,

Ozonoff & McMahon, 2005; Verte, Geurts, Roeyers, Oosterlaan & Sergeant, 2006a; Verte et al., 2006b) and the data here supports this notion. This also strengthens the idea that the communication cut-off may be relatively low compared to the other two cut-offs. The data from the 3Di will therefore mainly be used quantitatively for the remainder of this thesis.

2.1.4 Novel everyday behaviour questionnaire

In addition to the 3Di, a novel questionnaire was developed for the present study that aimed to tap behaviours indicative of mentalising, central coherence and executive function deficits, based loosely around an existing questionnaire (Frith, Happé & Siddons, 1994). This was sent to parents along with questionnaires regarding background data for the 3Di and can be found in Appendix 1. Each item in the questionnaire was designed to tap into a behaviour thought to be related to one, two or all three of the cognitive domains to be evaluated in this thesis. The items of interest however were those tapping the domains independently and only these items were analysed. As the executive function and mentalising domains were often difficult to separate, triplets of items were developed that referred to similar situations but differed only in their executive or mentalising requirements. For example, the item 'supplies important missing information' was accompanied by the mentalising item 'aware that each person's knowledge depends on their experience' and the executive item 'can solve puzzles with missing information eg. hangman'. Items were balanced to include equal numbers of positive and negative statements. Parents were asked to score each item on a three point scale: does not apply, applies somewhat, or definitely applies, producing an overall rating of the percentage of such behaviours shown by each child in each domain. Parents of children with ASD rated their children as showing on average 56%, 68% and 44% respectively of the theory of mind, central coherence and executive function impairments listed, whilst ratings from parents of control children were significantly lower at 8%, 39% and 7% ($t(78) > 6$, $p < 0.001$).

The high scores in the control group on the central coherence questionnaire may support the idea that this area of cognition is represented in a continuum across the general population (Happé, 1999). Additionally, the parents of control children may see the behaviours indicative of weak central coherence as positive assets and are therefore more likely to rate their children as showing these than the more negative behaviours in the mentalising and executive function questionnaires. For example, 'good at remembering lists of items, eg. phone numbers' is a central coherence item on which 'definitely applies' would indicate weak central coherence and which is generally seen as a socially appropriate asset to possess, whereas 'recognises embarrassment in others' is a mentalising item on which 'does not apply' would indicate mentalising problems and which is generally seen to be a negative quality to possess. The high proportion of behaviours indicative of weak central coherence in the controls may therefore be a result of the positive central coherence items being more common and more socially acceptable everyday behaviours in the general population.

2.1.5 Dysexecutive Questionnaire for Children

A further standardised questionnaire known as the Dysexecutive Questionnaire for Children (DEX-C) from the Behavioural Assessment of Dysexecutive Syndrome for Children (BADS-C: Emslie, Wilson, Burden, Nimmo-Smith & Wilson, 2003) was completed by the parents of each child. This questionnaire is designed to tap into the everyday behavioural problems displayed by individuals with executive dysfunction. This questionnaire has been validated in adult patients and has been shown to be related to performance on a number of executive tests, including some of those involved in the BADS (Burgess, Alderman, Evans, Emslie & Wilson, 1998). The questionnaire consists of 20 statements concerning specific areas of behaviour that must each be rated in terms of their frequency of occurrence on a 5 point scale, from 'never' to 'very often'. High scores are therefore indicative of the presence of dysexecutive symptoms. Out of a maximum

score of 80, the ASD group scored an average of 52 (SD 21) whilst the controls scored only 11 (SD 11), producing a robust group difference ($F(1,79)=65.62, p<.001$).

2.1.6 Comorbid diagnoses

The 3Di also contains some additional components that can be used to look for related or comorbid disorders, such as Attention Deficit Disorder (ADD), Attention Deficit Hyperactivity Disorder (ADHD), Oppositional Defiant Disorder (ODD) and Conduct Disorder (CD). One child met criteria for ADD, 14 for ODD, one for socialised CD, one for CD confined to the family, two for ADD and ODD, and one for ADHD and CD confined to the family. None of these children were excluded as it was felt that many of the behaviours necessary for such diagnoses could be explained by the social problems accompanying ASDs. Indeed, the majority of the children in the ASD group showed at least mild signs of inattention and impulsivity; additionally, DSM-IV (American Psychiatric Association, 2000) recognises this by discounting ADHD diagnoses when ASDs are present.

2.2 Test battery

All the children completed a battery of tests lasting approximately 3.5 hours, split into 3 sessions conducted over the course of a year for the majority of children, although some children had up to 7 testing sessions. This battery consisted of a range of tests tapping into mentalising, executive function and central coherence abilities (see chapters 3, 4 and 5 respectively), as well as testing general intelligence levels and recording the child's head circumference.

2.2.1 Intelligence testing

Given that the cognitive deficits present in ASD may affect performance, both positively and negatively, on IQ tests (Happé, 1994), reliability of such measures is obtained by combining results from a range of different tests rather than using a single test. As it was extremely important for this project to obtain reliable individual measures of verbal and performance IQ, the most obvious measure to use was therefore the Wechsler Intelligence Scale for Children III-UK (WISC: Wechsler, 1992). This measure includes 5 verbal and 5 performance subtests over which performance is averaged, as well as an additional subtest of verbal working memory (digit span), and was administered according to the manual. Performance IQ was tested in the first test session and verbal IQ in the second test session for the majority of children.

2.2.2 Head size

Head circumference was measured with a standard flexible tape measure and measurements were then converted to standardised z-scores, adjusted for age and sex according to available norms (Farkas, 1994). This measurement was performed in the last test session with each child. As only Caucasian norms were available, measurements were disregarded for any non-Caucasian children (2 controls and 5 children with ASD). Previous research has shown that macrocephaly exists in some children with ASD until at least the age of 12 years (Aylward, Minshew, Field, Sparks & Singh, 2002) and so it was expected that the range of ages of the children involved would still be sensitive to this measure.

2.2.3 Group matching

While the two groups of children were well-matched for age (age at time of first test session given in Table 2.3; $t(82)=.56$), gender ($\chi^2(1)=1.384$; 7 ASD girls, 6 control girls) and

Table 2.3 Descriptive statistics for age, intelligence and head circumference; means (and standard deviations) with *range*; head circumference only available for 25 control and 52 ASD Caucasian children.

	Control group (27)	ASD group (57)
Age (years)	9.1 (1.3) 6.6–11.3	8.9 (1.5) 6.8–12.0
Verbal IQ *	115 (16) 84–139	104 (19) 60–138
Performance IQ **	103 (12) 78–125	95 (14) 67–127
Full scale IQ **	111 (15) 83–134	100 (16) 65–131
Head circumference (z-scores)	.72 (1.07) -1.58–2.73	1.14 (1.29) -1.78–4.35

* $p < .05$, ** $p < .01$

ethnicity ($\chi^2(2) = .099$; 5 ASD non-Caucasians, 2 control non-Caucasians), the ASD group had lower verbal $t(82) = 2.41$, $p = 0.018$ and performance IQs ($t(82) = 2.77$, $p = 0.007$) and therefore lower full scale IQs ($t(82) = 2.96$, $p = 0.004$). Despite these differences, it can be seen from the means in Table 2.3 that the ASD group are performing at average IQ levels expected for the general population and therefore can be considered a high-functioning sample; rather, it is the controls who appear to be of above average intelligence, particularly in the verbal domain. However, it is quite possible that the WISC norms are out of date, having been collected over 10 years ago and before the advent of the literacy and numeracy hour initiative in UK schools in 1998; this is likely to have affected verbal more than performance IQ levels of even the oldest children in this sample given that both arithmetic and verbal subtests such as vocabulary load onto verbal IQ; this could explain why verbal IQ is so much higher than performance IQ in the controls and why they are performing on average one standard deviation above average. This would imply that the ASD group was instead performing below the level currently expected for verbal IQ.

In addition, observation of the ranges reveals that there are children in the ASD group spanning the same range of IQs as the controls but with an additional low-scoring tail of individuals below the range of the controls. Control children with lower IQs were actively sought for inclusion in the study; however, these were difficult to find as most children with below average IQs had some form of prior diagnosis. While it might therefore be valuable to exclude those children performing below the control range, it was considered vital to retain the large ASD group size in order to study individual profiles and possible subgroups; all 57 children were therefore retained in the sample. Furthermore, all except two children in the ASD group had one of verbal or performance IQ within the control range; as verbal and performance IQs are known to be more discrepant in ASD than control populations (Joseph, Tager-Flusberg & Lord, 2002), to exclude children with low scores on one of these may have introduced a sampling bias. Indeed, the discrepancies between verbal and performance IQ ranged from verbal IQ that was 49 points lower than performance IQ to verbal IQ that was 46 points higher than performance IQ in the ASD group whereas the control discrepancies were 12 and 36 respectively. Still, verbal and performance IQ were correlated in both groups (ASD: $r = .41$, $p = .002$; control: $r = .57$, $p = .002$).

Differences between the groups for head circumference were also examined and found to be non-significant ($t(75) = 1.42$), although there was a trend for the ASD group to include individuals with larger head circumferences than any of the controls, up to more than 4 standard deviations above the population mean. Twelve children with ASD, corresponding to 23% of this group, met clinical criteria for macrocephaly (more than 1.88 standard deviations above the population mean), similar to previous prevalence estimates (Bailey et al., 1995; Lainhart et al., 1997; Stevenson, Schroer, Skinner, Fender & Simensen, 1997).

2.3 Relating intelligence to symptom profiles and head size

Although an IQ profile in which performance IQ is higher than verbal IQ has long been associated with ASD (Lincoln, Courchesne, Kilman, Elmasian & Allen, 1988; Rutter, 1978), it is now known that the opposite profile is more commonly seen in high-functioning individuals (Klin, Volkmar, Sparrow, Cicchetti & Rourke, 1995; Manjiviona & Prior, 1995; Ozonoff, South & Miller, 2000). This difference is possibly due to the less frequent occurrence of language delay in high-functioning individuals. Indeed, when comparing those children in the ASD group with language delay (19) to those without (34), no differences were found in age ($t(51)=.05$) or performance IQ ($t(51)=1.44$); as might be expected however, those children with language delay had lower verbal IQs (mean of 97) compared to those without (mean of 109) ($t(51)=2.25$, $p=.029$).

A number of studies have looked at behavioural correlates of macrocephaly in ASD with little success (eg. Lainhart et al., 1997; Miles, Hadden, Takahashi & Hillman, 2000).

However, two recent studies have suggested that verbal and performance IQ and the difference between them may be related to both symptom severity and brain size (Deutsch & Joseph, 2003; Joseph et al., 2002). Specifically in these studies, low verbal IQ was found to be associated with greater social and communication problems, and relatively high performance IQ compared to verbal IQ (IQ discrepancy) was associated with greater social and communication problems and with larger head sizes (after correcting for absolute verbal IQ levels), whilst being otherwise unrelated to absolute verbal or performance IQ or to a number of aspects of language or executive function. In the present ASD sample, both verbal and performance IQ were correlated to all three symptom domains ($r > .30$, $p \leq .006$), with high IQ scores indicative of less severe symptoms. No correlations existed between IQ discrepancy and these symptoms ($r < .1$), or between head size and any IQ measure ($r < .1$) though.

These findings partially replicate those of Joseph et al. (2002) in that social and communication impairment were associated with low verbal IQ, as well as extending this relationship to repetitive behaviours. Low performance IQ was also related to impairment in all three domains, a finding that was also present in only the preschool subset of children in Joseph et al.'s study. However, no associations were found between IQ discrepancy and symptom severity, or between head size and any IQ measure. Although these latter results differ from those of Joseph et al. and Deutsch and Joseph (2003), two notable differences in methodology should be considered. Firstly, Joseph et al. used a more direct measure of symptomatology than here, using the Autism Diagnostic Observational Schedule (ADOS: Lord et al., 2000) rather than a parental interview such as the 3Di. More likely than this to affect the correlations, however, is the use of a different intelligence measure; the WISC is used here rather than the Differential Ability Scales (DAS: Elliott, 1990). It is possible that the DAS non-verbal measure is less sensitive to individual differences than the WISC, accounting for the lack of correlation between performance IQ and symptom severity in the older children in Joseph et al.'s study. Indeed, the DAS non-verbal measure is composed of only two subtests as opposed to five in the WISC. A final possibility is that, as the current sample of children with ASD did not contain many individuals with verbal IQ significantly lower than performance IQ, this subgroup may have been driving the results of Joseph et al. and Deutsch and Joseph by co-occurrence rather than association.

2.4 Relating cognitive impairments to intelligence and symptom profiles

The remainder of this thesis studies the relationship between the three main cognitive domains suggested to be impaired in ASD and intelligence and symptom profiles.

Chapters 3, 4 & 5 detail the tests used to tap into each cognitive domain and examine group differences over a range of tasks, assessing each theory within this population.

Chapter 6 considers the effect of these cognitive impairments on intelligence tests, explores the relationship between these different cognitive domains, examines the patterns of

impaired abilities found in different children, and investigates the ability of each cognitive domain to predict group membership through discriminant function analysis. Chapter 7 assesses whether different cognitive impairments can predict the presence of certain behavioural symptoms through comparison of children with ASD who did or did not show each cognitive impairment, regression analyses and structural equation modelling.

Chapter 3: Mentalising

3.1 Theory of mind battery

Since Baron-Cohen, Leslie & Frith's (1985) paper, there has been an immense interest in the theory of mind (ToM) theory of autism. In this first paper, individuals with autism were hypothesised to have specific problems in understanding other people's mental states, an ability that has come to be known as mentalising. This hypothesis was tested by use of a 'false belief' paradigm, designed by Wimmer and Perner (1983), in which a character (Sally) places an object (marble) in a location (basket) but a second character (Ann) moves the marble to a different location (box) while Sally is out of the room. This sets up a situation in which Sally holds a false belief about the current location of the marble and the critical test comes when the child is asked to predict where Sally will look for the marble when she returns. Story understanding is controlled by posing memory (where was the marble in the beginning?) and reality (where is the marble really?) questions. In line with the novel prediction made by these researchers, children with autism were more likely to say that Sally would look for her marble in the box, where it actually was, hence being unable to correctly predict Sally's behaviour on the basis of her (lack of) knowledge.

In general, studies employing such paradigms have focussed on this one task, occasionally pairing it with other tasks, such as the 'unexpected contents' task (Perner, Leekam & Wimmer, 1987), or using multiple trials of the same basic task. Since it is possible to pass these tasks 50% of the time by chance, using a single trial may give a false representation of an individual's ability (although this has been argued against by showing that children don't choose a third irrelevant location; Baron-Cohen et al., 1985). Theory of mind understanding develops over many years in normally developing children so that increasingly complex mental state representations can be understood (Wellman & Liu, 2004); indeed, it has long been known that false belief task performance is dependent on

verbal mental age (Happé, 1995), a strong correlate of chronological age in normally developing children, and that second-order false belief tasks are passed by normally developing children at a later age than first-order false belief tasks (Baron-Cohen, 1989b). Use of a single trial or a few task repetitions therefore limits any information that could be available about more subtle individual differences in performance and provides only a narrow window on a particular developmental level (Wellman, Cross & Watson, 2001).

Wellman & Liu (2004) have therefore recently proposed a battery of seven scaled ToM tasks that undergo an ordered developmental progression in normal development (see Hughes et al., 2005; Steele, Joseph & Tager-Flusberg, 2003 for similar batteries). In the majority (75%) of their young normally-developing children, they found a consistent pattern in the order of tasks passed across different children, whereby a diverse desires task was easier than a diverse beliefs task, followed by knowledge access, contents false belief, explicit false belief, belief emotion and real-apparent emotion in that order (see Appendix 2 for details of the tasks). It is possible that a similar progression is present in children with ASD if ToM development is delayed, but this may be different if understanding of others' mental states progresses along a deviant compensatory pathway. In either case, the idea that such progression takes place may therefore go some way to explain why, even in the initial experiment (Baron-Cohen et al., 1985), a minority of individuals with ASD (20%) was able to pass a false belief task.

Given that this thesis attempts to investigate individual differences in performance, these seven ToM tasks (Wellman & Liu, 2004) were collated with four additional tasks known to be sensitive measures of theory of mind ability in ASD (see Appendix 2). This whole set of tasks therefore ranges from an extremely simple diverse desires task, requiring the child to predict the action of a character who holds a different desire to their own, to more complex second-order false belief tasks, involving the understanding of what one person falsely thinks about another person's thoughts. A simple penny-hiding task (Baron-Cohen,

1992) was also added to this sets of tests, which requires the child to conceal a coin from the experimenter and is thought to be a more implicit on-line test of ToM abilities.

3.1.1 Method

The 12 ToM tasks were administered to each child in a randomised order during the first testing session (see Table 3.1). Seven of these tasks were taken from Wellman & Liu (2004) and five additional tasks were added, which were expected to be more challenging for the older age groups involved here and therefore increase variability in responses. These additional tasks consisted of an implicit false belief task (Baron-Cohen et al., 1985), an interpretation false belief task modelled on the 'Smarties' contents false belief task (Luckett, Powell, Messer, Thornton & Schulz, 2002), two second-order false belief tasks requiring a higher level of meta-representational skill (icecream: Baron-Cohen, 1989b);

Table 3.1 ToM tasks administered to all children ordered by expected task difficulty; the shaded tasks indicate those taken from Wellman & Liu (2004).

Task	Task demands
diverse desires	state another's desire contrary to own
diverse beliefs	state another's belief contrary to own
knowledge access	understand that seeing leads to knowing
contents false belief	predict knowledge on basis of false belief
explicit false belief	predict behaviour on basis of false belief
implicit false belief	predict behaviour on basis of false belief
interpretation false belief	predict behaviour on basis of knowledge
belief emotion	predict emotion on basis of false belief
real-apparent emotion	predict apparent false emotion
second-order false belief (puppy)	predict behaviour on basis of false belief about another's false belief
second-order false belief (icecream)	
penny hiding	actively conceal visual information

(puppy: Sullivan, Zaitchik & Tager-Flusberg, 1994) and a penny hiding task (Baron-Cohen, 1992). It was expected that the second order false belief tasks would be harder than all other tasks as they involved a higher order of meta-representation, that the additional implicit false belief task would be harder than the explicit false belief task as the false belief would need to be inferred from the situation, and that the interpretation false belief task would be harder than the contents, explicit and implicit false belief tasks as it involved a conceptual false belief rather than a physical false belief. The penny hiding task, whilst quite simple, was also expected to be challenging as it required the child to not only reason about another person's thoughts but also to actively perform in such a way as to take this reasoning into account.

The first 11 tasks consisted of situations in which: a character and the child had diverse desires or diverse beliefs; where a character had a lack of knowledge, a false belief or a false belief about another character's false belief; or where a character wanted to create a false belief in others. In all cases, the child was asked to predict the knowledge of, behaviour of or emotion felt by another character on the basis of their mental state; this answer was marked as a pass or fail (1 or 0). In all but the three simplest tasks, the child was also asked to justify why they had made that prediction; this was marked as a correct mental state justification (eg. 'because she doesn't *know* it's in the box'), a correct non-mental state justification (eg. 'because she left it in the basket'), or an incorrect justification (eg. 'because no-one stole it apart from Anne') (1, 0.5 or 0 respectively). This additional scoring aimed to check for false positive responses when the child was achieving the correct answer by guessing, and also was expected to increase the variation in responses with the aim of avoiding ceiling effects.

All of these tasks were presented with accompanying props or pictures to help the child to comprehend and engage with the scenarios. Control questions were administered (and prompt questions during the more complex tasks) in order to check for comprehension of

the situation. If a child failed a prompt question, the story was repeated from the beginning until the child gave the correct answer; this situation was extremely rare and comprehension was generally good. It was also rare for control questions to be failed; on the few occasions it did occur, it was on memory control questions that required the child to acknowledge that they themselves had initially had a false belief and therefore required a degree of mental state understanding; responses to mental state test questions were therefore not excluded on this basis.

The penny-hiding task involved 6 trials during which the experimenter modelled hiding the coin to the child, followed by 6 test trials during which the child was encouraged to hide the coin. The task was marked out of a total of 6, with one point for each trial performed by the child. One point was awarded for perfect performance, whereby the experimenter was unable to tell which hand the coin was hidden in. Half a point was given if the child made no obvious mistake but held their hands asymmetrically, in such a way to make it obvious which hand the coin was in. The child gained no points if they failed to deceive the experimenter, by either not hiding both hands behind their back whilst choosing which to hide it in, bringing only one hand out from behind their back, not closing the empty hand, or leaving the coin in sight of the experimenter during a critical part of the procedure. No points were deducted if the child played a trick on the experimenter, eg. by placing the coin on their chair and bringing out two empty hands, so long as the experimenter was unaware that a trick had been played until the end of the trial.

In total, the former 11 'reasoning' tasks were scored together out of 19 and the 'on-line' penny-hiding task was scored out of 6, hence a combined maximum score of 25 for the whole battery of tasks was possible. Given that the penny hiding task differed from the other tasks both in the nature of the task and in its task demands, it was of interest to see how results from this test differed from the remaining tasks and so scores for this task will be reported separately as well as in the combined total.

3.1.2 Results

As can be seen from Table 3.2, performance on the reasoning tasks differed between the groups with the ASD group performing more poorly than the controls. After accounting for age, verbal and performance IQ (in ASD group, total score x vIQ: $p < .001$; total score x pIQ: $p = .18$; total score x age: $p > .81$), this difference was found to be significant ($F(1,79) = 5.740, p = .019$). There was a trend for the performance of the ASD group to be lower than that of the control group on the penny-hiding task, although this difference did not reach significance ($F(1,79) = 3.263, p = .075$); unfortunately there was a floor and ceiling effect on this task in both groups. However, this test still added variance to the total ToM battery score as the group difference was stronger than for the reasoning tasks on their own ($F(1,79) = 8.195, p = .005$).

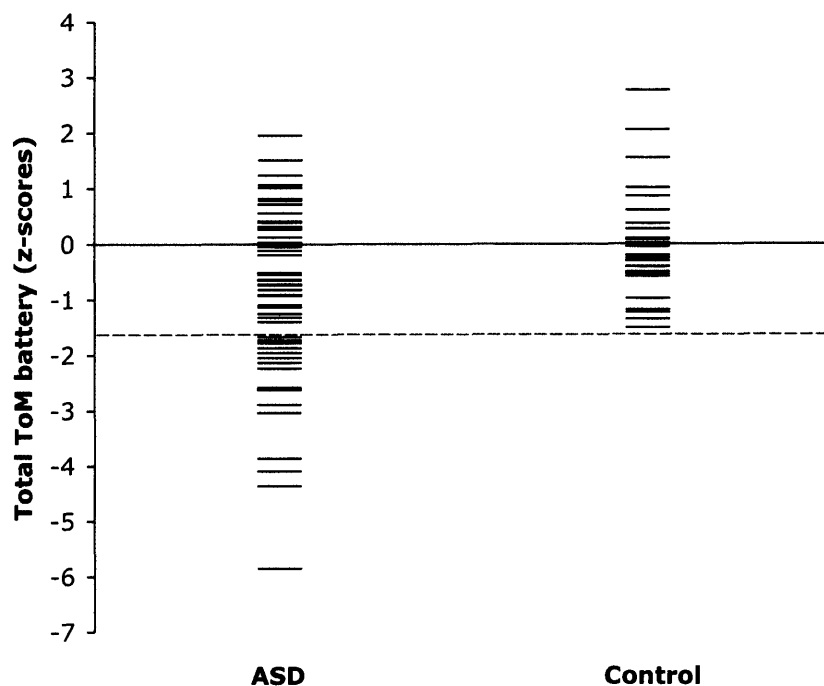
Observation of the range of scores on the reasoning tasks and total ToM battery indicates that these differences arise due to the ASD group having a broader range of scores, spanning that of the controls but extending lower with a tail of poor performers. This indicates that at least some children were performing at age and intelligence appropriate levels within the ASD group. Indeed, this is supported by the individual performances that can be seen in Figure 3.1; 32% of children in the ASD group could be categorised as

Table 3.2 Means (and standard deviations) with *range* for the ToM battery.

	Control group	ASD group
Reasoning tasks (19 max) *	15.1 (1.5) 11–17.5	12.5 (3.9) 2–17
On-line penny-hiding (6 max)	3.8 (1.7) 0–6	2.9 (1.7) 0–6
Total ToM battery (25 max) **	19.0 (2.5) 13.5–23.5	15.3 (4.5) 3–21.5

* $p < .05$, ** $p < .01$

Figure 3.1 Individual performance on the ToM battery after accounting for age and IQ; the position of the x-axis indicates the control mean and the dotted line illustrates the cut-off for the 5th percentile of control performance.



having deviant performance below the 5th percentile of control performance (see section 6.2 for further details of this methodology).

Given that the normally-developing children here were older than in Wellman & Liu (2004), they mostly answered the test questions correctly, making it difficult to see a developmental pattern in the results. Despite this, two tasks were answered correctly by less than 90% of the controls; real-apparent emotion (86%), and icecream second-order false belief (79%) and can therefore be considered the most challenging (see Table 3.3). Within the autism group, a higher proportion of incorrect answers was given to the test question, as expected. The tasks could therefore be ranked in order of difficulty according to the percentage of individuals who answered the test question correctly (see Table 3.3). The two hardest tasks for the controls were also the hardest for the ASD group. The overall order can be seen to be similar to that of the young normally-developing children in

Table 3.3 Percentage of correct responses to test question for the reasoning tasks, ordered by ASD group performance.

	Control group	ASD group
diverse desires	100	96
diverse beliefs	100	91
knowledge access	100	89
belief emotion	96	89
contents false belief	100	88
explicit false belief	96	84
implicit false belief	93	82
second-order false belief (puppy)	96	81
interpretation false belief *	96	77
real-apparent emotion	86	65
second-order false belief (icecream) *	79	58

* $p < .05$

Wellman & Liu (2004), with one notable exception: the children with ASD found the belief-emotion task to be as easy as the knowledge access task.

Comparisons between the numbers of correct responses for each test question in the two groups were calculated. χ^2 tests indicated that only two of the more challenging tasks differentiated the groups from each other (interpretation false belief, $\chi^2(1)=4.814$ $p=.028$; second-order false belief (icecream), $\chi^2(1)=4.509$, $p=.034$), although it should be noted that these did not take the IQ differences between the groups into account.

In order to investigate the number of children in the current sample who showed the expected developmental progression, only the seven tasks used by Wellman & Liu (2004) were studied. Only six of the 27 controls failed one of these tasks, of whom four failed the hardest real-apparent emotion task, conforming to the developmental pattern reported by Wellman & Liu, whilst the remaining two controls failed either the second or third hardest

task. When looking at all 11 reasoning tasks ordered by percentage of correct responses in the control group, 11 of the 27 children gave at least one incorrect answer with some children giving up to three incorrect answers, although only three of these children conformed to the task order (four children when optimally reordering tasks to give maximum number of children conforming). However, given that nine of these children failed only one task, this may simply be noise in the data or may indicate that the additional tasks were of similar difficulty making it hard to predict the order in which children would pass them.

Within the ASD group, 25 of the 57 children failed at least one of the seven tasks taken from Wellman & Liu (2004), with one child failing five out of seven tasks. Eight of these 25 children conformed to the pattern seen in Wellman & Liu, indicating that the normal developmental pattern is not seen in these children. However, even if the tasks are reordered so that the belief emotion task is given equal ranking to the knowledge access task as mentioned above, only 10 of the 25 children conform to this order and in fact this is the optimal order to which the highest number of children conform. When looking at the full 11 reasoning tasks, 38 children failed at least one task, with one child failing nine of the 11 tasks. 11 of these 38 children conformed to the task progression when ordered by difficulty (maximum of 13 children when optimally reordering tasks), indicating that the majority of children with ASD do not all follow the same pattern of developmental progression as each other.

The answers to the justification questions provided more information from both groups, as they did not explicitly ask for a mental state answer and so there was greater variation in responses. From easiest to hardest in the ASD group, these can be seen in Table 3.4. It can be seen that the order of difficulty differs between responses to the test and justification questions. Some tasks were highly likely to elicit a mental state response, for example the explicit false belief task in which the appropriate mental state answer was

Table 3.4 Percentage of mental state justifications given for the ToM tasks, ordered by ASD group performance.

	Control group	ASD group
explicit false belief *	93	65
real-apparent emotion	30	28
second-order false belief (puppy)	22	18
belief emotion	19	16
implicit false belief	7	14
interpretation false belief	22	11
second-order false belief (icecream) **	33	4
contents false belief	4	4

* $p < .05$, ** $p < .01$

integrated into the story, whilst others tended to produce non-mental state responses, for example the contents false belief task which has an obvious physical-state answer. The groups only differed on two tasks however; the explicit false belief task ($\chi^2(1)=4.514$, $p=.034$) and the second-order false belief (icecream) task ($\chi^2(1)=10.019$, $p=.002$). Again, it should be noted that these differences have not been corrected for intelligence levels.

When looking at all the data combined for each task, including the test and justification questions, the order of difficulty is similar to that expected from the predicted task difficulty and is more similar to the order for the test than justification question (see Table 3.5). The only exception is that the explicit false belief task was found relatively easy, mostly due to the high frequency of mental state terms used in the justification question. The control group consistently performed better than the ASD group across all tasks. Differences between the groups on this total score were found on three tasks: the explicit false belief task ($U=557.5$, $p=.008$), the interpretation false belief task ($U=566.5$, $p=.025$) and the second-order false belief (icecream) task ($U=427.5$, $p=.001$). Whilst these tests

Table 3.5 Mean total scores for each ToM task including a justification question (2 max).

	Control group	ASD group
explicit false belief **	1.89	1.49
belief emotion	1.54	1.38
contents false belief	1.50	1.31
implicit false belief	1.43	1.27
second-order false belief (puppy)	1.50	1.22
interpretation false belief *	1.50	1.18
real-apparent emotion	1.41	1.06
second-order false belief (icecream) **	1.30	0.80

* $p < .05$, ** $p < .01$

appear to be most sensitive in this sample of children, the consistently low performance across tests in the ASD group justifies the use of a combined battery of such measures.

3.1.3 Discussion

The use of a battery of theory of mind tasks, comprising of a range of more traditional tasks such as those involving false beliefs as well as a more naturalistic and on-line penny-hiding task, replicated the often reported finding of theory of mind difficulties in individuals with ASD. Whilst few individual tests in this battery produced group differences, the overall battery produced a strong effect, supporting its use. However, the distribution of scores on the overall battery indicated that only a proportion of children were performing outside the control range. This suggests that some children were performing at and above age- and intelligence-appropriate levels and therefore either had no mentalising problems or were compensating to such a degree that their performance was comparable to that of normally-developing control children.

Although few of the individual tests produced group differences, the tasks that did tend to were those that were hardest for the controls, indicating that there may have been a ceiling effect on other tasks. Interestingly, one of the easier tasks, explicit false belief, did produce a group difference, most likely because the control performance was so consistently high across participants that any differences in the ASD group were amplified. Given that the reasoning tasks are two alternate forced choice tasks, they are vulnerable to guessing and therefore passing by chance and so a battery of many such tasks is crucial to retain sensitivity.

The stable developmental progression seen by Wellman & Liu (2004) in younger children was present in very few of the children here with ASD who were not performing at ceiling, consistent with a more recent study by Peterson, Wellman & Liu (2005). After collecting the current data, Peterson et al. published a study in which they found that children with ASD did not conform to the order of task progression seen in normally-developing children. The convergence of this study with the current results is remarkable despite the broader age range, lower IQ levels, restriction to full autism diagnoses, and the use of only 5 of the tasks in Peterson et al.'s sample. Interestingly, exactly the same finding of relatively good performance on the belief emotion task was found by these authors as well. Furthermore, Peterson et al. found late-signing deaf children to progress through the same order of tasks as the normally developing children, despite being severely delayed in their ToM understanding, indicating that the abnormal progression seen in the ASD group is not just the result of delay.

This supports the idea that the development of the current ASD group's understanding of other people's behaviour in terms of mental states was unlikely to be only delayed, but also to follow a deviant developmental path. It is likely that other skills and abilities, as well as general knowledge and experience of how the social world works, can allow children to correctly solve some theory of mind scenarios, in the absence of a true understanding of

why this is the case. Indeed, few children with ASD who were able to give correct answers to the test questions were also able to spontaneously produce mental state justifications for these answers, although this was also the case to a lesser extent with the control children. Furthermore, examination of the task demands for the belief emotion task help to reveal why this task may have been relatively easy for the AS group; indeed, the ToM component of this task is actually only as complex as the knowledge access task. This task may be difficult for younger children due to the additional need to understand how emotions relate to mental states. For the current older children with ASD, as the understanding of basic emotions is thought to be intact in autism (Castelli, 2005), such understanding would have been acquired at the normal stage of development and this component of the task would therefore have been relatively easy for them. Hence, any problems encountered are likely to have been purely with the ToM component of the task, making it no more difficult for them than the knowledge access task.

If the ability of some children with ASD to pass theory of mind tests relies on other skills and abilities and more importantly on experience of how people behave in certain situations, this would differ wildly between different children depending on their individual circumstances. Some children may therefore be able to pass seemingly 'harder' tasks whilst failing easier ones and the particular pattern of performance would differ between different children. This reinforces the need for batteries of tests rather than single test scenarios in order to provide an accurate measure of individual performance.

Regardless of the ability of some children with ASD to pass a proportion of the tasks, it must still be acknowledged that a number of children were performing consistently well, indicative of a lack of mentalising problems. As already mentioned, it is possible that many of these tests produce ceiling effects and therefore are not sensitive enough to pick up more subtle problems in these children. More recently, new tests that tax such individuals' capabilities have therefore been sought in an attempt to reveal any mentalising problems

that do exist (Abell, Happé & Frith, 2000; Baron-Cohen, Jolliffe, Mortimore & Robertson, 1997b; Baron-Cohen, O'Riordan, Stone, Jones & Plaisted, 1999a; Channon, 2004; Heavey, Phillips, Baron-Cohen & Rutter, 2000; Kaland et al., 2002; Kleinman, Marciano & Ault, 2001; Klin, 2000; Rutherford, Baron-Cohen & Wheelwright, 2002). One such advanced test of mentalising, the Strange Stories (Happé, 1994), has revealed a deficit in the understanding of more complex mental states by high-functioning individuals with ASD, even those passing second-order false belief tasks. A number of studies have now replicated the finding that the Strange Stories reveal autism-specific impairments, even among high-functioning participants (Baron-Cohen et al., 1997b; Gillott, Furniss & Walter, 2001; Jolliffe & Baron-Cohen, 1999a; Kaland et al., 2005).

3.2 Strange Stories

The original set of 24 Strange Stories required the participant to answer mental-state questions relating to stories involving pretence, jokes, lies, white lies, misunderstandings, false appearances, sarcasm, figures of speech, double bluffs, forgetfulness and contrary emotions (Happé, 1994). These were accompanied by a smaller set of 6 control stories requiring understanding of physical states. However, all participants in Happé's study performed at ceiling on these control stories, which were not equated for difficulty with the mental state stories. It is therefore possible that the poor performance of the autism group on the mental state questions could have been due to general comprehension problems or the need to integrate information and make inferences across the text, which were not revealed by the easier physical state stories. In fact, comprehension problems and problems with inference are a common feature of high-functioning children on the autism spectrum (Norbury & Bishop, 2002) and may possibly be explained by the central coherence theory. Problems in integrating information, one aspect of so-called 'weak central coherence', are therefore an alternative explanation for failure on the mental state

stories, a possibility discussed by both Happé and Jolliffe & Baron-Cohen (1999a) in considering their findings.

In subsequent development of the Strange Stories for functional brain imaging (Fletcher et al., 1995), a subset of the 8 most demanding mental state stories was selected, involving understanding of double bluff, white lie, persuasion and misunderstanding. To test for the specificity of any difficulty with these stories, 8 control stories were created that required reasoning about physical states only, and were of comparable difficulty in healthy young adults. An additional set of 8 passages of unlinked sentences was also created; here participants were required to recall a specific fact from one sentence. All sets therefore involved people and required attention to sentence meaning, memory and question answering, while the mental and physical state sets also required the integration of information between sentences and inference from implicit information, and only the mental state set required mentalising.

These well-controlled stimuli enabled a 'mentalising network' in the brain to be revealed (see section 1.1.2). Reduced activation in this network was also seen in adults with Asperger syndrome (Happé et al., 1996). Here five adults were chosen to be extremely high-functioning and to perform well at these tasks, to allow valid scan comparisons. Despite this, the behavioural results still showed a significant difference between the groups on the mental state stories but not on the physical or unlinked stories. However on closer observation of the results, the adults with Asperger syndrome did perform non-significantly worse than controls on the physical state stories and better on the unlinked sentences. As participant numbers were small, these non-significant effects might have been more pronounced with a larger sample. In fact a recent study of 20 high-functioning children with autism spectrum disorders (Brent, Rios, Happé & Charman, 2004), found a similar pattern of performance in these mental and physical state story sets, showing significantly worse performance than age- and IQ-matched controls on the mental state set,

and non-significantly worse performance on the physical state set. Furthermore, an unpublished study with even larger group sizes has indicated that both children and adults with ASD have significant difficulties with both mental and physical state story types, although to a significantly greater extent with the mental state stories. Importantly, the groups performed equally well with the unlinked sentences (White, Hill, Happé & Frith, in prep).

Results from the use of these stories supports the idea that individuals with ASD have a specific difficulty in understanding other people's mental states, as evidenced by poor performance on the mental state stories. From the literature however, it is difficult to determine why individuals with ASD might also find the physical stories problematic. It is possible that, as already suggested, individuals with ASD have more general difficulties in integrating information and making inferences, possibly due to the role of weak central coherence in text comprehension (although there are also other possible reasons for a general comprehension deficit). This would affect performance on the physical state stories but also further affect mental state story performance. The unlinked sentences however, would be unaffected as these do not require information to be integrated between sentences or for inferences to be made.

A different interpretation could be that individuals with ASD find processing any information about animate beings (including animals) problematic; indeed, the recognition of biological motion from random dot kinematograms has been shown to be impaired in children with ASD (Blake, Turner, Smoski, Pozdol & Stone, 2003). Moreover, the superior temporal sulcus (STS), the main brain area thought to process biological motion (Bonda, Petrides, Ostry & Evans, 1996), has been shown to be functionally and structurally abnormal in individuals with ASD (Boddaert et al., 2004; Pelphrey, Adolphs & Morris, 2004; Waiter et al., 2004). Interestingly, the STS has been found to be activated in normal adults when reading both the mental and physical state Strange Stories (Fletcher et al.,

1995; Gallagher et al., 2000), even though these do not involve any direct visual biological motion. Furthermore, the same neural areas of medial prefrontal cortex are activated for both animals and humans when thinking about their mental as opposed to physical states (Mitchell, Banaji & Macrae, 2005), indicating that information about all animate agents may be processed in the same way. Indeed, one study looking at visual recognition memory in high-functioning adults with ASD found that impairments were present in this sample not just for human face stimuli but also for other potential agents including animals, and non-living agents such as vehicles, whilst showing retained memory for stimuli such as buildings (Blair, Frith, Smith, Abell & Cipolotti, 2002).

One further possibility is that the presence of humans in the physical stories might make physical state inferences slightly more difficult for individuals with ASD; the general population might use mentalising to aid their understanding of situations involving people, even when mental state processing is not explicitly required. Indeed, there is evidence to show that the network of brain areas thought to be involved in mentalising are also activated when making semantic judgements about people as opposed to objects (Mitchell, Heatherton & Macrae, 2002), although to a lesser extent than when performing an explicit mentalising task (Fletcher et al., 1995).

Table 3.6 Task requirements for the different story sets.

	Mental	Human	Animal	Natural	Unlinked
Explicit mentalising	✓	✗	✗	✗	✗
Thinking about humans	✓	✓	✗	✗	✗
Thinking about animate agents	✓	✓	✓	✗	✗
Integration of info across text	✓	✓	✓	✓	✗
Sentence comprehension	✓	✓	✓	✓	✓

Two new sets of physical stories involving either animals or natural events were therefore added to the original three sets of stories. Like the human-physical stories, these sets require the integration of information and inference but do not involve humans. They therefore allow us to investigate the level at which children with ASD have problems with these stories (see Table 3.6).

3.2.1 Method

The mental and human-physical story sets and the unlinked sentences (Fletcher et al., 1995), as well as the new animal-physical and natural-physical story sets each consisted of 8 stories. Full texts of all stories can be found in Appendix 3. A number of changes were made to the existing story sets in order to make them more appropriate for the age range and hypotheses. Firstly, only 4 of the human-physical stories from Fletcher et al. (1995) were used; a previous experiment found the other 4 to be too difficult for this age group (White et al., in prep). The 4 easiest stories were therefore selected, based on an independent sample of normally-developing children (White et al., in prep) and combined with 4 new easier stories. This modified set of human-physical stories was matched to the mental state set for difficulty. Similarly, the novel animal- and natural-physical stories were designed to match the other sets in difficulty, as confirmed by pilot testing.

In addition, the unlinked sentences used in Fletcher et al. (1995) differed from the other stories in the type of question that was asked; these involved closed (requiring the answer 'yes' or 'no') rather than open questions. This made it difficult to compare performance on these stories to the other story sets; if a child was guessing, they would perform at chance (half marks) on the unlinked sentences whilst if they were guessing on the other story sets, they would score at 'floor' level. The questions were therefore adapted from closed questions, such as 'Is Mary's birthday in February?' to open questions, such as 'When is Mary's birthday?'.

All stories were presented on a laptop computer using E-prime software, both as written and spoken text in order to aid concentration. Each story was pre-recorded by an adult male speaker and lasted approximately 30 seconds. The full text of each story was presented on the screen and the child was invited to follow the words whilst listening to the recording. 500 msecs after the spoken text had ended, a question about its content was presented on the screen and simultaneously spoken by the pre-recorded voice. As soon as the child began to give an answer, the experimenter pressed a button on the keyboard to record reaction time (the time from the end of the spoken question until the child began their response). In cases where the child began to speak but stopped immediately to think again before answering, a different button was pressed when they began their answer a second time, in order to indicate that this was the true reaction time. The same procedure was also available one additional time, in case a child made up to two false starts. The experimenter recorded the child's answer verbatim on paper. The child was then invited to press the spacebar to listen to the next story.

The stories were presented in blocks of 8 stories, one block for each story type. Other tasks were conducted between the blocks to aid concentration. The mental, human-physical and unlinked sentences were presented during the second testing session and the animal- and natural-physical stories during the third session. The order of the blocks within each session and the order of the stories within each block were randomised.

The accuracy of each verbal response was rated on a 0-2 scale by two researchers, with 0 given for incorrect answers or when the child did not know the answer, 1 for partially correct answers and 2 for correct answers (scoring schemes are given in Appendix 3).

Total scores for each story type were therefore marked out of 16. Good agreement (Cohen's kappa=.65, indicating substantial agreement; 78% of responses in agreement; only 3% of responses rated as 0 by one rater and 2 by other) was reached between the author and a co-rater blind to group based on a sample of answers to each question (20% of

answers selected randomly for each question). Only the author's scoring was used in the subsequent analysis.

3.2.2 Results

As can be seen from Table 3.7, accuracy across the different story sets was generally lower in the ASD group than the control group. Similarly to the false belief battery, the range of scores in the ASD group was wider, with an additional tail of lower scores compared to the controls. As before, on an individual level this indicates that only a proportion of the children had problems with these tasks. Interestingly, no such tail was seen for the jumbled stories but this may be due to floor effects. Figure 3.2 reveals the pattern of individual performance.

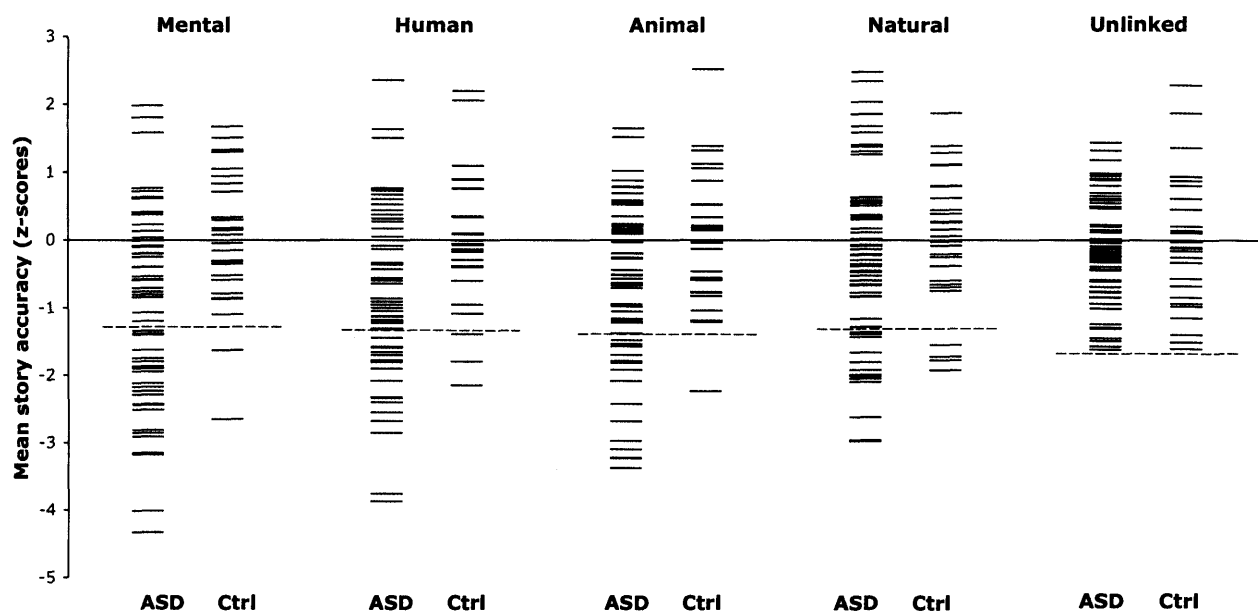
A 5x2 repeated measures ANCOVA, entering age, verbal and performance IQ as covariates (in ASD group, stories x vIQ: $p \leq .002$; stories x pIQ: $p \leq .056$; stories x age: $p > .22$), and comparing story type by group, revealed a main effect of story type ($F(3.9,$

Table 3.7 Means (and standard deviations) with *range* for the Strange Stories results.

	Accuracy (16 max)		Reaction time (secs)	
	Control group	ASD group	Control group	ASD group
Mental **Acc *RT	11.5 (2.8) 6–16	7.5 (4.2) 0–15	2.6 (2.3) .6–12.5	4.2 (4.4) .9–27.1
Human **Acc *RT	11.3 (3.1) 5–15	7.7 (4.1) 0–15	3.1 (2.4) .7–10.8	4.4 (3.4) .9–18.3
Animal **Acc	12.5 (2.8) 7–16	8.5 (4.7) 0–16	2.4 (1.1) .6–5.3	3.0 (1.8) .5–8.3
Natural	11.4 (3.6) 4–16	8.5 (4.9) 0–16	3.2 (1.9) .8–8.3	3.7 (1.9) .7–9.2
Unlinked	8.7 (3.4) 3–16	7.3 (2.7) 2–13	3.4 (3.0) .6–13.4	3.2 (2.0) .7–8.7

* $p < .05$, ** $p < .01$

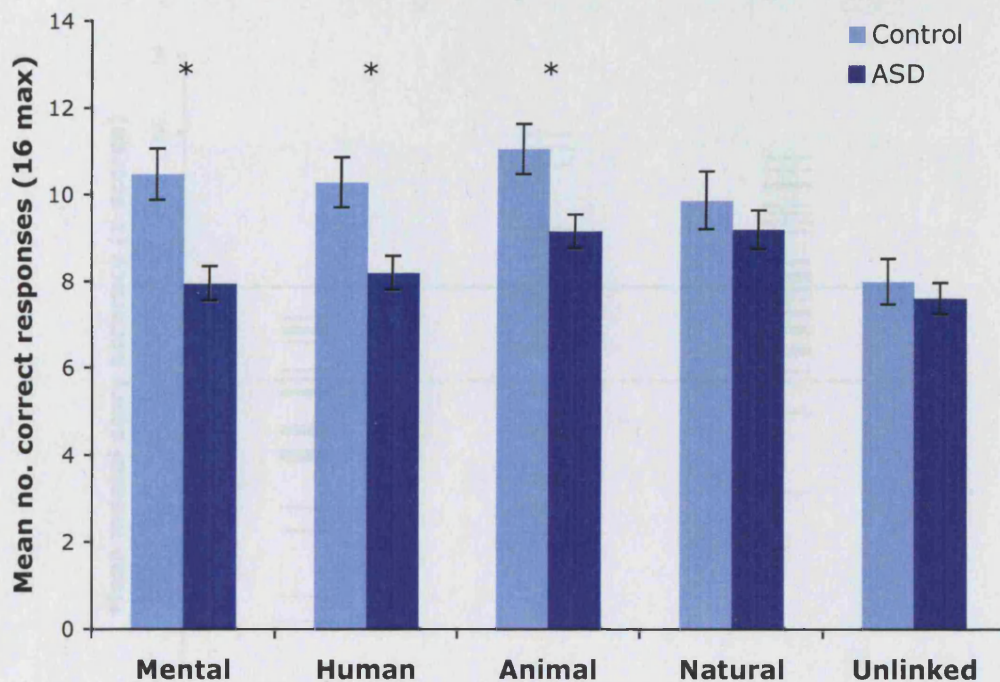
Figure 3.2 Individual performance on the different sets of Strange Stories after accounting for age and IQ; the position of the x-axis indicates the control mean and the dotted line illustrates the cut-off for the 5th percentile of control performance.



311.2)=4.81, $p=.001$), with the unconnected sentences being more difficult than the rest of the stories ($p<.03$). There was also a main effect of group ($F(1, 79)=9.59, p=.003$), with the ASD group generally performing worse than the controls. Furthermore, a significant interaction was found between story type and group ($F(3.9, 311.2)=2.41, p=.050$), with the ASD group performing worse than the control group only on the mental ($p=.001$), human-physical ($p=.004$) and animal-physical ($p=.009$) stories. Weak *post hoc* interactions (story type by group) were present between each of these three story sets and the two remaining story sets ($.030 < p < .096$). No other *post hoc* pairwise interactions were found (see Figure 3.3).

As the reaction time data were not normally distributed, non-parametric tests were used to compare the groups (it should be noted that age and IQ were therefore uncorrected in this analysis). As can be seen from Table 3.7 (p96), performance was similar to that seen in the accuracy data: slightly slower reaction times in general in the ASD group and a wider range of reaction times, including extremely slow performance. This slowness is indicative of

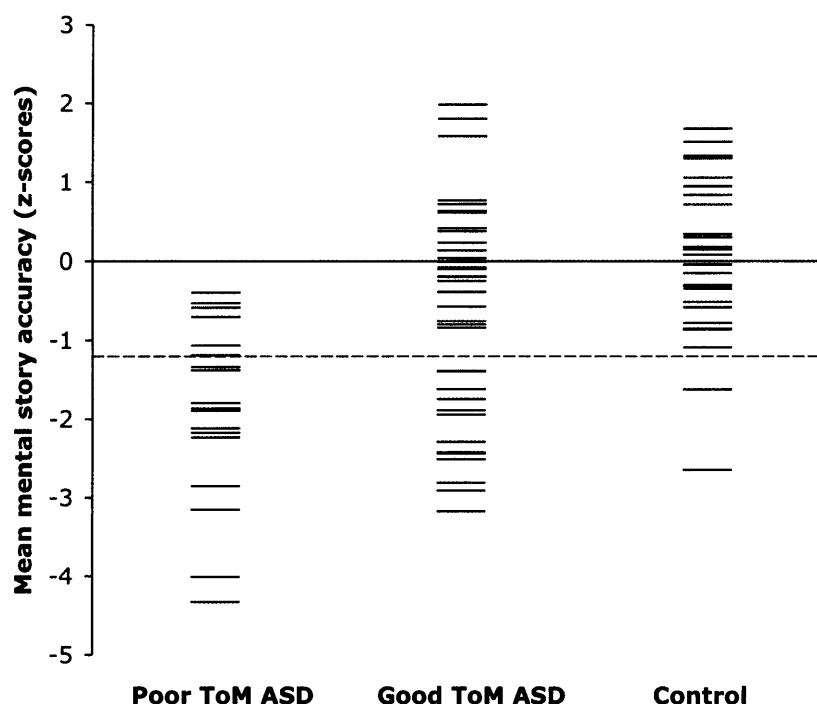
Figure 3.3 Performance on story sets by control and ASD children, corrected for age, verbal and performance IQ; error bars are standard errors; * indicates a significant group difference.



requiring a longer thinking time to produce an answer. Again, only a proportion of the children showed extreme reaction times, however. Mann-Whitney U-tests revealed group differences for only the mental ($U=526, p=.044$) and human-physical stories ($U=494, p=.020$).

In order to investigate further the problems that the ASD group had with the mental, human and animal stories, the ASD group was split into two subgroups on the basis of their performance on the ToM battery used in the first part of this chapter. Any child performing more than 1.65 SD below the corrected control mean was defined as having impaired ToM (18 children) and the remaining children as having relatively good ToM (39 children) (for further details of this methodology, see chapter 6). These groups did not differ from each other on age ($t(55)=1.124$), verbal IQ ($t(55)=.146$) or performance IQ ($t(55)=.346$). Individual performance split into these groups can be seen in Figure 3.4; while there is overlap between those children with ASD with poor or relatively good ToM,

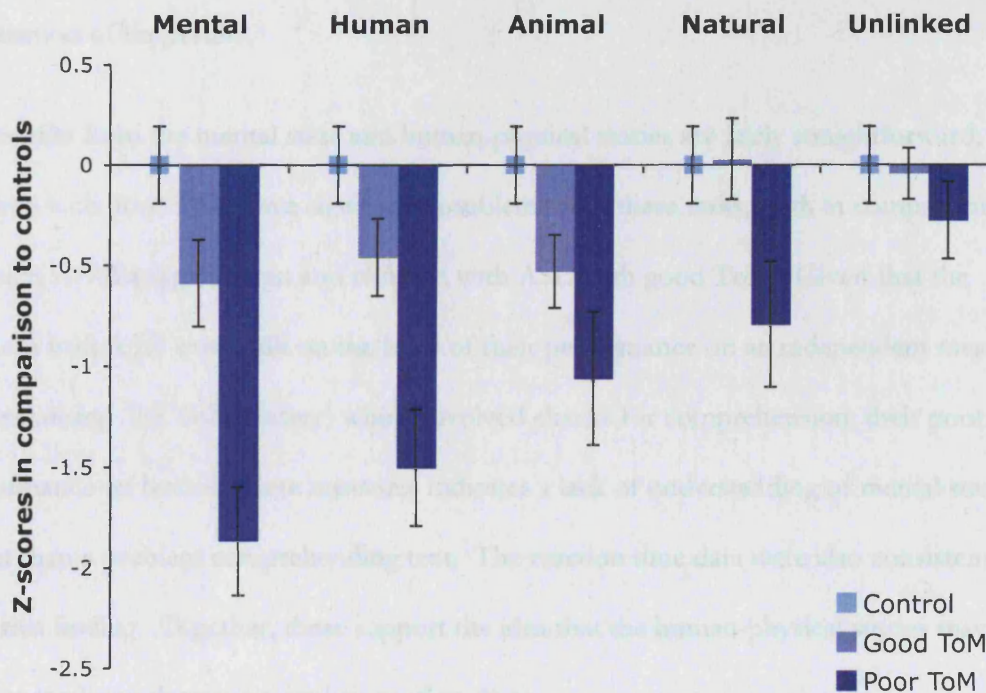
Figure 3.4 Individual performance on the mental state Strange Stories after accounting for age and IQ; the ASD group is split by performance on the ToM battery; the position of the x-axis indicates the control mean and the dotted line illustrates the cut-off for the 5th percentile of control performance.



it can clearly be seen that every child with poor ToM as defined by the ToM battery had performance below the control mean on the mental state Strange Stories, whilst the range of scores in the ASD group with relatively good ToM is similar to that in the control group. Indeed, the mental state Strange Stories were highly correlated with the ToM battery in the ASD group (partial correlations accounting for age & IQ: $r = .50, p < .001$).

A 5x3 repeated measures ANCOVA comparing story type by group was again performed. As before, this produced main effects of story type ($F(4.0, 309.8) = 5.02, p = .001$) and group ($F(2, 78) = 11.96, p < .001$), as well as an interaction ($F(8.0, 309.8) = 2.06, p = .040$). Tests of within-subjects contrasts indicated that the interaction produced a linear relationship between the groups across the different story types. *Post hoc* tests revealed that this interaction resulted mainly from the poor ToM subgroup performing worse than the controls on the mental ($p < .001$), human ($p < .001$) and animal ($p = .005$) stories, although from Figure 3.5 their performance was also slightly lower on the natural stories. The ASD

Figure 3.5 Performance on story sets by control and ASD children, corrected for age, verbal and performance IQ; scores are presented as z-scores relative to the control group; error bars are standard errors.



subgroup with relatively good ToM did not differ significantly from the controls in their performance although, as can be seen from Figure 3.5, their performance was slightly lower on the mental, human and animal stories. The relatively good and poor ToM subgroups differed significantly in their performance on the mental ($p=.001$) and human ($p=.006$) stories, as well as the natural stories ($p=.036$).

3.2.3 Discussion

Contrary to other studies using the Strange Stories, the current results do not provide support for the presence of a specific deficit on the mental state stories in ASD. Rather, the children with ASD were less likely to give accurate answers to the mental, human and animal stories, indicative of a more generalised impairment. An initial interpretation of these results might indicate that children with ASD have problems understanding stories which involve animate beings of any sort, as suggested in section 3.2.1 as one possible hypothesis. However, the second analysis, after dividing the ASD group into those with

good and poor performance on the theory of mind battery from the first part of this chapter, as well as the reaction time data, indicates that there may be an alternative explanation of the results.

The results from the mental state and human-physical stories are fairly straightforward; children with poor ToM have significant problems with these tasks, both in comparison to normally developing children and children with ASD with good ToM. Given that the children with ASD were split on the basis of their performance on an independent measure of mentalising (the ToM battery) which involved checks for comprehension, their poor performance on both of these measures indicates a lack of understanding of mental states rather than a problem comprehending text. The reaction time data were also consistent with this finding. Together, these support the idea that the human-physical stories may elicit mentalising despite no explicit need to do so.

Performance on the animal and natural stories is more difficult to interpret however, as the results produced a less consistent pattern. While the children with ASD and poor ToM appeared impaired relative to controls on the animal stories, their performance was not significantly worse than that of the children with ASD and good ToM. Despite this less clear cut result, the same pattern of performance can be seen on this set of stories as on the mental and human story sets. This indicates that any differences are likely to come from the same underlying deficit, a mentalising impairment, whilst possibly having a slightly lesser effect on this task relative to the mental and humans sets. This would suggest that it is normal to anthropomorphise non-humans in order to understand their actions in terms of mental states. Indeed, previous work from two independent groups of researchers has found that individuals with ASD have difficulties attributing mental states to animated triangles whilst controls appear to do this instinctively and with ease (Abell et al., 2000; Castelli, Frith, Happé & Frith, 2002; Klin, 2000).

Furthermore, the analysis of the interaction between story type and group revealed a linear relationship in performance differences between the groups across the story types, which can clearly be seen in Figure 3.5. This suggests that each consecutive set of stories, which were designed to be one more conceptual step away from the mentalising stories (see Table 3.6, p93), were less impaired in the poor ToM group than the previous set. Hence, the lower the likelihood that controls would use mentalising to aid their comprehension, the less impaired the poor ToM group were. Although not significant, this is reminiscent of the trend seen in previous studies for the mental state stories to be more difficult for individuals with ASD than the human-physical stories.

However, the fact that a similar pattern can be seen on the natural stories compared to the other stories is puzzling, this time with the children with poor ToM performing significantly worse than the remaining children with ASD and non-significantly worse than controls. Conversely, all the groups performed similarly on the unlinked sentences, revealing that sentence comprehension is intact in these children with ASD, regardless of ToM ability. This impairment on the natural stories is more difficult to explain as a mentalising deficit as no animate beings were involved in these stories; indeed, the children only needed to integrate information between sentences and make inferences from implicit information. One possible explanation could be that, despite similar verbal intelligence, the poor ToM group still had worse text comprehension in addition to their mentalising problems, possibly as a result of having poor ToM.

Overall, this experiment indicates that a mentalising impairment may have more widespread effects than previously thought. Indeed, mentalising ability appears to affect understanding of human and animal actions even when these do not explicitly require understanding of other people's mental states.

3.3 Conclusion

Both experiments together indicate the presence of a deficit in mentalising ability in some children with ASD. Two independent sets of ToM tasks were used and were found to discriminate the ASD from control group, as well as capturing the variance within these groups. While it was possible that ceiling effects may have been limiting performance in the ToM battery, it seems unlikely that this was the case with the Strange Stories as these stories were more complex and challenging and no ceiling effects were observed. The sets of Strange Stories also allowed for an extension and more detailed analysis of the impairments revealed by the ToM battery. Such a combined battery of tasks, rather than individual tasks, appears to be necessary to capture ToM impairment at an individual level and development of this sort of battery will be crucial for future clinical assessment.

The ToM deficit revealed by this battery of tasks appears to have a quantitative effect on mentalising ability as development is both delayed and deviant. A subgroup of children with ASD had distinct and severe ToM impairments, whilst other children appeared to have relatively preserved ToM task performance; those children with relatively good ToM performance, as measured by the false belief battery, were similarly unimpaired on the Strange Stories task and performance across the two sets of tasks was highly correlated. While this may indicate intact understanding of others' mental states, it may alternatively point to some form of compensatory learning in these children. Surprisingly however, those children with good ToM ability did not differ from the children with poor ToM on the basis of age, verbal or performance IQ. Given that past results have suggested that verbal ability may be the basis on which compensation is possible (Happé, 1995) and the fact that verbal IQ and performance on ToM tasks are strongly correlated in this sample, this would support the idea of a genuine lack of impairment in these children rather than compensation.

Chapter 4: Executive Function

4.1 Ecologically valid tests

Executive function (EF) encompasses a wide range of skills and abilities that have in common their involvement in the higher order control of behaviour in order to accomplish a goal. Whilst some researchers stress the componential nature of executive functions (Hughes, 1998; Ozonoff, 1997), others suggest that these are just different reflections of one system working towards a common goal. Specifically, the two main executive models involve a 'central executive' (Baddeley, 1986) or a 'supervisory attentional system' (Shallice, 1988), which fulfil this role. However, even these authors suggest that these central systems control peripheral modules with specific functions (Baddeley, 1996; Shallice, 1994). Whether executive functions can be fractionated into subcomponents and whether these or some central mechanism is impaired in ASD is an unresolved issue.

Furthermore, whilst many traditional tests of EF have been grouped under certain executive subheadings, many are more complex and involve multiple executive functions, making it difficult to interpret exactly what aspect of the task is problematic. For example, the Tower of London test, whilst usually considered a test of planning ability, also involves a significant degree of working memory and inhibition of prepotent responses. Moreover, the language used when describing such tests is ambiguous; for example, *flexibility* can be described in terms of an ability to flexibly switch to a new response, but also involves the *inhibition* of the current response, and the *planning* and *generation* of a novel response.

Additional problems with traditional tests are revealed when considering their efficacy in identifying executive dysfunction. These tests are frequently found to be insensitive to subtle impairments in patients with frontal lobe damage, whilst these patients have more obvious problems in everyday life, indicating that the tests do not provide an accurate and

reliable identification of the core EF problems (Shallice & Burgess, 1991)(Shallice & Burgess, 1991). Wilson, Evans, Alderman, Burgess & Emslie (1997) suggested that a reason for this disparity between laboratory and real-life performance is that these tests often require the individual to perform a single task with short trials, few rules, clear goals to be achieved, external support and explicit prompting from the experimenter, circumstances which are rarely true of everyday life. Indeed, it is just this aspect of executive control that is thought to be problematic in the executive dysfunction seen in frontal patients (Burgess, 1997). Problems may occur when multiple task demands are concerned, either due to a general information processing problem causing a break down in response under high load, or due to more subtle problems only being revealed under situations where compensation is more costly (Burgess, 2000). Similar proposals have been put forward for individuals with ASD. Garcia-Villamizar & Della Sala (2002) found that high-functioning adults with ASD were impaired on EF tasks that involved dual task performance but not when the individual task components were administered separately. Minshew, Goldstein & Siegel (1997) and Goldstein, Johnson & Minshew (2001) likewise found that individuals with ASD showed impairments only on tasks in which the complexity of the information processing demands was high, independent of the domain in which these were performed.

Further difficulties are encountered when attempting to test executive skill in children; as the majority of tests available were designed for use in adult patient populations, they are typically not suitable for use with children as they are not developmentally appropriate (Anderson, 1998). Firstly, the administration procedures may not be suitable for child populations due to underdevelopment of other skills, such as ability to read and remember complex instructions. Likewise, the use of such tests in child populations may tap into abilities unrelated to frontal lobe dysfunction and executive control. For example, tests such as the Stroop, which rely on relatively automatic reading skill, may be invalidated in

child populations when the child is still developing fluent reading and therefore identifies words laboriously.

These observations have led to the development of new tests that tap into more real-life scenarios, providing ecological validity of the tests; this allows the task to be both relevant to and representative of everyday behaviour (Burgess, Alderman, Evans, Emslie & Wilson, 1998; Burgess et al., 2006). A comprehensive neuropsychological test battery of six EF tasks suitable for children has recently been produced: the Behavioural Assessment of Dysexecutive Syndrome for Children (BADS-C: Emslie, Wilson, Burden, Nimmo-Smith & Wilson, 2003). The tasks in this battery were designed to be sensitive and suitable for high-functioning populations who may be compensating for any impairment. The requirement for reading, language skills and verbal short-term memory were also kept to a minimum by presenting the task instructions pictorially when possible. Furthermore, this test battery has been designed to assess everyday difficulties in more ecologically valid situations than traditional EF tests, such as finding a key in a field or planning a trip to the zoo.

Whilst the use of the BADS-C has not yet been reported in the literature, the adult version of the test has been utilised with a range of patient populations, including adults with Asperger Syndrome. The BADS therefore appears to be sensitive at detecting executive dysfunction in schizophrenic patients (Cools, Brouwer, de Jong & Slooff, 2000; Evans, Chua, McKenna & Wilson, 1997; Ihara, Berrios & McKenna, 2000, 2003; Krabbendam, de Vugt, Derix & Jolles, 1999), drug users (Zakzanis & Young, 2001), chronic alcoholics (Moriyama et al., 2002), patients with depression (Paelecke-Habermann, Pohl & Leplow, 2005) and patients with traumatic brain injury (Bach, Happé, Fleminger & David, 2006; Bennett, Ong & Ponsford, 2005a, 2005b). Most of the subtests also appear to have good test-retest reliability (Jelicic, Henquet, Derix & Jolles, 2001). Furthermore, the study by Bach et al. (2006) supports the idea that the BADS may detect EF impairments that more traditional tests do not.

The one study using the BADS in adults with Asperger Syndrome has also successfully detected executive dysfunction in this population (Hill & Bird, 2006). These adults were found to have impairments on the Zoo Map test, the Action Program task (equivalent to the children's Water task) and the Six Elements test (equivalent to the children's Six Parts test) from the BADS, as well as on an additional novel test, the Hayling Sentence Completion Test (Burgess & Shallice, 1997) (see section 4.2.1 for details of these tests). More traditional tests of EF were also administered but these failed to discriminate the adults with Asperger Syndrome from the controls. Furthermore, these authors studied the individual profiles of each of their participants and found all but one of their adults with Asperger Syndrome to be significantly impaired on at least one EF test. Relationships between performance on these tests and autistic symptomatology were also established, indicating that these tests are indeed ecologically valid.

Given that such a battery of tests now exists for children, it would be of interest to know whether high-functioning children on the autism spectrum show similar problems to adults with Asperger Syndrome across a range of ecologically valid tests of EF. One further study indicates that this may indeed be the case; a task similar to one of the tests from the adults version of the BADS (the 6 Elements test) has been developed to be suitable for children (Mackinlay, Charman & Karmiloff-Smith, 2006). This study compared a group of high-functioning boys with ASD to age and IQ matched controls and found impaired performance on this task. Participants in the ASD group were less likely than controls to plan a strategy to complete the task, showed less switching between tasks and made more rule-breaks. There was also limited evidence that the level of performance on this task was related to everyday behaviours, again supporting the ecological validity of the task.

4.2 Method

All EF tests were carried out during test session 3. The full BADS-C was administered and scored according to the user manual (Emslie et al., 2003), which consists of six tests tapping different aspects of executive functioning. In addition, a modified version of the Hayling Sentence Completion Test (Burgess & Shallice, 1997), adapted for children, was also administered.

4.2.1 Behavioural Assessment of Dysexecutive Syndrome for Children (BADS-C)

The *Cards task* taps into the ability to inhibit a prepotent response by flexibly shifting to a new rule and therefore avoiding perseveration. Here the participant responds twice to the same set of playing cards, firstly saying 'yes' to red cards and 'no' to black cards, and secondly saying 'yes' when the previous card was the same as the current one and 'no' when it was different. The second set of responses was critical as here the prepotent response set up by the previous rule must be put aside in order to answer according to the new rule. The number of errors made during the second set of responses was therefore recorded.

The *Water task* requires the child to plan and carry out a response in order to solve a multistep problem involving the retrieval of a cork from a tall tube; this can only be achieved by using a special tool to remove a lid from a pot of water, screwing a base onto a tube to form a container, filling the container with water twice and pouring these into the tall tube in order to make the cork float to the top. In the majority of cases, this requires the child to investigate possible responses at each stage and evaluate whether they have helped them get any closer to the solution. Two points were awarded for each stage successfully completed without prompting, with a one point penalty subtracted if the child perseverated in their response or took longer than 200 seconds.

The *Key Search task* examines the ability to plan an efficient solution to a problem that is implementable in everyday life. The child is presented with a picture of a birds eye view of a field (a square box) and asked to draw a line to indicate where they would walk in order to search it for their lost keys. Points were awarded for strategies that took into account the information provided (eg. searching only within the field), that were systematic, planned and efficient (eg. walking from side to side across the field) and for implementability (eg. strategies that did not rely on knowing your precise positioning in the field at an earlier point in time).

The *Zoo Map test* is split into two parts. Part 1 requires the child to plan a solution to a problem that requires the consideration of a number of rules. Here the child must plan a walk around a zoo, keeping to the paths, visiting only certain places and walking along certain paths only once. Points were awarded for visiting the places in the optimal sequence and were subtracted for breaking any of the rules. Part 2 requires the child to follow a set of instructions whilst obeying some simple rules. Here the child is presented with an identical map and asked to repeat the task obeying the same rules; however, this time the order in which to visit the places is provided, removing much of the need for planning.

The *Six Parts test* requires the child to plan a strategy in order to complete an overall task, involving carrying out six activities, without breaking any of the rules. The child must also monitor their own progress on each activity in order to ensure they keep to their plan. The six tasks consist of two picture naming tasks, two counting tasks and two sorting tasks. The child is given 5 minutes on a timer placed in front of them and is required to complete something from each task without performing two tasks of the same type consecutively. Here the child was awarded points for attempting all six parts, not breaking the order rule and for using specific strategies to achieve these two aims. Points were deducted if the child inefficiently returned to a particular task three or more times.

This battery of tasks also produces a *BADS-C total score* by combining the standardised scores from all 6 tests together to give an overall measure of EF performance.

4.2.2 Shallice Switching Sentence Completion Task (SSSCT)

The Hayling Sentence Completion Test was first adapted for use with children by Shallice et al. (2002). Shallice (unpublished) further adapted the task to make it even more challenging and this version, referred to here as the *Shallice Switching Sentence Completion Task* (SSSCT), was used in the current study. This test requires the generation and inhibition of verbal responses as well as task switching. It establishes a prepotent situation by making use of over-learned knowledge of sentence endings rather than creating the prepotent response during the test situation, thus making the prepotent response highly salient and so requiring a high degree of inhibition to overcome it. During task administration, the experimenter reads a sentence to the child with the last word missing and the child must complete the sentence with a single word. In the first condition (formerly Part A), the child must give the correct or most appropriate word, whilst in a second condition (formerly Part B), the child must produce a word that is unrelated to the sentence, to the missing word or to a previous answer. These two conditions were alternated, with the experimenter indicating 'correct' trials by placing their right hand on their lap and 'wrong' trials by holding their right hand up. For the 'correct' trials, the number of inappropriate responses given was recorded. For 'wrong' trials, three penalty points were awarded when the correct response was given or one point when the response was semantically related to the sentence or the missing word. After the task had been administered, children were asked how they had generated the 'wrong' words; specifically, whether they had used a strategy such as naming objects around the room or naming objects in a category, and whether they had thought of these words before or after they heard the sentence read to them.

4.3 Results

Raw scores rather than scaled scores were entered into all analyses for the BADS-C tests as these were felt to be more reliable and retained a higher degree of variation of responses. In addition, this allowed absolute age and IQ to be taken into account on an individual basis rather than on the basis of broader age and IQ bands. Performance on the Key Search task ($r = .48, p < .001$) and Zoo Map 2 ($r = .37, p = .004$) was found to increase with age in the ASD group, no test was related to performance IQ and performance on all of the subtests and the SSSCT was found to increase with verbal IQ ($r > .27, p < .05$). In the control group, performance on the Water task ($r = .50, p = .008$) and Zoo Map 1 ($r = .48, p = .012$) both increased with age; performance on the Cards, Water, Zoo Map 1 and Key Search tests increased with performance IQ ($r > .44, p < .03$); and the Cards test, Zoo map 2 and the Six Parts test all increased with verbal IQ ($r > .38, p < .05$).

The ASD and control groups were therefore compared on each test using a multivariate ANCOVA, entering age, verbal IQ and performance IQ as covariates (see Table 4.1). Group differences were found on the Key Search task ($F(1,79) = 6.99, p = .010$), the Six Parts test ($F(1,79) = 5.24, p = .025$) and the SSSCT ($F(1,79) = 4.85, p = .031$). In addition, a weak trend towards a significant group difference was observed for the Zoo Map 2 test ($F(1,79) = 2.79, p = .099$), which was found to be significant once the lack of equality between the variances in the two group had been accounted for ($p = .047$). No other comparisons on EF tests were significant. An overall difference on the BADS-C total score was also found ($F(1,79) = 7.16, p = .009$). Observation of the group means revealed that the ASD group were consistently performing worse than the controls across all measures, even those on which significant group differences were not found.

Those tests that produced significant differences were studied in more detail. Differences between the groups on the Key Search task were found for the ability to produce a

Table 4.1 Means (and standard deviations) for performance across the EF tests; note that negative scores are possible on some tests.

	Control group	ASD group
Cards task (errors in set 2)	.93 (1.44)	2.02 (2.99)
Water task (max 10)	7.81 (2.39)	6.42 (3.30)
Key Search task (max 14) **	9.11 (4.15)	5.49 (4.51)
- task understanding (max 5)	4.07 (.47)	3.72 (.90)
- efficient planning (max 8) *	4.44 (3.65)	1.47 (3.85)
Zoo Map 1 (max 8)	1.74 (4.45)	-.21 (4.86)
Zoo Map 2 (max 8) *	7.56 (1.16)	5.16 (4.56)
- no. moves in correct sequence (max 8) +	7.93 (.38)	7.30 (1.68)
- no. rule breaks	.37 (1.11)	2.05 (3.29)
- time before making first move	4.63 (5.62)	5.93 (8.09)
Six Parts test (max 16) *	12.59 (2.79)	9.96 (3.43)
- max time on any subtask (secs) +	75.89 (52.08)	126.52 (78.79)
- no. subtasks attempted (max 6)	5.59 (1.12)	4.86 (1.84)
BADS-C total score (mean 60) **	61.74 (10.61)	51.47 (11.76)
SSSCT (total errors) *	6.15 (2.92)	9.75 (6.14)
- 'correct' trial errors	1.22 (1.25)	2.33 (2.09)
- 'wrong' trial errors	4.93 (2.84)	7.42 (5.78)
- strategy use (% of children) ***	63	23
- generating word before sentence (%) **	44	16

* $p < .05$, ** $p < .01$, *** $p < .001$, + $p < .06$

systematic, planned and efficient search strategy ($F(1,79)=6.21, p=.015$) with the ASD group performing worse than controls, whilst the ASD group was as good as the controls in understanding the task requirements ($F(1,79)=.804$). On the Zoo Map 2, the number of moves made by the child in the correct sequence neared significance after correcting for inequality of variances in the two groups ($t(80.3)=1.92, p=.058$) whilst no differences were found on the number of rule breaks made by the child ($F(1,78)=1.70$) or the time taken before the first move ($F(1,78)=1.49$). For the Six Parts test, the group difference arose from the ASD group having a slightly longer maximum time on any one subtask than the controls ($F(1,78)=3.87, p=.053$), whilst not differing from them on the number of subtasks attempted ($F(1,79)=.724$). Whilst the overall score on the SSSCT produced group

differences, these were only seen as trends in the separate conditions ('correct' $F(1,79)=3.45, p=.067$; 'wrong' $F(1,79)=2.63, p=.109$). However, the ASD group was significantly less likely than controls to use a strategy (eg. naming objects in the room) to help them think of words in the 'wrong' trials ($\chi^2(1)=12.87, p<.001$) and were more likely than controls to think of these words after they had heard the sentence than before ($\chi^2(1)=8.02, p=.005$).

From Figures 4.1 and 4.2, it can be seen that most of the tests produced a tail of outliers in the ASD group (Zoo Map 2, Six Parts, SSSCT) and this was the case even for some tests that did not produce significant group differences (Cards, Water, Zoo Map 1). The Key Search test, however, appeared to produce different distributions in the two groups despite similar ranges, skewed towards poor performance in the ASD group; this is likely to be produced from a floor effect in the marking scheme for this test. This latter pattern was also seen in the BADS-C total score, although here it is likely to be due to inconsistency in individual performance across the different tests. Despite this, it is notable that in both

Figure 4.1 Performance of each individual across the subtests of the BADS-C; scores are shown as z-scores based on control performance and the dotted line illustrates the cut-off for the 5th percentile of control performance.

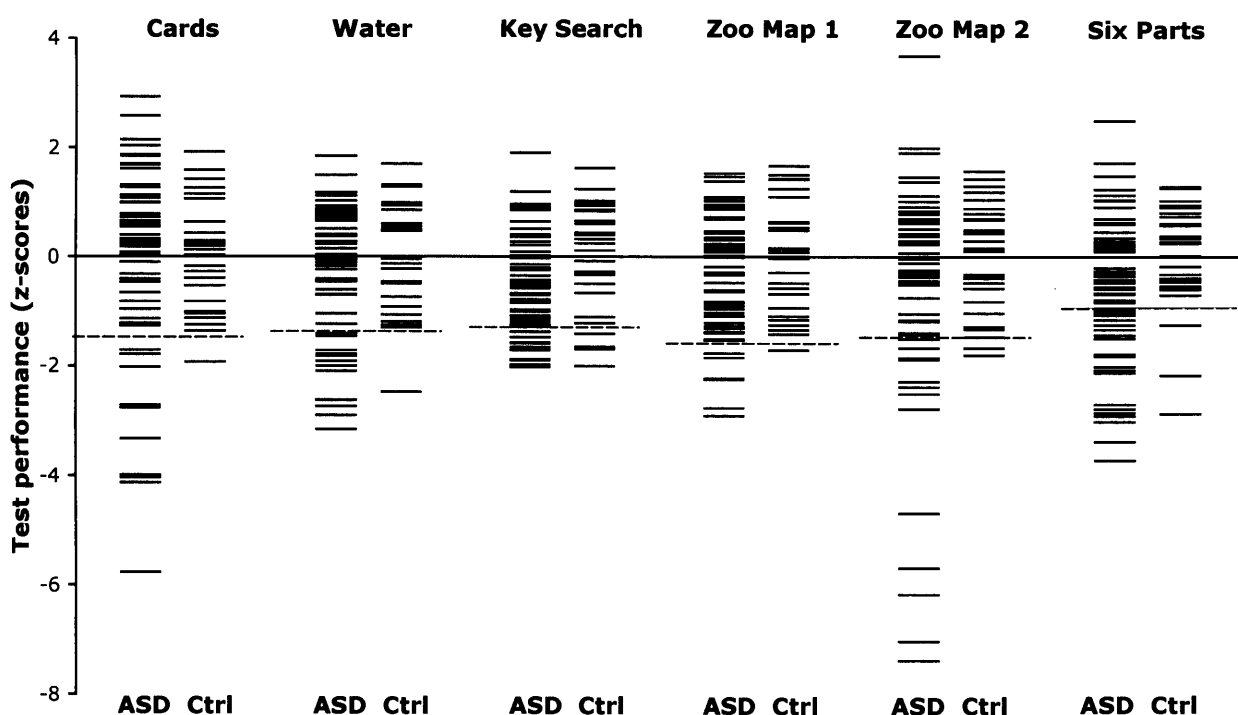
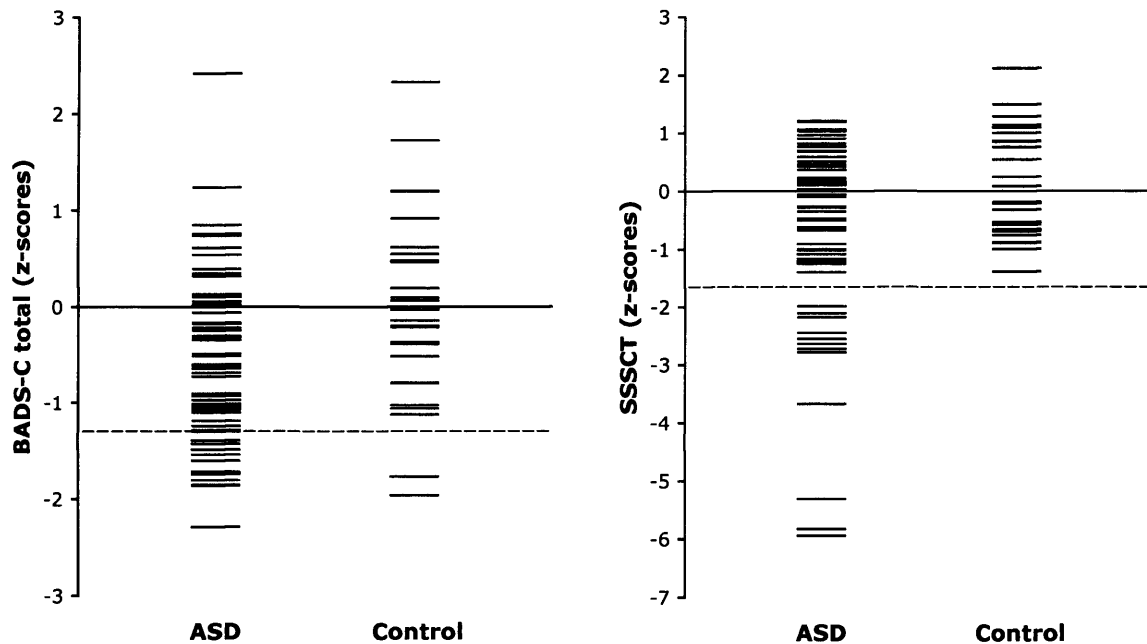


Figure 4.2 Performance of each individual on the BADS-C total score and the Shallice Switching Sentence Completion Task; scores are shown as z-scores based on control performance and the dotted line illustrates the cut-off for the 5th percentile of control performance.



patterns, only a small proportion of the children in the ASD group could be classified as showing deviant performance, performing below the 5th percentile of control performance (see section 6.2 for more details of this methodology). This proportion ranged from 12% (Zoo Map 1) to 37% (Six Parts test). 10 children showed intact performance on all 7 tests, 20 children showed impairment on 1 test, 17 on 2 tests, 4 on 3 tests, 5 on 4 tests and 1 on 5 tests. Furthermore, performance in the ASD group spanned the full range of the control performance, with some children performing particularly well above the top of the control range.

A few specific cases in the ASD group are worth mentioning. CB performed extremely well across a number of tests, with performance above the control range on 4 of the 7 tasks (Cards, Water, Key Search and Six Parts). This particular child was one of the youngest in the study (6 years 10 months at the first test session) and performed on the IQ test in the low-average range (verbal IQ 87; performance IQ 69). Despite this, he performed all the EF tests intuitively and with ease and, after correcting the scores for age and IQ, his

performance was therefore found to be exceptionally high. A similar case to this, albeit to a lesser extent, is AS who performed above the control range on 2 of the 7 tasks (Cards and Six Parts); she was only 7 years 4 months at the first test session with verbal and performance IQs of 98 and 91 respectively and performed well on the EF tests. Two particularly interesting cases both showed extreme performance on two tasks, with performance on one task being exceptionally high and performance on the other being exceptionally low (RW Cards low, Zoo Map 2 high; YA Cards low, Six Parts high). Both children were again young (7y7m and 6y11m respectively) although only RW had low IQ scores (verbal IQ 66, performance IQ 77; YA verbal IQ 98, performance IQ 108). It seems likely that both children had specific problems inhibiting the first rule and moving onto the second rule in the Cards test. Still, such variable performance across a single battery of tests warrants caution.

Relationships between the different EF tasks were examined in both of the groups separately. After accounting for age and IQ, only the Water task and Zoo Map 1 were correlated in the ASD group ($r = .37, p = .004$). This same relationship was also seen in the controls ($r = .57, p = .002$) and correlations in this group were also found between Zoo Map 2 and the Cards test ($r = .46, p = .016$), Zoo Map 2 and the Key Search task ($r = .52, p = .005$), Zoo Map 2 the Six Parts test ($r = .40, p = .041$) and Zoo Map 2 and the SSSCT ($r = .40, p = .040$).

4.4 Discussion

Consistent with many previous studies assessing EF in ASD (Hill, 2004a), executive dysfunction was found in the ASD group across a number of tasks from a novel ecologically valid neuropsychological battery suitable for children (Emslie et al., 2003), as well as a sentence completion task. Those tests showing significant group differences were the Key Search test, Zoo Map 2 test and Six Parts test from the BADS-C, as well as the

SSSCT. These findings closely parallel those found on the adult version of the battery in adults with Asperger Syndrome, particularly on the Six Parts test and the SSSCT (Hill & Bird, 2006).

While significant group differences were observed on only on a few tests, the ASD group means were consistently lower than those of the control group across all tasks. From examination of individual performance, a fairly consistent pattern was seen across the tests: a tail of children in the ASD group were performing below the control range but the majority of children with ASD performed similarly to the controls. Whilst it can be fairly certain that these deviant performers had problems with the tasks, it is possible that some other children with ASD, who were not classed as performing poorly, also had executive dysfunction but were compensating to an extent on these tasks. Remarkably, some children in the ASD group were performing well above the control range of scores, indicating that executive abilities were truly intact in at least some children. Furthermore, 18% of children showed intact performance across all 7 tests, again a remarkable achievement on their part suggestive of truly retained ability.

Those group differences that were found to be significant were those with the greatest number of deviant performers (with the exception of the Key Search task; this is likely to be due to a floor effect in the task), indicating that these children were driving the group differences and that some tests appeared to be more sensitive to executive dysfunction than others. There was little consistency, however, in the children performing poorly across tests, with different children finding different tasks difficult, supported by the lack of significant correlations between the tasks. While this warranted caution when considering any hypothesis attempting to pinpoint a single cause of poor performance across these tasks, it is still advantageous to look for a common denominator that could then be used to direct and focus future tests.

4.4.1 A common denominator

A more detailed analysis of those tasks showing significant group differences revealed that some children in the ASD group did not spontaneously produce an efficient strategy in the Key Search test, whilst they were relatively unimpaired in their understanding of the task requirements and took these into account in their responses (see Table 4.1, p112). This indicates problems with planning or with impulsivity and lack of inhibition; lacking an efficient strategy could result from either an inability to make such a plan or an inability to stop oneself from beginning the task immediately, thus not allowing time to plan. On the SSSCT, some children in the ASD group were more likely both to give incorrect answers when they were meant to be finishing the sentence appropriately and to give correct or semantically related answers when they were asked to supply unrelated words. The first of these appeared to occur as a form of perseveration as the child became stuck in the ‘wrong’ condition, responding with unrelated words regardless; the latter can be interpreted as an inability to inhibit the correct sentence ending. In addition, some children with ASD showed a lack of planning in their wrong answers, being less likely to produce a strategy to aid them in the ‘wrong’ condition and more likely to generate their answers after they heard the sentence.

For the Zoo Map 2 test, some children with ASD made fewer moves in the correct sequence. This is curious for two reasons: firstly, because the groups did not differ significantly on this measure for the more difficult Zoo Map 1; secondly, because the sequence was actually provided for them in the Zoo Map 2 test, whilst it was not for Zoo Map 1. The control children found this additional information a great aid, performing Zoo Map 2 close to perfectly, whilst finding Zoo Map 1 similarly challenging to the ASD group. The children in the ASD group could obviously follow the task instructions, as shown by a lack of group difference on the number of rule breaks and similar performance to the controls in Zoo Map 1. It was noted in a number of cases, however, that some children

repeated the route they have used in Zoo Map 1 when completing Zoo Map 2, making the same mistakes again rather than improving their performance (this could be obviously seen in 9 cases); this indicated that their poor performance on the easier Zoo Map 2 may be due to perseveration. This hypothesis would be easily testable by presenting the Zoo Map 2 condition without Zoo Map 1; those children with ASD who had previously performed poorly on Zoo Map 2 should then perform as well as controls.

In the Six Parts test, the maximum time spent on any one subtask was longer in the ASD group than in the controls, although they attempted as many subtasks. Rather than spending longer across all subtasks and therefore not managing to complete as many subtasks, it therefore appears that some children in the ASD group were spending less time on some tasks in order to spend more time on others. Exactly this same pattern of performance was also reported in the previous study with adult with Asperger Syndrome using this test (Hill & Bird, 2006). Anecdotally, a few children in the ASD group verbalised this intention during the test session, stating that they were uninterested in spending time on tasks they didn't enjoy, preferring to focus on one favourite task. Whilst this strategy is not maladaptive and complies with the task instructions, it is obviously a consistent difference between the two groups; it appeared that some children with ASD had a different understanding of the appropriate behaviour during the test situation, with their own wishes predominating and the experimenter's wishes not taken into account. While this may be indicative of an executive failure to comply with the *implicit* purpose of the test and to monitor one's own progress, it may be exactly this *implicit* information that is not available to or less salient for some children with ASD; if they were less likely to model another's mental states, they would have less access to the experimenter's expectations of them. In the same vein, these children may not have knowledge of or the desire to conform to social scripts of how to behave appropriately in certain social situations. Such behaviour could be described as egocentric.

This hypothesis could also explain the mixture of EF impairments seen in other tasks; while some tasks appeared to reveal planning impairments, others seemed to be better characterised by inhibition and perseveration problems. It may be that an explanation concerning a lack of adherence to implicit task demands more successfully encompasses the poor performance seen in a proportion of the children with ASD across these tasks, with performance being driven by the child's own desires rather than the experimenter's expectations.

4.4.2 Implicit task demands

This hypothesis of a lack of understanding of the implicit task demands could also be seen anecdotally in other tests, such as the Zoo Map test, in a few cases when children spontaneously remarked on their own performance. For example, after visiting the animals in the wrong order, one child commented that he would always go and see the lions first as he was a cat lover. The same child, when visiting only 5 of the 8 specified locations, explained that he would save the others for another day, and didn't need to visit the café as he'd been organised and brought a picnic with him. A different child similarly explained that he had visited the café last as he would be hungry then, after having walked all the way round the zoo visiting the animals. Whether these are *post hoc* justifications after having forgotten the rules is unclear, however.

This leads to a consideration of the use of ecologically valid tasks in populations with ASD. Why do such tests appear to be sensitive in picking up group differences between individuals with and without ASD when traditional tests are not sensitive? It appears that the ecologically valid, uncontrolled and open-ended nature of such tasks allowed a minority of the current children with ASD the freedom to impose their own task demands on the test situation. Indeed, the absence of prompting in ecologically valid tasks, which is present in more traditional tasks (Wilson et al., 1997), may provide enough information for

certain children with ASD to comply with the task demands. Furthermore, as Russell suggests (Biro & Russell, 2001; Russell, 1997), EF tasks commonly impose arbitrary rules on individuals and it appears to be with these that some individuals with ASD struggle most. While this may be due to a lack of verbal self-prompting as suggested by Russell, it is also possible that certain individuals with ASD are not aware of the social expectation to comply with such arbitrary rules, instead imposing their own desires. In support of this possibility, Ozonoff (1995) found that individuals with autism performed as well as controls on an EF test when presented in a computerised format and therefore presumably seen as away from the experimenter's intentions, than when presented by an experimenter in a normal testing situation.

This suggestion may even go some way towards explaining the associations seen between mentalising and EF tasks in ASD samples (Joseph & Tager-Flusberg, 2004; Ozonoff, Pennington & Rogers, 1991a). Whether such a relationship is indeed present in the current sample of children with ASD will be addressed in chapter 6. It would also be possible to test this suggestion more directly by running an 'ecologically valid' test of the sort used in the current study and closely matching a control condition with explicit prompting and instruction to it. It would be expected that certain children with ASD would find only the experimental condition hard relative to controls; furthermore, it would be expected that those with the greatest disparity between the control and experimental conditions would have the most severe mentalising problems.

4.5 Conclusion

The current findings support the idea that ecologically valid tests of EF do indeed detect abnormalities in task performance by a proportion of individuals with ASD. Indeed, it appears that it is the very nature of these open-ended and unguided tasks that is a challenge to such individuals. These tasks require the child to respond to implicit demands laid down

by the experimenter and it has been proposed here that it is this implicit information that may be unavailable, less salient or less desirable to certain children with ASD. While this tendency not to conform to the implicit task demands may be due to an executive failure to prompt oneself during unguided tasks in order to stay on task, it is also possible that implicit information is less available to these children due to their social communication problems, leading to a poor understanding of the appropriate behaviour during the test situation. It is clear that such a problem does not affect all children with ASD however, as a proportion of children displayed intact task performance, even with these more sensitive tasks and even across a whole battery of tests.

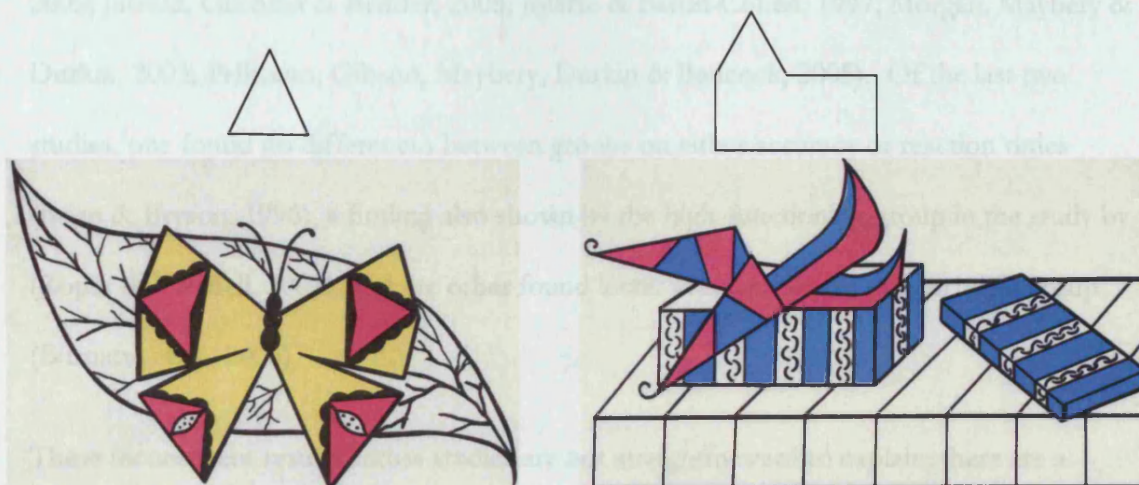
Chapter 5: Central Coherence

Weak central coherence is currently thought to result from a superiority in, a bias towards or a preference for local stimuli, rather than an impairment in global processing (Happé, 1999; Happé & Frith, 2006). Hence, global processing is generally thought to be intact in individuals with ASD (Mottron, Dawson, Soulieres, Hubert & Burack, 2006) although local processing may take precedence over global processing in tasks where the two are in competition. Those tasks thought to be most sensitive in detecting weak central coherence either pit global and local processing against each other or require fast online responses that are able to pick up this bias; the ability of one such task to detect this bias will be investigated in the first part of this chapter. Recent research has also begun to suggest a mechanism for this bias, in terms of an inability to shift out of local processing and therefore into global processing (Mann & Walker, 2003; Rinehart, Bradshaw, Moss, Brereton & Tonge, 2001). Whether this supposedly directional switching problem can be explained by a more general problem with switching between stimuli will be further explored in the second part of this chapter. Lastly, the hypothesis that macrocephaly may be a biological marker of weak central coherence will be examined.

5.1 Embedded Figures Test

The traditional test used to tap central coherence, and the first used in an autistic population, is a visuo-spatial test called the children's Embedded Figures Test (EFT: Witkin, Ottman, Raskin & Karp, 1971). This test requires the child to find a figure, such as a triangle, hidden as a small element in a much larger image. The child must therefore disregard the whole image and the semantic information it carries with it and focus in on the detail of the shapes and lines present in the picture, searching for the hidden element. This is a complex process requiring the child to ignore and inhibit Gestalt principles, such

Figure 5.1 Examples of stimuli used in the Embedded Figures Test; the child is required to find the shape (above) in the picture (below). © Consulting Psychologist Press, Inc., Palo Alto, CA.



as the continuity of lines, foreground and background information, and texture and colour cues. Performance is known to increase with age (Witkin et al., 1971) and is also likely to depend on the individual child's experience, intelligence and perceptual abilities, as well as their processing bias in terms of central coherence. Examples of some of the stimuli are given below in Figure 5.1.

Typically, children with ASD have been found to perform better than control children on this task and there is a reasonably pervasive sense not only that children with ASD are guaranteed to show superior performance on the EFT, but also that this test is a marker of weak central coherence. However, in reality, results are equivocal (see Table 5.1 for a summary of all studies). The initial study using this task found increased levels of accuracy in children with ASD compared to controls matched either on non-verbal mental age or on both non-verbal mental- and chronological age (Shah & Frith, 1983). Unfortunately, no control task was presented with this or any of the subsequent studies using the EFT. Since then, nine different papers using the task have been published, only two of which has replicated this accuracy difference (Ropar & Mitchell, 2001; van Lang, Bouma, Sytema,

Kraijer & Minderaa, 2006b). Of the remaining seven studies, five found that the ASD group located the embedded figures more quickly than controls (Edgin & Pennington, 2005; Jarrold, Gilchrist & Bender, 2005; Jolliffe & Baron-Cohen, 1997; Morgan, Maybery & Durkin, 2003; Pellicano, Gibson, Maybery, Durkin & Badcock, 2005). Of the last two studies, one found no differences between groups on either accuracy or reaction times (Brian & Bryson, 1996), a finding also shown by the high-functioning group in the study by (Ropar & Mitchell, 2001), and the other found lower accuracy scores in their ASD group (Burnette et al., 2005).

These inconsistent results across studies are not straightforward to explain; there are a number of different reasons why they might have occurred. These include the use of different ages, different levels of general ability and different administration techniques. Any of these factors could reduce the likelihood of identifying a true group difference through interference from variables other than central coherence and the presence of floor or ceiling effects. For example, it is likely that Edgin & Pennington (2005) had a ceiling effect in accuracy due to a large age range spanning up to 16 years, when the children's version of the test is designed for 5-12 year olds. Similarly, Jarrold et al. (2005) had a ceiling effect in their accuracy data. It is also of interest that both studies detecting group differences in accuracy used lower-functioning groups (Ropar & Mitchell, 2001; Shah & Frith, 1983); it is possible that these individuals had a more severe form of ASD and so differences in central coherence were more enhanced.

A related issue, and one that pervades the literature on ASD, is how to match control groups to ASD groups when the latter are of below average intelligence. In a paper discussing such methodological issues, Mervis & Klein-Tasman (2004) argue against the use of younger mental age- or raw score-matched control groups as different abilities develop at different rates; it is therefore possible that the use of younger controls might affect differences between the groups. As can be seen from Table 5.1, such comparison

Table 5.1 Studies using the Embedded Figures Test in individuals with ASD.

	Comparison group	Age (ASD) & task version	Ability level	Average accuracy (ASD; ctrl)	Group diff (RT=reaction time)
Shah & Frith (1983)	CA, performance IQ	8-18 years Children's	Low	21/25; 14/25	Accuracy
Brian & Bryson (1996)	Verbal raw score; non-verbal raw score	10-27 years Children's	Low	8/8; 8/8	No acc or RT diff
Jolliffe & Baron-Cohen (1997)	CA, verbal & performance IQ	18-49 years Adult	High	11/12; 11/12	RT
Ropar & Mitchell (2001)	Verbal MA	9-18 years Adult	Low	7/12; 3/12	Accuracy & RT
	CA	8-15 years Adult	High	8/12; 9/12	No acc or RT diff
Morgan et al. (2003)	CA, performance IQ	3-5 years Preschool	High	18/24; 19/24	RT
Jarrold et al. (2005)	Non-verbal raw score	8-15 years Children's part A only	Low	not given	RT
Pellicano et al. (2005)	CA, performance IQ	8-12 years Children's	High	not given	RT
Edgin & Pennington (2005)	CA, verbal IQ & Block Design	7-16 years Children's	High	20/24; 21/24	RT
Burnette et al. (2005)	CA, verbal & performance IQ	Mean 11y Children's	High	25/32; 29/32	Accuracy; ASD worse!
van Lang et al. (2006b)	CA, verbal & performance IQ	10-20 years Children's	Low	13/25; 11/25	Accuracy

groups were used in the studies by Jarrold et al.(2005) and by Ropar & Mitchell (2001), both of which did find significant group differences in reaction times, with the older children with autism performing perhaps unsurprisingly better than the younger control children. Furthermore, matching groups on the basis of verbal ability, on which ASD populations may score below the expected level, may lead to respectively higher scores on

non-verbal tasks and a false interpretation that this is an area of superiority when it is in line with non-verbal abilities. This was certainly a possibility in Ropar & Mitchell (2001).

Furthermore, when analysing reaction time differences between groups, different authors have used different techniques, each of which produce quite different results. Some authors have calculated the average reaction time for correct responses only (Morgan et al., 2003; Pellicano et al., 2005), whilst others have calculated the reaction time to all stimuli, either substituting the maximum time allowed for incorrect trials (Jolliffe & Baron-Cohen, 1997) or using search time regardless of whether the response was correct or not (Edgin & Pennington, 2005). Burnette et al. (2005) recently combined reaction time with accuracy by awarding an extra point for each trial in which reaction time was faster than a cut-off.

When accuracy is at ceiling, such as in Jolliffe and Baron-Cohen (1997), the method of reaction time analysis is of little significance as there are few incorrect trials; however, when considering non-significant differences between groups in accuracy, this issue is of high importance. As the children's EFT consists of two parts, one easier and one more difficult, average reaction times may not be comparable between low and high accuracy scores.

Children who have lower accuracy are likely to give correct answers to the easier test items; the target stimuli are therefore likely to be identified more quickly than children with higher accuracy who correctly identify more difficult targets. A group with a slightly lower accuracy may therefore have a faster reaction time when reaction times for only correct responses are included. This effect could be further amplified by the classic speed-accuracy trade-off seen in many psychological experiments. Studies using this method may therefore artificially produce group differences and, indeed, this exact pattern of results is seen in Morgan et al. (2003).

Similarly, using time spent searching for the target, regardless of trial accuracy, could lead to differences in reaction times between groups due to different search strategies on incorrect trials; it is possible that one group might give up searching for the target more

quickly than others. The alternative method of substituting the maximum time limit for reaction time on incorrect trials also seems inappropriate; this would mask any group differences in the data, particularly for individuals with lower accuracy, as reaction time would basically reflect accuracy scores. One way of dealing with all these problems would be to take accuracy into account in each individual's average reaction time score for correct trials.

However, there is a second possible reason for inconsistent findings which is related to heterogeneity; specifically, only a subset of individuals with ASD may exhibit weak central coherence. For example, it has been stated that weak central coherence is applicable mainly to savants. Savant abilities may occur in rote memory, art, mathematics, calendrical calculations, music, perfect pitch and a variety of more specialist areas (Hermelin, 2001). These abilities have been explained in terms of central coherence as the style of information processing concerned with detail and specific exemplars rather than meaning or generalisations (Heaton, Hermelin & Pring, 1998; Mottron & Belleville, 1993). The occurrence of savants in the autistic population is thought to be about 10% (whilst only about 1% in the normal population; Rimland & Hill, 1984) and this lends support to the idea of heterogeneity, with central coherence only being present in a minority of individuals.

Individual data are rarely seen in the autism research literature but are vital to understand how prevalent weak central coherence is in ASD. Three authors of papers using the Embedded Figures Test do provide such information. Edgin & Pennington (2005) reveal similar distributions of performance in the ASD and control groups but with a slow tail in the controls and three extremely fast performers in the ASD group, indicating that the ASD group reaction time distribution may be shifted towards faster responses. On the other hand, Jarrold et al. (2005) and Pellicano et al. (2005) both show an almost complete separation in the distributions of the two groups, indicating that fast performance may be

universal in ASD (although it should be noted that the method of reaction time analysis will affect this). However, the means and standard deviations given in other papers support the idea that the distributions of the two groups tend to overlap.

The first part of this chapter will therefore examine group and individual differences in performance on the EFT by the current sample of children with ASD, as well as assessing the different methods of analysing reaction time data within the same participants.

5.1.1 Method

All items in the Children's (test session 2) and Adult's (test session 3) Embedded Figures Tests (Witkin et al., 1971) were administered to each child, thus providing a greater range of possible scores and avoiding ceiling effects in the older or more able children. The tests were presented in a series of three laminated picture books and the child was instructed to search for the target shape in each picture (either a triangle, a house shape or a mixture of designs) and to draw over the target with a pen when they had found it. This encouraged the child not to be ambiguous or vague in their response and so allowed for more accurate scoring and timing. Accuracy and reaction times for all stimuli were recorded. A maximum of 60 seconds was allowed for each picture, rather than the recommended 180 seconds in the manual, as most children were found to give up looking within this period during piloting and the shorter time limit also helped to maintain their attention on the task.

5.1.2 Results

From the means in Table 5.2, it can be seen that the ASD group were in general performing slightly worse in terms of accuracy than the control group, although this could be explained by differences between the groups in IQ. The relationship between task performance and age and IQ was therefore investigated. In both groups, EFT total

Table 5.2 Means (and standard deviations) of Embedded Figures Test results.

		Control group	ASD group
Accuracy	Child: Triangle (11 max)	8.93 (1.90) 4–11	7.88 (2.49) 0–11
	Child: House (14 max)	8.11 (3.59) 1–12	6.28 (3.59) 0–13
	Adult (12 max)	3.81 (3.15) 0–9	3.00 (3.02) 0–10
	Total (37 max)	20.85 (7.68) 7–31	17.16 (7.88) 3–32
	Reaction time 1: correct responses only (seconds)	Child: Triangle 8.26 (2.88) 2.79–13.80	7.99 (3.86) 1.93–17.43
	Child: House ** 16.92 (7.39) 6.37–40.55	13.25 (7.39) 1.50–30.47	
	Adult 19.58 (6.28) 8.49–31.31	17.48 (8.97) 2.56–44.71	
	Total 12.83 (3.66) 4.88–20.65	11.25 (3.89) 3.33–19.04	
Reaction time 2: 60s for incorrect trials (seconds)	Child: Triangle 18.07 (8.82) 6.33–43.20	22.94 (11.37) 3.18–60.00	
	Child: House 34.17 (12.80) 14.03–58.61	39.13 (12.46) 11.34–60.00	
	Adult 46.97 (11.22) 25.05–60.00	49.31 (11.11) 19.82–60.00	
	Total 33.53 (9.84) 17.51–51.27	37.62 (9.84) 16.78–56.55	
Reaction time 3: correct responses accounting for accuracy (z-scores; -ve numbers indicate fast responses)	Child: Triangle 0.00 (1.00) -1.45–2.49	-0.12 (1.30) -2.00–3.86	
	Child: House ** 0.00 (1.00) -1.40–3.17	-0.54 (0.82) -2.27–1.80	
	Adult 0.00 (1.00) -1.75–1.78	-0.36 (1.44) -2.84–3.94	
	Total 0.00 (1.00) -1.68–2.14	-0.30 (1.03) -2.16–1.92	

** $p < .01$

accuracy increased with age (ASD: $r = .48, p < .001$; control: $r = .55, p = .003$), verbal (ASD: $r = .37, p = .005$; control: $r = .49, p = .010$) and performance IQ (ASD: $r = .27, p = .039$; control: $r = .65, p < .001$), although the strength of this relationship differed between the groups, particularly for performance IQ. An ANCOVA comparing group accuracy performances, after factoring out the variance in age, verbal and performance IQ, found no significant

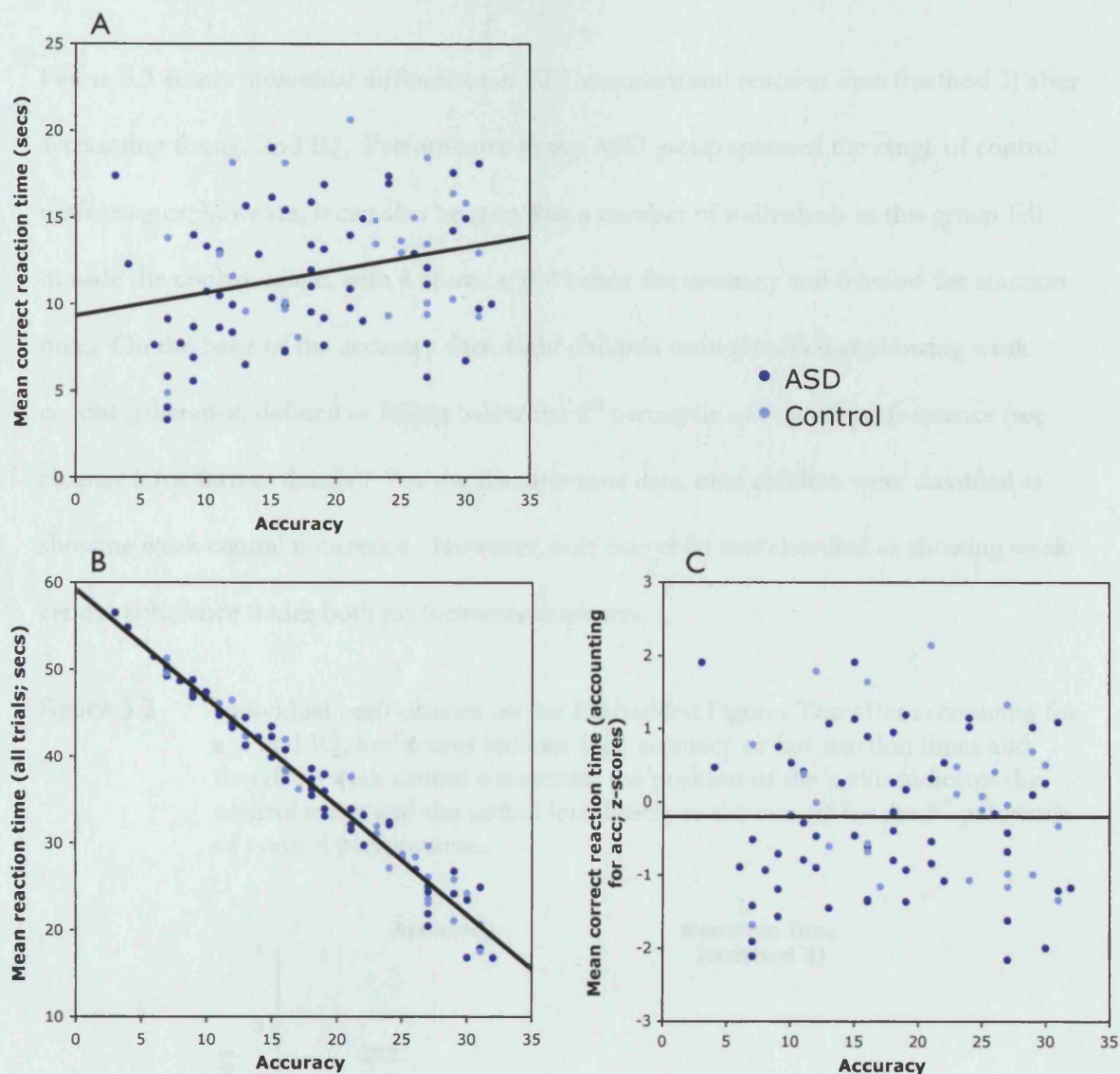
differences in any condition ($F(1,79) < .5$). Indeed, the corrected means indicated that the groups were performing extremely similarly in all conditions.

Reaction times were calculated in three ways; firstly reaction times to correct trials only, secondly reaction times with 60 seconds entered for incorrect trials, and lastly correct reaction times with accuracy scores covaried out of them (shown in Table 5.2). It should be noted that for the first and last methods, not all children have reaction time data as this relies on them having correctly located at least one target shape. One child with ASD therefore has missing data from the tent condition, one child with ASD from the house, and 15 children with ASD and 6 controls on the adult version; this is a serious floor effect but can be taken into account in the Embedded Figures total.

From the table, the different patterns of results produced by the two reaction time calculation methods can clearly be seen. For the first method, the ASD group performed more quickly than controls (although this was only significant for the house condition once age, verbal and performance IQ had been accounted for: $F(1,78) = 8.06, p = .006$) whilst, for the second method, they performed more slowly (although these differences were not significant: $F(1,79) < 1$). This illustrates the differential effect that group differences in accuracy have on reaction time and can be seen clearly in graphs A and B of Figure 5.2 below. This is especially striking for the second method, in which the majority of variance in reaction time is eliminated as it is almost perfectly linearly related to accuracy (with a negative gradient), particularly at low accuracy levels. Both methods can therefore be seen to be consistently biased by the accuracy of the individual.

Given this thesis addresses individual differences in performance, not just group differences, it is important to account for individual accuracy levels within the reaction time data; this will also eradicate the issue of accuracy affecting reaction time data. This can be done by the third method of calculation mentioned above. An ANCOVA was therefore performed on the mean reaction time for correct responses, with accuracy, age, verbal and

Figure 5.2 Relationship between Embedded Figures Test accuracy and reaction time, using three different methods to calculate reaction time: A, reaction time for correct responses only; B, reaction time with 60 seconds assigned to incorrect trials; C, reaction times accounting for accuracy. Line of best fit shown for total sample.



performance IQ entered as covariates. A significant group difference was found only for the house condition ($F(1,77)=8.24, p=.005$), with the ASD group performing faster than the controls (estimated marginal means of 12.95 seconds and 17.53 seconds respectively).

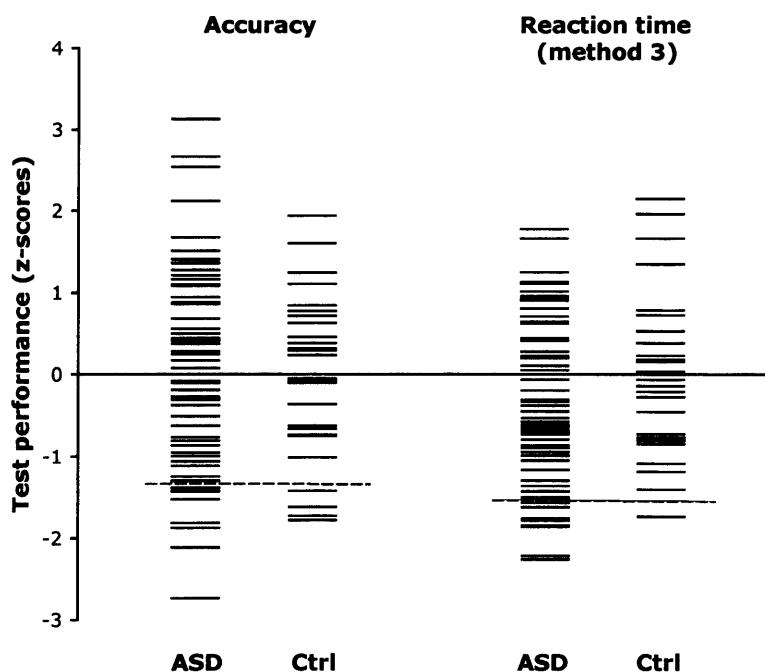
From graph C in Figure 5.2, it can be seen that accuracy is of course no longer reflected at all in the reaction time data.

As so many children performed at floor levels on the adult version of the test, a composite of scores from the two child subtests was also produced. This also allowed for comparison

with previous findings using only the child version of the test. Neither accuracy ($F(1,79)=.38$) nor reaction time (method 3; $F(1,78)=2.64$) produced significant differences between the groups.

Figure 5.3 shows individual differences in EFT accuracy and reaction time (method 3) after accounting for age and IQ. Performance in the ASD group spanned the range of control performance; however, it can also be seen that a number of individuals in this group fell outside the control range, with 4 above and 4 below for accuracy and 6 below for reaction time. On the basis of the accuracy data, eight children were classified as showing weak central coherence, defined as falling below the 5th percentile of control performance (see chapter 6 for further details). For the reaction time data, nine children were classified as showing weak central coherence. However, only one child was classified as showing weak central coherence under both performance measures.

Figure 5.3 Individual performance on the Embedded Figures Test after accounting for age and IQ; low scores indicate high accuracy or fast reaction times and therefore weak central coherence; the position of the x-axis indicates the control mean and the dotted line illustrates the cut-off for the 5th percentile of control performance.



5.1.3 Discussion

Contrary to received wisdom that individuals with ASD are bound to show superior performance on the Embedded Figures Test in comparison to controls, past studies have produced varying results for different research groups. It is the case that there has been a trend towards finding group differences in reaction time but not accuracy though. Due to problems with group matching and data analysis techniques, some of these results may be spurious however. From the information available in the various papers, three studies definitely address these problems (Jolliffe & Baron-Cohen, 1997; Shah & Frith, 1983; van Lang et al., 2006b), two showing group differences in accuracy with low-functioning children, the other in reaction time with high-functioning adults. Indeed, in Jolliffe & Baron-Cohen (1997), accuracy differences may have been observed if the test had not produced a ceiling effect.

It is therefore surprising that the current experiment finds little support for these results, revealing no differences between the groups in accuracy or reaction time on the total measures. A difference was seen however in the reaction times to the house condition of the children's version of the test, with the children with ASD performing on average faster than the control children. This difference was not strong enough to produce group differences on the combined children's measure used in previous studies though.

The most notable differences between the present experiment and the three mentioned above is in the use of higher-functioning children compared to Shah & Frith (1983) and van Lang et al. (2006b) and in using children rather than adults compared to Jolliffe & Baron-Cohen (1997). From these three studies, neither level of functioning nor age appear to affect the presence of group differences and it seems unlikely that the particular combination of being high-functioning and being a child would cause the present results to differ from previous findings.

However, the fact that the ASD children did perform faster as a group than the controls on the house subtest indicates that they were performing in line with previous studies, albeit to a lesser extent (see van Lang et al., 2006b for similar results in accuracy). The house subtest is designed to be more challenging than the triangle subtest and it is possible that the triangle condition produced a slight ceiling effect in the ASD group, allowing the control group to perform very similarly to them. Indeed, the ASD group was non-significantly faster on this subtest. A very similar performance can be seen on the adult version of the test, this time possibly being limited by a floor effect as the test was too hard for many children. The performance displayed by the ASD group on the triangle subtest is therefore rather reminiscent of Jolliffe & Baron-Cohen's (1997) results, given that they used only the 12-item adult version, most appropriate for their population.

Another reason why the results may not have been as clear cut as expected is based on the heterogeneity within the particular sample of individuals used. It can be seen from Figure 5.3 that both the accuracy and reaction time measures in the ASD group spanned the range of control performance, with only a few children performing outside the control range. If only a proportion of the ASD population display weak central coherence or if the distributions for the ASD and control populations are overlapping but displaced from one another, it is possible that a different sample of the ASD population would by chance contain more children with performance outside the control range, producing a stronger group difference. It should be noted that the children falling outside the control range do so in both directions for the accuracy data; this may indicate that both extreme weak and strong central coherence exists in this sample of children. However, it is important to consider here that weak central coherence is shown by extremely good accuracy on this test, making it a very conservative test of the theory; those children showing good performance are likely to really possess a locally biased processing style. Consequently, strong central coherence would be defined as poor accuracy on this test. There are of course a number of different reasons why a child might perform poorly on any test but the

lack of control task here makes it difficult to discount factors which are not specific to the task.

It would therefore be advantageous to further investigate the nature of this processing style using a separate central coherence task in the same population in order to control more tightly for general rather than specific poor performance through task design, and to explore the nature of the mechanism that underlies the purported difference in information processing style in ASD. In addition, this will allow the pattern of individual differences in performance to be further investigated.

5.2 Local-Global Switching task

The other main visuo-spatial task designed to tap central coherence that has been used with individuals with ASD involves Navon figures. In 1977, Navon investigated local and global processing within the normal population using what he termed 'hierarchical stimuli'; large letters made up of smaller letters. These were used to tap into the processing of global and local visual information as both were contained in these stimuli, the large letter being the global element and the small letters being the local elements. Navon created both congruent stimuli, in which the global letter was the same as the local letter and incongruent stimuli, in which the global letter was different from the local letter. These stimuli are therefore referred to as Navon figures (Figure 5.4). Within the normal

Figure 5.4 An example of some Navon figures. A congruent figure is shown on the left, in which the global and local letters are both H; on the right, an incongruent figure is shown, in which the global figure is H whilst the local figure is A.

HH	HH	AA	AA
HH	HH	AA	AA
HH	HH	AA	AA
HHHHHHHH		AAAAAAAA	
HH	HH	AA	AA
HH	HH	AA	AA
HH	HH	AA	AA

population, Navon found that global information was processed faster and often at the expense of local information, a phenomenon he termed 'global precedence'. Specifically, participants were both slower to report the local than the global elements of incongruent stimuli, indicating a *global advantage*, and were slower to report the local elements of incongruent than congruent stimuli, indicating *global interference*.

Navon figures therefore lend themselves ideally to studying different hypotheses about the nature of local and global processing in ASD, such as whether global processing is intact or not, and whether processing biases are present despite intact abilities. A number of authors have utilised these stimuli through a variety of different methods and with varying results (see Table 5.3). Typically, an individual is shown a Navon figure for a short period of time (200-4000 msecs) and is asked to identify which of two target letters was displayed (or sometimes whether or not a particular target letter was displayed). Congruent stimuli only involve a single letter at both levels, whereas incongruent stimuli involve two target letters, only one of which the participant must respond to (see Figure 5.4). As two different letters are present within incongruent stimuli, these are thought to compete and hence increase processing time. Neutral stimuli consist of a target letter paired with a non-target letter, which may be perceptually similar or not to the target letter; these are likely to cause less interference when they are similar to the target stimulus. Both accuracy and reaction times are used as measures of information processing.

The main difference between methods used in different studies concerns whether the participant is informed beforehand which level of processing (local or global) to attend to. Selective attention paradigms provide this information for the participant whereas divided attention paradigms do not; in the latter, individuals may therefore have to quickly switch their attention between levels of processing if the target stimulus is not present in the first level they attend to. In selective attention tasks, the normal global precedence effects are generally seen in everybody, including individuals with ASD (Mottron, Burack, Stauder &

Robaey, 1999b; Ozonoff, Strayer, McMahon & Filloux, 1994; Plaisted, Swettenham & Rees, 1999; Rinehart, Bradshaw, Moss, Brereton & Tonge, 2000), whereas they are either absent in individuals with ASD or replaced by local precedence effects in divided attention tasks, particularly for interference effects (Behrmann et al., 2006; Plaisted et al., 1999; Rinehart et al., 2001). These results indicate that global and local information are

Table 5.3 Studies using a Navon paradigm in individuals with ASD

	Comparison group	ASD age & ability level	Task version	ASD results (G=global, L=local)
Ozonoff et al. (1994)	CA, verbal, non-verbal & full-scale IQ	8-16 years High	Selective attention (was it H or S?)	G advantage & interference
Mottron et al. (1999b)	CA, non-verbal IQ	7-19 years High	Selective attention (was it H or S?)	G advantage & interference (but not in controls!)
Plaisted et al. (1999)	CA, non-verbal IQ	6-16 years High	Selective attention (was it H or S?)	G advantage & interference
			Divided attention (was A present?)	L advantage & interference
Rinehart et al. (2000)	CA, full-scale IQ	6-20 years High	Selective attention (was it 1 or 2?)	G advantage & interference, also L interference
Rinehart et al. (2001)	CA, full-scale IQ	6-20 years High	Divided attention (neutral stimuli only) (was it 1 or 2?)	Slower for G targets after a L target
Mottron, Burack, Iarocci, Belleville & Enns (2003)	CA, full-scale IQ	10-21 years High	Divided attention (neutral stimuli only) (was it H or S?)	No group diff
			Fragmented letters (global level only) (was it H or S?)	No group diff
			Disembedded letters (local level only) (was it H or S?)	ASD group relatively fast
Behrmann et al. (2006)	Age, education level	19-53 years High	Divided attention (was it H or S?)	No advantage, L interference (neither found in controls)

processed normally when the participant is explicitly directed towards a particular processing level, but that a natural bias towards or inability to inhibit local processing is observed when attention is undirected.

At first glance, Mottron et al.'s (2003) results appear to contradict the results from other authors using divided attention paradigms as they found no evidence for a local bias using the same technique. A closer scrutiny of their stimuli is necessary to interpret this difference. Mottron et al. required their participants to indicate whether an H or an S was present in the Navon figure; figures were constructed in rectangular configurations similar to those shown in Figure 5.5 (page 140). They used neutral congruent stimuli in which the second letter present was perceptually similar to the target. As these letters were consistently paired together (an A always with the target H and an E with the target S), the non-target letters can be seen as representing the target that is present, and processing at both levels is therefore not necessary. Consequently, the participant does not need to attend to both levels of the stimuli and so this task should not be considered a divided attention task.

An alternative interpretation of the divided attention results is that children with autism are poor at switching between levels of processing (Plaisted et al., 1999), producing no precedence effects in either level. This could be seen as a problem with executive functioning (see Chapter 4). For example, if an individual has a bias towards global processing and always attends first to the global level, they would be faster on trials in which the target was present at the global than local level. This pattern of performance is certainly the case with normally developing participants. However, if an individual more generally has problems switching and gets 'stuck' within a level, they will always attend first to the level they are currently in, that of the target in the previous trial; this would predict that they would be of similar speeds in both local and global incongruent conditions and slower at both of these than congruent conditions. This is indeed the pattern of results

shown in the reaction time results for the ASD group when performing the divided attention condition in Plaisted et al.'s study.

More recently, Rinehart et al. (2001) have employed a Navon task in which they compared performance across consecutive pairs of trials in order to analyse the ability to stay within a processing level or switch between levels, thus pitting an executive switching problem against a local or global processing bias. They found that individuals with autism, but not Asperger Syndrome, were non-significantly slower to identify a global target stimulus that came after a local target stimulus than vice versa. This borderline result is of interest as it supports the idea of intact global and local processing with a natural bias towards local processing, but also implicates a mechanism for this problem, specifically in the switching from global to local levels.

It would be of interest to pursue Rinehart et al.'s (2001) finding for a number of reasons: to clarify their borderline result and to refine their methodology. Two issues are included in this latter point: there was no control over the inter-stimulus interval and it is unclear how data from incorrect responses were analysed. Furthermore, divided attention paradigms have their own caveats as it is difficult to know how the task is being performed and what strategies are being used. Therefore, rather than group differences reflecting any underlying processing differences, it is possible that differences may reflect differences in strategy use, unrelated to central coherence.

Indeed, Rinehart et al.'s (2001) results prompted Mitchell Valdés, a researcher at the Cuban Neuroscience Centre in Havana, to design a novel task based around the same principles, which will be investigated here (personal communication). His Local-Global (LG) Switching task employs Navon figures composed of only global or local components in an attentional blink paradigm (Raymond, Shapiro & Arnell, 1992). This is the phenomenon whereby participants often have trouble reporting a second stimulus that appears shortly (300-400 msecs) after a first, as they have few attentional resources available to direct

towards it. Moreover, if the participant is required to switch between two different modes of processing for the two stimuli, an additional attentional cost is required, further reducing the probability of correctly reporting the second stimulus (Ward, 1982).

This LG Switching task is well suited to testing individual differences in global and local processing and has been developed and piloted in control adults and individuals with ASD in an as yet unpublished study (Torres, Valdés, Lopez, Garcia & Manzano, 2004). Firstly, it would be expected that a switch from local to global processing or vice versa would be more costly than staying within a mode of processing. Secondly, it would be expected that switching into a less preferable processing mode would be more costly than switching into a favoured mode; if the cost of switching processing mode is low, the participant will therefore be able to process the second stimulus whilst, if the cost of switching processing mode is high, the second stimulus may fall within the attentional blink and so will not be processed. Individuals with ASD would therefore be expected to find it easier to switch from global to local processing and normally developing children would be expected to find it easier to switch from local to global processing.

Furthermore, this paradigm avoids one of the problems of divided attention paradigms; namely that both global and local information are presented at the same time, making it is difficult to know what strategy a participant is using on any one trial. For example, one reason why people with ASD may not show the usual global precedence effects is that they may inefficiently attend to both levels of processing before responding. In the current study, global and local information were presented separately and therefore the only competition is temporal, in terms of the cost of switching and the demands on attention.

Lastly, this paradigm enabled issues relating to the crossover between the executive function and central coherence theories to be addressed. An executive theory would predict that individuals with ASD would show impairments on all switching conditions

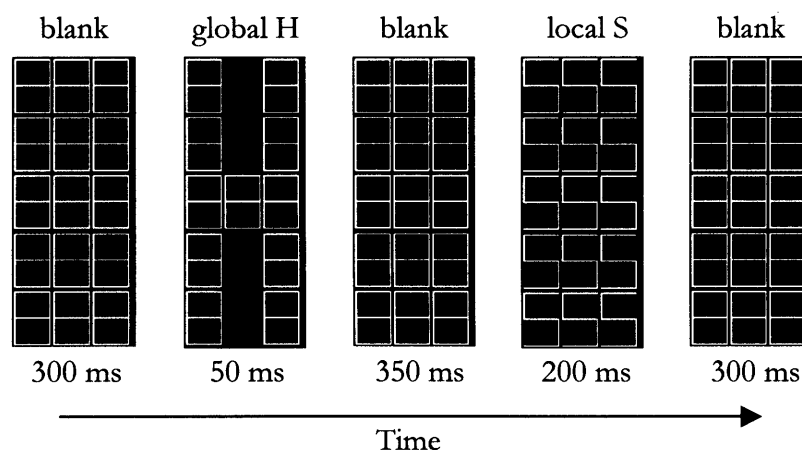
whereas the central coherence theory would predict problems in ASD specifically when switching from local into global but not in the opposite direction.

5.2.1 Method

This experiment was run during test session 3 on a laptop computer using E-prime software (Psychology Software Tools, Inc.). Blank stimuli were constructed from a 5 by 3 grid of elements each consisting of two rectangles, one above the other, similar to a square number 8. Global stimuli were letters constructed from different combinations of these blank elements. Local stimuli were a grid of 15 small letters, each constructed from the relevant subcomponents of a blank element. The letters involved in the task were E, H, P, S and U. Examples of both local and global stimuli can be seen in Figure 5.5.

The participant was first introduced to the same global and local stimuli that they would see during the task. Once they had correctly identified each of the letters, the participant completed 4 blocked conditions in a set order; global-global (GG), local-local (LL), global-local (GL) then local-global (LG). Each block consisted of 5 slow practice trials followed by 25 test trials with corrective feedback after each trial. Each trial consisted of two consecutive letters with an inter stimulus interval of 400 ms between their onsets. Global stimuli had a duration of 50 ms whilst local stimuli had a longer duration of 200 ms; the

Figure 5.5 Time line for a single trial of the global-local condition in the LG Switching task.



latter were much harder to identify during piloting (the normal global advantage) and these durations were designed to produce similar levels of accuracy across the stimulus types.

Accuracy data for the two letters in each trial were recorded and the proportion of responses on which the response to the second letter was correct was calculated for only those trials on which the first response was correct. This was necessary in order to ensure that the first stimulus had been processed and there had been a chance for the attentional blink to occur. Chance performance would therefore result in correct responses 20% of the time. As reaction time data was not of interest, it was also stressed that the participant was under no time constraints.

Figure 5.5 gives the time-line for each trial; in this example, a switch from global to local processing is required. The child must respond to each trial by saying which two letters they saw (here, H and S). This paradigm has been trialed with small samples of low-functioning adolescents and adults with autism (Torres et al., 2004), revealing that those with autism were indeed worse at switching from local to global stimuli while controls (normally developing adolescents and adults) were worse at the condition which required a switch from global to local processing.

5.2.2 Results

Data were not available for three children in the ASD group as they were unable to complete the task; all three thought the task was impossible and therefore became frustrated and upset that they had been asked to do something that they thought they could only fail at. Two of these children were young with intelligence in the normal range, one boy and one girl; the third child was an older girl with particularly low performance IQ. All three children completed all other tasks.

As mentioned in section 5.2.1, only those trials on which a child responded correctly to the first letter were suitable for analysis of the attentional blink paradigm, resulting in different

children having different numbers of trials analysed. However, in order to ensure that the children in both groups found the task of equal difficulty and any further analyses involved the same mean number of trials in both groups, responses to the first letter were also analysed. While there was a slight trend for the children with ASD to respond correctly to fewer first letters than the controls, group differences were not significant after accounting for the effects of age and IQ ($F(4,73)=1.36$).

In order to calculate each individual's ability to switch into a level, the difference between the relevant non-switching and switching condition was calculated; for switching into global, each participants score was calculated as GG-LG, whilst for switching into local, LL-GL was used. This gave a measure for each child of the cost of switching into a level in comparison to that child's own ability of processing within that same level.

As can be seen from Table 5.4, group means indicate that both groups found the switching conditions (GL and LG) harder than the non-switching conditions (GG and LL). The

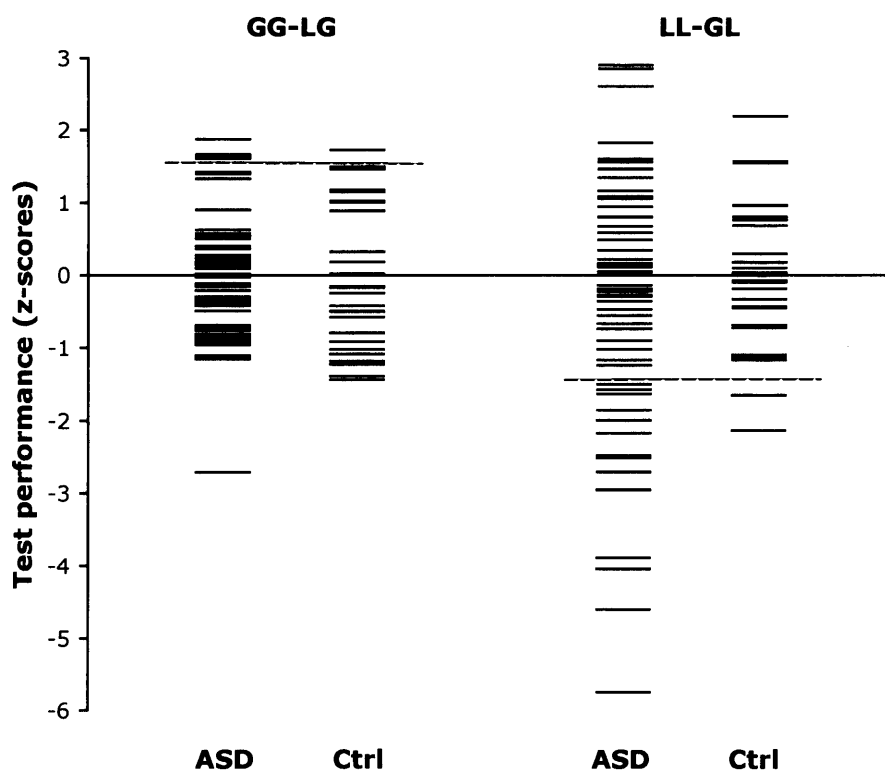
Table 5.4 Means (and standard deviations) *with range* of LG Switching task results; scores reflect the proportion of correct responses to the second stimulus for trials on which the first stimulus was correctly identified.

	Control group	ASD group
Global-Global (GG)	0.70 (0.22) .00–1.00	0.63 (0.26) .00–1.00
Local-Local (LL)	0.81 (0.18) .43–1.00	0.78 (0.24) .00–1.00
Global-Local (GL)	0.51 (0.17) .20–.91	0.55 (0.23) .00–1.00
Local-Global (LG)	0.55 (0.31) .05–1.00	0.51 (0.27) .10–.96
Global switching cost (GG-LG)	0.15 (0.26) -.25–.56	0.12 (0.25) -.67–.68
Local switching cost (LL-GL)	0.30 (0.14) .07–.65	0.23 (0.24) -.50–.71

ASD group were performing slightly worse across all conditions except the GL condition, on which they performed slightly better than controls, as predicted. In general, their switching costs were also lower in both conditions (GG-LG and LL-GL), although as expected this was more pronounced in the local condition. However, a 2x2 repeated measures ANCOVA, entering age, verbal and performance IQ as covariates, revealed that there was no main effect of condition ($F(1,76)=.58$) or group ($F(1,76)=.63$) and no interaction between the two ($F(1,76)=.55$).

Individual differences in performance on both switching conditions were examined. Five children were found to have particular difficulty (below the 5th percentile of control performance) switching into the global level of processing and 14 children were particularly good at switching into the local level of processing (see Figure 5.6).

Figure 5.6 Individual performance on the local and global switching conditions of the LG Switching task after accounting for age and IQ; high scores indicate high cost of switching (high GG-LG scores and low LL-GL scores therefore indicate weak central coherence); the position of the x-axis indicates the control mean and the dotted line illustrates the cut-off for the 5th percentile of control performance.



5.2.3 Discussion

The results from the LG Switching task reflect those from the Embedded Figures Task in terms of a lack of results. Children with ASD did not differ significantly from normally developing children despite a trend towards a relatively lower switching cost in the ASD group on the local condition. While the failure to replicate the borderline result of Rinehart et al. (2001) and the Cuban pilot study (Torres et al., 2004) was disappointing, this again reveals the inconsistency of results in the central coherence literature on local and global processing. The fact that similar results were found here, across two quite different tests tapping into local and global processing, indicates that this may indeed be a result dependent on the particular sample due to heterogeneity within the ASD population.

How is the subgroup of children with ASD to be identified, who are responsible for the marginal group differences in experimental studies of local processing bias? One hint is given by the hypothesis of under-connectivity due to pruning failure (see section 1.3.2): is there therefore a relationship between head size and weak central coherence? The suggestion is that the increased head or brain size seen in a small proportion of individuals with ASD may result from a lack of early neuronal pruning leading to diffuse and inefficient feedback connections. A lack of top down modulation might then result in good exemplar-based or local processing and poor generalisation or global processing, as is currently assumed to be characteristic of weak central coherence. To this end, the effect of head size on performance on tests of central coherence was therefore examined.

5.3 Head size and central coherence

Head z-scores were available only for those children who were Caucasian, which reduced the size of the ASD and control groups to 52 and 25 respectively for the Embedded Figures Test and 49 and 25 for the LG Switching Task.

5.3.1 Results

Partial correlations between these variables, accounting for age, verbal and performance IQ, were calculated and revealed that no relationship existed between performance on the Embedded Figures Test and head size in either group (ASD: $r < .26$; control: $r < .28$).

However, within the ASD group only, head size was correlated to the cost of switching into the global mode of processing in the LG Switching task ($r = .36$, $p = .014$). As Figure 5.7 shows, children with larger heads had a greater cost of switching into a global mode of processing than children with smaller heads. Head size was unrelated to the cost of switching into a global mode of processing in the control group ($r = -.22$). Furthermore, it was unrelated to the cost of switching into local processing in either group (ASD $r = .02$; control $r = -.06$).

Group differences between those children in the ASD group who could be classed as having macroencephaly (head circumference z-score greater than 1.88, equivalent to the

Figure 5.7 Relationship between head circumference and cost of switching into global processing in both groups; line of best fit is represented for ASD group only.

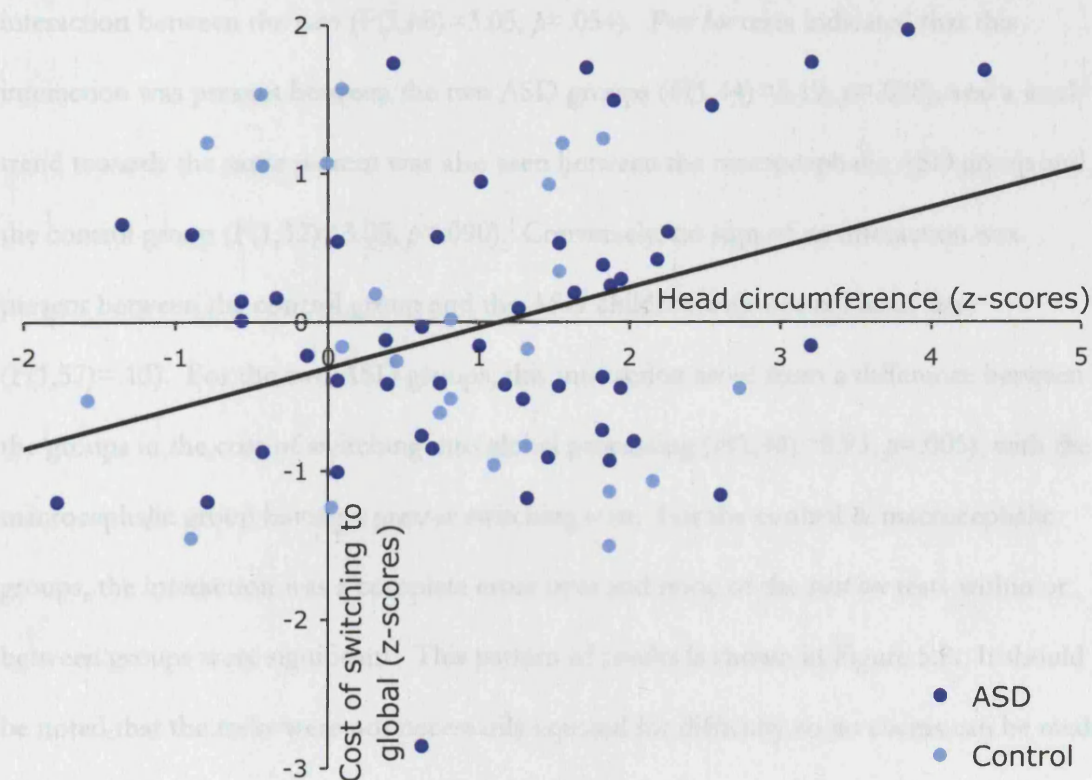


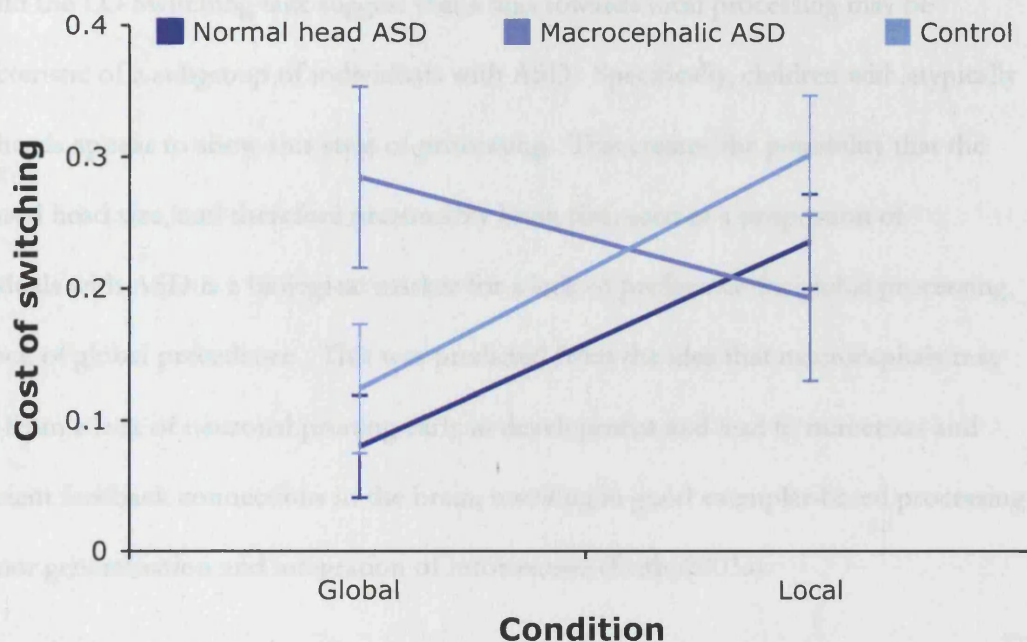
Table 5.5 Means (and standard deviations) *with range* for LG Switching task results, with the ASD group split into those with or without macrocephaly.

	Control group (25)	ASD group (49)	
		Normal head size (37)	Macrocephalic (12)
Global switching cost (GG-LG)	0.15 (0.26) -.25-.56	0.08 (0.22) -.67-.60	0.26 (0.29) -.18-.68
Local switching cost (LL-GL)	0.30 (0.14) .07-.65	0.24 (0.23) -.30-.71	0.18 (0.30) -.50-.66

top 3% of the normal population, here 12 children or 24% of the ASD group), those with head sizes in the normal range, and the controls (2 children or 7% of whom could be classed as macrocephalic) were therefore analysed. Table 5.5 shows the means and standard deviations for the three groups and indicates that the macrocephalic ASD group did indeed show a greater cost when switching into global processing than the remaining children with ASD and the controls.

A 2x2 repeated measures ANCOVA comparing all three groups revealed no main effect of either condition ($F(1,68)=1.06$) or group ($F(2,68)=1.48$) but a borderline significant interaction between the two ($F(2,68)=3.05, p=.054$). *Post hoc* tests indicated that this interaction was present between the two ASD groups ($F(1,44)=5.19, p=.028$), and a weak trend towards the same pattern was also seen between the macrocephalic ASD group and the control group ($F(1,32)=3.05, p=.090$). Conversely, no sign of an interaction was present between the control group and the ASD children with normal head sizes ($F(1,57)=.13$). For the two ASD groups, this interaction arose from a difference between the groups in the cost of switching into global processing ($F(1,44)=8.93, p=.005$), with the macrocephalic group having a greater switching cost. For the control & macrocephalic groups, the interaction was a complete cross over and none of the *post hoc* tests within or between groups were significant. This pattern of results is shown in Figure 5.8. It should be noted that the tasks were not necessarily equated for difficulty so no claims can be made

Figure 5.8 Interaction between the cost of switching into global versus local processing in the macrocephalic ASD group versus the remaining children with ASD and the controls. Error bars represent standard error of the mean.



about the absolute cost of switching across the two conditions within a group; for example, it may be that the local conditions were generally harder and therefore that the control and normal head size ASD groups found both types of switching to be of equal difficulty.

Given this LG Switching task has not been used previously in populations with ASD, partial correlations between performance on this task and the Embedded Figures Test were examined. In the ASD group, the cost of switching into either the global or local level was unrelated to Embedded Figures performance (accuracy and reaction time, $r < 0.15$).

Additionally, when comparing those children with extremely good and extremely poor EFT accuracy, the children with good performance had slightly but non-significantly bigger head z-scores than those with poor performance ($t(12) = .83$). Furthermore, differences between those children in the ASD group with and without macrocephaly on the different tests in the ToM and EF domains were also explored but revealed no significant differences ($F(9,39) = 1.11$).

5.3.2 Discussion

While such a novel result needs replicating, the results from the interaction between head size and the LG Switching task suggest that a bias towards local processing may be characteristic of a subgroup of individuals with ASD. Specifically, children with atypically large heads appear to show this style of processing. This creates the possibility that the increased head size, and therefore presumably brain size, seen in a proportion of individuals with ASD is a biological marker for a lack of preference for global processing, or a lack of global precedence. This was predicted from the idea that macrocephaly may result from a lack of neuronal pruning early in development and lead to numerous and inefficient feedback connections in the brain, resulting in good exemplar-based processing but poor generalisation and integration of information (Frith, 2003a).

The high level of control of the LG Switching task conditions allows a careful examination of exactly where the difference in this processing style lies. Individuals with macrocephaly appear to have difficulties switching into global processing mode; they show a greater cost and presumably have to use more resources to overcome this attentional barrier. There was no evidence however that individuals with macrocephaly have difficulty processing global stimuli *per se*; rather they have difficulty when having to switch out of local processing and into global processing. This would predict that individuals with macrocephaly would have problems on any task in which they have to switch into global processing under pressured conditions; this may indeed be the problem in divided attention Navon tasks whilst selective attention tasks require processing only within the current level. This could be described as a local bias or preference for local over global processing, in the same way that Happé & Frith (2006) use the term (see section 1.3.6), and is consistent with suggestions made by Plaisted (Plaisted, Dobler, Bell & Davis, 2006). These results could also be considered consistent with Mottron's (Mottron et al., 2006) Enhanced Perceptual Function theory if a local enhancement can be conceptualised as a local bias, although the

fact that the significant result was in an inability to switch into global processing departs from this theory (see section 1.3.7). The important difference to these theories being made here, however, is that this processing style is not characteristic of ASD but rather of ASD plus macrocephaly.

These results have implications for the executive function theory of autism. Whilst this theory would have predicted impaired performance across both switching conditions, the results find little support for this idea. As already mentioned, those individuals with macrocephaly had difficulties specifically with switching in a particular direction, from local into global processing, but not vice versa. Furthermore, those individuals with ASD but normal head size were performing similarly to controls across all conditions, if anything performing slightly more efficiently on the switching conditions. This would indicate that any problems seen with switching set are likely to occur at a conscious level as this task was assessing the capacity of the brain to switch quickly between levels. Indeed, the problem in executive tasks may be more to do with prompting oneself to switch levels or choosing when to do so, whilst possessing an intact ability to perform the switch when prompted externally as in the LG Switching task.

It appears from these results however that the LG Switching task is tapping into a slightly different ability to the Embedded Figures Test; correlations were not seen between these two tasks or between the Embedded Figures Test and head size. This might be because of the complex, uncontrolled and open-ended nature of the Embedded Figures Test. In this task, participants are instructed to ignore the global image and search for the local image within it, hence a switch between local and global processing is not necessary. The advantage that individuals with ASD sometimes show in this task may come from the controls finding it hard to ignore the global image and slipping out of local processing rather than from individuals with ASD finding it hard to switch into global processing. Furthermore, there is a high degree of conscious control over processing in this task,

making it difficult to assess the strategies used by different participants to achieve good performance. The LG Switching task, on the other hand, is a more implicit task, putting the attentional system under maximum stress or pressure whilst measuring its efficiency.

5.4 Conclusion

Both the Embedded Figures Test and the LG Switching task results together indicate that findings supporting the presence of weak central coherence in autism are not unequivocal. In this population of high-functioning children with ASD, there was little evidence of this processing style in the whole group. However, when the presence of macrocephaly was used to split the group, a clear pattern of results emerged; those children with ASD and macrocephaly showed a bias towards local processing on the LG Switching task, portrayed in a greater processing cost when switching from local into global processing. If these results are replicated, this would imply that weak central coherence is restricted to a subgroup of individuals with ASD and should therefore not be considered as a universal property of ASD. The tentative hypothesis can therefore be proposed that head size may be a biological marker of weak central coherence and may provide a useful endophenotype for investigating the genetic basis of a subgroup of individuals with ASD.

Chapter 6: Cognitive impairments

Whilst intended to assess general cognitive ability, by design intelligence tests also rely on a number of different cognitive skills. It has been suggested that some of these skills may be similar to those cognitive abilities proposed to be causal in ASD (Frith, 1989). The first part of this chapter therefore assesses the relationships between the three cognitive impairments involved in this thesis and performance on the various subtests of the measure of intelligence used here. Furthermore, it has been suggested that the underlying cognitive impairments in autism may be related in a variety of ways (eg. Perner & Lang, 1999; Russell, 1997). The second part of this chapter will therefore clarify the relationships between theory of mind, central coherence and executive function through the various tests used here, designed to tap into each of these domains.

6.1 The relationship between cognitive impairments and intelligence test performance

Although intelligence tests are designed to tap into general cognitive ability, intelligence test performance in ASD is typically characterised by an uneven profile, particularly on the Wechsler IQ tests. This profile contains distinctive peaks and troughs, most notably with a peak on the Block Design subtest and a dip on the Comprehension subtest (Lockyer & Rutter, 1970). This pattern has been observed both in individuals and across whole groups of individuals with ASD (Happé, 1994). It has also been shown to be independent of: overall IQ level, being present in both high- and low-functioning populations (Shah & Frith, 1993); the discrepancy between verbal and performance IQ, being present even when verbal IQ is higher than performance IQ (Lincoln, Allen & Kilman, 1995); the specific diagnosis of the individual within the spectrum, being present for both autism

(Shah & Frith, 1993) and Asperger Syndrome (Bowler, 1992); and of age, being present in children (Siegel, Minshew & Goldstein, 1996) and adults (Dennis et al., 1999).

It has been suggested that different cognitive impairments may explain this typical pattern of IQ test performance (Frith, 1989). The peak of performance seen in the Block Design subtest is thought to result from weak central coherence (CC). This test requires the child to recreate a given geometric pattern using blocks which have two coloured sides, two white sides and two sides diagonally split between colour and white (see Figure 6.1). In order to succeed at this task, the child must focus in on the local detail of the pattern, mentally dividing the pattern into components the size of an individual block, and not be swayed by cohesiveness of the overall pattern. The presence of a bias towards local processing would therefore be an asset in such a test.

This suggestion has been supported by a study using the Block Design Task and a modified novel 'segmented' version (Shah & Frith, 1993). By presenting the geometric pattern to be

Figure 6.1 The Block Design subtest from the Wechsler Intelligence Scales.

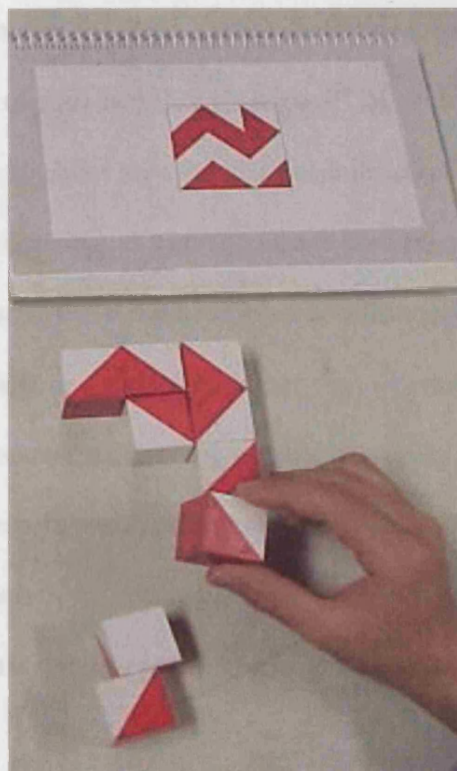


Figure 6.2 The novel segmented (left) and the original un-segmented (right) version of the Block Design test (taken from Caron, Mottron, Berthiaume & Dawson, 2006).



constructed in a segmented display, with each block shown separately, the child no longer needs to mentally divide the pattern and so is not affected by the global design (see Figure 6.2). Indeed, in this study the control children were just as fast as the age and ability matched children with autism in this segmented version, but when presented with the original un-segmented version, the control children fell behind the children with autism. This result has now been replicated (Caron et al., 2006).

Similarly to Block Design, the trough on the Comprehension subtest is thought to arise from the cognitive impairment in Theory of Mind (ToM) present in ASD (Frith, 1989). This subtest consists of questions about how to behave in certain life situations and why certain aspects of our society function in the manner they do, which the child must answer. For example, one question asks the child, “What should you do if you see thick smoke coming from the window of your neighbour’s house?”. A correct answer to this question would require the child to consider why the experimenter asked the question and the kind of answer they might be expecting, as well as to think about the question in terms of other people in addition to himself. So in this example the child must infer that the experimenter is really asking for a response to a house fire rather than to smoke, and that the appropriate response would involve consideration of the neighbour’s safety (calling the fire brigade) whilst maintaining his own safety (not going into the house to investigate). In this manner this task is likely to load heavily on an understanding of other people’s (the experimenter’s and the neighbour’s) mental states.

Evidence to support the idea that a ToM impairment may be causal of the Comprehension trough comes from a study by Happé (1994). Individuals with autism were divided into those who failed at least one of two standard first-order false belief tasks and those who passed both tasks. The results indicated that those individuals who failed the false belief tasks had poorer performance on the Comprehension subtest and were more likely to have a personal Comprehension trough relative to their overall verbal level. This indicates that the inability to pass false belief tasks has a detrimental effect on Comprehension subtest performance.

Although the classic uneven profile on IQ subtests can possibly be explained by the two cognitive theories of weak CC and ToM, it is less clear how the third candidate theory of executive function (EF) would affect intelligence test performance in ASD; this has not been investigated and no specific predictions have previously been made. One possibility is that executive dysfunction might adversely affect accomplishment of the performance IQ subtests. It is well documented that patients with acquired frontal lesions are poor both on tests of EF and fluid intelligence but show retained performance on tests of crystallised intelligence (Duncan, Burgess & Emslie, 1995). Fluid intelligence tests are thought to involve novel 'online' problem solving and reasoning, as do many tests of executive function, whereas crystallised intelligence tests require reiteration of previously learnt information. In the Wechsler Intelligence Scales, fluid intelligence therefore seems to be most reflected in the performance rather than verbal subtests and therefore leads to the hypothesis that performance IQ will be lower in individuals with autism displaying executive dysfunction.

The analysis presented in the first part of this chapter therefore aims to investigate how WISC subtest performance is affected by the cognitive skill of children with ASD in the three cognitive domains under consideration in this thesis, by comparing performance on the WISC to performance in these three cognitive domains. In addition it will examine

whether the distinctive IQ test profile that has been previously documented is present in all children with ASD.

6.1.1 Method

In order to investigate the presence of the characteristic WISC IQ profile, the following method was used to define the presence of subtest peaks and troughs in each individual in the current sample. Verbal and performance IQ subtests were treated independently as the discrepancy between them is known to vary widely in either direction in ASD (Lincoln et al., 1995; Manjiviona & Prior, 1995). Within both verbal and performance IQ therefore, the mean subtest scaled score and standard deviation were computed and one standard deviation above and below the mean was calculated. Subtest scores outside this range were then considered to be peaks or troughs.

It is important to note here that it is possible to do well on a subtest without it being a personal peak and, similarly, that it is possible to perform poorly on a subtest without it being a personal trough. Peaks and troughs are therefore relative to the overall ability level of an individual. Another important observation to make is that this method is more conservative than that used previously (see Happé, 1994) in which all tests can be seen as peaks and troughs away from verbal or performance mean.

6.1.2 Results

In order to compare the overall group profile in the ASD group to that in the control group, the IQ differences between the groups first needed correcting; this is important given that the control group had higher verbal and performance IQs than the ASD group so any subtle differences in profile would be masked by overall IQ differences. In order to do this, the discrepancy between the two groups in terms of the mean subtest scaled score for verbal IQ and performance IQ separately was calculated and subtracted from each

control participant's score for each subtest. This retained the relative pattern of performance across subtests whilst lowering the absolute level of subtest performance in the control group in a consistent manner. Each verbal IQ subtest was lowered by 1.54 scaled score points and each performance IQ subtest was lowered by 1.22 points in the control group. Group differences on each subtest were then examined and a trend towards a significant difference was found only for the Comprehension subtest ($t(82)=1.95$, $p=.054$). However, as Figure 6.3 indicates, the ASD group were performing relatively well on the Block Design, Similarities, Information and Object Assembly subtests and poorly on the Coding and Picture arrangement subtests.

The relationship between individual differences on the Block Design (BD) subtest and weak CC was first explored. Fifteen children with ASD were located with personal BD peaks and were compared to the remaining children with ASD. These groups were matched on age ($t(55)=.57$), verbal IQ ($t(55)=.17$) and performance IQ ($t(55)=.08$) but, as

Figure 6.3 WISC subtest profile; 'corrected' relative performance levels shown for control group (with absolute performance levels superimposed as paler bars).

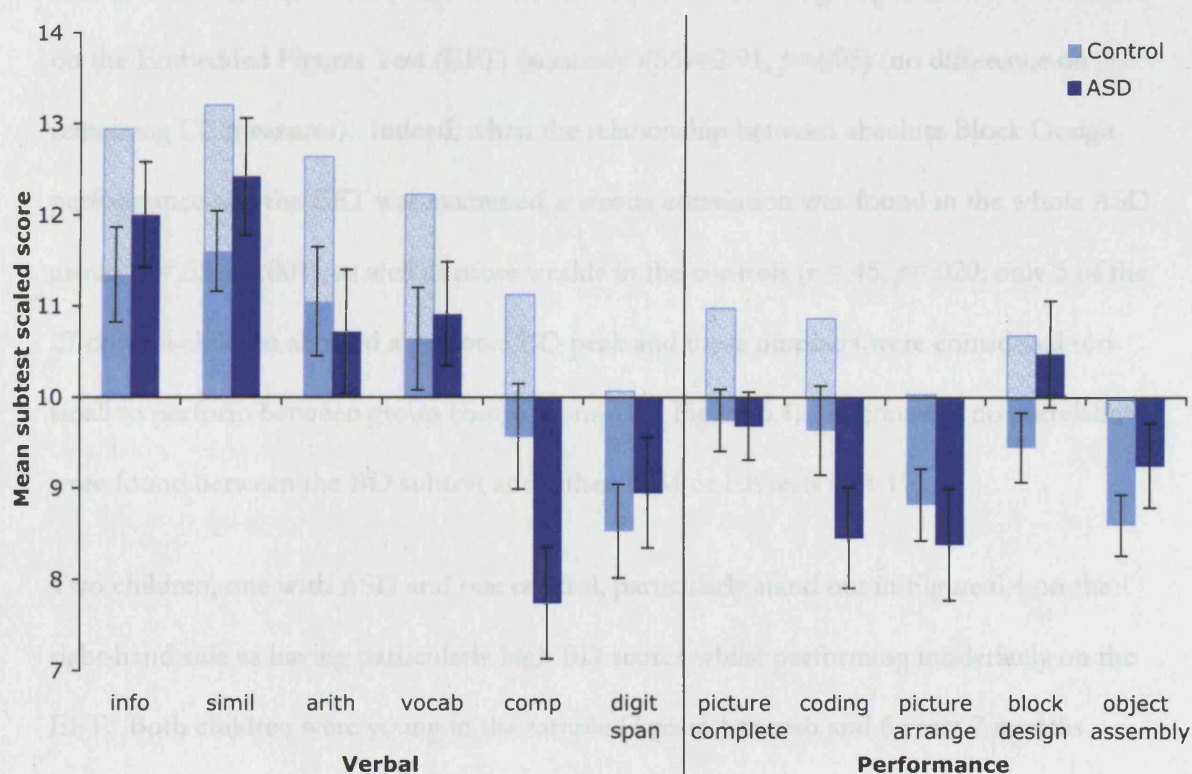
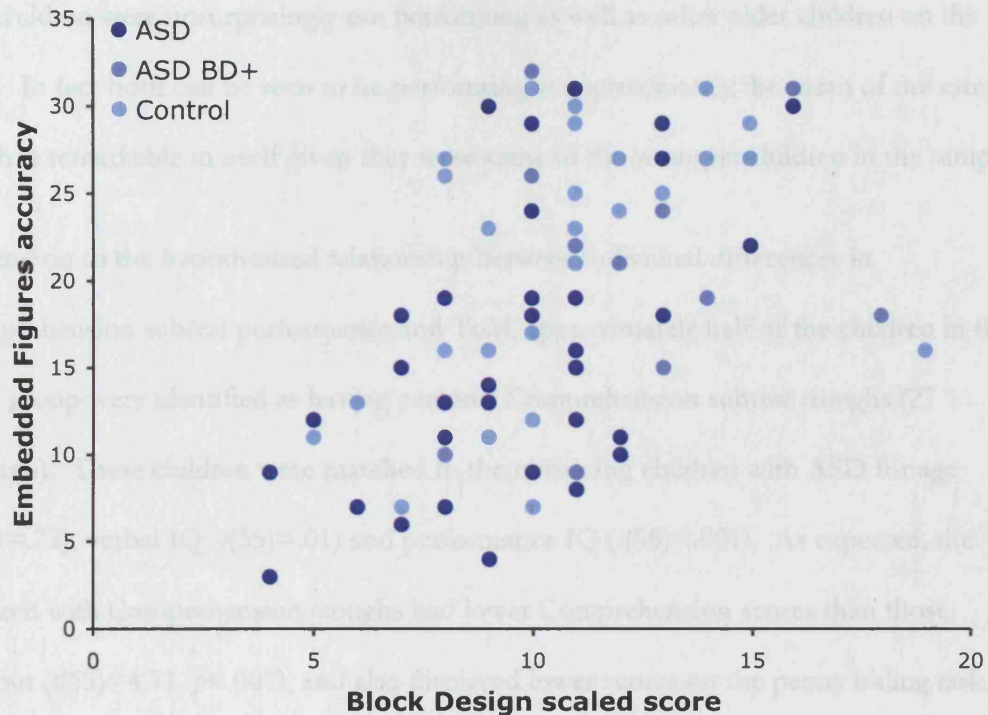


Figure 6.4 Performance on the Block Design subtest and Embedded Figures Test in the ASD and control groups; ASD group divided by presence of personal Block Design peak (BD+).



would be expected, the group with personal BD peaks showed higher Block Design scaled scores than the remaining children ($t(55)=3.97, p<.001$). No group differences were found between these groups on any ToM or EF test ($t(55)<1.8$) but a group difference was found on the Embedded Figures Test (EFT) (accuracy $t(55)=2.91, p=.005$) (no difference on remaining CC measures). Indeed, when the relationship between absolute Block Design performance and the EFT was examined, a strong correlation was found in the whole ASD group ($r=.55, p<.001$), as well as more weakly in the controls ($r=.45, p=.020$; only 5 of the 27 control children showed a personal BD peak and these numbers were considered too small to perform between group comparisons) (see Figure 6.4). In contrast, no correlations were found between the BD subtest and either ToM or EF tests ($r<.17$).

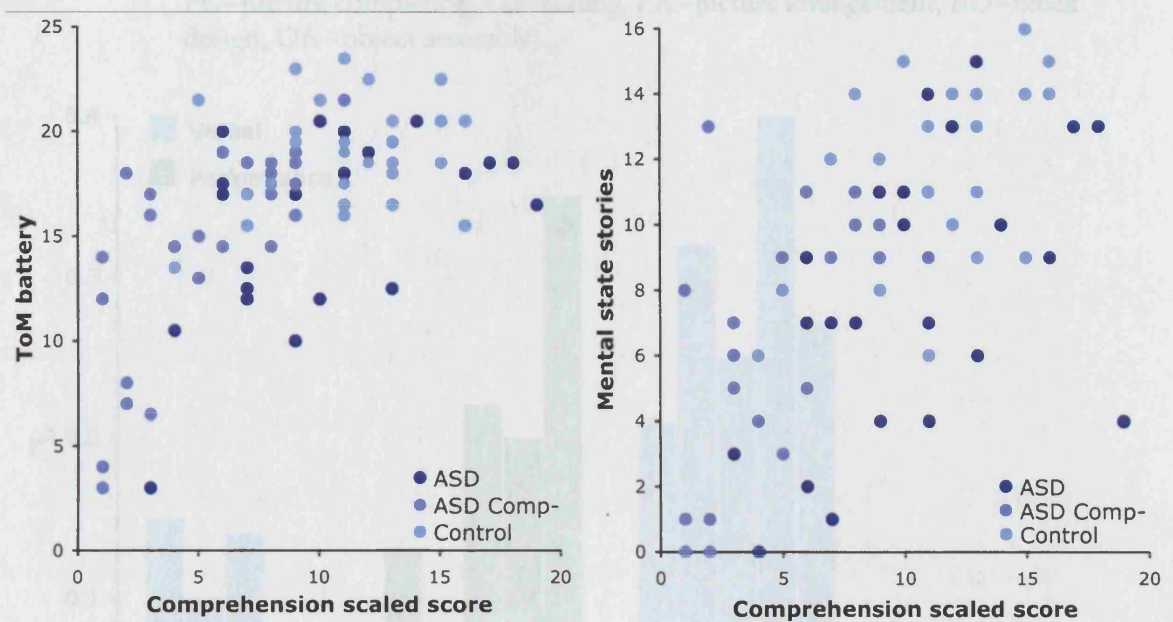
Two children, one with ASD and one control, particularly stand out in Figure 6.4 on the right-hand side as having particularly high BD scores whilst performing moderately on the EFT. Both children were young in the sample (7 years 1 month and 6 years 7 months

respectively at the first test session). Whilst the BD scaled score accounts for age, the EFT does not; consequently, whilst performing outstandingly well for their age on BD, these two children were unsurprisingly not performing as well as other older children on the EFT. In fact both can be seen to be performing at approximately the mean of the sample, which is remarkable in itself given they were some of the youngest children in the sample.

Moving on to the hypothesised relationship between individual differences in Comprehension subtest performance and ToM, approximately half of the children in the ASD group were identified as having personal Comprehension subtest troughs (27 children). These children were matched to the remaining children with ASD for age ($t(55)=.22$), verbal IQ ($t(55)=.01$) and performance IQ ($t(55)=.001$). As expected, the children with Comprehension troughs had lower Comprehension scores than those without ($t(55)=4.71$, $p<.001$), and also displayed lower scores on the penny hiding task ($t(55)=2.54$, $p=.014$) but not on the false belief tasks ($t(55)=.32$) or mental state Strange Stories ($t(55)=1.13$). No group differences were found on the CC or EF tests, except the Navon task where the group with Comprehension troughs appeared to find it easier to switch into global processing ($t(52)=2.12$, $p=.039$) and harder to switch into local processing ($t(52)=2.33$, $p=.024$) than the remaining children (indicative of a preference for global processing or strong CC); this finding was unexpected. Correlations were found between Comprehension subtest scores and performance on all ToM tests ($r>.36$, $p\leq.006$; see Figure 6.5) but also with the tests of EF ($r>.34$, $p\leq.009$). In the controls, correlations were found only for the mental state Strange Stories ($r=.50$, $p=.009$).

Given that a specific hypothesis concerning a particular subtest had not been made for the EF domain, personal peaks or troughs in performance could not be evaluated. In order to investigate the effect of EF on IQ test performance therefore, correlations between the IQ subtests and the BADS (total score) were examined. As expected, correlations were found mainly between the performance IQ subtests and the BADS in the control group

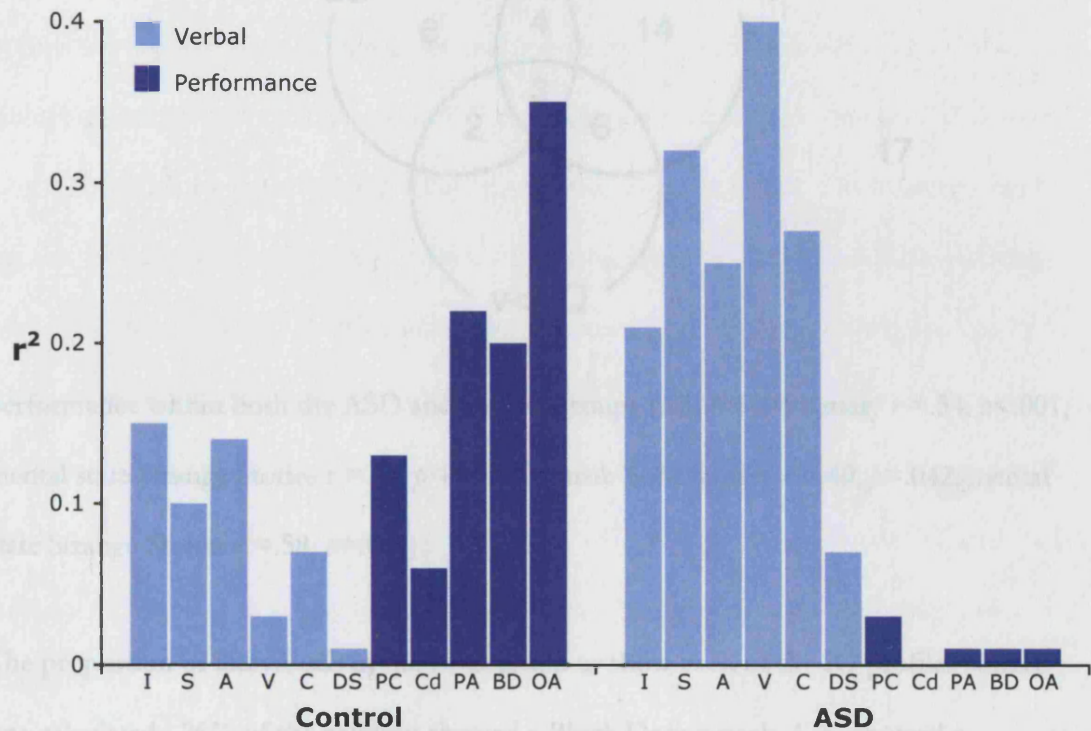
Figure 6.5 Performance on the Comprehension subtest and the ToM tests in the ASD and control groups; ASD group divided by presence of personal Comprehension trough (Comp-).



(performance IQ x BADS total: $r = .69, p < .001$), particularly for the Object Assembly subtest ($r = .59, p = .001$) but also for Picture Arrangement ($r = .47, p = .013$), Block Design ($r = .44, p = .021$) and Information ($r = .39, p = .045$). Contrary to this finding and to expectations, no correlations were seen between the performance IQ subtests and the BADS in the ASD group (performance IQ x BADS total: $r = .14$). Instead, strong correlations were found between all the verbal IQ subtests and the BADS ($r > .46, p < .001$) with the exception of digit span ($r = .27, p = .040$) (verbal IQ x BADS total: $r = .41, p = .002$) (see Figure 6.6). A similar pattern of results was seen for the Shallice Switching Sentence Completion Task (SSSCT), with significant correlations with all verbal IQ subtests ($r > .26, p < .05$) except arithmetic and digit span but not with performance IQ subtests.

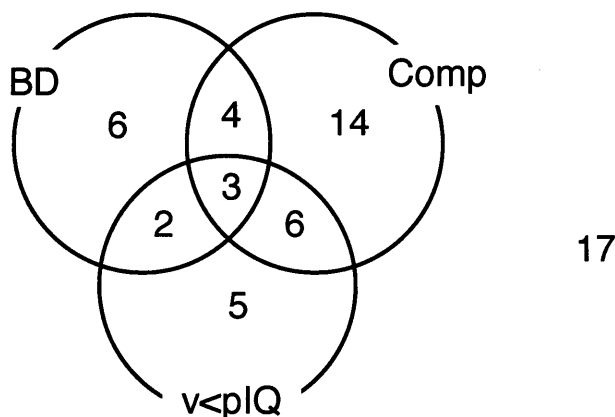
Given that executive function appears to be related to verbal IQ as a whole in the ASD group, those children with relatively low verbal IQ in comparison to their performance IQ were compared to the remaining children with ASD. As it was reasonably rare for children in this sample to show verbal IQ lower than performance IQ (see section 2.2.3 for a

Figure 6.6 Correlations between IQ subtests and the BADS total score in each group; correlations given as r^2 values, signifying proportion of shared variance; each bar represents a different subtest (I=information, S=similarities, A=arithmetic, V=vocabulary, C=comprehension, DS=digit span, PC=picture completion, Cd=coding, PA=picture arrangement, BD=block design, OA=object assembly).



discussion of why this might be), all 16 of the children whose verbal IQ was lower than their performance IQ were compared to the 41 children with verbal IQ greater than performance IQ. These groups were matched for age ($t(55)=.09$) and performance IQ ($t(55)=.97$) but the group with verbal lower than performance IQ had lower verbal IQs ($t(55)=5.97, p<.001$). These groups differed on the BADS overall score ($t(55)=5.63, p<.001$), but also more specifically on the Cards test, Water test, Key Search test, Zoo Map 1 and the Six Parts test ($t(55)>2.23, p<.029$) with the relatively low verbal IQ group performing worse (but not on the Zoo Map 2 or SSSCT). While these groups did not differ on the CC tests ($t(55)<1.54$), they differed both on the ToM battery ($t(17.6)=3.69, p=.002$) and the mental state Strange Stories ($t(21.6)=3.59, p=.002$), again with the low verbal IQ group performing worse. Verbal IQ was found to be correlated to ToM

Figure 6.7 Venn diagram to show the number of children with ASD who displayed Block Design (BD) peaks, Comprehension (Comp) troughs or verbal IQ lower than their performance IQ ($v < pIQ$); the 17 children outside the diagram represent those with none of these profiles.



performance within both the ASD and control groups (ASD: ToM battery $r = .54, p < .001$, mental state Strange Stories $r = .64, p < .001$; Control: ToM battery $r = .40, p = .042$; mental state Strange Stories $r = .58, p = .002$).

The proportion of individuals in the ASD group to show each of the IQ profiles studied was calculated. 26% of the children showed a Block Design peak, 47% showed a Comprehension subtest trough, and 28% showed lower verbal than performance IQ. In combination with Figure 6.7, this indicates that any of these profiles can exist in any combination. Furthermore, 30% of children did not show any of these profiles.

6.1.3 Discussion

With respect to IQ profiles in ASD, these results suggest that children on the autism spectrum show an uneven profile in the majority of cases. Exactly where this unevenness was found in any one individual was more variable however; approximately a quarter showed a Block Design peak, half showed a Comprehension trough and a quarter showed relatively low verbal IQ. These different profiles were also found to overlap, occurring in all possible combinations. However, approximately a third of children with ASD showed no evidence of any of these profiles.

The second aim was to investigate the relationship between the three cognitive impairments studied in this thesis and IQ subtest performance. The cognitive impairments suggested to be core to ASD do relate to the IQ profile even in high-functioning children; in fact all three appear to be independently necessary to interpret the profile. Specifically, both group differences and correlations indicated that the ToM deficit was revealed in the Comprehension subtest trough, weak CC through the Block Design peak and EF through a broader effect across verbal IQ. In terms of assessment therefore, Block Design can be considered a possible indicator of weak CC and so children with a personal Block Design peak may have weak CC; children with this profile should therefore be followed up. Similarly, children with a personal Comprehension subtest trough are likely to have a ToM impairment and should be followed up. Furthermore, children with ASD with relatively low verbal IQ in comparison to their performance IQ may show EF deficits. These subtests can be seen in some sense to have a screening function for certain cognitive impairments.

The finding that children with ASD who had Comprehension troughs had a bias towards global processing indicative of strong CC was unexpected. While it is difficult to interpret these weak group differences, at very least it indicates that poor ToM and weak CC are likely to be independent cognitive impairments in ASD.

The presence of some null results in the ToM domain (see page 158) in relation to the Comprehension subtest may indicate that more than one factor is contributing towards performance. The fact that correlations between each ToM test and Comprehension were strong but group differences were not significant for the false belief tasks and the mental state Strange Stories suggests that the range of ToM scores in the two groups were large and overlapping. Indeed, Figure 6.5 (p160) indicates that there are some children with poor Comprehension scores but relatively good ToM performance, but not vice versa. It is possible that a child might be able to reason about another's mental state when explicitly

asked to do so but, when given a more real-life scenario, would not think to consider another's point of view. This was shown in one such child's answer to the Comprehension question about thick smoke given in the introduction to this chapter; he simply stated that he would close his own windows to keep the smoke out. Such a tendency to overlook another's point of view despite being capable of mental state reasoning in a simpler and more explicit scenario indicates that ToM is fragile and that performance is dependent on other cues. A further reason for the absence of significant differences within the ASD group on some of the ToM tests despite correlations with the Comprehension subtest is due to ceiling effects, particularly in the false belief tasks. This would also explain the lack of correlation in the controls for the false belief tasks but not the mental state Strange Stories. Despite these criticisms, these tests were still powerful discriminators between the control and ASD groups (see chapter 3).

Low ToM task performance in the presence of relatively low verbal IQ may be the result of particularly low Comprehension subtest scores pulling overall verbal IQ down. However, this relationship has been seen many times before, using simpler vocabulary based measures of verbal IQ (eg. Happé, 1995). It has frequently been argued {Baron-Cohen, 1997 #12a; Bloom, 2000 #13} that an impairment in ToM would affect verbal IQ more generally, as well as specifically having a more pronounced effect on the Comprehension subtest. One explanation for this relationship might be that children with higher verbal IQs use this ability to compensate for their ToM impairments (Tager-Flusberg, 2000), at least in the test situation. Alternatively, language learning may be restricted by a lack of understanding of the intentions behind a speaker's vocalisation (Bloom, 2000). Then again language may provide the environment in which to learn about mental states and therefore be a necessary precursor (Astington & Jenkins, 1999). The latter two suggestions seem less likely as unitary explanations given the presence of ToM impairments in individuals with ASD compared to controls matched for verbal IQ; however, it is possible that these different explanations may interact together over time.

It is interesting to note single cases when this relationship between verbal IQ and ToM task performance is not so obviously related. One child in the current sample stands out in this respect. CH was 11 years 8 months at the first test session and was capable only of understanding diverse desires, diverse beliefs and that seeing leads to knowing from the false belief battery, scoring 3/25, lower than every other child in the sample. Similarly, he had a scaled score of 1 on the Comprehension subtest and scored 0/16 on the mental state Strange Stories. Despite this profound impairment in ToM and the effect it had on his Comprehension subtest score, CH had a verbal IQ of 90, within the average range (performance IQ 86). This case illustrates the far from straightforward relationship between verbal ability and ToM understanding.

The relationships between Block Design and the EFT, and Comprehension and the ToM tasks were found in the control group as well as the ASD group, although to a lesser extent. It has been suggested that CC may exist as a continuum in the general population from weak to strong, with children with ASD clustered at the weak end (Happé, 1999). The results from the current study support this idea of a continuum for CC, with a larger proportion of individuals clustering at the weak extreme in the ASD than the control group. Whether the similar results in the ToM domain are also indicative of a separate continuum or whether this is an artefact of the nature of the tasks is a matter for future research.

More striking than these results, however, was the stark difference in the relationship of EF performance to the IQ subtests between the two groups. While the directionality of the cause and effect in this relationship is unclear, the difference between the two groups was striking. The control children showed the expected relationship between EF and performance IQ, particularly for the Object Assembly subtest which involves novel problem solving and a high degree of flexible thinking to generate and try out new ideas, abilities that are commonly associated with EF. In contrast, the children in the ASD group

showed no relationship between these tasks but instead between EF and all of the verbal IQ tasks.

Why might EF be related to verbal IQ in ASD? Although unexpected, this result is consistent with one study by Liss et al. (2001) showing correlations between verbal IQ and EF performance. The authors refer to a theory put forward by Russell, Jarrold & Hood {, 1999 #42a}, which suggests that the nature of the EF deficit in ASD may be in verbally encoding rules. Specifically, Russell et al. stated that children with ASD have problems verbally encoding arbitrary rules in tasks that do not have any verbal output. Verbal encoding allows the child to rehearse the rules as internal speech and therefore to self-monitor their own task performance; these rules must be arbitrary as logical rules would not need to be rehearsed; the task must not have a verbal response otherwise interference between the internal and vocalised speech will occur and the internal speech will be lost. Presumably Liss et al. were extending this idea to state that having a lower verbal IQ would make a child less proficient at using such internal speech. In this sense, lower verbal IQ could be thought to be causing the poor performance seen by some children on executive tasks. Zelazo, Jacques, Burack & Frye (2002) make an alternative but similar hypothesis that executive tasks are best solved by complex verbal hierarchical (if-if-then) rules and therefore that low verbal IQ may prevent use of such rules.

One problem with either of these hypotheses is that there seems little reason to expect a lack of relationship between the performance IQ tests and the BADS test in the ASD group, given their similarities in terms of novel problem-solving. It is likely however that the performance IQ subtests put less demand on this verbal self-monitoring system as the tasks may involve fewer arbitrary rules. Even if this is the case, Joseph, McGrath & Tager-Flusberg (2005) have recently suggested that Russell et al.'s {, 1999 #42a} theory would predict poor use of verbal self-monitoring in individuals with ASD despite intact verbal skills and therefore a lack of relationship between the executive deficits seen and verbal

ability, the opposite of what was found in the current study. Indeed, when considering the verbal skills involved in verbal IQ tests, these seem a long way from the skills that would be required for verbal self-monitoring; the skills involved in self-monitoring are likely to relate most strongly to verbal working memory and this ability, reflected most strongly in the digit span subtest of the WISC, was only weakly correlated with EF.

Rather than verbal ability limiting executive skill, is it possible that the executive component of the verbal IQ tasks is relatively high and therefore that executive ability is limiting verbal ability? The similarities subtest certainly requires a high degree of flexible thought but it is less clear how the other tests might relate to executive abilities. A further possibility is therefore that executive dysfunction causes poor language learning; in order to understand and learn the meaning of words, it may be necessary to have a fairly flexible understanding of their usage in order to understand them in different situations and use them appropriately. However, it really seems that there is a completely different relationship between executive skills and IQ in the control and ASD groups, which indicates that the children with ASD are performing the BADS in a totally different manner to the controls.

On further observation of the ASD IQ profile, one thing becomes evident: children with ASD may, if anything, be performing relatively well on some verbal tasks, particularly information and similarities. An alternative idea therefore is that children with exceptional verbal skills but with executive dysfunction might use their verbal skills to help them in EF tasks, possibly through internal verbal self-prompting or other verbal strategies; indeed, this suggestion fits with the pattern of results found here. In contrast, normally developing children would not need to use such strategies as they could use their executive skills to succeed on such tasks. Verbal skills can thus be seen not as causal to the executive problems in ASD but as a compensatory mechanism for overcoming the executive

problems. This may therefore explain why different relationships are seen between EF and IQ in the two groups.

To bring another component into the discussion, the similarity of the relationship between ToM with verbal IQ and EF with verbal IQ should be noted. It is therefore possible that a more complex relationship exists between EF and verbal IQ, as ToM and EF may also be related. This possibility will be explored further by examining the relationships between the three cognitive domains in section 6.2 and chapter 7.

Regardless of the exact causal pathway linking each of the cognitive impairments to IQ subtests, the fact that they are linked is of importance when considering group matching.

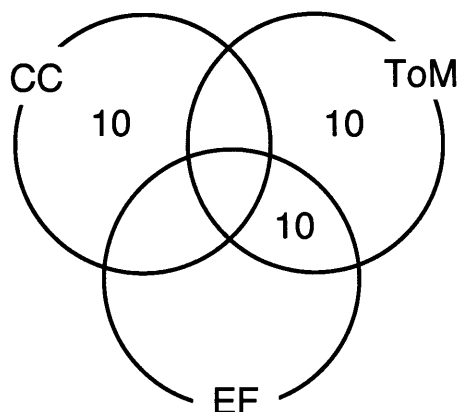
Particularly for ToM and EF, their correlations with verbal IQ indicate that matching groups for verbal IQ may actually reduce group differences on ToM and EF tests, and possibly mask ASD specific variation. On the other hand, when group differences are found under such strict matching conditions, they can be considered strong effects.

Similarly, the link between weak CC and Block Design performance may result in an overestimate of ability when such IQ tests are used in isolation. This is particularly relevant therefore for short forms of the WISC and WAIS that estimate performance IQ on the basis of two subtests, one being Block Design. Block Design may have less effect however in a larger battery of tests, such as the full WISC performance IQ scale.

6.2 Relationships between cognitive domains

One proposed approach to tackling the problem of heterogeneity in the search for the underlying biological and genetic basis of autism has been to look for subgroups of individuals showing traits associated with autism (Bailey, Phillips & Rutter, 1996). These are known as endophenotypes and tend to focus on the defining behavioural symptoms. As yet, however, cognitive endophenotypes have not been used in the search for the genetic basis of ASD, mainly due to the lack of consensus about the universality of these

Figure 6.8 Venn diagram showing hypothetical results.



impairments and their relationships to each other. One of the major aims of this thesis was therefore to assess the relationships between the three different cognitive explanations of ASD examined here through tests specifically designed to tap into these domains. Of particular interest is the relationship between ToM and EF, given that both domains seem to correlate with verbal IQ whilst CC does not. For example, in the hypothetical situation given in Figure 6.8, it is proposed that all cases of ASD with ToM impairment also have executive dysfunction, whilst the converse is not the case. This figure illustrates the possibility that EF is primary to ToM, whilst CC is separate and unrelated to these two domains (see sections 1.2.4 and 1.3.4 for a theoretical discussion of the relationships between these domains and of evidence supporting different claims). Is the pattern present in the current sample similar to this idealised case or different?

6.2.1 Method

In order to investigate the relationship between ToM, EF and CC, composite measures were produced for each of these domains, calculated by averaging z-scores (calculated in relation to control performance) for each participant across all tasks in that domain, giving equal weighting to each task. In the ToM domain, the tasks entered were the ToM battery and the mental state Strange Stories; in the CC domain, EFT reaction time (method 3) and a measure combining the two switching conditions in the attentional switching task (the

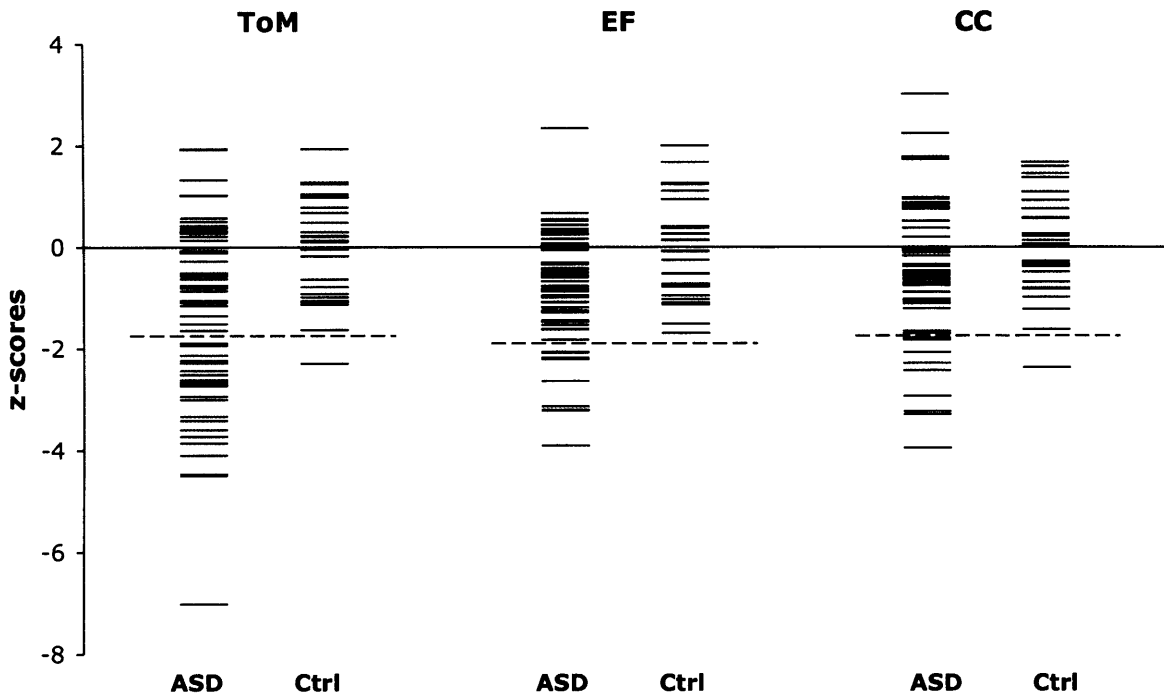
average of LL-GL and of GG-LG inverted) were used; and in the EF domain, all 7 tests were used (inverted for the Cards test and SSSCT as these were error scores). Each task entered into the composite measure had already had age, verbal and performance IQ taken into account by entering the task into a regression analysis as the dependent variable with age and IQ as the independent variables and collecting the residuals. Positive scores therefore always represent good performance and negative scores indicate poor performance.

Correlations were examined between the different domains, as well as examining the pattern of impairments across the different individuals. Of particular interest here were individual differences in performance and individuals with abnormally low performance were therefore identified. Deviant performance was defined as below the 5th percentile of control performance. To detect individuals with deviant performance on each task or composite, any control outliers performing more than 1.65 standard deviations (SDs) below the control mean were removed in order to obtain a better estimate of normal performance, regardless of controls who might have performed abnormally on any one task. The control mean and SD were then recalculated and outliers were defined as those lying more than 1.65 new SDs below this new control mean (procedure as in White et al., 2006).

6.2.2 Results

Individual performance across the three cognitive domains can be seen in Figure 6.9. The groups differed significantly only on the ToM ($t(80.3)=4.35, p<.001$) and EF composites ($t(82)=3.30, p=.001$). The overall distributions are quite striking, however. In the ToM domain, scores can be seen to span the whole range of the control performance but with an elongated tail of individuals performing particularly badly outside of the control range; indeed, 46% of children in the ASD group fell below the 5th percentile cut-off.

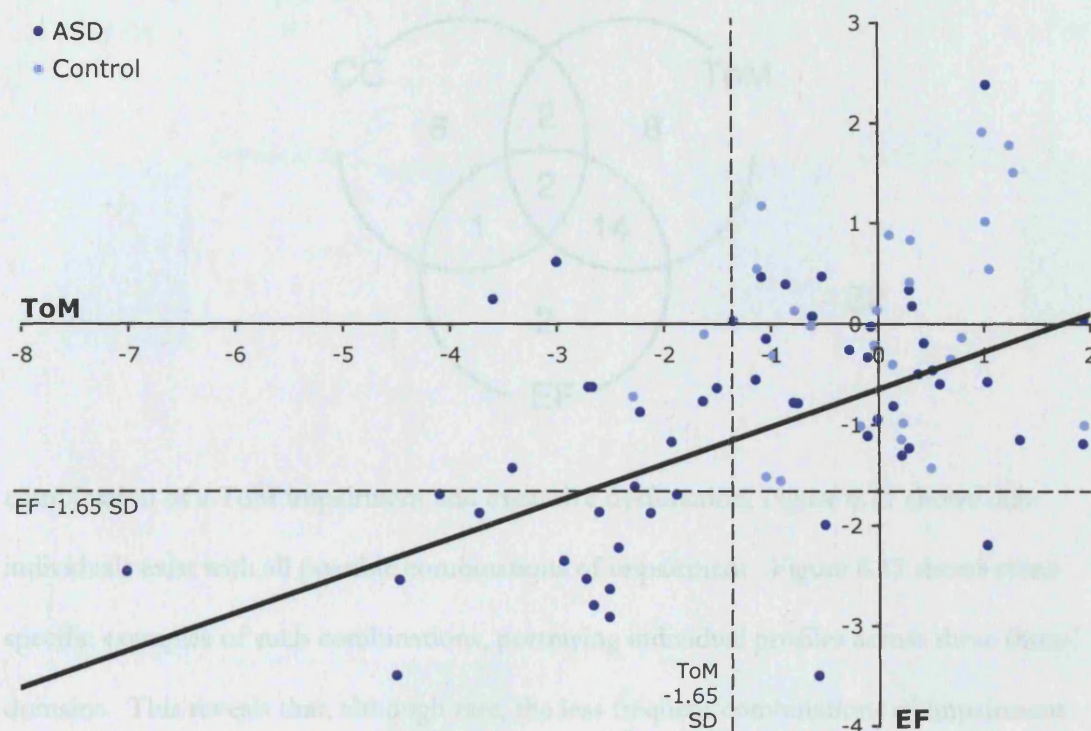
Figure 6.9 Individual performance on the composites for each of the three cognitive domains; the x-axis represents the control means and the dotted line represents the 5th percentile cut-off; impairment and therefore *poor* task performance is generally reflected by low z-scores, except for the CC domain in which weak CC and therefore *good* performance on the EFT would be represented by low z-scores.



Performance was similar in the EF domain with few children performing above the control mean, although slightly fewer children (33%) fell below the 5th percentile cut-off; instead the majority of the remaining children (47%) fell between the control mean and this cut-off. In the CC domain, only 20% of the children in the ASD group showed a profile indicative of weak central coherence, falling below the 5th percentile cut-off; however, the opposite extreme of performance was more noticeable than in the other cognitive domains, with four children performing above the control range. As noted in section 5.1.3, poor performance on the EFT is reflected by high CC z-scores here and so poor performance by some children on this task may have resulted from difficulty with non-specific task factors; this may explain the extreme outliers with positive z-scores.

Correlations between the three domains were examined for the whole sample and found to hold only between the ToM and EF domains ($r = .56, p < .001$), but once the groups were examined separately, this relationship was only found within the ASD group (ASD: $r = .52,$

Figure 6.10 The relationship between Theory of Mind and Executive Function performance in both groups. The regression line is for the ASD group only, whilst the broken lines represent the -1.65 SD cut-off for deviant performance based on the control distributions.

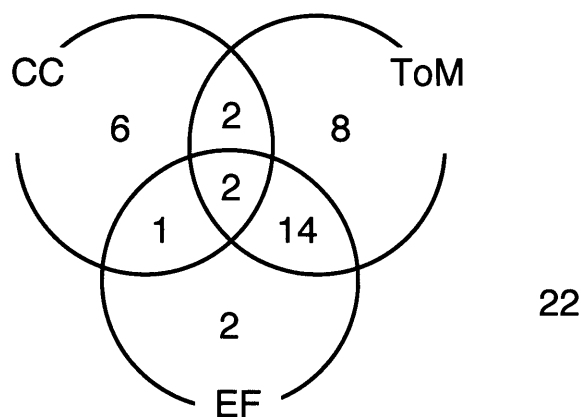


$p < .001$; control: $r = .31$). However, as can be seen from Figure 6.10, there seemed to be a trend towards individuals with ASD and low ToM to be less affected in the EF than ToM domain. Indeed, there were a number of children who had performance below the deviance cut-off on the ToM composite whilst performing above the cut-off on the EF composite. No other correlations were significant. Given that the different executive tests were found to be poorly related to each other (see section 4.3), this correlation between ToM and EF in the ASD group was further investigated by looking at relationships between each of the tests in the two domains. Both the ToM battery and the mental state Strange Stories were found to be related to the SSSCT ($r > .41$, $p \leq .001$), whilst only the ToM battery was related to the Cards test ($r = .43$, $p = .001$). No further correlations were significant.

Patterns of impairment in each individual were studied and can be seen in Figure 6.11.

While the majority of children either had a ToM impairment only, weak CC only or a

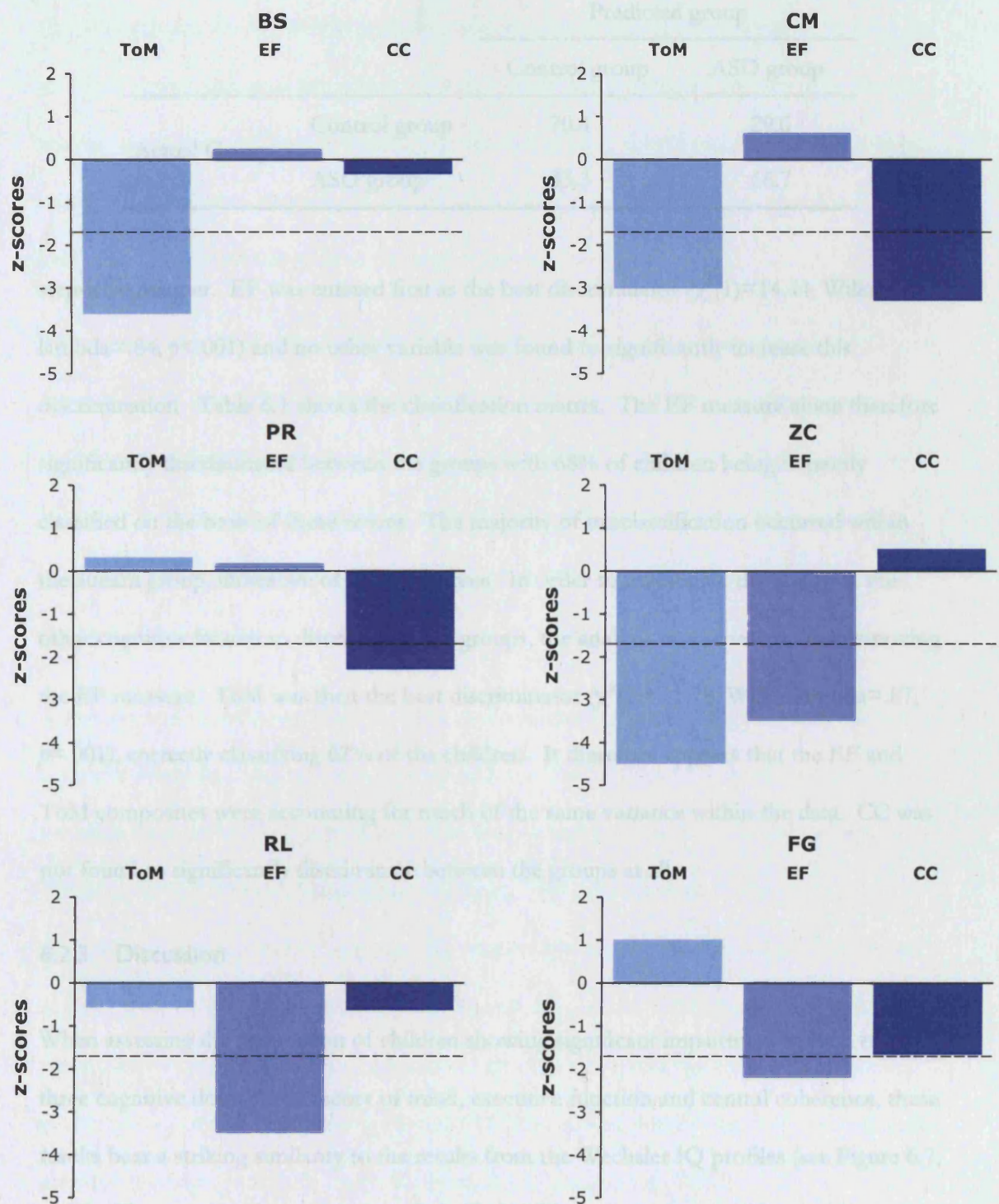
Figure 6.11 Venn diagram to show the number of children with ASD who displayed significant ToM, CC or EF impairments; the 22 children outside the diagram represent those in which none of these impairments were detectable.



combination of a ToM impairment and executive dysfunction, Figure 6.11 shows that individuals exist with all possible combinations of impairment. Figure 6.12 shows some specific examples of such combinations, portraying individual profiles across these three domains. This reveals that, although rare, the less frequent combinations of impairment are not an artefact of the cut-off methodology used; children exist with significant impairment in each possible combination of domains whilst having retained performance in the remaining domains. Figure 6.11 further reveals a high proportion of individuals with ASD who appear to have no significant cognitive impairments on the tests used here; 39% of children fell into this category.

Those children who did not display any cognitive impairment were compared to the remaining children with ASD on age and IQ measures. Whilst no significant differences were found (age: $t(55)=1.06$; verbal IQ: $t(55)=1.23$; performance IQ: $t(55)=1.73$, $p=.090$), a trend was found for the group without cognitive impairment to have lower performance IQs; in fact both verbal and performance IQ scores were 6 points below the remaining children with ASD (verbal IQ: 100.5 and 106.9; performance IQ: 90.8 and 97.1). In addition, only one of these 22 children had positive z-scores across all three cognitive domains whilst six of the 27 control children did ($\chi^2(1)=3.09$, $p=.079$).

Figure 6.12 Profiles for six children with ASD showing the presence of all possible combinations of impairment and double dissociations between these three cognitive domains.



Finally, Wilks' step-wise discriminant function analysis was used to investigate which cognitive factors (from ToM, CC or EF) were best able to discriminate the groups and predict group membership (ASD or control). Variables were entered and removed in a

Table 6.1 Classification matrix for the discriminant function analysis, showing the percentage of correct classifications according to EF composite scores.

		Predicted group	
		Control group	ASD group
Actual Group	Control group	70.4	29.6
	ASD group	33.3	66.7

step-wise manner. EF was entered first as the best discriminator ($\chi^2(1)=14.44$, Wilks' lambda=.84, $p<.001$) and no other variable was found to significantly increase this discrimination. Table 6.1 shows the classification matrix. The EF measure alone therefore significantly discriminated between the groups with 68% of children being correctly classified on the basis of these scores. The majority of misclassification occurred within the autism group, indicative of false negatives. In order to investigate the ability of the other cognitive factors to discriminate the groups, the analysis was repeated after removing the EF measure. ToM was then the best discriminator ($\chi^2(1)=11.78$, Wilks' lambda=.87, $p=.001$), correctly classifying 62% of the children. It therefore appears that the EF and ToM composites were accounting for much of the same variance within the data. CC was not found to significantly discriminate between the groups at all.

6.2.3 Discussion

When assessing the proportion of children showing significant impairment in each of the three cognitive domains of theory of mind, executive function and central coherence, these results bear a striking similarity to the results from the Wechsler IQ profiles (see Figure 6.7, p162). They suggest that approximately half of children showed mentalising impairment, a third of children showed executive dysfunction and a fifth showed a detail focussed processing style; furthermore, these impairments can occur in any combination. The similarity between Figures 6.7 and 6.11 (p162 & 173) are remarkable given the differences in tests used and the more strict cut-off here of 1.65 SD (as opposed to 1 SD). This

supports the idea that the Block Design and Comprehension subtests as well as the presence of low verbal IQ map closely onto these three cognitive domains. Furthermore, the large proportion of children with mentalising impairment indicates that this may be the most consistent and sensitive deficit present in ASD; this may be an artefact of the lower internal consistency of the EF and CC domains, however. By no means all children have mentalising problems though; the individual results clearly show that a proportion of individuals have superior skills in all three domains.

These proportions are also similar to those from the only previous investigation of all three domains (Pellicano, Maybery, Durkin & Maley, 2006), although the proportions were slightly higher there due to a more lenient cut-off of 1 SD (ToM 68%; EF 42%, CC 74%). The most notable difference was in the CC domain, with the present study producing many fewer deviant performers; this may in part be due to the tests used but also the method of data analysis.

A primary aim of the present analysis was to assess the relationships between the three cognitive domains. The correlations seen between the ToM and EF domains support previous findings in ASD populations that these domains are related (Joseph & Tager-Flusberg, 2004; Ozonoff, Pennington & Rogers, 1991; Zelazo et al., 2002). The discriminant analysis further supports this relationship, indicating that EF and ToM have similar power in terms of discriminating between the groups. Furthermore, the specific correlations with individual EF tests indicate that the tests most closely related to ToM performance were those involving prepotent inhibition and set-shifting, which also parallel previous findings (Joseph & Tager-Flusberg, 2004; Zelazo et al., 2002).

Correlations provide little information about the direction of the causal relationship between these domains, however; one may be primary to the other or they may both be related through a third factor. Perner & Lang (1999) argue that one way of informing this issue is to look at dissociations between domains. How then do the results here compare

to the theoretical position shown in Figure 6.8 (p169)? Whilst some authors would argue that the greater number of outliers in the ToM domain would indicate primacy of ToM over EF {Ozonoff, 1991 #39a}, others would argue that the pattern of impairments seen here, with most cases of executive dysfunction also having impaired ToM, would indicate that EF is primary to ToM. The reasoning behind this latter stance suggests that, if executive dysfunction is primary, it will always result in impaired ToM, whilst ToM impairment may result from alternative causes in other cases (Perner & Lang, 1999). However, despite their scarcity there are three children with ASD in the current study who showed the opposite pattern of intact ToM despite executive dysfunction. Indeed, the individual profiles shown in Figure 6.12 (p174) reveal double dissociations between all three cognitive domains. While this is difficult to reconcile with the correlation seen in Figure 6.10 (p172), this hints that the relationship between ToM and EF is unlikely to be causal. Instead it seems more likely that a third factor, such as verbal ability, may be associated with them both, or that one is involved in the process of compensation of the other.

Indeed, it is possible that the relationships between verbal IQ and both ToM and EF task performance seen in section 6.1 could partially explain the relationship between ToM and EF in terms of compensation. Verbal ability may act as one compensatory method of solving otherwise difficult ToM and EF tasks for children with ASD; it is unlikely to be the sole compensational method however, given that age and IQ were factored out of the correlational analyses. The trend seen in Figure 6.10 for children with ASD and low ToM task performance to be less affected in the EF than ToM domain may imply that this compensation is more successful for EF tasks. It is interesting that EF was the best discriminator between the groups, however, indicating that many of the ASD children not classed as showing deviant performance below the cut-off still had mildly impaired performance in comparison to the controls. This supports the idea that compensation may have a differential effect in these two cognitive domains, or possibly indicate a lower

sensitivity for the EF measure, particularly given that internal consistency between the different EF tests was low.

A further idea relating to findings in chapter 4 is that children with ToM impairments have problems with ecologically valid EF tasks, such as the ones used here, which require an understanding of implicit task demands, rather than with EF itself. This would explain both why ToM impairments appear to be more severe than EF impairments and why the relationship between these domains as shown in Figure 6.10 appears to be strongest in the children with severe impairments below the 5th percentile cut-off and non-significant in controls.

Remarkably, using the cut-off criteria defined in section 6.2.1, 39% of the children diagnosed with ASD tested here showed no signs of impairment in any of the three cognitive domains and 33% were classified as controls in the discriminant function analysis. An important phenomenon to mention here is regression towards the mean; any factor that is used as the basis on which to select groups is likely to be a better discriminator than any further factor tested; cognitive tests will therefore naturally produce some wrongly classified cases. Another factor to mention again is the relationship between verbal IQ and some of these variables; by accounting for verbal IQ levels in the composite scores, some of the ASD-specific variation may have been lost as the IQ tests are likely to reflect some aspects of these cognitive domains (see section 6.1). Furthermore, the slightly lower IQs of these children without detectable cognitive impairment may have led to their impairments being masked, particularly if these lower IQs resulted from an underestimation of true ability.

While these suggestions may go part way towards explaining these diagnosed children with intact cognitive performance, the large number of children involved here is still somewhat unexpected and puzzling. There are three possible reasons for this: the tests used here were not sensitive to the impairments tested; even in combination, these three cognitive

deficits are not sufficient to explain autistic behaviour and therefore these children had other causal impairments untested for here; or these children had been misdiagnosed. While these three possibilities are difficult to differentiate without additional investigation, a number of findings begin to inform these issues. The distribution of EF scores in the ASD group reveals that, whilst only a minority of individuals performed below the deviance cut-off, the majority were performing below the control mean. This may indicate that the tests were not sensitive enough to discriminate the groups, possibly due to floor and ceiling effects. Indeed, there were fewer children with ASD and intact performance than controls who showed performance above the control mean across all three domains. Moving to the issue of misdiagnosis, it is possible that these children either have other disorders and have received the wrong diagnostic label, or that the problems they displayed at diagnosis were transient and have since recovered. The possibility that these 'cognitively intact' cases may in fact be mild or misdiagnosed cases will be assessed in the following chapter when comparing behavioural with cognitive data.

Considering the possibility that these children with intact cognitive performance may have other undetected impairments, what might these be? Given that many of the children in this sample did not fulfil full criteria in all three behavioural domains for ASD, it is possible that some of them may have primary communication problems rather than those that are secondary to social difficulties. The idea that some of these children may have had pragmatic language impairments without social impairment will also be assessed in the next chapter. Other possible cognitive impairments that have been suggested in autism are, for example, attentional problems (Allen & Courchesne, 2001) and memory problems (Ben Shalom, 2003), but these have not been assessed in the current test battery.

6.3 Conclusion

An analysis of task performance in the 3 main cognitive domains implicated in ASD has revealed that all three are related to IQ subscale scores in different ways, whilst mentalising and executive skill also appear to be related to one another. Both mentalising and executive skill are related to verbal ability whilst central coherence appears to be independent of both verbal ability and these two cognitive domains, rather being related to a specific aspect of performance IQ revealed in the Block Design subtest of the WISC. This indicates that at least two distinct cognitive subtypes exist within the autism spectrum. Whilst firm conclusions about the nature of the relationship between ToM and EF cannot be made at this stage, observation of individual profiles across these domains indicates that neither impairment may be primary to the other and therefore that they may be related through a currently unspecified factor or that one may be involved in the compensation process for the other. A further unresolved observation is the high number of children with ASD who showed no cognitive impairment by the definition applied here. Both of these issues are further investigated in chapter 7.

Chapter 7: Relating cognition to behaviour

7.1 Heterogeneity at three levels

It is already clear from the previous chapter that there is considerable heterogeneity at the *cognitive* level in the group of children with ASD in the current study; some children were distinctly impaired while others were not and some of these latter children were performing well above average, indicating intact cognitive function. That *behavioural* heterogeneity also exists within the autism spectrum has long been acknowledged, even with the use of standardised diagnostic tests (Folstein, Bisson, Santangelo & Piven, 1998). Indeed, Chapter 2 shows different children in the current sample to have problems in different areas of the behavioural triad. Furthermore, it is possible for two children with the same degree of impairment in a particular area of the triad to show variability in their behavioural presentation of that aspect of the triad as they may display behaviour characteristic of different items composing that area.

In addition to cognitive and behavioural heterogeneity, there is also *genetic and biological* heterogeneity in ASD, which became obvious when the genetic basis could not be identified in the same way as for simple genetic disorders (Spence, 2001). There is therefore a need for genetically predictive phenotypic markers or endophenotypes on which to subgroup individuals, allowing more uniform populations within ASD to be studied when searching for genetic risk factors for ASD. Examples of dimensions of phenotypes previously suggested and used include IQ (Spiker, Lotspeich, Dimiceli, Myers & Risch, 2002), the behavioural symptoms (Ronald, Happé & Plomin, 2005; Shao et al., 2003; Silverman et al., 2002), language (Pickles et al., 2000; Shao et al., 2002) and savant skills (Nurmi et al., 2003). However, none of these have proved especially fruitful, and researchers continually search for new subgroups of children within the spectrum with similar characteristics to each other.

Probably the first attempt to locate behavioural subtypes in the autism spectrum was presented by Wing & Gould (1979). These authors noted different types of social impairment in different subgroups of individuals; some being described as aloof, others as passive and still others as odd. However, children were observed to shift between these behaviours over time and these subgroups were typical of different levels of functioning; thus, the subtypes were considered different behavioural manifestations of the same underlying problem, expressed in different ways by children at different ages and levels of abilities. This sort of grouping is therefore less useful at a biological level, whilst it may be of use at a more applied level when considering the child's needs.

Prior et al. (1998) also looked for subgroups in the spectrum on the basis of diagnostic behaviours using a cluster analysis technique. The subgroups they located differed in terms of the severity of behaviours, rather than showing qualitatively different patterns of impairment in the different clusters. The authors also found the most severely affected group to have lower verbal IQs and worse mentalising ability. However, as the clusters differed only in severity, this is likely to have created artificial groups that do not really exist from a continuous spectrum of impairment.

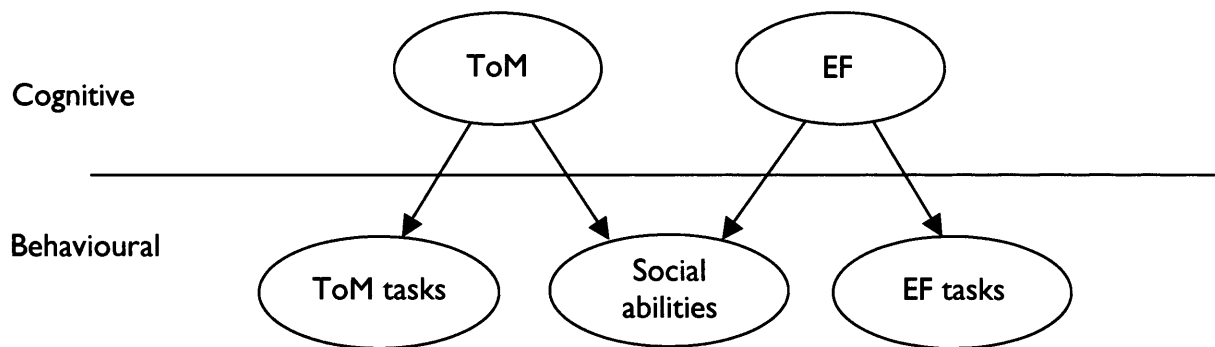
Given that the current behavioural triad has only limited use for ASD genetics, a number of studies have attempted to reclassify the behavioural symptoms empirically rather than theoretically, grouping behaviours in different ways from the traditional triad (Robertson, Tanguay, L'Ecuyer, Sims & Waltrip, 1999; Szatmari et al., 2002; Tadevosyan-Leyfer et al., 2003; Tanguay, Robertson & Derrick, 1998). One recent study found that a model involving three new symptom categories (make believe and play skills, social communication skills, and stereotyped language and behaviours) fitted the behavioural data from a large sample of children better than the DSM diagnostic triad (van Lang et al., 2006a). This study used the sub-items within each area of the DSM behavioural triad to produce these new categories, combining these sub-items in a different way. As the

current conceptualisation of the triad appears not to be optimal, this may explain why the different areas of the DSM triad are commonly found to overlap so heavily, particularly the social and communication areas (Volkmar et al., 1994). The novel behavioural groupings in the study by van Lang et al. were suggested as one possible marker to further genetic research. It will therefore be important in the present analyses to not be restricted to the triad of behaviours but to look at the sub-items that the triad is composed from.

If the cognitive domains thought to be causal in ASD are to be useful as potential endophenotypes on which to subgroup individuals for genetic studies, it is of interest how the cognitive heterogeneity relates to the behavioural heterogeneity; this is the main aim of this chapter. In addition, given that autism is currently defined behaviourally, it is important to be able to relate any factor thought to be causal in its aetiology back to the diagnostic symptoms. As reviewed in Chapter 1, previous findings seem to indicate that theory of mind (ToM) impairment is related to the social (Frith, Happé & Siddons, 1994) and communication (Capps, Kehres & Sigman, 1998) impairments but not to the presence of repetitive behaviours (Turner, 1996). Weak central coherence (CC) does not seem to be related to any of the triad of symptoms (Teunisse, Cools, van Spaendonck, Aerts & Berger, 2001), instead being related to other features not currently part of the diagnostic criteria (Mottron & Belleville, 1993). Poor executive function (EF) seems to be related to all symptom areas (Happé, Booth, Charlton & Hughes, 2006; Lopez, Lincoln, Ozonoff & Lai, 2005) although results are more mixed in this domain (Ozonoff et al., 2004). One aim of this chapter is therefore to systematically assess the relationship of each of these cognitive domains to everyday behavioural symptoms as assessed through parental report.

A major question that arose from the analysis of cognitive impairments is whether there are any behavioural differences between those children who appear to have no cognitive impairments in the domains studied here and those who have at least one. Are those children with more clearly identifiable cognitive deficits also more clearly identifiable in

Figure 7.1 Illustration of convergence from the cognitive to the behavioural level; here, ToM and EF both affect social interaction skills.



parental reports as showing autism-specific features? How does IQ interact between the cognitive and behavioural factors? Do we see convergence from any of the cognitive impairments onto the same behavioural symptoms, do the different cognitive impairments affect different aspects of the symptom triad, or might they affect different aspects within one of the behavioural groups of symptoms? The illustrations here depict some of the possible outcomes.

Figure 7.1 illustrates the idea that there could be more than one cause of, for example, the social problems in autism, modelled according to the notation of Morton & Frith (1995).

In this example, both ToM and EF are shown to independently affect social abilities, with a ToM impairment being the primary impairment for some children whilst the EF deficit is the primary deficit for others.

Figure 7.2 Illustration of independence of cognitive impairments in terms of their effect on behaviour; here, ToM and EF are shown to produce different aspects of the diagnostic triad.

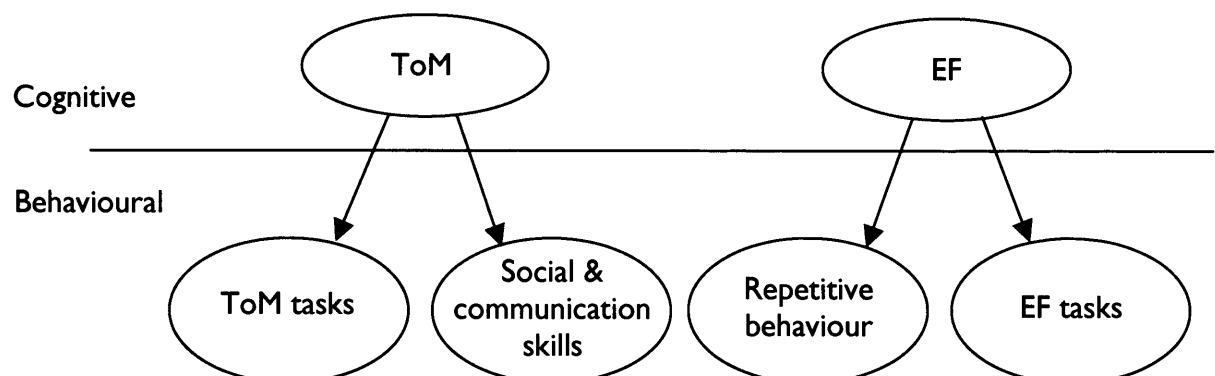
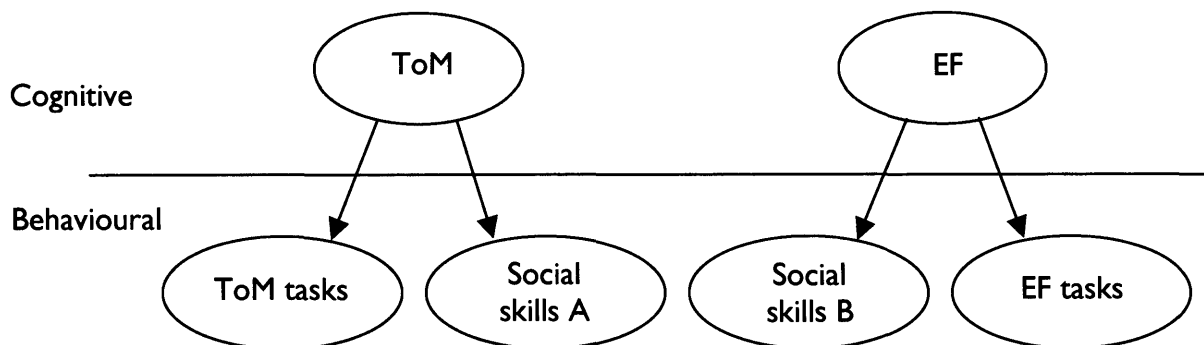


Figure 7.3 Illustration of different cognitive impairments affecting different aspects of the same symptom category; here ToM and EF both affect different aspects of social interaction.

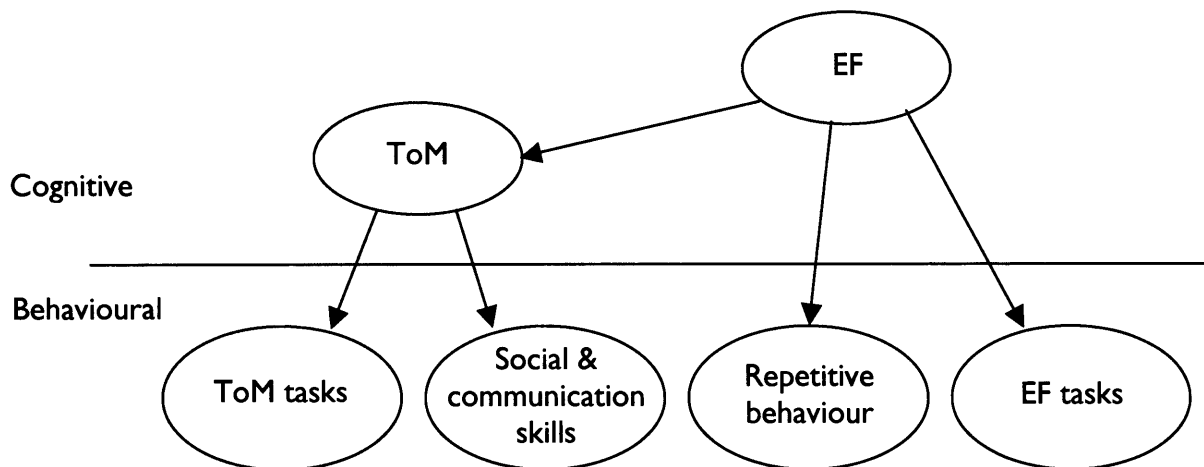


Another possibility is that different cognitive impairments could result in different aspects of symptomatology which together make up the typical pattern of behaviours seen in ASD. The cognitive impairments would therefore have independent effects on the behavioural level and different children might have different combinations of those impairments. This is modelled in Figure 7.2.

A further possibility that encompasses both of the previous ideas is that convergence of two cognitive impairments may occur onto one set of behaviours but that those two cognitive impairments have independent effects on different aspects of that set of behaviours. In the example below in which both ToM and EF impairments result in problems with social interaction, children with a ToM impairment could still be distinguished from children with an EF impairment on the basis of the particular social problems they were displaying. This idea is modelled in Figure 7.3.

Furthermore, the results from chapter 6 already illustrate that ToM and EF are related to each other. It is possible that one of these impairments relates to all aspects of the triad, some directly and some indirectly through the mediation of the other cognitive impairment. This idea is illustrated in Figure 7.4, with an EF impairment resulting in both the presence of repetitive behaviours and a ToM impairment, which in turn results in social and communication difficulties. This particular illustration would also imply that it would

Figure 7.4 Illustration of different cognitive impairments affecting each other as well as different aspects of the diagnostic triad; here EF affects ToM, and EF and ToM both affect behaviour in different ways.



be possible to have a ToM impairment alone (through some other cause) and that only social and communication difficulties would then be seen in this case.

The main aim of this chapter was therefore to assess how the cognitive domains related to each other in terms of their effect on behaviour.

7.2 Method

Behavioural measures were defined as those detecting everyday behaviours thought to be typical of individuals with ASD (3Di triad of behaviours, ToM, CC and EF novel questionnaires, and DEX-C), while cognitive measures were defined as tests specifically designed to tap into a particular cognitive domain (ToM, CC & EF composites plus IQ measures); the latter were therefore theoretically motivated while the former tended to have been derived from clinical observation of symptoms that commonly co-occurred across different individuals with ASD.

A break down of the components of the 3Di can be found in Table 7.1. Each area of the triad was composed of 4 categories, each of which was made up of a number of items.

Although the number of questions within each item varied (see number in parentheses in

Table 7.1 Structure of the 3Di in terms of areas of the triad, categories within each area and items within each category.

Area of triad	Category	Items (no. questions)
Social	Using non-verbal social cues	Eye contact (3) Smiling (4) Facial expression (9)
	Peer & sibling relationships	Imaginative play with others (8) Playing together (3) Others' perception (3) Social interest (8)
	Shared enjoyment	Sharing own enjoyment (1) Sharing material things (5) Sharing others' enjoyment (2)
	Emotional reciprocity	Picking up on and responding to others with comfort (4) Using others as tools (1) Responsiveness to others (2) Laughing inappropriately (2) Picking up on facial expression and tone of voice (5)
Communication	Use of conventional gestures	Pointing (3) Gestures (6) Positive head nodding (2) Negative head shaking (1)
	Conversational interchange	Purposeful conversation (4) Social conversation (3)
	Stereotyped, repetitive or idiosyncratic speech	Stereotyped, egocentric and repetitive speech (9) <i>Faux pas</i> (2) Pronominal reversal (3) Neologisms (1)
	Imaginative play	Imitating (2) Pretend play on own (5) Imitative social play (2)
Repetitive behaviours	Circumscribed interests	'Normal' intense interests (4) 'Odd' intense interests (2)
	Ritualistic behaviour	Word repetitions (1) Ordered routine (1)
	Mannerisms	Simple mannerisms (1) Complex mannerisms (1)
	Non-functional object use	Repetitive object use (3) Unusual sensory interests (2)

table), these were averaged so that each item was scored out of a total of two points. Each question therefore has equal weighting within each item and each item has equal weighting within each area of the triad. When analysing the relationships between the cognitive measures and behaviour on the 3Di, the three areas of the triad were explored. In order to guard against false positive results due to multiple comparisons, only the categories within those areas of the triad that produced significant effects were further explored and similarly, only those items within categories which produced significant effects were further explored.

Within the ASD group, group differences on measures of everyday behaviour between those with and without deviant performance on tests tapping into each cognitive domain were examined, as well as between those children showing no cognitive impairment and those with deviant performance in at least one domain (see section 6.2.1 for a definition of deviant performance). Correlations between the cognitive and behavioural measures were also calculated. Finally, a model was fitted to the data using a structural equation modelling programme (AMOS 6.0, © Arbuckle 1983-2005).

7.3 Results

7.3.1 Relationship between IQ & age and behavioural measures

Whilst IQ was correlated to some of the behavioural measures in the control group (vIQ & social: $r = -.46, p = .017$; vIQ & EF questionnaire: $r = -.61, p = .001$; pIQ & EF questionnaire: $r = -.47, p = .013$; vIQ & DEX-C: $r = -.51, p = .001$), with high IQs being associated with fewer deviant behaviours, this was only the case in the ASD group for the EF questionnaire (vIQ: $r = -.30, p = .030$) and a trend towards an association for the communication behaviours was also seen (vIQ: $r = -.27, p = .051$). This relationship was explored further in the ASD group by looking at the categories constituting the communication scale. A strong association was found between verbal IQ and the

‘conversational interchange’ category ($r = -.48, p < .001$). Both items making up this category also produced similar relationships (purposeful conversation: $r = -.35, p = .010$; social conversation: $r = -.46, p < .001$). These items cover the ability not to talk about topics that are well-known to the conversational partner, not to ask questions when the answer is already known, not to talk repetitively about one’s own interests, to take turns at making sounds as an infant prior to acquiring speech, to make ‘small talk’ rather than just talking in order to meet one’s own needs, to engage in conversation that is interesting and enjoyable for the conversational partner, and to respond to other people’s conversational cues. Age, on the other hand, was not related to any of the behavioural measures in the control group and only associated with the EF questionnaire in the ASD group ($r = -.27, p = .048$), with older children showing fewer deviant behaviours.

Given that associations between the behavioural and cognitive measures were to be studied further only in the ASD group, there was no need to enter age or IQ as covariates of the behavioural measures (the results remained the same when these were covariates). For the cognitive measures however, both the composites from chapter 6 (with age and IQ as covariates) and composites with only age entered as a covariate were used; this allowed the influence of each cognitive domain on behaviour, both independent of IQ and through the mediating effect of IQ, to be assessed. These will now be referred to as composite (age & IQ) or composite (age).

7.3.2 Relationship between ToM and behavioural measures

Individuals with ASD classified in chapter 6 as having significantly deviant performance on the ToM composite (age & IQ) were compared to the remaining children with ASD; the former children had more severe problems on the 3Di social scale ($t(51) = 2.02, p = .048$), whilst no differences were found in the communication ($t(51) = 1.19$) or repetitive behaviour ($t(51) = 1.33$) scales. Interestingly, despite being reported as less severe, the mean

Table 7.2 Means (and standard deviations) for performance across the behavioural measures in the ASD group, divided by performance on the ToM composite; high scores indicate deviant performance.

	ASD group	
	Deviant ToM (31)	Remaining children (26)
3Di social scale (30 max; 10 cut-off) *	14.5 (4.4)	12.1 (4.3)
- peer & sibling relationships (8 max) **	4.4 (1.4)	3.1 (1.6)
- imaginative play with others (2 max) **	1.4 (0.5)	0.9 (0.6)
- playing together (2 max) *	1.2 (0.6)	0.8 (0.7)
3Di communication scale (26 max; 8 cut-off)	15.4 (3.9)	14.3 (3.0)
3Di repetitive behaviour scale (12 max; 3 cut-off)	5.6 (2.5)	4.7 (2.5)
ToM questionnaire (%)	58 (20)	56 (18)
CC questionnaire (%) *	73 (18)	63 (18)
EF questionnaire (%)	42 (21)	46 (18)
DEX-C (80 max)	9.75 (6.14)	6.15 (2.92)

* $p < .05$, ** $p < .01$

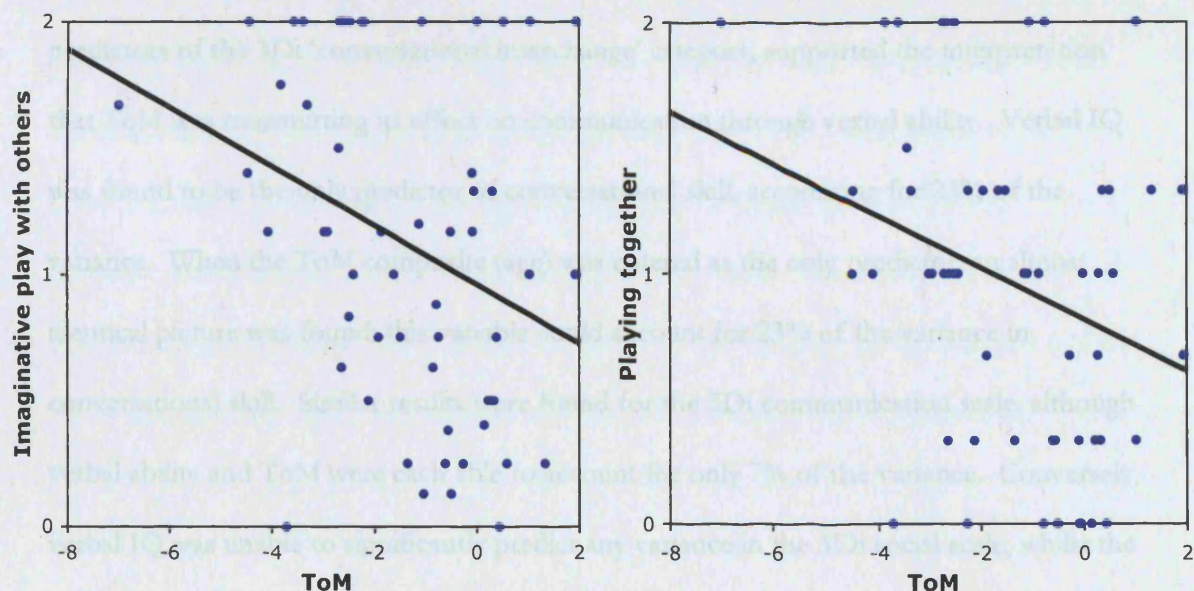
score on the social scale of the children with non-deviant ToM task performance was still above the recommended 3Di cut-off of 10 (see Table 7.2). In order to explore further the difference between the groups on the social scale, the four categories making up the social measure were examined. This revealed that the difference stemmed from the category regarding peer and sibling relationships ($t(51)=3.00, p=.004$). Again, the four items within this relationships category were examined and two produced significant group differences: imaginative play with others ($t(53)=3.37, p=.001$) and playing together ($t(53)=2.67, p=.010$). The first of these measures consisted of questions concerning whether the child ever played imaginative games with siblings and other children and, if so, what their level of understanding of the game was, whether they contributed ideas to the game and whether the game tended to follow the same story line every time. The second measure was concerned with the child's ability to play with, as opposed to alongside, another child and

whether they tended to wander off and leave the other child alone. This indicates that the ToM measures were able to capture the severity of certain social impairments.

The novel questionnaire tapping into behaviours thought to stem from ToM, EF and CC produced surprising results. Children with deviant ToM task performance were rated as showing more behaviours thought to be characteristic of weak CC than the remaining children with ASD ($t(51)=2.02, p=.048$). No group differences were found for the ToM ($t(51)=.34$) or EF ($t(51)=.77$) questionnaires however, or on the DEX-C ($t(51)=.83$).

The pattern of correlations across the whole of the ASD group supported the group comparison results, with a trend towards a relationship between the 3Di social behaviours and ToM composite (age & IQ) ($r=-.25, p=.068$) and a significant relationship between the CC questionnaire and ToM composite (age & IQ) ($r=-.31, p=.026$), whilst no other relationships were significant. The correlation between the 3Di social scale and ToM task performance was further examined and a relationship was found for the 'peer and sibling relationships' category ($r=-.34, p=.013$) and within this for both the 'imaginative play with others' ($r=-.36, p=.007$) and 'playing together' ($r=-.32, p=.019$) items as before. These two relationships are shown in Figure 7.5.

Figure 7.5 The relationship of ToM test performance to two of the items in the 3DI social scale in the ASD group: 'imaginative play with others' and 'playing together'; high scores on the 3DI items indicate impairment.



Correlations were also examined between the ToM composite (age) and the behavioural measures. All three areas of the 3Di triad of behaviours were found to be significantly related to ToM task performance (social: $r = -.33, p = .017$; communication: $r = .27, p = .049$; repetitive behaviour $r = .27, p = .049$), with poor ToM performance indicative of greater severity of abnormal behaviour, as well as a trend again towards a relationship between the CC questionnaire ($r = -.26, p = .057$) with ToM task performance. These previously unseen relationships between the ToM composite (age) and the 3Di communication and repetitive behaviour scales indicated that IQ, most likely verbal IQ for the communication area, was having a mediating effect between ToM task performance and these behaviours.

Specifically, the effect that ToM ability had on communicative skill could be explained through the effect of ToM on verbal IQ. As with the relationship between verbal IQ and the 3Di communication scale, the relationship between ToM (age) and this scale was only present within the 'conversational interchange' category ($r = -.48, p < .001$), as well as the 'purposeful conversation' ($r = -.36, p = .006$) and 'social conversation' ($r = -.43, p = .001$) items. The relationship between the ToM composite (age) and the 3Di repetitive behaviour scale was present in both the 'ritualistic behaviour' ($r = -.34, p = .014$) and 'non-functional object use' ($r = -.30, p = .025$) categories and within these in the 'word repetitions' ($r = -.39, p = .004$) and 'repetitive object use' ($r = -.30, p = .025$) items respectively.

A step-wise linear regression analysis, entering the ToM composite (age) and verbal IQ as predictors of the 3Di 'conversational interchange' category, supported the interpretation that ToM was transmitting its effect on communication through verbal ability. Verbal IQ was found to be the only predictor of conversational skill, accounting for 23% of the variance. When the ToM composite (age) was entered as the only predictor, an almost identical picture was found: this variable could account for 23% of the variance in conversational skill. Similar results were found for the 3Di communication scale, although verbal ability and ToM were each able to account for only 7% of the variance. Conversely, verbal IQ was unable to significantly predict any variance in the 3Di social scale, whilst the

ToM composite (age) predicted 11% of the variance. For the 'peer and sibling relationships' category, only the ToM composite (age) was found to predict 19% of the variance; when verbal IQ was entered alone, it could predict only 7% of the variance.

It was less clear why repetitive behaviour should be related to the ToM composite only when IQ was not accounted for, however; a regression analysis revealed that the ToM composite (age) was the only significant predictor of repetitive behaviour, accounting for 7% of the variance (11% of variance for ritualistic behaviour and 9% for non-functional object use), whilst verbal or performance IQ were unable to significantly predict any variance in these measures. These inconsistent results may be the product of the correlation being close to the significance threshold, as it would not remain after correction for multiple comparisons.

7.3.3 Relationship between EF & CC and behavioural measures

Children classified as having weak CC through deviant test performance were found to have no behavioural differences from the remaining children with ASD on the 3Di diagnostic triad ($t(51) < 1.58$); this was expected given that CC does not attempt to explain behaviours included in the triad. More surprisingly, similar results were found in the EF domain, with no group differences on the 3Di diagnostic triad between those identified with or without deviant performance on the EF composite ($t(51) < 1.2$). Similarly, no differences were found between the groups on the ToM, CC or EF novel questionnaires either in the CC ($t(51) < .77$) or EF cognitive domains ($t(51) < .15$), except for a trend towards a difference between those with and without EF impairment on the CC questionnaire ($t(51) = 1.95, p = .057$) and group differences were also absent on the DEX-C in either cognitive domain ($t(51) < 1.65$). Furthermore, no correlations between the CC and EF composites (age & IQ, or age only) and the behavioural measures were found.

7.3.4 Comparison of children with and without cognitive impairment

Those children classified as displaying no cognitive impairment in any domain were compared to the remaining children with ASD. Those children with no detectable impairment in any of the three cognitive domains were reported to display fewer behaviours characteristic of weak CC (CC questionnaire) than the children classified as showing one or more cognitive impairment ($t(51)=2.55, p=.014$). Otherwise, none of the behavioural measures distinguished these groups. The proportion of children with or without any detectable cognitive impairment were compared across the different 3Di diagnostic groups (autism, Asperger Syndrome, PDD-NOS). Whilst no significant differences were found ($\chi^2(1)=2.37, p=.124$), there was a slight trend towards children with autism and Asperger Syndrome (impairment in all three areas of the triad) to be more likely to show a cognitive impairment than those with PDD-NOS (69% vs 48%). Similarly, there was no difference in the proportion of children with or without a secondary diagnosis on the 3DI (ADD, ADHD, ODD, CD) to display a cognitive impairment (56% vs 63%; $\chi^2(1)=.27$).

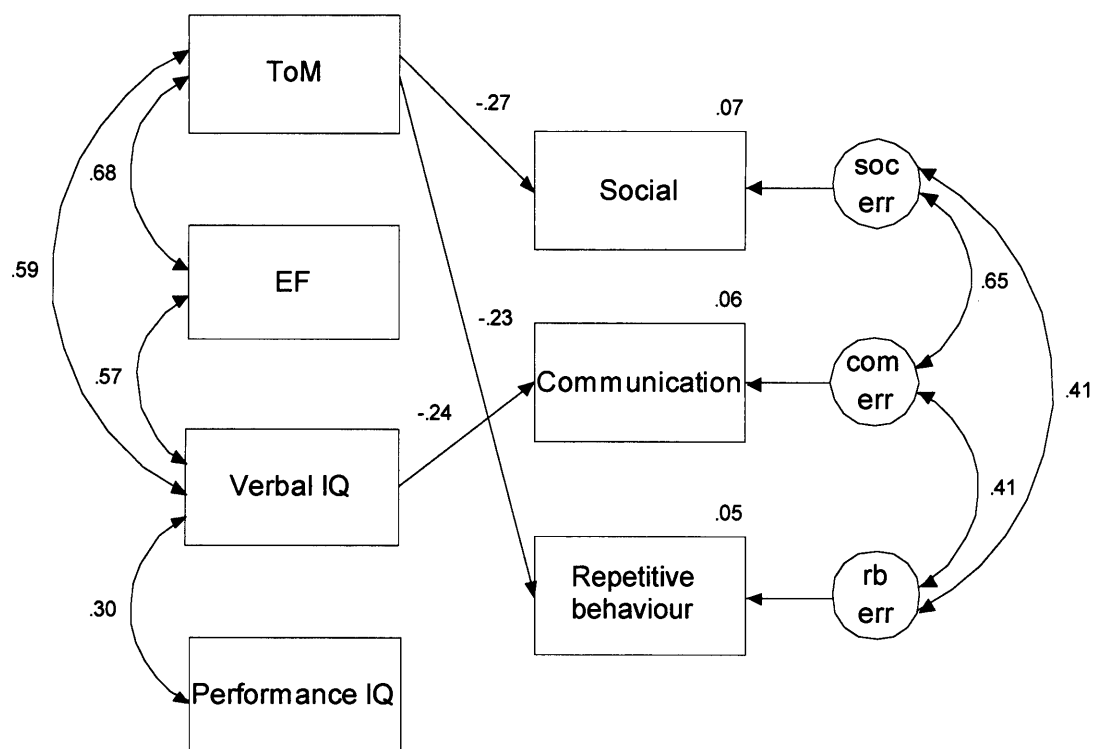
7.3.5 Modelling the relationship between cognitive measures and the behavioural triad

In order to assess the relationships between and within the cognitive and behavioural levels simultaneously, structural equation modelling was employed. Maximum likelihood estimation procedures were used to analyse the variance/covariance matrix of the observed variables; latent variables were not used due to the relatively low number of participants involved here and so this was an observed variable model. As the relationship between IQ and the cognitive measures was also of interest, verbal and performance IQ were entered into the path analysis as predictors of the autistic triad of behaviour (3Di scales), along with the ToM, CC and EF composites (age). All possible correlations between the five

predictor factors were initially present in the model, as were all possible correlations between the three outcome behavioural measures and all possible paths between the predictors and outcome measures. Non-significant correlations and paths were then dropped to produce a much simplified model (one marginally significant path was not dropped from the model as this greatly reduced the final model's fit; ToM & repetitive behaviour: $p=.071$).

The final path model is shown in Figure 7.6. CC was dropped from the model as it was found to be unrelated to any other variable and was therefore not contributing to the model. Only ToM was a predictor of both social skill and the presence of repetitive behaviours, whilst verbal IQ was a predictor of communication ability. EF did not predict any of the outcome measures however. While the model was a good fit to the data ($\chi^2(11)=10.72, p=.467, RMSEA=.00, CI=.00-.138, CFI=1.00$), only a small proportion of

Figure 7.6 Path model representing relationship between cognitive and behavioural measures resulting from structural equation modelling; error terms (err) for each of the predicted variables are necessary to account for noise in the predictive paths; the numbers on arrows denote path weights (r) and the numbers on boxes denote the proportion of unique variance accounted for (r^2).



the variance for each of the observed behavioural variables was explained by the cognitive measures ($r^2=.053$ to $.070$).

The path analysis was repeated substituting the 'peer and sibling relationships' category for the social scale from the 3Di, the 'conversational interchange' category for the communication scale from the 3Di and the 'ritualistic behaviour' category for the repetitive behaviour scale from the 3Di. Almost identical results were found ($\chi^2(13)=13.91, p=.381, RMSEA=.035, CI=.00-.14, CFI=.992$), although ritualistic behaviour was unrelated to the other two measures of everyday behaviour. The most noticeable difference was the greater proportion of variance in the behavioural measures that was explained by the cognitive measures, this time between 11% and 22%.

7.4 Discussion

In support of the cognitive results from the previous chapter, the structural equation model indicated that ToM, EF and verbal ability, as assessed by the present range of tests, are all related to each other and further, that they are all independently related to each other, each accounting for unique variation in the other two domains. Similarly in support of the findings in section 2.1.3, behavioural symptoms as assessed by the three scales on the 3Di were all intricately related, each accounting for unique variation in the others. While the relation between the cognitive and behavioural data was much weaker producing very few associations, a few notable relationships were revealed.

7.4.1 Relating cognition to behaviour

There were indications that ToM ability predicted parental reports of skill in social interaction, specifically in the way the children related to other children in play situations, including those involving imaginative games. This fits with previous findings (Frith et al., 1994; Hughes, Soares-Boucaud, Hochmann & Frith, 1997b) and makes intuitive sense

since the ability to decouple representations is thought to be necessary in order to understand and therefore to participate normally in pretend play (Leslie, 1987); also, the inability to understand another's point of view is likely to be most obvious in unstructured social interactions with other children who may not compensate for the autistic child's difficulties in the way adults might. These situations are therefore likely to be the most markedly impaired in a child with a ToM impairment. Additionally, ToM ability also weakly predicted the presence of repetitive behaviours. While this association has not been found in previous studies (Turner, 1996), it may be that such behaviours are more noticeable in the absence of normal social and symbolic activities, acting as a replacement activity; this is similar to the original conceptualisation of the third member of the triad by Wing & Gould (1979). This would also explain why a specific aspect of repetitive behaviour associated with ToM ability (ritualistic behaviour) was not directly related in terms of severity to the aspects of social and communicative impairment also associated with ToM ability (relationships & conversation).

Parental reports of communication abilities appeared to be related both to ToM and verbal ability, consistent with previous findings (Capps et al., 1998; Hale & Tager-Flusberg, 2005). Both verbal IQ and ToM task performance explained almost identical variance in the communication scale, both affecting conversational abilities in particular. This category of behaviour particularly covered the ability to respond in socially appropriate ways in a conversational exchange, a skill likely to depend on an understanding of a conversational partner's mental processes. Conversational ability also logically depends on verbal ability, requiring a high degree of fast online verbal production, a large vocabulary and knowledge of appropriate verbal responses in order to flexibly respond to the conversational partner. Furthermore, the path analysis suggested that ToM affects conversational abilities indirectly, mediated through its effect on verbal IQ. As previously mentioned, one of a number of different theories has proposed this relationship to be causal in this direction,

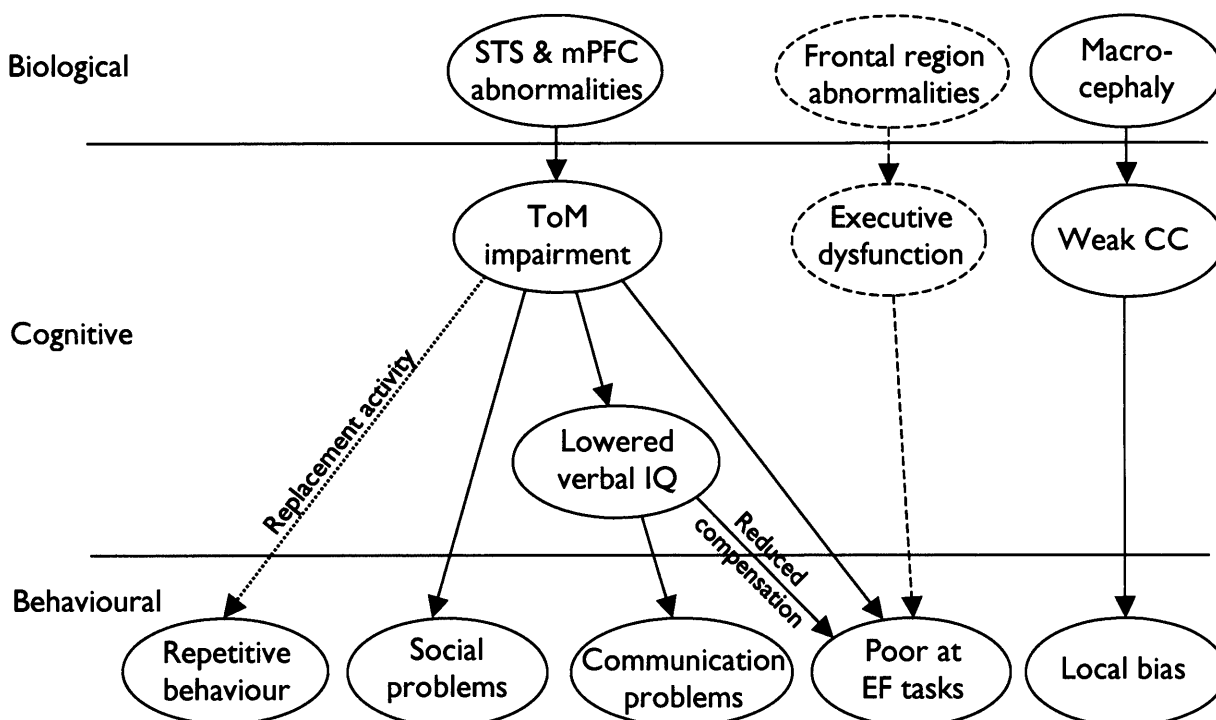
with ToM development as a necessary precursor for normal language development to occur (Baron-Cohen, Baldwin & Crowson, 1997a; Bloom, 2000).

Behaviours thought to be characteristic of weak CC do not play a role in current diagnostic schemes; here, CC did not appear to have any affect on the triad of autistic behaviours. In addition, the finding from section 6.2.2 that only a minority of individuals with ASD show an extreme tendency towards weak CC may indicate that it is not a causal factor in the currently defined autistic behavioural profile but an additional feature which accompanies ASD in some cases. The association of weak CC to ASD may result from genetic or biological factors that are far back in the causal chain and therefore difficult to identify at the cognitive or behavioural levels. Such factors will warrant careful study in the future.

Surprisingly, the EF composite was unrelated to any of the behavioural measures and therefore did not play a causal role in terms of explaining the behaviours in the structural equation model. However, both from chapter 6 and the path analysis, EF task performance was seen to be related to both verbal ability and ToM task performance in the children with ASD involved in this study. In this way, it appears that ToM and verbal ability are related to EF independently of their relationship with autistic symptomatology and therefore that EF cannot be causal to the relationship between these cognitive and behavioural variables. It seems most likely therefore that the relationships between ToM and verbal ability with EF are in this direction, with ToM and verbal ability both affecting EF task performance independently.

Figure 7.7 illustrates a hypothesis of a new causal model involving all of these cognitive and behavioural factors, as well as the biological measure of head size from section 5.3. Here, a ToM impairment is shown to result in social problems and repetitive behaviour, as well as lowering verbal ability (although this does not rule out the possibility that absolute verbal ability could still be high in a particular child). In addition, a ToM impairment results in communication problems through this effect on verbal ability and is also shown to lead to

Figure 7.7 Hypothesis for a new causal model of ASD; the dashed pathway indicates that executive dysfunction may not be the cause of poor EF task performance in ASD. STS=superior temporal sulcus; mPFC=medial prefrontal cortex.



poor performance on certain executive tasks. Verbal ability is characterised as having a compensatory effect on EF task performance, although this effect will be smaller for those children with more severe ToM impairments and generally lower verbal IQs. Executive dysfunction is illustrated as possibly not being a causal factor in poor EF task performance in children with ASD; it could be the cause of poor performance in other children, however. In addition, weak central coherence may result from an increase in neurones producing macrocephaly, and this is shown to result in a local processing bias; it would be expected that certain behaviours not currently included in an ASD diagnosis would also result in this case. This latter area of impairment is shown to be independent of the former one.

7.4.2 Children with no detectable cognitive impairment

An important aim of this chapter was to determine whether behavioural measures could differentiate those individuals with a prior diagnosis of ASD who were classified as not

showing impaired performance in any cognitive domain in the previous chapter. These children were reported as showing fewer behaviours indicative of weak CC, and there was a slight trend towards these children not being rated above the 3Di cut-off on at least one domain; these may both indicate slightly milder impairments. Nonetheless, these children's difficulties were severe enough to warrant a diagnosis on the autism spectrum so their problems must be taken seriously. Their lack of impairment may be related to the use of the 5th percentile cut-off for deviant performance imposing a categorical approach to impairment on these children rather than a continuous one. The correlational analyses between cognitive and behavioural measures seems to support this view, with degree of impairment at least in the ToM domain relating to severity of autistic symptoms as measured by the 3Di.

It may be that this group of children with no detectable cognitive impairments may not be a homogeneous group and so different explanations may be appropriate for different children. Whilst some may have milder impairments that were not detected by the 5th percentile cut-off, others may have been misdiagnosed (particularly given the high proportion of other non-ASD diagnoses present in this sample), others may have been compensating for and masking their difficulties, and still others may really have been unimpaired on these measures with impairments in other untested domains (see section 6.2.3).

7.4.3 Limitations of cognitive and behavioural measures

Does the general paucity of relationship between the cognitive and behavioural levels mean that the cognitive measures used here were irrelevant to the way in which ASD manifests itself? This seems unlikely given that significant group differences were present in both the cognitive and behavioural data. It seems likely that the poor correlations were due to noise in the data instead, making it difficult to pick up valid information about true ability.

A number of factors may have contributed to this within both the cognitive and behavioural levels.

Concerning the behavioural level, it is possible that the current diagnostic triad may not be the best way to categorise the behavioural symptoms present in ASD or that certain behaviours may not be causally related to ASD. This was supported by the finding that ToM was related only to certain categories within each of the 3Di scales and is consistent with previous findings that the behavioural symptoms of ASD are better classified in alternative ways (van Lang et al., 2006a).

Furthermore, it may be that parental rating scales and interviews are not the best way to collect accurate individual data on behaviour that is comparable across individuals. As each parent rates only one child, it may be extremely difficult to acquire good inter-rater reliability. It seems unlikely that each parent rated their child at a severity consistent with all the other children in the study; rather, parental ratings are likely to be dependent on what the parent was rating their child against and what they used as a comparison, and this is likely to depend on the other children that they see their child with. The parents, of course, were also not blind to the child's prior diagnosis; both this knowledge and the parent's opinion of the diagnosis may have affected ratings. Indeed, it has previously been found that parents of normally developing children tend to overestimate their abilities (Miller & Davis, 1992), and that parents of children with severe autism were likely to overestimate their child's abilities whilst parents of children with milder PDD-NOS did not (Hughes et al., 1997b), possibly due to the need to maintain a positive outlook on a difficult situation. Moreover, Hughes et al. also found that parents of children with ASD were unable to identify subtle social differences that teachers and therapists could, indicating that parents may be insensitive to detecting certain behaviours. Teacher and therapist ratings may be more objective measures but would also be systematically biased by prior knowledge of diagnoses.

Direct behavioural observation is an alternative that could be used in a future follow-up of the present cases. This eliminates the problem of poor inter-rater reliability but is carried out at a single time point so can only provide a snap shot of behaviour which may not be very representative. The essence of developmental disorders is perhaps grasped best by the dynamics of repeated testing over time; this may also be true for the cognitive measures. Indeed, it has previously been found that a standard parental report and behavioural observation measure have poor agreeability (Norbury & Bishop, 2002), indicating that both are limited in their representation of the child's behaviour.

In relation to the cognitive measures adopted in this study, any group differences may have been less pronounced and therefore not detected on some of the EF and CC tests as there were fewer deviant performers in these domains; this was less the case in the ToM domain however. Additionally, within the EF and CC composites, the tests were poorly correlated, indicating that they might have represented more than one concept. Conversely, the poor relationship between the cognitive and behavioural measures may actually reflect the true state of this relationship; it has previously been shown that intervention, compensation and training at either that cognitive and behavioural level do not transfer to improved performance outside of the specific area of training (Frith et al., 1994; Ozonoff & Miller, 1995; Swettenham, 1996). Particularly, cognitive tasks performed within a controlled environment may prove less difficult for highly intelligent individuals whilst, outside the laboratory, such problems may be more pronounced.

Similar results were reported in the one previous study to look at all three cognitive domains and relate them to behaviour; no significant relationships between the cognitive measures and behavioural parental ratings were found (Pellicano, Maybery, Durkin & Maley, 2006). One other study looking at ToM and EF, as well as verbal ability, and comparing these to behaviour as measured through behavioural observation (Joseph & Tager-Flusberg, 2004) did find some similar relationships, with verbal ability relating to

communication skills, and ToM task performance relating to all three areas of the triad (only to communication skill after accounting for verbal ability though). However, differing from the present findings, they also found verbal ability to relate to social skills and repetitive behaviour, and EF task performance to relate to communication skills (but not after accounting for verbal ability). These discrepant findings may in part be the result of using different measures to assess verbal ability, the cognitive domains and behaviour, as well as a consequence of recruiting a more variable sample in terms of age and ability.

7.5 Conclusion

The extensive battery of cognitive and behavioural measures applied to a large sample of children with ASD allowed a large number of detailed comparisons to be made in order to address the issue of causality from the cognitive to the behavioural level in ASD. This is a complex endeavour given the extensive heterogeneity in any ASD sample. Through a series of analyses culminating in structural equation modelling, a number of complex relationships were identified. It appears that ToM, EF and verbal IQ are all intricately related and further that ToM affects communication skills, social interaction and repetitive behaviours, the former indirectly through its effect on verbal ability. EF appears to have no effect on autistic symptomatology in this sample however, implying that it is not involved in the same causal pathway as ToM and verbal ability. A model was proposed in which poor performance on EF tasks may result from poor ToM abilities, whilst verbal ability may be a compensatory factor in EF task performance. Weak CC was found to be independent of the other cognitive factors and of the measures of autistic symptomatology used here, supporting the idea that this processing style represents a distinct subtype in the autism spectrum. Using both ToM test performance, and CC test performance or macrocephaly as endophenotypic markers of ASD may provide a way forward in the search for the genetic basis of autism.

Chapter 8: Conclusions

This thesis has focussed on three of the main theories that have attempted to characterise autism at the cognitive level: theory of mind, executive function and central coherence. These theories all share a common goal in attempting to find a unitary cause by which a range of autistic behaviours can be drawn together. Relationships between behavioural tests thought to tap into these cognitive abilities and reports of everyday behaviours were therefore explored, along with relationships between these cognitive domains, in a group of high-functioning junior-aged children with ASD in comparison to a similar group of children with no such diagnosis.

8.1 Summary of main findings

The first aim of this thesis was to assess each of the three main cognitive theories of autism and their relationship to each other with selected tests. Support for the relevance of all three theories to ASD was found and in general, results were consistent with previous findings. Thus, tests that were designed to tap into the cognitive processes that each theory considers a primary abnormality differentiated the present samples of children diagnosed with ASD and typically developing children. Furthermore, at the level of individual differences, subgroups were found characterised by extreme scores (less than the corrected 5th percentile of control performance) on tests tapping each of these theories.

Within the theory of mind (ToM) domain, a battery of tasks was selected, tapping mainly into understanding of false beliefs, as well as a range of other mental states. These tasks produced consistent and highly significant differences between the control and ASD groups. A closer analysis of the tasks revealed that the development in the understanding of others' minds is both delayed and follows a deviant course in many children with ASD

in the present sample, and that this ToM impairment may have wider effects on the understanding of biological agents in general.

In the domain of executive function (EF) too, significant group differences were found. A common denominator in the impaired performance seemed to be in the child's understanding of the task demands when specific prompts were absent. An implicit understanding of the experimenter's expectations did not appear to be available to at least some children with ASD, resulting in performance that appeared idiosyncratic or egocentric. Such behaviour may result from an executive failure in self-monitoring or may be the consequence of a failure in perspective taking.

Group differences were less clear-cut in the central coherence (CC) domain; whilst trends towards a bias for local processing could be demonstrated in both a well known and a novel test, evidence for significant group differences was scant. Observation of the individual profiles revealed a small number of children in the present sample to be particularly good at the more difficult component of the Children's Embedded Figures Test, indicative of weak CC. More importantly, it was predicted that the processing style of weak CC would be found in those children with macrocephaly and the results from a novel Local-Global Switching Task supported this hypothesis. As this is a novel finding, it has to be treated with caution before it has been replicated. Speculatively, this finding might provide a starting point for a neurobiologically-based endophenotype. This test also allowed the nature of the locally-biased processing style to be further examined; the results indicated that those children with macrocephaly could be described as finding it difficult to switch from local level and into global level processing, whilst performing as well as controls when switching in the opposite direction. This can be visualised as a gradient between local and global processing: the slope for individuals with macrocephaly would be higher at the global end, making it more of an uphill struggle to switch from local to global

than the converse, whilst the gradient would slope in the opposite direction for the majority of the population.

When assessing the relationships between the three cognitive domains in the children with ASD, CC emerged as an independent factor from the other domains, whilst ToM and EF were closely related, indicating that at least two independent cognitive subtypes can be found in the autism spectrum as represented by the present sample. This finding also supports the hypothesis that ToM impairments may adversely affect performance in unstructured EF tasks through a lack of understanding of the implicit task demands. Furthermore, ToM and EF task performance were both independently related to verbal ability, although only the relationship between EF and verbal ability appeared to be qualitatively different to that seen in the normally developing children. The emerging hypothesis was therefore that the present ASD group were performing the EF tasks in a qualitatively different way to the control group. This finding led to the proposal that a compensatory mechanism may be involved to overcome problems with EF tasks, possibly through internal verbal self-prompting.

Another aim of the thesis was to examine the pattern of these three types of cognitive impairment in different children with ASD. Using a strict technique to detect deviant performance in each domain (less than the corrected 5th percentile of control performance), between 20% and 50% of children with ASD were identified in each case; these children can be assumed to be impaired in the relevant domain with certainty. Within different children, all possible combinations of impairment were found, although it was most common to see CC or ToM impairment alone or ToM and EF impairments in combination. Whilst many children were therefore classified in each domain as not showing such deviant performance, it cannot be assumed that all of these individuals were unimpaired; their impairments may have been milder or they may have been able to perform the tasks by some other strategy.

Despite this, a proportion of individuals in each domain could be seen to show normal or even superior performance, indicating that none of these cognitive impairments were present in all children with ASD in this sample. Indeed, 39% of children were identified as possibly having intact abilities in all three domains simultaneously. Whether these children had been diagnosed through over-inclusive application of the present criteria is a question that the present study cannot resolve.

The final aim of the thesis was to relate performance on the tasks tapping these cognitive domains to real-life behaviour, as indexed by parental interview. This proved a difficult task. Structural equation modelling, correlations and regression analyses confirmed intricate relationships between both ToM and verbal ability and specific aspects of the behavioural triad.

8.2 Strengths and limitations of study

The work covered in this thesis has been a multiple case study design. This has allowed a range of detailed data to be collected from different individuals in an attempt to assess the impairments present within each individual as accurately as possible and then to compare the pattern of these impairments across different individuals. The relatively large number of children with ASD included here is therefore an asset of the study, as is the quantity of data collected on each child. The inclusion of multiple measures relating to each cognitive domain sought to increase sensitivity and reliability of the overall measure of ability in each domain. Whilst many studies in this field assess one particular aspect of cognition in ASD, this design in which data has been collected relating to three different areas of cognition within the same children has also enabled the relationships between the different theories to be systematically explored, uniting or separating them. Furthermore, by including measures of everyday behaviour, this study has allowed the adequacy of current behavioural tests to be explored, that is the extent to which they tap into these underlying

cognitive impairments, and the extent to which they explain the symptoms of ASD, assessed through parental report.

Despite the strength of the experimental design, a number of limitations in the methodology exist. Firstly, the ASD and control groups were not matched for verbal and performance IQ levels. Even though such matching was originally intended and could have been achieved by excluding participants in the lower IQ band, it seemed most profitable for the particular design employed in this study to include as many children with ASD as possible.

All those children who were included in the ASD group were functioning at a level that allowed them to comply with the task demands, and the majority had either verbal or performance IQ within the control range. While they might therefore be considered high-functioning relative to the autism spectrum as a whole, not all children here were functioning at a level equal to the controls. Still, as already mentioned, individual verbal and performance IQ levels were accounted for in all data analyses in order to correct for this disparity between the groups. As the relationships between most of the experimental variables and IQ scores were similar between the two groups for the majority of cases, the increased range in the ASD group would therefore have little effect on this method of correction. However, this was not the case for the EF tests; it is possible that in some cases, the children with lower IQs may have affected this correction method in minor ways. This had the effect of making children with low verbal IQs appear slightly more competent and children with low performance IQs appear slightly less competent than without these children. On the other hand, the inclusion of only relatively high-functioning children in the ASD group will have led to a sample that is not representative of the ASD population even in the present geographical area. Obviously, the present findings cannot be assumed to be applicable to the ASD population as a whole. For this, an epidemiological study with a representative sample would be necessary.

Whilst the behavioural tests used here were designed specifically to tap into particular cognitive domains, all such tasks will require more than one cognitive ability in order to perform the task, as well as being affected by age and general ability. This is inevitable with almost all cognitive measures but makes task performance difficult to interpret. A number of steps were taken in order to attempt to minimise this problem. Firstly, individual age and IQ levels were accounted for by entering these variables into each analysis as covariates or by using the residuals from regression analyses with these variables entered as predictor variables; this was carried out in order to control for the effect that these factors may have had on task performance. In addition, to avoid task specific factors and increase the likelihood of the intended underlying cognitive factor being tapped, performance was averaged across a number of diverse tests within each domain. This required a child to perform consistently across a number of tasks in order to be categorised as showing deviant performance. Furthermore, some of the tasks involved control conditions to check that poor performance was not the result of more general task difficulties; in particular, there were questions in the ToM battery to control for comprehension, carefully matched physical stories and unlinked passages in the Strange Stories and non-switching conditions in the Local-Global Switching task.

Despite these attempts to ensure the data reflected specific cognitive abilities, it is still possible that other factors may have been contributing to performance. Particularly in the CC and EF domains, control conditions were missing and performance across the different tasks was inconsistent. This made it difficult to assess whether task performance truly reflected these particular cognitive domains. In the ToM domain, consistency may have been overestimated because of the use of only verbal tests; if non-verbal ToM tests had been used as well, the results might have been less consistent. The possibility that some children may be compensating for their impairments cannot be ruled out and makes good performance, or at least performance not classified as deviant, difficult to interpret.

Compensation has been suggested frequently in the literature, particularly when untimed

tasks are involved which may allow task completion through slow deliberation. Again, applying control conditions and accounting for IQ helps to minimise these possibilities but is unlikely to cover all the different possible compensatory mechanisms that could be employed.

Given that the motivation of the children to perform well was high throughout and that they were given constant encouragement during the testing sessions, task failure is more straightforward to interpret than task success. Indeed, the use of the 1.65 SD cut-off to detect individuals with deviant performance was useful in order to explore the pattern of impairments present in each individual. While this cut-off represents performance below the 5th percentile of control performance, a common requirement for clinical significance, this boundary is in some senses artificial. Particularly, the fact that the distribution of scores in the ASD group was sometimes shifted down from the control distribution indicates that all children may have been performing slightly below the expected level on certain tests, rather than some children showing intact performance and others showing deviant performance. Obviously, a bimodal distribution would be the clearest evidence in support of the hypothesis that only a proportion of children were impaired on a particular test or domain, but such idealised data is rarely seen in practice; this is in part the consequence of using quantitative behavioural techniques to tap into cognitive systems, as well as the noise present in any data set. Throughout the thesis, interpretation of the data has therefore attempted to reflect the limitations of this methodology.

Similarly to the cut-off used for the cognitive tests, the cut-offs recommended on the 3Di were found to be rather arbitrary; the data seemed better represented as a continuum. Again, this does not mean to say that children with ASD are only quantitatively different from normally-developing children, rather that it is difficult to locate qualitative differences at the behavioural level and that the 3Di is designed to be a quantitative tool. Since the data from the children in the ASD group did seem to indicate that the cut-off for the

communication domain was relatively low in comparison to the social and repetitive behaviour domains, it may well have been that some children who met criteria only on the basis of communication impairments were actually rather mild cases.

Another limitation of the data must be the sole use of parental report to assess everyday behaviour; such data may produce high consistency between measures within each child, possibly artificially high, but high variability between different children. This would result from each child being rated by a different person and from different parents perceiving the same behaviour differently. If this was the case, this would help to further explain the paucity of relations between the reports of everyday behaviour and the test measures. Ratings by teachers and direct behavioural observation would have been useful supporting evidence, but were not possible due to time constraints.

8.3 Implications of results

The finding that at least two distinct cognitive subgroups of relatively able junior-aged children with ASD exist and that only a proportion of children show each of these profiles has implications for ASD research methodology and theory, for genetic studies, and for clinical diagnostic and interventional strategies. Methodologically, the multiple case study and analysis of individual data design begins to address issues of causality which group studies are not able to approach. It also highlights the need to treat ASD as a heterogeneous group of individuals with different underlying cognitive impairments that cannot be assumed to be universal; this is important for both experimental design and data interpretation.

Cognitive heterogeneity may also have implications for identifying distinct endophenotypes within the autism spectrum, which could aid in the search for candidate genes. Although ASD is known to be a highly heritable condition (Folstein & Rosen-Sheidley, 2001), specific genetic loci for the disorder have still to be located and reliably replicated

(Bacchelli & Maestrini, 2006), most likely because genetic and therefore neurobiological heterogeneity exist in ASD in addition to cognitive heterogeneity; different samples of individuals with ASD may contain individuals with different underlying genetics, increasing noise in the data and making it unlikely that the same results would be replicated. The use of cognitive subtypes as endophenotypes may therefore aid in the search for the genetic basis of autism; different genes or groups of genes may be related to different cognitive subtypes. The finding in the central coherence domain of a subgroup of individuals with weak central coherence and macrocephaly is encouraging in this respect; it may well indicate that head size is one potential biological endophenotype that could be explored.

Given that children on the spectrum are currently being grouped under a single label (or categorised in terms of severity and IQ) but differ enormously both in the behavioural manifestations they display (Wing & Gould, 1979) and the cognitive underpinnings of the disorder, any interventional approach is unlikely to benefit every child (Hutchins & Prelock, 2006). Klinger & Renner (2000) reported that the assessment of cognitive impairments is currently useful for treatment planning. Having a better understanding of the underlying problems present in different children should enable clinicians to plan interventions appropriate to an individual child and allow parents and teachers to respond more directly to the child's needs. It would therefore be beneficial for diagnostic assessment to recognise cognitive subtypes in addition to assessing behavioural symptoms.

8.4 Future directions

The outcomes of the present research highlight a number of avenues for future exploration. Firstly, it would be useful to follow-up and replicate the finding that weak central coherence is found in individuals with autism with macrocephaly. Specifically, it would be of interest to recruit two new groups of children with ASD, one with and one without macrocephaly and measure their performance across a variety of different central

coherence tasks, particularly those with strict control measures that also require fast on-line processing and including the Local-Global Switching task used here. Furthermore, it would be crucial to extend this causal chain to the behavioural level, exploring those non-diagnostic behaviours specifically thought to result from weak CC, in order to assess the effect that this biological abnormality and cognitive impairment would have on everyday behaviours. Current diagnostic measures pay little attention to behaviours such as rote memory, hyper- and hyposensitivity, islets of ability, literal understanding of language and poor comprehension of text relative to word decoding ability. Whilst the CC questionnaire included in this thesis failed to relate directly to the present CC measures, it was only a first attempt to identify such behaviours and did in fact produce significant group differences; this would need to be refined and developed.

The relationship between EF and ToM also deserves further investigation. The proposed hypothesis that a poor understanding of implicit task demands in certain executive function tasks results from a lack of understanding of the experimenter's expectations would predict problems specifically on tasks without explicit prompting. It would therefore be beneficial to explore performance of individuals with ASD on a task in which either implicit or explicit instructions were given in different matched conditions.

A big question mark hangs over those children who showed apparently intact performance across all three cognitive domains. In what other way can their reported behaviours, which led to their diagnosis, be explained? Were these children in fact able to compensate for their difficulties so that they performed abnormally well on tasks that should have presented a challenge to them? Firstly, it would be crucial to check on the reported behaviours by utilising a more explicit means of assessing everyday behaviours through a direct observational method. For instance, the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 1999) could be used to ascertain whether these children differ at all in the severity or profile of autistic symptoms and to further assess whether they truly fit an

autistic profile. It would be helpful to know whether these children were wrongly diagnosed as having ASD, but may be merely delayed in certain aspects of their development and have outgrown earlier difficulties. Alternatively, they may have hidden impairments not assessed at present. Whilst explicit behavioural measures designed to tap into cognitive processes may be insensitive to such impairment, it is possible that more implicit measures, such as eye-tracking technology, could be used to detect residual cognitive differences.

The use of brain imaging techniques could aid in clarifying the reason for these children not displaying significantly deviant performance on cognitive tasks. An informative way of differentiating between children with ASD but with intact cognitive abilities and those with impaired cognitive abilities but intact performance through compensation is to measure brain activity using functional Magnetic Resonance Imaging. This would involve comparing those children with ASD and intact performance in a particular cognitive domain to control children, matched on the basis of their performance on cognitive tasks. When comparing brain activity whilst performing experimental tasks designed to tap into that cognitive domain as well as control tasks, it would be expected that children with ASD with intact performance would differ from control children if they were somehow compensating for a hidden impairment, showing similar brain activity to those children with ASD who showed impaired task performance. However, if brain activity were similar in the children with ASD plus intact performance and in the control groups, this would indicate truly intact cognitive abilities.

This thesis has attempted to advance the understanding of how the cognitive impairments in ASD relate to one another and to behaviour. As a developmental disorder, any investigation conducted at a particular point in time is therefore certain to fall short of providing a full understanding of how these factors interact. A future aim would therefore be to follow up these children in order to study the developmental course and stability of

their impairments and investigate whether the severity and presence of each cognitive deficit was predictive of future outcomes. This would be useful for providing appropriate resources and interventional strategies and for planning for the individual's future. An additional avenue that would also have direct clinical relevance would be to develop a standardised battery of tests tapping into these cognitive domains, for use in clinical and educational assessment.

8.5 Concluding thoughts

Is it really plausible to suggest that we can unite so many different individuals under the 'autism spectrum' label? Can the child who hides his head under his coat in order to avoid a testing session be reconciled with the child who claimed that 'G' was the capital of Greece, with the child who wondered whether Christopher Columbus was the Christopher in his class, with the child who only eats yellow food when he goes out to restaurants, with the child who is indifferent to the many people who want to be his friend, with the child who destroys his school work when it lacks perfection, with the child who can't understand why other children don't enjoy reading an encyclopaedia, with the child who walks directly behind someone they're talking to, with the child who calls everyone 'he' regardless of their gender?

There is no simple answer to this question but it is hoped that, as with the multitude of other research in this field, this study will be a small step forward in understanding this complex disorder. While each child may indeed appear different on the surface, clinical intuition has led us to search at a deeper level for the 'something' in common. Indeed, much of the analysis presented here points towards a continuum of severity. Nonetheless, the multiple case study approach undoubtedly indicates that children with ASD differ from each other in terms of their underlying problems and therefore that the continuum of severity is likely to be composed of a number of dimensions. The notion of 'autism' is

obviously still highly relevant and valuable but the reality of its manifestation may be broader than our current conceptualisation of the disorder permits.

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Appendices

Appendix I: Novel everyday behaviour questionnaire

Theory of Mind items

- understands the concept of pretending, eg. someone acting on TV isn't that person in real life
- unaware that different people enjoy different things, eg. food, work
- lacks the understanding of how to behave appropriately with different people, eg. teacher / brother / stranger
- aware that a person's knowledge depends on their experience, eg. asks questions to people who are likely to know the answers
- unaware that adults have limited knowledge, eg. surprised when parent makes a mistake
- understands other people's long term intentions, including failed attempts to get something, eg. someone may return to the library every week in order to take out the latest bestseller but come home without the book as it's out on loan
- aware that young children may need things explained more simply
- has difficulty recognising surprise in others
- recognises embarrassment in others
- shows pride after an achievement and looks for public praise
- lacks the knowledge of how to conceal information from others
- confides in others and understands that people keep secrets in order to keep information private
- apologises for hurting others' feelings; not just a routine "sorry"
- lacks the understanding of how own behaviour affects others, eg. seems unaware that, if slow getting ready for school, this will also make parents late
- responds to being called or looked at with eagerness to listen

Executive Function items

- finds it hard to think up new ideas on own, eg. a new game to play
- can make hypothetical inferences about factual information eg. if it's snowing in Scotland, can infer it must be cold there
- when drawing, only uses a single colour and does not change colours
- can make sensible decisions for self, eg. puts raincoat on when going out in the rain
- prevaricates at every decision, eg. can't chose what flavour of icecream to have
- appears unable to control own actions; acts without thinking
- once understood, is able to follow the rules of a game without having to be reminded
- obeys parents/teachers; if told not to do something, will not do it
- unaware of personal safety, eg. when crossing the road or taking part in physical activities
- happy to wait for rewards; doesn't need them immediately, eg. will tidy room today when reward is going to the zoo next week

Central Coherence items

- notices small details when others do not, eg. small sounds, small changes in the position of furniture
- has strong interests which he/she gets upset about if he/she can't pursue them
- usually concentrates more on the whole picture rather than the small details
- good at remembering lists of items, eg. phone numbers

- poor at remembering facts, eg. dates of birth, general knowledge
- has a hobby involving collecting items in a particular category or making lists, eg. stamp collecting, trains
- hypersensitive to certain noises and objects, eg. agitated by the sound of the vacuum cleaner, dislikes the feel of certain materials

Theory of Mind & Executive Function items

- attends appropriately to others' emotional states, eg. celebrates successes, consoles failures
- finds it hard to initiate flexible small talk, eg. unlikely to chat to newcomers or make them feel at home
- volunteers important missing information, eg. tells someone, who has missed the beginning of a TV programme, what has happened so far
- expresses ideas in more than one way, eg. if someone doesn't understand something, he/she can say it again in a different way to make him/herself clear
- has difficulty when choosing appropriate presents for others, eg. likely to choose something he/she likes rather than something the other person likes
- says and does things that might embarrass others, eg. scratching/yawning in public, saying they don't like a present to the person who gave it to them
- reveals private information to inappropriate people, eg. might give phone number to a stranger
- poor at playing games such as hide & seek or cheat, eg. reveals his/her hiding location, reveals the fact he/she is cheating
- keeps secrets for as long as appropriate
- weighs consequences of own actions on others, eg. doesn't eat all the crisps so there are some left for others, even though he/she wants them all
- dislikes sharing with others, eg. toys, sweets, pens
- commits 'faux pas' without noticing and does not notice this in others, eg. saying something tastes awful in the presence of the person who cooked it
- has realistic long-range goals and plans that take others into account, eg. if wants to go to friend's house, considers whether parent would be free to drive them there that day

Executive Function & Central Coherence items

- repeatedly says/does the same thing
- plays creatively, eg. will build different kinds of things from lego rather than building the same model with slight variations
- gets caught up in minor details or a single topic
- can solve puzzles with missing information, eg. hang man, crosswords
- gets so absorbed in one thing that loses sight of other things
- doesn't grasp the gist of stories or films; has to ask what's going on
- copes well with changes to daily routine eg. a different route to school
- has favourites and won't try alternatives, eg. piece of music, food, clothes
- likes to keep things in certain places or in a certain order and gets upset if they are rearranged
- likes to have every detail of an activity he/she is involved in carefully planned (by self or someone else)
- can think ahead/plan logically for self without help eg. what books needed for school that day

Theory of Mind, Executive Function & Central Coherence items

- responds to hints and indirect cues in conversation, eg. takes shoes off in response to someone saying "I've just cleaned the floor"

- initiates conversation of interest to others, eg. asks friends about their hobbies as well as talking about own
- engages in a range of elaborate make-believe activities, eg. can act different characters
- tends to take things that are said literally, eg. "you're a pain in the neck"
- manipulates others by positive means of flattery, bribery and getting on the right side of someone rather than by physical or negative means
- understands irony and jokes, eg. "well that's very neatly put away" when the room is a mess

Appendix 2: Theory of Mind Tasks

1) Diverse Desires: Snacks (sheet needed)

This is Mandy. It's snack time so Mandy wants something to eat. Here are two different snacks: carrots and cakes.

Own desire: Which snack would you like best? Carrots / Cakes

Well, Mandy really likes [the other one]. She doesn't like [child's choice]. She likes [the other one] best.

So, now it's time to eat. Mandy can only chose one snack.

Test: Which snack will she chose? Carrots / Cakes

Reality control: Which snack does Mandy like best? Carrots / Cakes

2) Diverse beliefs: Cat (sheet needed)

This is Linda. Linda wants to find her cat. Her cat might be hiding in the tree or it might be hiding in the garage.

Own belief: Where do you think her cat is hiding? Tree / Garage

Well, that's a good idea but Linda thinks her cat is hiding in the [other place].

Test: Where will she look for her cat? Tree / Garage

Reality control: Where does Linda think her cat is hiding? Tree / Garage

3) Knowledge access: Bear (props needed)

Show child closed box.

Own belief: What do you think is inside this box?

Open box and show the bear to the child.

Put bear back in box and replace lid.

Own knowledge: What's inside the box?

Polly has never seen inside this box before. Now here she comes.

Test: Does Polly know what's inside the box? Yes / No

Reality control: Has Polly seen inside the box? Yes / No

Memory control: When I first showed you the box, what did you think was inside?

4) Contents False Belief: Smarties (props needed)

Show child sealed Smarties tube.

Own belief: What do you think is inside?

Open tube and show that it actually contains a pencil.

Put pencil back in tube and replace lid.

Own knowledge: What's inside the tube?

In a minute your friend X is going to come in. He hasn't seen this tube yet. When he comes in I'm going to show him this tube, closed up just like this. I'm going to ask him 'What's in here?'

Test: What will X say? Smarties / Pencil

Why will they say that?

Reality control: What is really inside?

Memory control: When I first showed you the tube, what did you think was inside?

5) Explicit False Belief: Gloves (sheet needed)

This is Scott. Scott wants to find his gloves. They might be in his rucksack or they might be in his drawers. Scott's gloves are really in his rucksack but Scott thinks they're in his drawers.

Test: Where will Scott look for his gloves? Rucksack / Drawers

Why will he look there?

Reality control: Where are his gloves really? Rucksack / Drawers

6) Implicit False Belief: Sally-Ann (props needed)

This is Sally and this is Ann. Sally has a basket and Ann has a box. Sally has a marble and she puts her marble in her basket to keep it safe when she goes out. But while Sally is out, naughty Ann takes Sally's marble out of her basket and she puts it in her box.

Test: When Sally comes back, where will she think her marble is?

Basket / Box

Why?

Reality control: Where is the marble really? Basket / Box

Memory control: Where did Sally put the marble in the beginning?

Basket / Box

7) Belief-Emotion: Cereal (props needed)

Show child closed cereal box

Own belief: What do you think is in here?

This is Jane. Rice crispies are her favourite cereal. "I love rice crispies" says Jane. Then off she goes to have a wash before breakfast.

Let's have a look inside. There are really stones inside, and no rice crispies.

Memory control: What is Jane's favourite cereal? Rice crispies

Now remember, Jane has never seen inside this cereal box. Now, Jane comes back in and it's breakfast time now. Let's give the box to Jane.

Test: How does Jane feel when she gets the box? Happy / Sad?

Why?

Open the box.

Emotion control: How does Jane feel when she looks inside the box?

Happy / Sad?

8) Real-Apparent Emotion: Jokes (2 sheets needed)

Show pictures of faces and check child knows that they're happy, sad and OK.

Show picture of girl. "This is a story about Penny. I'm going to ask you about how Penny really feels inside and how she looks on her face. She might really feel one way inside but look a different way on her face. Or she might really feel the same way inside as she does on her face. I want you to tell me how she really feels inside and how she looks on her face. Penny's friends were playing together and telling jokes. One of the older boys called Matt told a mean joke about Penny and everyone laughed. Everyone thought it was very funny but Penny didn't. BUT, Penny didn't want the other children to see how she felt about the joke because they would call her a baby. So Penny tried to hide how she felt.

Memory control: What did the other children do when Matt told the mean joke about Penny?

What would the other children do if they knew how Penny felt?

Test-feel: So how did Penny really feel inside when everyone laughed?

Happy / OK / Sad

Test-look: How did Penny look on her face when everyone laughed?

Happy / OK / Sad

Why?

9) Ice-Cream Story (props needed)

This is John and this is Mary. They live in this village.

Naming: Which is John/Mary?

Here they are in the park. Along comes the ice-cream man. John would like to buy an ice-cream but has left his money at home. He is very sad.

"Don't worry," says the ice-cream man, "you can go home and get your money and buy some ice-cream later. I'll be here in the park all afternoon".

"Oh good," says John, "I'll be back in the afternoon to buy an ice-cream".

Prompt: Where did the ice-cream man say to John he would be all afternoon?

So John goes home. He lives in this house. Now, the ice-cream man says to Mary, "I am going to drive my van to the church to see if I can sell my ice-creams outside there".

Prompt: Where did the ice-cream man say he was going?

Prompt: Did John hear that?

The ice-cream man drives over to the church. On his way he passes John's house. John sees him and says "Where are you going?" The ice-cream man says "I'm going to sell some ice-cream outside the church". So off he drives to the church.

Prompt: Where did the ice-cream man tell John he was going?

Prompt: Does Mary know that the ice-cream man has talked to John?

John goes to buy an ice-cream.

Now Mary goes home. She live in this house. Then she goes to John's house. She knocks on the door and says "Is John in?"

"No", says his mother, "he's gone to buy an ice-cream".

Test-belief: Where does Mary think John has gone to buy an ice-cream?

Justification: Why?

Reality control: Where did John really go to buy an ice-cream?

Memory control: Where was the ice-cream man in the beginning?

10) Birthday Puppy (props needed)

Tonight is Peter's birthday and Mum is surprising him with a puppy. She has hidden the puppy in the basement. Peter says "Mum, I really hope you get me a puppy for my birthday".

Remember, Mum wants to surprise Peter with a puppy. So, instead of telling Peter she got him a puppy, Mum says, "Sorry Peter, I did not get you a puppy for your birthday. I got you a really great toy instead".

Prompt: Did Mum really get Peter a toy for his birthday?

Prompt: Did Mum tell Peter she got him a toy for his birthday?

Prompt: Why did Mum tell Peter that she got him a toy for his birthday?

Now, Peter says to Mum, "I'm going outside to play". On his way outside, Peter goes down to the basement to fetch his roller skates. In the basement, Peter finds the birthday puppy! Peter says to himself, "Wow, Mum didn't get me a toy, she really got me a puppy for my birthday". Mum does **not** see Peter go down to the basement and find the birthday puppy.

Nonlinguistic control: Does Peter know that his Mum got him a puppy for his birthday?

Linguistic control: Does Mum know that Peter saw the birthday puppy in the basement?

Now the telephone rings, ding-a-ling! Peter's grandmother calls to find out what time the birthday party is. Grandma asks Mum on the phone, "Does Peter know what you really got him for his birthday?"

2nd order ignorance: What does Mum say to Grandma?

Now remember, Mum does not know that Peter saw what she got him for his birthday.

Then, Grandma says to Mum "What does Peter think you got him for his birthday?"

2nd order false belief: What does Mum say to Grandma?

Justification: Why does Mum say that?

11) Penny Hiding

We're going to play a simple hiding game, one that they probably know already.

Hide a coin behind your back and bring hands out again as two closed fists. Ask child to guess which hand has the coin. Always use RLRRLR.

Now it's your turn. See if you can trick me. Hide it really well, just like I did.

Get child to do this 6 times, and note down how successful each trial is.

	Trial1	Trial2	Trial3	Trial4	Trial5	Trial6
Which hand?						
Does child hide <i>both</i> hands behind back? (-1)						
Does the child bring <i>both</i> hands forward? (-1)						
Are hands closed? (-1)						
Is the coin hidden? (-1)						
Asymmetric hands? (-0.5)						
Tricks used?						

12) Cow task

Show child the degraded picture of the cow.

Own belief: What can you see in this picture?

Allow 10 seconds to look at picture then place transparency over picture.

Own knowledge: What can you see in this picture now? Cow / Cow with help

Help child to recognise it as an animal of some sort.

Remove transparency.

**Can you show me the *animal's* ears?
eyes?
nose?**

If answer is negative, replace transparency and repeat process.

“In a minute, one of your friends is going to come in. If I show him this picture, just like I showed you the first time, and I'll ask him what's in the picture.”

Test: What will he say?

Justification: Why?

Reality control: What's really in the picture?

Memory control: When I first showed you the picture, what did you say was in the picture?

Appendix 3: Strange Stories

Mental state stories

Simon is a big liar. Simon's brother Jim knows this, he knows that Simon never tells the truth! Now yesterday Simon stole Jim's ping-pong paddle, and Jim knows Simon has hidden it somewhere, though he can't find it. He's very cross. So he finds Simon and he says, "Where is my ping-pong paddle? You must have hidden it either in the cupboard or under your bed, because I've looked everywhere else. Where is it, in the cupboard or under your bed?" Simon tells him the paddle is under his bed.

Q: Why will Jim look in the cupboard for the paddle?

2 points reference to Jim knowing Simon lies

1 point reference to facts (that's where it really is) or Simon hiding it without reference to implications of lying

0 points reference to general non-specific information (because he looked everywhere else)

During the war, the Red army captures a member of the Blue army. They want him to tell them where his army's tanks are; they know they are either by the sea or in the mountains. They know that the prisoner will not want to tell them, he will want to save his army, and so he will certainly lie to them. The prisoner is very brave and very clever, he will not let them find his tanks. The tanks are really in the mountains. Now when the other side ask him where his tanks are, he says, "They are in the mountains".

Q: Why did the prisoner say that?

2 points reference to fact that other army will not believe and hence look in other place, reference to prisoner's realisation that that's what they'll do, or reference to double bluff

1 point reference to outcome (to save his army's tanks) or to mislead them

0 points reference to motivation that misses the point of double bluff (he was scared)

Brian is always hungry. Today at school it is his favourite meal - sausages and beans. He is a very greedy boy, and he would like to have more sausages than anybody else, even though his mother will have made him a lovely meal when he gets home! But everyone is allowed two sausages and no more. When it is Brian's turn to be served, he says, "Oh, please can I have four sausages, because I won't be having any dinner when I get home!"

Q: Why does Brian say this?

2 points reference to fact that he's trying to elicit sympathy, being deceptive

1 point reference to his state (greedy), outcome (to get more sausages) or factual

0 points reference to a motivation that misses the point of sympathy elicitation/deception, or factually incorrect

Jill wanted to buy a kitten, so she went to see Mrs. Smith, who had lots of kittens she didn't want. Now Mrs. Smith loved the kittens and she wouldn't do anything to harm them though she couldn't keep them all herself. When Jill visited she wasn't sure she wanted one of Mrs. Smith's kittens, since they were all males and she had wanted a female. But Mrs. Smith said, "If no one buys the kittens I'll just have to drown them!"

Q: Why did Mrs. Smith say that?

2 points reference to persuasion, manipulating feelings, trying to induce guilt/pity

1 point reference to outcome (to sell them or get rid of them in a way which implies not drowning) or simple motivation (to make Jill sad)

0 points reference to general knowledge or dilemma without realisation that the statement was not true (she's a horrible woman)

One day Aunt Jane came to visit Peter. Now Peter loves his aunt very much, but today she is wearing a new hat; a new hat which Peter thinks is very ugly indeed. Peter thinks his aunt looks silly in it, and much nicer in her old hat. But when Aunt Jane asks Peter, "How do you like my new hat?", Peter says, "Oh, its very nice".

Q: Why does he say that?

2 points reference to white lie or wanting to spare her feelings; some implication that this is for aunt's benefit rather than just for his, desire to avoid rudeness or insult

1 point reference to trait (he's a nice boy) or relationship (he likes his aunt); purely motivational (so she won't shout at him) with no reference to aunt's thoughts or feelings; incomplete explanation (he's lying, he's pretending).

0 points reference to irrelevant or incorrect facts/feelings (he likes the hat, he wants to trick her)

Helen waited all year for Christmas, because she knew at Christmas she could ask her parents for a rabbit. Helen wanted a rabbit more than anything in the world. At last Christmas Day arrived, and Helen ran to unwrap the big box her parents had given her. She felt sure it would contain a little rabbit in a cage. But when she opened it, with all the family standing round, she found her present was just a boring old set of encyclopaedias, which Helen did not want at all! Still, when Helen's parents asked her how she liked her Christmas present, she said, "It's lovely, thank you. It's just what I wanted".

Q: Why did she say this?

2 points reference to white lie or wanting to spare their feelings; some implication that this is for parent's benefit rather than just for her, desire to avoid rudeness or insult

1 point reference to trait (he's a nice girl) or relationship (she likes her parents); purely motivational (so they won't shout at her) with no reference to parent's thoughts or feelings; incomplete explanation (she's lying, she's pretending).

0 points reference to irrelevant or incorrect facts/feelings (she likes the present, she wants to trick them)

Late one night old Mrs. Peabody is walking home. She doesn't like walking home alone in the dark because she is always afraid that someone will attack her and rob her. She really is a very nervous person! Suddenly, out of the shadows comes a man. He wants to ask Mrs. Peabody what time it is, so he walks towards her. When Mrs. Peabody sees the man coming towards her, she starts to tremble and says, "Take my purse, just don't hurt me please!"

Q: Why did she say that?

2 points reference to her belief that he was going to mug her or her ignorance of his real intention

1 point reference to her trait (she's nervous) or state (she's scared) or intention (so he wouldn't hurt her) without suggestion that fear was unnecessary

0 points factually incorrect/irrelevant answers; reference to the man actually intending to attack her

A burglar who has just robbed a shop is making his getaway. As he is running home, a policeman on his beat sees him drop his glove. He doesn't know the man is a burglar, he just wants to tell him he dropped his glove. But when the policeman shouts out to the burglar, "Hey, you! Stop!", the burglar turns round, sees the policeman and gives himself up. He puts his hands up and admits that he did the break-in at the local shop.

Q: Why did the burglar do that?

2 points reference to belief that policeman knew that he'd burgled the shop

1 point reference to something factually correct in story

0 points factually incorrect/irrelevant answers

Human physical state stories

Two enemy powers have been at war for a very long time. Each army has won several battles, but now the outcome could go either way. The forces are equally matched. However, the Blue army is stronger than the Yellow army in foot soldiers and artillery. But the Yellow army is stronger than the Blue Army in air power. On the day of the final battle, which will decide the outcome of the war, there is heavy fog over the mountains where the fighting is about to occur. Low-lying clouds hang above the soldiers. By the end of the day the Blue army has won.

Q: Why did the Blue army win?

2 points reference to both weather conditions and either relative ground superiority or inability of other army's planes to be useful in fog (names of armies unimportant)

1 point reference either to weather or relative superiority on ground versus air (because it was foggy); nothing about why weather makes it especially difficult for planes or nothing about planes being affected more than tanks; reference to fog to justify incorrect response (the aeroplanes won because the fog meant they could hide from the tanks)

0 points reference to irrelevant or incorrect information (they won because they had better planes); justifications for why tanks are better than planes

A burglar is about to break into a jewelers' shop. He skillfully picks the lock on the shop door. Carefully he steps over the electronic detector beam. If he breaks this beam it will set off the alarm. Quietly he opens the door of the store-room and sees the gems glittering. As he reaches out, however, he steps on something soft. He hears a screech and something small and furry runs out past him, towards the shop door. Immediately the alarm sounds.

Q: Why did the alarm go off?

2 points reference to animal which the burglar disturbed setting off alarm by crossing beam (type of animal unimportant)

1 point reference to burglar setting off alarm (he was startled by the animal so crossed the beam); reference to animal setting off alarm without explaining it crossed the beam (he trod on a cat and it set off the alarm)

0 points reference to irrelevant or incorrect factors (the animal's screech set off the alarm); alternative reasons for alarm going off (a security camera saw him and set the alarm off)

Old Mrs. Robinson is very frail. One day she slips on her icy door step and falls on her side. She gets up right away, although she feels quite bruised and shaken. The next day her leg feels very stiff and she can scarcely walk. She makes her way to the doctors. As soon as the doctor hears about the fall, and sees her swollen side, he says, "Go immediately to the hospital". At the hospital they take an X-ray.

Q: Why did they take an X-ray?

2 points reference to possibility that she has fractured/broken her hip/leg; reference to wanting to know or trying to find out (i.e. 'it was broken' is not enough); must refer to fact that X-rays are for broken things or bones (to see if there's any damage to the bone)

1 point reference to general aim (to see what's wrong, because of her fall she might have damaged something) or factually correct (it's bruised and stiff)

0 points reference to irrelevant (because she fell) or incorrect factors (that's what doctors do) or to X-rays being cures themselves (to mend her leg)

John is going shopping. He buys a nice new desk lamp, for his study. He needs a light bulb for his new lamp. He goes from the furniture department to the electrical department. In the electrical department he finds that there are two brands of light bulb of the right kind. Everbrite light bulbs cost less in single packs than Literite bulbs. However, only Literite bulbs come in multi-packs of six. John buys the multi-pack, even though he

only needs one bulb.

Q: Why does John buy the Literite bulbs?

2 points reference to saving money by buying the multipack

1 point reference to convenience of having more bulbs, or future need for more than one bulb; no mention of saving money

0 points reference to irrelevant or incorrect factors (Literite bulbs are brighter)

Bob and Jim are best friends. They are both ten years old. Bob has brown hair, green eyes and is over 5 foot tall. Jim looks very different to Bob. He has blonde hair and blue eyes and he is much smaller than Bob. Bob and Jim go on an outing to the fun fair. They go on lots of rides. For the last ride of the day they decide to go on the big rollercoaster. But there is a sign which says: For safety reasons no persons under 5 foot are allowed on.

Q: Why does only Bob go on the rollercoaster?

2 points reference to Jim being too short for the ride or Bob being tall enough (Jim's less than 5 foot)

1 point reference to Jim being short or Bob being tall or both; no reference to height in comparison to the limit (Jim's shorter than Bob)

0 points reference to irrelevant or incorrect factors (Jim doesn't like rollercoasters)

Rupert has never been skiing before and is looking forward to his first skiing holiday this winter. All his kit for the holiday has been well prepared; his mum has bought him a pair of goggles and she has thoroughly waxed and polished the bottom of his skis to protect them. On the first day of Rupert's holiday his skis keep slipping from underneath him, making him fall over into the snow.

Q: Why does Rupert keep falling over?

2 points reference to Rupert's Mum having waxed the skis, making them slippery

1 point reference to Rupert's never having skied before

0 points reference to irrelevant or incorrect factors (his skis are loose)

Clare is having her room redecorated; her mother is painting the walls and having new curtains hung. Before, Clare's room was pink and white with thin net curtains but now the walls are dark red, and brand new thick and expensive velvet curtains have been put up. On the first morning in her new room, Clare fails to wake up at the normal time. As her mother rushes to get her out of bed for school, Clare says it must be too early to get up because it 'feels like the middle of the night'.

Q: Why did Clare oversleep?

2 points reference to the room being darker after redecoration (her room is dark now that she has thicker curtains)

1 point reference to redecoration; no reference to this making the room darker

0 points reference to irrelevant or incorrect factors (she's too tired, she doesn't want to go to school)

Sam decides to go on a long walk to get some fresh air. Unfortunately, just after leaving the house, the wind begins to pick up and it starts to rain. Luckily Sam always has an umbrella with him. He quickly puts up the umbrella and wraps his coat tightly around him. Suddenly a gust of wind blows the umbrella straight out of Sam's hand and it lands in a large, very prickly bush. Sam manages to run and fetch it before it blows off again and is pleased to find it all in one piece. As he walks home, he notices that his head is starting to get wet despite the umbrella.

Q: Why is Sam getting wet?

2 points reference to the bush making holes in the umbrella

1 point reference to either the bush or to holes in the umbrella

0 points reference to irrelevant or incorrect factors (it was raining, he hasn't got an umbrella)

Animal physical state stories

Emperor penguins live in the Antarctica, where it is extremely cold. There is always snow on the ground and ice on the surface of the sea. Emperor penguins can often be found standing clumped together in huge, huddled masses. Every few minutes, a penguin in the middle of the huddle moves to the edge of the huddle, changing places with one of the penguins on the outside of the group.

Q: Why do the penguins keep changing places?

2 points reference to the middle penguin being the warmest or to taking turns at being warm

1 point reference to keeping warm without relating this to the huddle

0 points reference to irrelevant or incorrect factors

Snakes are remarkable animals. They have very stretchy skin, which they shed once a year and can also separate their upper and lower jaws and open their mouths really wide. The anaconda is an example of a very large snake. One day, a deer gallops under a tree, from which an anaconda is hanging, as it makes its way towards a lake. Later that day, the snake is lying on the ground with a huge bulge in its middle. The deer however is nowhere to be seen.

Q: Where is the deer?

2 points reference to the snake having eaten the deer (in the snake's tummy)

1 point reference to the deer having been eaten or being dead without reference to the snake

0 points reference to irrelevant or incorrect factors

It has been raining for days and days and there are no signs that it is going to stop anytime soon. A little island lies in the middle of a huge river. The water in the river has been slowly rising each day and it has nearly reached the top of the river banks. The otters swim around in the water and the field mice run about the island gathering food. Five days later, the rain has finally stopped. The otters still swim in the water, but there are no signs of the field mice.

Q: What has happened to the field mice?

2 points reference to the field mice having drown or being dead because of the water

1 point reference to them being dead without reference to water or to the water taking them away without reference to them being dead

0 points reference to irrelevant or incorrect factors

Lions are fierce hunters. They can run as fast as a car when they are young and fit but they get very slow and weak when they are old. One very hot day, an old and hungry lion is standing at the mouth of a cave, watching a herd of zebras moving across a large open plain. When the herd has passed by, the lion begins to chase a small zebra at the back of the herd. One by one, the zebras nimbly jump across a river. But the lion returns to the cave, still hungry.

Q: Why is the lion still hungry?

2 points reference to the lion being old or slow or weak and so not being able to catch the zebras (he was too old and weak to jump over the river)

1 point reference to the lion being unable to catch the zebras without reference to him being old or slow or weak (he couldn't jump across the river to catch the zebras)

0 points reference to irrelevant or incorrect factors

Some types of birds, like geese and swallows, only like very warm weather. When it is winter in England, it is still very warm in other countries that are further south. Last autumn, flocks of swallows could be seen flying in huge groups in the same direction away from England. At the beginning of summer, these swallows flew back to England.

Q: What were the swallows doing?

- 2 points reference to migration or to flying to the place where it is currently warm
- 1 point reference to specific examples without a general understanding of finding warm places (they were going away from the cold weather in England; they were going away to a hotter country) or to keeping warm without any explanation
- 0 points reference to irrelevant or incorrect factors

Swordfish come in many different colours; black, greyish blue, brown, purple and bronze. They often live in tropical places where the water is very warm. In these tropical places, the weather sometimes gets so hot that thunderstorms occur. When this happens, huge waves crash onto the beaches and travel a long way up the land. On one very hot day in Hawaii, a swordfish is lying on the beach.

Q: Why is the swordfish lying there?

- 2 points reference to the swordfish having been washed up onto the beach by a huge wave or the storm
- 1 point reference to the storm or sea or waves without reference to the swordfish
- 0 points reference to irrelevant or incorrect factors

Seals have very big eyes and long whiskers that help them to sense tiny movements. Underwater they use their whiskers to find fish so they can then catch the fish and eat them. A seal without any whiskers at all, is lying on a rock in the North Sea. This seal is very, very thin and tired.

Q: Why is this seal so thin?

- 2 points reference to the seal having no whiskers and therefore not being able to find fish
- 1 point reference either to not having eaten or to not having whiskers
- 0 points reference to irrelevant or incorrect factors

Animals that live in groups often have an order of importance within the group. The strongest male is the leader of the group. This leader will often attack other animals in the group who are not as strong as this leader. This shows the other animals how important the leader is. In a group of chickens a very small chicken hasn't got many of its feathers left.

Q: Why hasn't this chicken got many feathers left?

- 2 points reference to the chicken having been attacked by the leader/a larger chicken or having been attacked because of its size
- 1 point reference either to the chicken's size or to having been attacked (reference to it being young rather than small do not count)
- 0 points reference to irrelevant or incorrect factors

Natural physical state stories

In stormy weather, rocks often fall from the top of mountains. One day on a mountain in the Dolomites, a very large boulder becomes loose and starts rolling down the mountain. It rolls and rolls and rolls, gathering speed and spinning and bouncing off the mountain side. Suddenly, there is a very noisy splash.

Q: Why is there a loud splash?

- 2 points reference to the boulder falling into water to make the splash (the boulder must have fallen into a lake)
- 1 point reference to water without reference to the boulder (there was a pool at the bottom of the mountain)
- 0 points reference to irrelevant or incorrect factors (it's very big so it's very noisy)

A storm is building up over a little village in the mountains. There is thunder and lightening. The trees sway in the heavy gusts of wind, and the rain is pouring down.

Leaves and even some branches are falling from the trees. After one extremely bright flash of lightning, there is a loud crashing noise and the lights go out in all of the houses in the village.

Q: Why did the lights go out?

2 points reference to the lightning hitting a tree which fell onto a power line and cut the electricity (the lightning hit a tree which crashed into the electricity wires)

1 point reference to lightning hitting power lines

0 points reference to irrelevant or incorrect factors

It is a very cold winter and has been snowing for days and days. The snow has covered everything; the trees, the houses, the hilltops, even the fences are covered in a thick layer of snow. Everything looks completely white apart from the dull grey sky. One morning, the skies are blue and the sun comes out. The sun beats down on the houses, the trees, the hilltops, and the fences. Puddles start to form at the edges of the fields.

Q: Why are there lots of puddles?

2 points reference to the snow melting or the effect of the sun on the snow (the sun makes the snow turn into water)

1 point reference to the snow without reference to melting or the sun (because of the snow)

0 points reference to irrelevant or incorrect factors

The little village of Littlehurst is close to the river Worrow. A year ago, a wall was built all the way round the edge of the village. The river floods its banks in April every year and, in the past, water would flow into many houses and cause lots of damage. For three weeks now the rain has been pouring down. However, this year, all the houses in Littlehurst are perfectly dry inside.

Q: Why were all the houses dry?

2 points reference to the wall stopping the water entering the village

1 point reference either to the wall or to the water not getting in but no connection between the two

0 points reference to irrelevant or incorrect factors

The summer has been long and very warm, just the right conditions for producing lots of apples. All summer long the orchard has been quiet and peaceful. Now, at the end of summer, the apples hang from the trees, glistening in the bright sun, all ripe and rosy. And every now and then in the orchard, little thumps can be heard.

Q: Why are there little thumps?

2 points reference to the apples falling from the trees or hitting the ground

1 point reference to the apples without mention of them falling or hitting the ground

0 points reference to irrelevant or incorrect factors

One of the huge parks in the middle of London has stone statues all around the edge of it. It also has lots of trees, which drop their leaves in autumn every year. On a cold, dry morning in November, a huge bonfire is burning all the leaves in one corner of the park and the statues in that corner of the park can't be seen. But in the afternoon, it is clear that they are still there.

Q: What had happened in the morning?

2 points reference to the smoke from the fire covering the statues

1 point reference to the bonfire or smoke without explaining how this affected the statues

0 points reference to irrelevant or incorrect factors including incorrect reference to the fire (it burnt down the statues)

It is late in April and the sky has been clear and blue all morning. An empty fountain sparkles in the middle of the park. As the day goes on, the sun occasionally disappears behind little white fluffy clouds, soon to appear again on the other side. However in the late afternoon, the sky becomes dark and filled with lots of grey clouds. A little after this, the fountain starts to spout water.

Q: Why did the fountain spout water?

2 points reference to the rain filling up the empty fountain

1 point reference to either to rain or to the fountain being filled up

0 points reference to irrelevant or incorrect factors

Iceland is a country where earthquakes often occur. They happen very suddenly when big rocks under the ground suddenly move, making the ground shake. One day last year, the ground started to shake near a mountain in the south of Iceland. As the ground shook more and more, a large cloud of smoke appeared above the mountain and huge flames roared from the mountain top.

Q: Why did this happen?

2 points reference to a volcano or to an earthquake starting a volcano (reference to larva or similar is also fine)

1 point reference to the earthquake (because the ground under mountain was skaking)

0 points reference to irrelevant or incorrect factors

Unlinked sentences

The two countries had been at war. A housewife is about to enter the super-market. Today he is going to buy an expensive new stereo. Mrs. Brown, the post-mistress, receives a special parcel. Mrs. Pearson wouldn't harm a fly. Mary's birthday is in February. Late one evening the old man was watching television.

Q: When is Mary's birthday?

2 points February

0 points Anything else

Young Simon is very robust. She sees that Fred cannot play. Jeremy is always laughing. Ruth sees her uncle very often, but today he has gone to Brazil. Richard is packing up to go away. Today, at college, it is Jim's worst lecture - statistical mechanics. She has only one dollar left, which she must keep for her bus fare. He buys a bright tie, to go with his new shirt.

Q: How many dollars does she have left?

2 points One

0 points Anything else

Simon takes the special butter from the refrigerator. Each boxer has won several fights. He skillfully picks out the imperfect items. They are either in Boston or in New York. She has to cut the grass and find somewhere to plant the bay tree. The conductor sees that the cellist has broken a string. Tracy took the bus to the station.

Q: Where did Tracy take the bus to?

2 points The station

1 point Anything that seems part way there (eg. the bus station)

0 points Anything else

The four brothers stood aside to make room for their sister, Stella. Gill repeated the experiment, several times. The name of the airport has changed. Louise uncorked a little bottle of oil. The two children had to abandon their daily walk. She took a suite in a grand hotel. It was already twenty years since the operation.

Q: Who abandoned their daily walk?

- 2 points The two children
- 1 point Anything that seems part way there (eg. the children)
- 0 points Anything else

One day Uncle Simon came to visit Alex. The first part of the performance had come to an end. He put away the letter and stuck his hands in his pockets. She was still holding her umbrella. The cats ran back to the boy. Flora came into the middle of the square. The little island had a high rocky shoreline.

Q: Where did Flora go to?

- 2 points The middle of the square
- 1 point Anything that seems part way there (eg. the square)
- 0 points Anything else

At the edge of the road a little grass was growing. He reaches out to find the light switch. A sailor who has just left his ship is walking to the town. She has to decide where to keep the pasta. At last daylight came, and Tommy got out of bed to open his presents. Jim knows all about investing money, as he works in a large bank. They exchanged a few brief words about the weather.

Q: Why did Tommy get out of bed?

- 2 points To open his presents
- 1 point Anything that seems part way there (eg. coz daylight came/it was morning)
- 0 points Anything else

She is always saying that someone will eventually find the treasure. Everyone is allowed two visits and no more. At the psychiatry department they were interviewing the new nurses. Jim will win the first race of the meeting. She has taken all the children to visit the zoo today. Simon's uncle is wearing a new suit. The same phrase of twenty three notes recurred throughout.

Q: What will Jim win?

- 2 points The first race of the meeting/the first race
- 1 point Anything that seems part way there (eg. the race)
- 0 points Anything else

He needs a new engine for his old car. The prize is an immediate lump sum of \$20,000 tax-free. Japan is stronger than Italy in economic terms. The mother is very brave and long suffering. The new book is about statistics and experimental design, and contains many graphs. The front room contained a little bird in a cage. Although Jim is only twenty one years old, he has an income of \$20,000 per year. There are not many people this evening in the large rectangular dining room.

Q: Who is brave and long suffering?

- 2 points The mother
- 0 points Anything else

