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Executive Function and Autistic Symptomatology in Very Young Children

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submitted for the degree of PhD

University of Durham

Department of Psychology

2001

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Acknowledgements

Firstly I would like to thank the children, families and clinicians based in the North-East of England without whom this project would never have begun. Thanks also go to Michelle Turner and Ann Le Couteur for their valuable guidance and advice, to Robert Drewett for statistical advice, to mum and dad for picking me up when the going got really tough, to Helen McConachie for encouraging me to stick it out and her knack of saying the right thing at the right time and, of course, to James whose continual support helped make sure the going wasn't too tough too often!

Abstract

The Executive Function hypothesis proposes that the behavioural aspects of autism can be explained by impairment in executive function skills. The term 'executive function' refers to several cognitive skills including inhibitory control, generation of novel responses, working memory and planning. Many studies have demonstrated that school-aged children, adolescents and adults with autistic spectrum disorders are impaired on tasks designed to measure these skills (Pennington & Ozonoff, 1996). However, despite the early onset of this developmental disorder, little work has explored the executive functioning of pre-school children with autism. In a developmental context, the executive function hypothesis would predict early executive deficits for children with autistic spectrum disorders, an association between executive function skill and symptom severity, and a relationship between the developmental trajectories of executive function and behavioural profiles.

The present study recruited three-year-old children referred for autistic spectrum disorders or speech and language delay. A battery of executive function tasks measuring inhibition, working memory and planning was administered on recruitment and one year later. Detailed behavioural information was also gathered at both time points.

Cross-sectional group comparisons revealed little evidence for an executive deficit in children with autism at either age. Similarly, there were no reliable relationships between executive performance and symptomatology. At a

group level behavioural scores appeared to change little over the year. However, within each group there was evidence for both positive and negative change. Moreover, executive function performance did not change reliably across individuals over the year, nor was there any consistent relationship for individual children between performance on one executive function task and one behavioural domain. These findings are discussed in relation to the psychological theories of autism, and implications for intervention approaches are considered.

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Chapter 1

An Introduction to Autistic Spectrum Disorders and the Executive Function Construct

Autism is a behaviourally defined developmental disorder characterised by social and communicative impairments and repetitive behaviours. The primary cause of autistic spectrum disorders is as yet unknown. Explanatory hypotheses have been constructed at the genetic, neurobiological and neuropsychological levels but no individual hypothesis has yet stood up to rigorous evaluation. Given the developmental nature of these disorders, research following the maturation of very young children with autism is likely to provide crucial information. Unfortunately, thus far little research of this nature has been conducted.

There are currently three major neuropsychological accounts of autistic spectrum disorders: Theory of Mind, Central Coherence and Executive Function. Executive function is an umbrella term referring to specific cognitive skills implicated in goal-directed, problem solving behaviour. Over the last 15 years, a large body of experimental evidence has reported executive dysfunction in both children and adults with autism. This evidence has lead to the development of the executive dysfunction hypothesis of autism that proposes impairment in these skills plays an important role in the development of autistic symptomatology, in particular repetitive behaviours. Despite the influence of this hypothesis, relatively little work has explored the executive function skill of young children with autism.

This thesis sets out to examine the Executive Function hypothesis in relation to the cognitive and behavioural presentation of very young children with autistic spectrum disorders. In addition, a longitudinal component is to be included

whereby the development of both executive function skills and repetitive symptomatology can begin to be explored. It is beyond the scope of the current thesis to describe or evaluate the other accounts of autism.

The first chapter will describe the clinical picture of autism and then provide an introduction to the executive function construct. This chapter will focus on executive function skill in young children with and without autism and adopts a plausible developmental path to provide the framework for the literature review. The second chapter discusses the executive function hypothesis of autism in more detail and builds towards the specific aims of the thesis. Chapter Three details the experimental work concerning executive function skill in preschoolers with autistic spectrum disorders and Chapter Four presents repetitive behaviour information collected on these children. Chapter Five will consider the relationship between executive function task performance and repetitive behaviour. Finally, Chapter Six provides a discussion of all the work carried out and concludes the thesis.

1.1 Autism: The Clinical Phenomena

1.1.1 Behavioural Presentation

Autism is a developmental disorder independently recognized by Leo Kanner (Kanner, 1943) and Hans Asperger (Asperger, 1944). These early reports of autism described individuals for whom social and communication skills proved problematic. An influential epidemiological study in Camberwell, London, (Wing & Gould, 1979) identified a distinct group of children between the ages of 0 and 14 years of age characterised by the presence of three impairments: poor communication skills, poor social understanding and a lack of imagination. These impairments have become known as the triad of impairments in autism.

Social impairment is often identified by an individual's difficulty in forming and maintaining social relationships. Typically, an individual with autism may fail to appreciate the reciprocal nature of social relationships and be apparently unable

to understand and follow social rules (e.g. Lord, Storoschuk, Rutter & Pickles, 1993; VanMeter, Fein, Morris, Waterhouse & Allen, 1997).

Verbal and non-verbal communication is characterised by delay and deviance. Most individuals with autism are profoundly delayed in their acquisition of language and many never achieve spoken language. Deviance in the understanding and use of eye gaze and gesture as communicative functions is common (e.g. Camaioni, Perucchini, Muratori & Milone, 1997; Leekam, Hunnisett & Moore, 1998a; Leekam, Lopez & Moore, 2000; Stone, Ousley, Yoder, et al, 1997). For example, children with autism rarely or never point to a distant object in order to show it to another although they may learn to point to an object that they need. Even in individuals who do develop language skills, communicative efforts are often marked by pragmatic difficulties, pronoun reversal, echolalia and stereotyped language (e.g. Baron-Cohen, 1988; Eales, 1993).

The lack of imagination noted by Wing and Gould can be most clearly seen in the development of play skills. In particular, the distinction of manipulative and functional play from symbolic or more imaginative play is important. Whilst functional play may or may not be impaired (Baron-Cohen, 1987; Jarrold, Boucher, & Smith, 1996; Lewis & Boucher, 1991; Sigman, Ungerer, Mundy, & Sherman, 1986) individuals reliably show deficits in the capacity to pretend (Atlas, 1990; Baron-Cohen, 1987; Jarrold et al, 1996; Libby, Powell, Messer, & Jordan, 1995; Sigman & Ungerer, 1984; Wing, 1978; Wing, Gould, Yates, & Brierly, 1977).

Although the quantity of social and communicative interaction may be reduced in autism, the cardinal descriptions of this disorder refer to the *quality* of behaviour rather than quantity. An individual with autism may make as many bids for communication as another individual but these bids may take an inappropriate form (e.g. Capps, Kehres & Sigman, 1988). This distinction is often referred to as the delay versus deviance distinction. For a diagnosis of autistic spectrum disorders, deviant behaviour must be observed; delayed behaviours alone are not sufficient to warrant this diagnosis.

A restricted repertoire of activities and interests is essential for a diagnosis of autism (e.g. American Psychiatric Association, 1994; Kanner, 1943; Turner, 1999b; World Health Organisation, 1992). Stereotyped movements (such as hand flapping or finger twisting) or manipulation of objects (such as spinning the wheels on a car repeatedly or lining toys across the room) are regularly reported, as is insistence on sameness in routine or surroundings (such as taking the same route to the shops everyday or showing distress when furniture is rearranged). Unusual reactions to sensory stimuli can also exist (such as marked negative reaction to their mother singing – even when the singing is acceptable to other people!). In some cases the restricted quality of behaviour becomes apparent through a circumscribed interest (such as a fascination with lampposts or gratings). The interest is often marked by a selective and exclusive focus (for example a fascination in train timetables together with a complete disinterest in locomotives).

Much research uses autistic spectrum disorders as a categorical label to describe a single group of individuals, however there is an increasing body of evidence to support marked heterogeneity within the label (e.g. Le Couteur, Bailey, Goode et al, 1996) and within the broader description of the autistic phenotype (Bailey, Palferman, Heavey, & Le Couteur, 1998). Although this is often masked by the need to use consistent diagnostic labels, the developmental approach taken by this thesis has the potential to offer fascinating descriptions of individual profiles of development within the autistic spectrum.

1.1.2 Identification and Diagnosis of Autistic Spectrum Disorders in Young Children

Historically, there has been some disagreement over the precise behavioural criteria for a diagnosis of autism. However, the recent ICD-10 (World Health Organisation, 1992) and DSM-IV (American Psychiatric Association, 1994) diagnostic criteria have agreed upon the presence of at least six symptoms across the three domains of social interaction, communication and repetitive behaviours (at least two of which must occur in the social domain). Additionally, evidence of delay or deviance must be apparent before the age of three years in at least one area (social interaction, language as used in social

communication or symbolic and functional play). Current clinical and research practice is to use the term autistic spectrum disorders (ASD) to refer to a continuum of disorders associated with social and communicative deficits including core autism, PDDNOS, Asperger Syndrome, childhood disintegrative disorder and atypical autism. Although the exact nature of the relationship between these disorders is debated (Freeman, 1997; Frith, 1989; Leekam, Libby, Wing, Gould, & Gillberg, 2000; Lord, Risi, Lambrecht et al, 2000; Miller & Ozonoff, 2000; Ozonoff, Rogers, & Pennington, 1991b; Prior, Eisenmajer, Leekam, et al, 1998; Robertson, Tanguay, & L'Ecuyer, 1999; Szatmari, Bartolucci, & Bremner, 1989; Szatmari, Tuff, Finlayson, & Bartolucci, 1990; Wing, 1981) the main issue seems to be whether a categorical or a dimensional approach best suits the disorder.

A diagnosis of autistic spectrum disorders is usually based upon a combination of a developmental history and direct observation provided by a multidisciplinary team of specialists (including parents) across a variety of settings. Standardised diagnostic instruments also play a vital role in maintaining the validity and reliability of diagnostic decisions. At present, most children in the UK receive a clinical diagnosis of autism around the age of five years, and of Asperger's around 11 years (Howlin & Moore, 1997). However, in many cases parents have reported concerns about their child several years earlier (e.g. De Giacomo & Fombonne, 1998; Howlin & Asgharian, 1999; Vostanis, Smith, Corbett, et al, 1998). It is clear that ASD can be identified at younger ages than it is currently diagnosed; indeed the diagnostic criteria require that behavioural abnormalities are present before the age of three years.

A growing body of evidence supports diagnostic decisions made before the age of 5 years. Retrospective coding of home video recordings has highlighted reliable behavioural characteristics in very young children who go on to receive a diagnosis of autism (Dahlgren & Gillberg, 1989a; Gillberg, Ehlers, Schaumann, et al, 1990; Maestro, Casella, Milone, Muratori, & Palacio-Espasa, 1999; Osterling & Dawson, 1994). A review concluded that autism may be screened for around 18 months and reliably diagnosed clinically by 30 months (Gillberg, Nordin, & Ehlers, 1996). Accurate early identification of autism is

clearly important. For example, children identified at young ages may benefit substantially from early treatment programmes (Dawson & Osterling, 1997; Hoyson, Jamieson, & Strain, 1984; Jordan, Jones, & Murray, 1998; Lovaas, 1987; Ozonoff & Cathcart, 1998; Rogers, 1998; Rogers & Lewis, 1989; Sheinkopf & Siegel, 1998).

Clinical judgement appears to be more reliable at diagnosing ASD in children under three years old than standardized developmental history instruments. In differentiating autistic spectrum disorders from non-spectrum diagnoses, one study reported inter-clinician reliability of 88% for decisions at two years old and an 80% agreement on diagnosis at two and three years old. Discriminations between autism and pervasive developmental disorder not otherwise specified were less reliable and stable (Stone, Lee, Ashford, et al, 1999).

The Autism Diagnostic Interview Revised (ADI-R) is a standardized investigator-based interview yielding a detailed developmental history of the child that can be coded and subjected to a statistically derived diagnostic algorithm (Le Couteur, Rutter, Lord, et al., 1989; Lord, Rutter, & Le Couteur, 1994). Lord (1995) reported that this instrument, when used in isolation, both over- and under-diagnosed autism at the age of 2 years compared to clinical diagnoses made 12 to 15 months later. This study suggested that expert clinical decisions are most reliable for differentiating autism from speech and language delay at two years old, and that a combination of the ADI-R and clinical judgment is best at three years old. It should be noted that the Lord study made differential diagnoses between autism and speech and language delay whilst Stone et al (1999) compared autistic spectrum disorders and non-autistic spectrum disorders. Since it seems likely that differentiating two groups of children who both have communication difficulties is the more difficult task, this may explain the slightly reduced success of the ADI-R at the age of 2 years (Lord, 1995).

Other work has also demonstrated the value of administering the ADI-R when diagnosing young children. The ADI-R has been shown to reliably differentiate children with autism from developmental delay when the child has a mental age

of over 18 months (Lord et al, 1993; Pilowsky, Yirmiya, Shulman, & Dover, 1998; Sigman & Ruskin, 1999). The combination of standardised assessment and clinical judgment has been shown to lead to sensitive clinical diagnoses of autism that are reliable over a period of up to 8 years (Cox, Klein, Charman, et al, 1999; Sigman & Ruskin, 1999). In the Cox et al study, the ADI-R demonstrated good specificity but poor sensitivity at 20 months and good stability from 20 to 42 months. Discrimination between autistic spectrum and non-autistic spectrum disorders was particularly good, but both clinical and ADI-R diagnoses of pervasive developmental disorders and Asperger's syndrome lacked sensitivity at 20 months.

The Autism Diagnostic Observation Schedule – Generic (ADOS-G) is a standardized instrument designed to elicit socio-communicative behaviours that can be reliably coded to assist diagnoses of autistic spectrum disorder (DiLavore, Lord & Rutter, 1995; Lord et al, 2000). Although good agreement has been reported between parental reports of behaviour and direct observation (Stone, Hoffman, Lewis & Ousley, 1994) a combination of information sources is crucial for a diagnosis of autism in very young children.

In summary, expert clinical judgement based upon information from several sources, and combining clinical and standardised assessments involving developmental history and direct observation across multiple settings, seems to form the most reliable and stable basis for an early diagnosis. Preschool categorical diagnoses are best when differentiating autistic spectrum disorders from non-spectrum diagnoses since the clearest markers are severity rather than quality of delay (Lord et al, 2000). However it is possible to make diagnostic decisions between autistic spectrum disorder and speech and language delay or developmental delay at this young age (Lord, 1995; Lord et al, 1993).

1.1.3 Epidemiology of Autistic Spectrum Disorders

Core autism is a relatively rare, well validated, child psychiatric disorder (Bailey, Phillips, & Rutter, 1996). Reported prevalence rates have varied substantially according to the precise diagnostic criteria applied (e.g. Bryson, Clark, & Smith, 1988; Gillberg, Steffenburg, & Schaumann, 1991b). A recent

estimate of prevalence in children referred to clinical services between the ages of 2 and 6 years was 16.8 per 10,000 for autistic spectrum disorders and 45.8 per 10,000 for other pervasive developmental disorders (Chakrabarti & Fombonne, 2001). Gillberg and Coleman (2000) reported similar rates. Other estimates have varied from 2-5 per 10, 000 for core autism (Cialdella & Mamelle, 1989; Gillberg & Gillberg, 1989) and as over 20 per 10, 000 for autistic spectrum disorders (Gillberg & Coleman, 1992; Gillberg & Gillberg, 1989; Kadesjö, Gillberg & Hagberg, 1999; Wing & Gould, 1979).

A striking sex difference in prevalence rates for autistic spectrum disorders exists; males are more likely to be affected by autistic spectrum disorders than females. The male: female ratio affected by core autism is about 4:1 (Gillberg & Coleman, 1992) whilst the male excess appears to increase in the normal range of ability and reports of ratios as high as 10:1 have been made (Gillberg et al, 1991a; Wing & Gould, 1979).

Autism can occur at all ability levels. Although some 70% of people diagnosed with autism are described as learning disabled and have intelligence quotients below 70 (Gillberg, Steffenberg, Wahlstrom, et al, 1991a; Rutter, 1979; Wing & Gould, 1979), some autistic individuals have exceptionally high ability levels. Similarly, almost 50% of individuals with autism show no useful speech at the age of five years indicating a poor outcome (Lord & Schopler, 1989a; Nordin & Gillberg, 1998; Venter, Lord, & Schopler, 1992), whilst other individuals can be very loquacious. However, even in persons with moderate language skills the quality of communication is often stilted and limited.

The heterogeneity of ability and communicative skill in autism is also reflected in repetitive behaviour. This is an important aspect of autistic spectrum disorders and has yet to be adequately acknowledged in experimental work. Statistical procedures require sample groups of a notable size and therefore the inter-group variation in cognitive or behavioural variables is rarely explored.

1.1.4 Levels of Explanation

Authors within many disciplines have addressed the possible causes of an autistic disorder. These include genetic, neurobiological and neuropsychological accounts. So far no single level of explanation has provided a satisfactory account of autism. In fact, the complex nature of the disorder strongly suggests a multidimensional approach would be most productive (e.g. Bailey et al, 1996; Minshew, Goldstein & Siegal, 1997). Even within the neuropsychological discipline there is good reason to think that one single account is unlikely to explain those aspects of autism that are explicable in terms of neuropsychology.

> "*Each [psychological] hypothesis addresses part of the puzzle of autism, none would claim to have the complete story, and it may be that autism is the result of abnormalities in the development of several distinct systems*" (Happé, 2000, p.203).

Although an integrated approach may be the best way to explain autism, the elements of any multi-disciplinary account must be rigorously examined and evaluated. Similarly, within the neuropsychological discipline the competing theories of autism need to be individually examined and assessed before a sound integrative approach can be taken. This thesis concentrates on one account of autism within the neuropsychological discipline. The remainder of this first chapter introduces the executive function construct that underlies the executive function hypothesis of autism.

1.2 Executive Function

The so-called 'executive function' skills involved in problem solving and goal directed behaviour have been studied over the last fifty years (e.g. Luria, 1966; Piaget, 1954). Whilst uncertainty remains about the precise nature of the component skills, the general consensus is that executive function skills facilitate future, goal-directed behaviour, by allowing for planning, flexible strategy employment, impulse control and organized search relatively independent of IQ (e.g. Duncan, 1986; Welsh, Pennington, & Groisser, 1991). Key behaviours include the ability to shift from one concept to another; the

ability to modify behaviour in response to new or modified information about task demands; the ability to synthesise and integrate isolated details into a coherent whole; the ability to manage multiple sources of information; and the ability to make use of relevant acquired knowledge (e.g. Stuss & Benson, 1987).

The executive function construct was coined to represent the separate cognitive factors that are implicated in everyday problem solving. Separation of individual skills may facilitate the understanding of component skills and in turn allow reflection about how they may interact in the pursuit of a problem solution.

Many authors have favoured this fractionation approach to executive function. Fuster (1985) distinguished three components: future oriented planning, retrospective working memory and interference control. More recently, Welsh and colleagues (Welsh et al, 1991) also proposed a three-factor solution of executive function reflecting fluid and speeded responding, hypothesis testing and planning. Similarly, Hughes (1998a) implicated working memory, attentional flexibility and inhibitory control as the major components of executive function. The terms lack theoretical precision (see also Pennington et al, 1997). For example, the term attentional flexibility invokes inhibitory and generative processes whilst problem solving involves error detection, inhibition and shifting of action. Furthermore, the difficulty in identifying executive demands of an experimental paradigm *a priori* (Burgess, 1997) has hindered the careful exploration of individual component skills.

Conversely, Fodor has proposed that executive function skills do not fractionate (e.g. Fodor, 1983). More recently, his premise has been reworked to focus upon the *interaction* between several different skills in order to solve a problem. For example, it is proposed that the probability of making an erroneous prepotent response depends upon the interaction of working memory and the strength of prepotency (Rogers, Bertus & Gilbert, 1994).

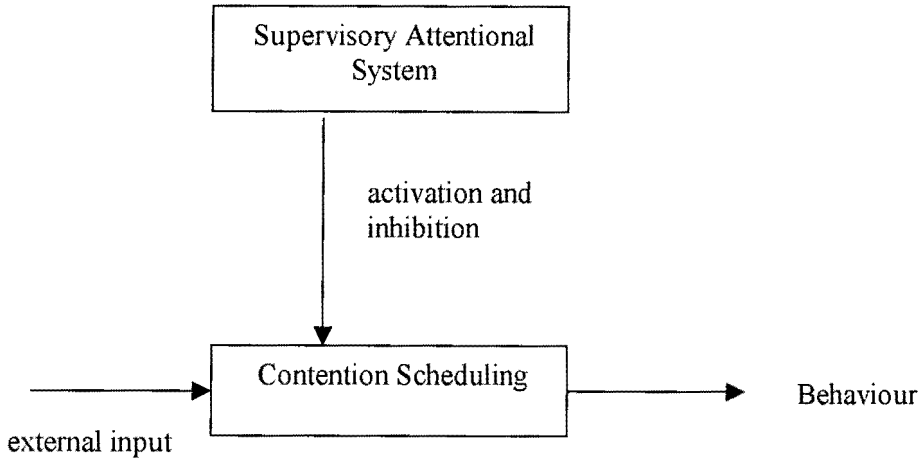
In fact, most current research considers executive function to comprise separable yet integrated skills. The Supervisory Attentional System model (Norman & Shallice, 1986; Shallice, 1988, Shallice, 1994; Shallice & Burgess, 1991) describes a system where the **supervisory attentional system** (SAS) provides

top-down control over the process of action selection. The model assumes that behaviour can be divided into two streams: one controlled by conscious processes (SAS) and the other driven by automatic processes in the absence of conscious control (**contention scheduling**). The same behaviour or action could occur as a result of either system, the difference lies in the exercise of conscious control. For example, a driver might turn the car steering wheel 'automatically' when approaching a bend and make the same turning action 'consciously' when choosing to pull out of the road. In other words, the role of the SAS is to form representations of goal states and to monitor the planning of actions in order to achieve these goals. The contention scheduling process is responsible for carrying out actions that have been initiated by external stimuli or the SAS. In this way, the two streams of behaviour are separable yet integrated.

The model predicts that a malfunction in the SAS would lead to a failure of inhibitory control or a failure to generate appropriate behaviour (See Jarrold, 1997, for more detailed discussion of this theory). An external stimulus may 'automatically' trigger an action but if this behaviour is not appropriate the SAS must inhibit the activation levels of the schema. Without this top-down processing, inhibitory control would fail. In addition, the SAS is used to generate action when there is no external trigger. A malfunction in the SAS would lead to little or no generation of activity. In this way, inhibition and generation of behaviour can be seen as separable yet integrated executive function skills. Figure 1.1 presents a simple version of the SAS model of executive control.

The SAS model is not the only theory that has postulated a close relationship between inhibition of response and generation of response. Luria (1966) had already identified two ways in which inhibitory control could break down: the compulsive repetition of a behaviour (failure of inhibition) and inertia of program (failure of generation). Inhibition of response and generation of a novel response can be seen as two sides of the same coin: a response must be generated before it can be inhibited, and in turn an ongoing action must be inhibited before a new response can be generated.

Figure 1.1 The SAS model of executive control (adapted from Jarrold, 1997).



Based upon work by other authors who have previously argued for different levels of inhibitory control (e.g. Freeman & Gathercole, 1966; Luria, 1965; Sandson & Albert, 1984) the current thesis proposes inhibitory control can be described at three levels. By distinguishing types of inhibition we can hope to understand more fully the nature of executive control and the associated perseverative errors when the control fails. This thesis does not set out to rigorously evaluate the developmental progression of individual aspects of executive control, but it does adopt the three levels as a framework for the literature. The first level is simple inhibition of response (motor or verbal) where the executive processes require simply the cessation of a response which has been activated. The second level concerns the simultaneous (or near-simultaneous) inhibition of one response and the implementation of another. The third level requires the ability to flexibly shift from one conceptual set to another where correct responding according to the set is not directly associated with one specific response

It also seems likely that generativity must be considered at different levels, although there has so far been little literature concerned with this (see Turner, 1999a; Turner, 1999b for exceptions). Turner (1999a; 1999b) describes at least two types of generation: selection and implementation of an appropriate strategy (for example, trawling one's lexicon to identify words beginning with the same letter) and the generation of a novel, unconventional response (for example,

generating an original idea for how to use an object). There is no obvious developmental pathway between these two levels. The acquisition of a repertoire of strategies to adapt and apply for generating responses may be more cognitively complex than generation of novel responses. Alternatively, it may be that responses that are not instigated by a common strategy make more complex demands on cognitive processing. For this reason no developmental pathway for generation is proposed in this thesis.

1.3 A Common Biological Basis for Executive Function and Autistic Spectrum Disorders?

Observations of 'higher-order' cognitive deficits that reflect failure of executive control in patients with damaged frontal lobes (Milner, 1963; Milner, 1964; Passler, Issac & Hynd, 1985; Stuss & Benson, 1983) have implicated this region as the seat of executive function skills (e.g. Cohen & Servan-Schreiber, 1992; Goldman-Rakic, 1987; Jones-Gotman & Milner, 1977; Kagan, Rosman, Day, Albert, & Phillips, 1964; Milner, 1964; Shallice, 1988; Shue & Douglas, 1992; Welsh, Pennington, Ozonoff, Rouse, & McCabe, 1990; see also Pennington & Ozonoff, 1996 for a review and Benton, 1991a; Luria, 1973; Parkin, 1996; Stuss & Benson, 1986).

Prefrontal cortex has been implicated in inhibitory control (e.g. Ciesielski & Harris, 1997; Diamond, 1991a; Diamond, 1991b; Incisa Della Rochetta & Milner, 1993), as the basis for goal planning (e.g. Luria et al, 1967; Teuber, 1967) and in rigidity of behaviour (Stuss & Benson, 1983). Given the developmental progression of executive control that is discussed in more detail elsewhere (section 1.4), the fact that prefrontal cortex matures relatively late in development and may not be completely mature until age 7 (Luria, 1966) or even age 12 (Golden, 1981 cited in Passler, 1985) further implicates this cortical region in executive control.

There is also evidence for abnormality or dysfunction in the frontal regions in autistic spectrum disorders. Twenty years ago, Damasio and Maurer (Damasio & Maurer, 1978) hypothesised that the mesolimbic cortex of the medial frontal

lobes was dysfunctional in autism. More recent studies using SPECT and PET techniques have shown reduced blood flow to frontal areas in autism (George, Costa, Kouris, et al, 1992; Pennington & Ozonoff, 1996).

The possibility of abnormal frontal cortex in autism has also been considered in a developmental context. A SPECT scanning study of five autistic children at the age of three to four years and then again three years later described a delayed maturation of activity in the prefrontal cortex at first that was normal by the second scan (Zilbovicius, Garreau, Samson, et al, 1995). This study concurs with others reporting disordered development of the frontal lobes in autism (Bailey, 1993; Bishop, 1992; Piven, Berthier, Startstein, et al, 1990b).

The frontal cortex is not the only brain area to be implicated in autism. Some studies have indicated abnormalities in the medial temporal lobe (Bachevalier, 1994; Bauman & Kemper, 1985; DeLong, 1992) and in cerebellar and neocerebellar areas (Courchesne, 1995; Courchesne, 1991; Courchesne, Hesselink, Jernigan, & Yeung-Courchesne, 1987; Courchesne, Yeung-Courchesne, Press, et al, 1988; Gaffney, Kuperman, Tsai, & Minchin, 1989; Gaffney, Tsai, Kuperman, & Minchin, 1987; Hashimoto, Tayama, Murakawa, et al, 1995). However CT and MRI scanning techniques have not reported structural changes in the medial temporal lobe (Bailey et al, 1996; Courchesne, Press, & Yeung-Courchesne, 1993; Saitoh, Courchesne, Egaas, et al, 1995) and cerebellar abnormalities have not been replicated (e.g. Ekman, de Chateau, Marions, et al, 1991, and Minshew et al, 1999).

An argument has been made that cerebellar deficits correspond to attentional deficits seen in autism (Courchesne, Townsend, & Saitoh, 1994a) and that dysregulation of this system may have implications for the prefrontal regions of medial temporal and fronto-striatal cortex (Robbins, 1997; Robbins & Everitt, 1995). In support of this idea, Diamond (2000) proposed a neural circuit linking the prefrontal cortex and cerebellum in executive function development (Diamond, 2000). It is possible that this circuit is disrupted in autism. Diamond summarises,

> *"the cerebellum and prefrontal cortex participate as critical parts of a neural circuit that is important when (1) a cognitive task is difficult as opposed to easy, (2) a cognitive task is new as opposed to familiar and practiced, (3) conditions of the cognitive task change, as opposed to when they remain stable and predictable, (4) a quick response is required, as opposed to longer response latencies being acceptable, and (5) one must concentrate instead of being able to operate on 'automatic pilot'."* (Diamond, 2000, p.45).

One study of young children with autistic spectrum disorders reported a stronger relationship between socio-communicative measures and a task purported to tap the medial temporal lobe rather than the dorsolateral prefrontal cortex (Dawson, Meltzoff, Osterling, & Rinaldi, 1998). The methodological approach to this study is interesting because the correlational relationship between performance on cognitive tasks and socio-communicative measures was used to further the neurobiological account. This approach requires significant assumptions about the relationship between cortical regions and task performance and therefore the conclusions must be cautious. However, using correlational techniques to assess the possible contribution of certain cognitive skills to behavioural presentation could be valuable to the understanding of autism. Although the research is not conclusive, it does seem that the frontal lobes may be disordered in autistic spectrum disorders and are important in the development of executive function skills.

1.4 Nature and Timing of Developments in Executive Function

Converging evidence from neuropsychological and cognitive studies has confirmed the developmental nature of executive function skill (Passler, Isaac, & Hynd, 1985; Welsh & Pennington, 1988; Welsh et al, 1991). Luria (1959, cited in Passler et al, 1985) observed developmental progressions in the ability to control motor responses between the age of 3 and 4½ years that lagged behind the ability to respond verbally. He (Luria, 1966) concluded that the prefrontal regions of the cortex do not mature until between 4 and 7 years old.

Passler and colleagues (1985) administered a battery of verbal and non-verbal inhibition tasks to children between the ages of 5 and 12 years and reported a multi-stage developmental process throughout this age range. Similarly, after assessing children aged 3 to 12 years old on a battery of tasks, Welsh and colleagues (1991) observed that adult-level performance was achieved at different ages for different subsets of the tasks. For example, organized and planful behaviour was detected as early as 6 years old but had not reached adult levels by the age of 12 years. Both these studies relied heavily upon tasks that had originally been developed for use in adult neuropsychology, however the review by Welsh and Pennington (1988) argues strongly for the development of tasks appropriate for children and the recognition of partially separable executive skills.

Inhibition and generation are skills that have been explored in some detail by authors. Working memory is identified as a separable skill by others who have argued for the fractionation of the executive function construct (Fuster, 1985; Hughes, 1998a). Planning can be viewed as an executive function skill that provides a prime example of the interaction between component skills in the pursuit of a solution but has also been identified as a component of executive function in its own right (Fuster, 1985; Welsh et al., 1991). Following from these preliminary, cross-sectional, studies of the developmental trajectories of executive function skill the current thesis focuses upon the development of these skills in very young children. As such, the following literature review concentrates upon the developmental trajectories of inhibition, generation, working memory and planning skills in both typically developing children and children with autistic spectrum disorders.

1.5 The Development of Inhibitory Control

1.5.1 Inhibition of a response

As inhibition of one response without the implementation of another would seem to represent the most basic level of inhibitory control it is perhaps surprising that few tasks have directly assessed the ability of young children to stop a response. Most tasks have instead focussed on the ability to implement an alternative response whilst inhibiting a previous or particularly salient response.

Typical development

Indirect evidence to suggest response inhibition emerges in infancy and early childhood comes from the child's ability to exercise self-control. The typical developmental path is that children are able to delay gratification (i.e. a reward) for longer periods of time when there is a perceived benefit (i.e. more rewards!) as they develop through infancy and early childhood (Golden, Montare, & Bridget, 1977; Lee, Vaughn, & Kopp, 1983; Vaughn, Kopp, & Krakow, 1984; Zelazo & Reznick, 1991). The strategies used by children to control their own behaviour also become more sophisticated with age (Mischel & Mischel, 1983) and individual differences in the desire to delay gratification become more apparent at the age of 30 months (Logue, Forzano, & Ackerman, 1996; Vaughn et al., 1984).

Development in autistic spectrum disorders

Response control has, however, been assessed in autistic spectrum disorders through two tasks developed from the information processing literature. In the first condition of the Go-NoGo task (Ozonoff et al, 1994) the participant is asked to press a button when a square is shown on a screen but has to withhold that response when a circle is displayed. A group of children with autism (mean age = 12 years) performed as well as normally developing children and clinical controls matched for age and ability. Similarly, high-functioning children with autistic spectrum disorders (mean age = 13 years, mean IQ = 100) were unimpaired, compared to typically developing children, in the ability to withhold a motor response when a tone was sounded during the Stop Task (Ozonoff & Strayer, 1997).

In contrast, one study has shown impairment in self-control for children with autism. Children aged nine to 17 years were given the opportunity to increase their reward of one sweet if they waited. Fewer children with autism chose to wait than children with moderate learning disability, and fewer of those who did wait chose to manage the delay by hiding the sweet (Hughes, 1996). This study indicates that children with autism were less likely to place themselves in a situation where they had to control their behaviour and were less likely to employ distraction techniques to facilitate self-control.

1.5.2 Inhibition-and-Implementation

Most tasks that have been used as inhibitory control tasks require inhibition of one response and implementation of an alternative response. Perseveration to the first response is interpreted as inhibitory control failure.

Typical development

By the age of about 12 months an infant is able to *inhibit* the prepotent response to reach directly towards a reward that is visible through a transparent box and can instead *implement* an indirect reach to the side of the box that enables retrieval of the reward (e.g. Diamond, 1988; Diamond & Goldman-Rakic, 1985; Diamond, Prevor, Callender & Druin, 1997). During the first year of life the infant is also able to retrieve a reward from one of two or more locations. However, when the object is then hidden in a different location (a reversal trial) the infant will tend to continue to reach incorrectly (perseverate) to the first location (e.g. Bremner, 1978; Diamond, 1985; Diamond et al, 1997; McEvoy, Rogers, & Pennington, 1993; Piaget, 1954; Wellman, Cross, & Bartsch, 1986; but see Sophian & Wellman, 1983 for a non-replication in nine-month-old infants). This failure to stop a previously correct response and implement a new response has become known as the A-not-B error.

Performance on A-not-B tasks improves between the ages of six and 12 months (see Marcovitch & Zelazo, 1999 and Wellman et al, 1986 for meta-analyses of A-not-B tasks). Typically, shorter delays between hiding and retrieval reduce errors, and infants can succeed at longer delays with increasing age (e.g. Diamond, 1985; Diamond, Cruttenden, & Neiderman, 1994; Diamond et al,

1997; Gratch, Appel, Evans, et al, 1974). At seven-and-a-half months of age a delay of less than two seconds will produce the A-not-B error, but by the age of 12 months the delay needs to be over 10 seconds (Diamond, 1985). It should be noted that this pattern of development implicates both inhibitory control and working memory in resisting the A-not-B error.

Distinguishing inhibitory (response) control problems from other executive or cognitive problems is difficult. For example errors may result from a child's lack of understanding of the rules they are expected to follow, or they may result from the working memory demands of the task rather than a failure of inhibitory control. However some authors have argued convincingly that children demonstrate inhibitory failure independently of other problems.

Firstly, children appear to understand simple sorting rules by the age of two-and-a-half years, but do not develop the response control required to execute a given sorting rule whilst inhibiting other responses until about six months later (Luria, 1961; Luria, 1982; Zelazo & Reznick, 1991; Zelazo, Reznick, & Pinon, 1995). These observations of performance on sorting tasks in different conditions have been used to argue that children as young as two-and-a-half-years old have the cognitive capacity to implement rules but have not yet developed sufficient motor response control to execute the rules. Additional evidence that implicates inhibitory control as the source of error comes from a report that incorrect perseveration in two-year-olds only occurs when they had successfully *actively* searched at the location. On a multi-location A-not-B style task, children who had simply seen the reward hidden in the first location did not perseverate to that place but children who had searched the first location did perseverate (Zelazo, Reznick, & Spinazzola, 1998)[2].

Similarly, an elegant study by Diamond and colleagues (Diamond et al, 1994) confirmed inhibitory control was independent from working memory in successful performance on a multiple location A-not-B task by presenting three conditions in which the covering of the locations was manipulated. This

[2] Children in the active condition received a reward during preswitch trials whilst children in the see-only condition did not. This may have adversely affected the motivation of the active children and therefore the results on which the authors base their conclusions.

provides a telling example of the interplay between executive skills even in tasks that appear simple, and also how careful analysis of the individual's response can help clarify component executive skills.

The ability to inhibit a response and simultaneously implement a conflicting response following explicit instruction continues to develop throughout childhood and adolescence. Stroop-like tasks measure an individual's ability to inhibit the conventional response and produce a conflicting response. For example, children would be asked to respond 'day' when presented with a picture of a moon and 'night' when presented with a sun (e.g. Diamond et al, 1997) or to tap twice when the experimenter taps once and to tap once when the experimenter taps twice (e.g. Passler et al, 1985). Common dependent variables of interest are the time taken to make a response and the number of errors made on the task. Cross-sectional studies have suggested that children become increasingly sophisticated on these measures of inhibitory control between the ages of 3½ and 10 years with a significant period of development between 6- and 8-years although the precise ages may differ across verbal and non-verbal domains (Diamond et al, 1997; Diamond & Taylor, 1996; Passler et al, 1985).

Task structure and presentation has a notable impact upon the nature of inhibitory demands made on the participant. The A-not-B and Stroop tasks provide the child with substantial information about the alternative response. In the A-not-B tasks the child watches the toy being hidden and in the Stroop tasks the child is told the required response for each type of stimuli. In contrast, the Spatial Reversal task provides the child with less supporting information. Like the A-not-B tasks, the child is required to find a reward hidden in one of two boxes. However, the placement of the reward in one box now occurs out of sight of the child. Therefore, on the first trial the child has no information about which box contains the reward on which to base their response: they must select a box to search. Following the first search, the child has more information: either they had picked the successful box or they had chosen the empty box. The reward is repeatedly hidden in the same box until the child makes several correct consecutive responses. At this point the reward is placed in the other box, unbeknownst to the child who continues to check the first box. Crucially, the

child must use the feedback from their unsuccessful search to realise the reward is now hidden in the other box. Then they have to inhibit the first response and implement the new response. Whilst the inhibitory demands placed upon the child are very similar to the A-not-B inhibition-and-implementation demands, the amount of available supporting information is reduced. Perhaps it is useful to make this distinction by describing performance on the Spatial Reversal task as *inhibition* of one response and *initiation* of a second.

The Spatial Reversal task was administered to 12 typically developing children with a mean chronological age of 38 months as part of a study of children with autism (McEvoy et al, 1993). Although the main focus was the performance of the children with autism, the authors did note that the typically developing children made 'few perseverative responses'. This study demonstrates that by the age of three years children can inhibit one response and initiate a second in a structured situation where explicit information is not forthcoming.

Development in autistic spectrum disorders

There are mixed findings about the early inhibitory control of children with autistic spectrum disorders. Many studies show that children with autism have inhibition-and-implementation skills that are appropriate for their mental age but other studies report these skills are deficient in autism.

The early acquisition of object permanence is thought to be undisturbed in very young children with autism[3] (Abrahamsen & Mitchell, 1990; Curcio, 1978; Dawson & McKissick, 1984; Lancy & Goldstein, 1982). Correspondingly, two studies failed to report an autism specific impairment in inhibition-and-implementation skill for four- and five-year-olds (Griffith, Pennington, Wehner & Rogers, 1999; McEvoy et al., 1993). McEvoy and colleagues reported ceiling effects for five-year-olds with autism and developmentally delayed controls on two A-not-B tasks. The second study, using the same tasks, without the complication of ceiling effects, reported that four-year-olds with autism (mean

[3] Hammes & Langdall (1981), however, reported difficulty in using this object knowledge for children.

verbal mental age = 22 months, mean nonverbal mental age = 34 months) performed as well as developmentally delayed controls (Griffith et al, 1999).

In contrast, some studies report inhibitory problems for five-year-old children with autism. Adrien and colleagues (Adrien, Martineau, Barthelemy, et al., 1995) administered a series of increasingly complex object permanence measures designed to assess to Piaget's last three stages of sensorimotor intelligence to young children with autism (mean chronological age = 59 months, mean global developmental age = 19 months). They reported pervasive difficulty in maintaining set, more perseverative errors and more variable performance in the children with autism than age- and ability-matched controls. Dawson and colleagues (Dawson, Meltzoff, Osterling & Rinaldi, 1998) also reported that children with autism aged five were less successful on the reversal trials of an A-not-B response task compared to Down syndrome and typically developing samples when non-verbal mental age was taken into account. However, it is interesting to note that all groups in this study failed to perform above chance on this task.

Performance on the Spatial Reversal task has demonstrated a similar mixture of inhibition-and-initiation skill. Three-and-a-half-year-olds (Wehner & Rogers, 1994 cited in Griffith et al, 1999) and four-year-olds (Griffith et al, 1999) with autism are as able as comparison groups to generate an appropriate response set. These studies also reported equivalent rates of perseverative responses when that set is no longer appropriate and, notably, fewer errors in the maintenance of a response set. On the other hand, McEvoy et al (1993) reported that five-year-old children with autism showed more perseveration to the previous rule than the comparison group.

Two other tasks have revealed an impairment in inhibiting a prepotent response and implementing an alternative response in children with autism. The Windows task assesses a child's ability to inhibit the prepotent response to reach directly towards a reward and to instead implement an alternative rule. Two boxes are displayed, one of which contains a chocolate. The child is told that pointing to the box containing chocolate means they do not receive the chocolate, whilst

pointing to the empty box means they receive the chocolate (Hughes & Russell, 1993; Russell, Mauthner, Sharpe & Tidswell, 1991). Seventy percent of children with autism (mean verbal mental age = four years) repeatedly indicated the baited window compared to 37% in the non-autistic mentally handicapped controls (Hughes & Russell, 1993). Following on from this finding, Hughes and Russell developed a novel piece of apparatus - the Detour Reach Task - designed to test inhibition-and-implementation with minimal interaction with an examiner.

In the Detour Reach task a marble is placed inside an aluminium box with a circular opening at the front. A direct reach towards the marble breaks an infrared beam and causes the marble to drop out of sight. A yellow light signals that turning the knob on the right side of the box can retrieve the marble. When a red light is lit, this route is no longer available. Instead the child has to flip a switch on the left side of the box and then make a direct reach in through the opening to retrieve the marble (Hughes & Russell, 1993). The knob route requires the child to inhibit a direct reach and turn the knob instead. The switch-reach route requires the child to firstly inhibit the direct reach, flip the switch and then implement the reach into the box. Because the switch-reach route was always presented after the knob route this second rule not only required sufficient inhibitory control to sequence the responses but also to inhibit the knob rule administered earlier. Since both rules were explained to the participants this task was assessing inhibition-and-implementation rather than inhibition-and-initiation.

All children with autism (mean verbal mental age = 6.6 years, nonverbal mental age = 6.7 years) were successful on the knob route, as were children with mental handicap matched for ability and younger typically developing preschoolers with equivalent mental ages (mean chronological age = 3 years 8 months). Success rates on the switch-reach rule were still over 90% for the two comparison groups whereas only 55% of the children with autism were now successful. Analysis of the relationship between mental age and performance on the switch-reach task revealed that whilst it was trivially easy for typically developing four-year-olds, success for autistic individuals only begins to emerge

at a mental age of over four years and failure is not uncommon with a mental age of almost seven years. Error analysis indicated that children with autism were more likely to perseverate to the knob route during administration of the switch-reach route (23% of the autistic subjects touched the knob at least once compared to 4% of the mentally handicapped children) and were more likely to make at least two consecutive direct reach errors (65% compared to 32%). In spite of the ability of autistic children to inhibit the direct reach and generate one action (knob rule), they displayed difficulty inhibiting this reach when the rule demanded a sequence of two actions (switch-reach). This may reflect either an inhibitory problem or a difficulty in correctly sequencing two actions.

Studies with older children have not reported severe inhibition-and-implementation impairments of motor or verbal responses. Twelve-year-olds with autism showed only a mild impairment on the second condition of the Go No Go Task (Ozonoff et al, 1994) that required the inhibition of a previous response and the implementation of a motor response that conflicted with the first condition. High-functioning children with autism (mean age 13 years, mean IQ 100) were also as able as comparison groups to inhibit irrelevant distractor items that had previously been relevant (Ozonoff & Strayer, 1997). In addition, verbal responses on Stroop tasks seem to be unimpaired in autism (Bryson, 1983; Eskes, Bryson, & McCormick, 1990; Ozonoff & Jensen, 1999). Specifically, 13-year-old children with autism (mean verbal mental age 7 years) made as many correct responses, with a similar reaction time, as developmentally delayed and typically developing children matched for chronological age and ability level on the Day/Night Task (Russell, Jarrold & Hood, 1999). Therefore, older children with autism do not appear to have an inhibition-and-implementation deficit compared to matched control groups.

Additional support for intact inhibition in autism comes from an interesting study of inhibitory skill in a play situation (Jarrold, Boucher, & Smith, 1994). This elegant study assessed children's (mean chronological age = nine-years-old, mean verbal mental age = four years six months) choice of placeholder prop when given a selection of non-conventional objects with different degrees of perceptual similarity to the target and a counter-functional object (i.e. an object

with which an irrelevant conventional action was associated). The authors predicted that a difficulty in inhibiting the conventional action associated with the counter-functional object would lead to the non-conventional perceptually dissimilar items being preferred[4]. However, the group of children with autistic spectrum disorders did not support the prediction, nor did the choices they made differ from typically developing children. This study is particularly notable for its stringent structure within a setting more representative of childhood activities than standard cognitive tasks.

There is a complex pattern of development of inhibition-and-implementation skills in autistic spectrum disorders. Five-year-olds with autism seem to display inhibition-and-implementation deficits on a variety of tasks with differing amounts of information support. In contrast older individuals and younger individuals with autism do not display these deficiencies in comparison to ability-matched control groups. Because of the subtle differences in task structure and demands across studies, and the fact that the studies recruited different samples with slightly different ages, conclusions must be tentative. However, the studies may suggest that, in contrast to comparison groups, the developmental progression of inhibition-and-implementation slows or stops for children with autism around the age of five years and then catches up in older childhood and adolescence. The proposal is that this developmental trajectory is distinct from that seen in other populations.

Preliminary longitudinal data supports this proposed developmental trajectory. A subset of the Griffith sample (Griffith et al, 1999) attempted the Spatial Reversal task twice (mean age at first attempt = 39 months and at second attempt = 55 months). Performance in the autism group remained the same over time, whilst there was a non-significant trend for the developmentally delayed group to commit fewer perseverative errors over time. Although unconfirmed, these data suggests that the comparative impairments on tasks involving inhibition of one response, initiation and implementation of another in older children with autism may reflect a lack of skill improvement over time whilst

[4] This assumes that individuals with autism are as likely as typically developing children to make the association between an object and its conventional use.

other groups make developmental progress. In other words, children with autism may 'grow into' an executive problem.[5]

Clearly further longitudinal work is essential to explore this possibility. One important task would be to apply the Detour Reach task to a younger sample of children with autism and identify if they exhibited impairments of inhibition-and-implementation in comparison to age- and ability-matched children. Tracking the development of inhibition-and-implementation skill (through the Detour Reach and A-not-B tasks) in this young sample would provide an opportunity to directly test and extend the developmental explanation.

1.5.3 Flexible Set Shifting

More advanced inhibitory skill is demanded by tasks or situations where flexible shifting between two or more cognitive sets is required. At this level there is no one-to-one association between a response and success, instead the participant must be able to respond flexibly according to one cognitive set and then inhibit reference to that set in order to follow another.

Typical development

The most familiar test of cognitive flexibility is the Wisconsin Card Sorting Task (WCST: Grant & Berg, 1948). In the original version the cards varied on three dimensions: number, colour and shape. The participant's task was to use feedback from the experimenter to learn the correct sorting rule. Once the participant had deduced the first rule the experimenter changed the rule unannounced and the participant was required to deduce the new rule. This required an ability to monitor feedback, inhibit the previously correct sorting strategy and generate one of the two possible remaining strategies. Although designed for use with adults, Welsh and colleagues (Welsh et al, 1991) showed that children as young as seven-years old could attempt the WCST, ten-year olds performed almost at adult level and 12-year olds had reached adult performance levels.

[5] This same explanation may help to reconcile the conflicting reports concerning A-not-B tasks noted earlier.

Claire Hughes adapted the WCST for use with preschoolers (Hughes, 1998a; Hughes, 1998b). She asked preschoolers to perform colour and shape card sorts under the guise of giving teddy the cards that he liked. Feedback was given to the child until they reliably gave cards to teddy on the basis of the dimension that he 'liked'. The set shift was signalled by a complete change of stimuli (cards and bear) however the child was still expected to generate the new rule on the basis of experimenter feedback. Hughes defined rule success as six consecutive correct sorts. At an average age of 3 years 11 months, the mean number of rules solved was 1.48 (out of a maximum 2) (Hughes, 1998a). This indicates that some children were able to switch from the first to second rule but others could not. The same children were also assessed 13 months later (Hughes, 1998b). In this study, Hughes reported that the mean number of trials required to pass a rule had reduced, significantly, from 13.3 to 12.4 over the year. Although the two studies do not report the proportion of children who were successful in switching rules, nor the pattern for individual members of the sample, there is some suggestion of a developmental increase in the ease with which the child can shift rules.

The Dimensional Change Card Sort (Bialystok, 1999; Frye, Zelazo, & Palfai, 1995; Zelazo et al, 1996) also tests the ability to switch from one rule to another. The task requires the child to sort cards according to a rule. Each test card matches one target card on one dimension (e.g. colour) and the other on a second dimension (e.g. shape). The child must sort the cards first according to one dimension (preswitch) and then by the second dimension (postswitch). A major difference between this task and the WCST-like tasks is that the child is provided with the sorting rules. Rather than having to choose and initiate a sorting rule and then switch to sorting by an alternative rule, this task merely requires the child to implement a given rule and implement the second rule. Typically, three-year-olds continue to use the preswitch rule on the postswitch trials. Because three-year-olds were unable to detect a puppet's errors on the sorting task (Jacques, Zelazo, Kirkham, & Semcesen, 1999), it was concluded that the failure to swap rules at this age is a result of immature cognitive flexibility rather than a simple motor response control difficulty.

Development in autism

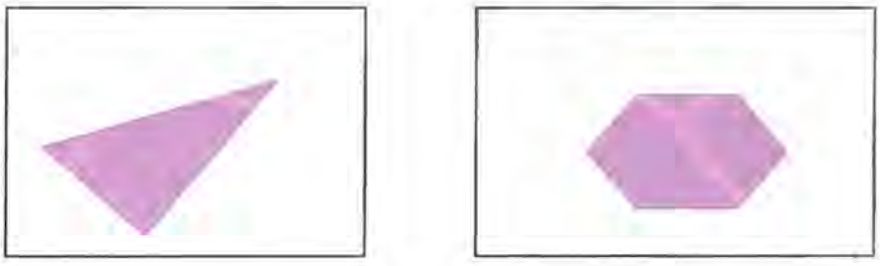
There have been no studies looking at this type of set-shifting ability in young children with autism. In contrast, several studies have explored flexible set shifting in older children and adults with autistic spectrum disorders. The Wisconsin Card Sorting Task is one of the most widely used tests of executive function in autism (for a review see Pennington & Ozonoff, 1996). Overall, individuals with autistic spectrum disorders sort fewer categories (Rumsey, 1985; Rumsey & Hamburger, 1988; Szatmari, Tuff, Finlayson & Bartolucci, 1990) and make more perseverative errors than comparison groups (Bennetto, Pennington, & Rogers, 1996; Ozonoff, 1995; Ozonoff & Jensen, 1999; Ozonoff & McEvoy, 1994; Prior & Hoffman, 1990; Rumsey & Hamburger, 1988; Rumsey, 1985; Szatmari et al, 1990; but see Minshew, Goldstein, Muenz, & Payton, 1992 and Schneider & Asarnow, 1987 for non-replications). A similar, but rarely used, object-sorting task has also demonstrated set shifting impairments in autism (Minshew et al, 1992).

The presence of set shifting impairments in adolescents with autistic spectrum disorders has also been demonstrated on the Go-NoGo information-processing paradigm (Ozonoff et al, 1994). In the third and most difficult condition of this task the response-stimuli associations are switched unexpectedly during a block of trials. The subject has to identify this switch and then alter their responses accordingly. Twelve-year-olds with autism were substantially impaired in their ability to shift flexibly from one stimuli-response association to another compared to controls. The deficit was particularly striking given the comparable performance of the groups on the inhibition of response and inhibition-and-implementation conditions.

A further source of investigation of set-shifting impairment in autism comes from the administration of the carefully constructed ID/ED task to a group of seven to 18 year old children with autism with a mixture of ability levels and developmentally delayed and typically developing matched groups (Hughes, Russell, & Robbins, 1994). This is particularly notable since most of the studies that have sought to assess cognitive functioning in autistic spectrum disorders have recruited high-functioning individuals. Since autistic spectrum disorders

presents across the ability range, including less able individuals in a testing sample increases the generalisability of the findings. The task takes the participant stage by stage through a series of discrimination tasks (Figure 1.2 provides an example of the experimental stimuli). Most children with autism could transfer learning within one dimension to new stimuli (the ID shift) but could not transfer learning to the previously irrelevant dimension (the ED shift). Both clinical groups were less skilled than their typically developing controls at transferring learning to a new set of exemplars but were only separable from each other on the final stage of the task where novel stimuli were introduced and the contingency changed to the previously irrelevant dimension (Hughes et al, 1994). This pattern of results suggests that individuals with autism may be differentiated from another clinical group by their difficulty in shifting set across stimuli and rules. Individuals with autistic spectrum disorders may be particularly impaired in their ability to respond appropriately when required to switch from one cognitive set to a previously irrelevant set.

Figure 1.2. Example of the ID/ED task stimuli



1.5.4 Summary

The emerging argument is that inhibitory control may be usefully considered at three levels of complexity: inhibition of response, inhibition of one response and implementation of another, and flexibly shifting between cognitive sets. Typically developing children have been shown to develop these skills throughout childhood and adolescence. There is some evidence to suggest that the component skills of executive function develop contemporaneously but independently (to a certain extent). The developmental pattern in autism has suggested a lack of impairment in very young children with autism on simple

inhibition tasks, and a lack of impairment in older children on simple inhibition-and-implementation tasks. However, impairment in four- and five-year old children with autism has been noted at the inhibition and implementation level. Furthermore the most complex stage of flexible shifting between responses seems to be substantially impaired in autism.

1.6 The Development of Generative Skill

The generation of responses can be categorised into two different types. The first is the ability to generate and implement an appropriate strategy or a conventional response. These strategies may include sorting by colour or number, or searching through a semantic lexicon. A conventional response may be the generation of a typical picture in response to the request to draw a picture. The second is the ability to generate a non-conventional or imaginative response. Going beyond a stored lexicon of knowledge to generate a creative strategy plays an important role in providing innovative solutions. To date, there has been little debate about the relative complexity of the categories or a possible developmental pathway for generative skill. The ability to generate a strategy or conventional response may be more cognitively demanding and therefore might be thought to follow the generation of novel responses. Alternatively, it might be argued that novel responses make more substantial demands on cognitive capacity and therefore might follow the ability to apply simple strategies.

The structure of a task places constraints on the level of generation that a participant can demonstrate. Certain fluency tasks or problem solving tasks may require the participant to think laterally or imaginatively. The actual responses generated by a participant can be coded as following a given strategy, applying a standard or contextualised strategy or producing a novel response. Similarly, experimental instructions can forbid the use of certain strategies or encourage the use of imagination.

Typical development

Standard tests of generativity have been fluency tasks. There are three main types of fluency task: word, ideational and design fluency. All tasks require the participant to provide as many appropriate responses as they can. Individual task structure and instruction can provide the participant with a strategy or explicitly request non-conventional responses. Typically, word fluency tasks provide a letter or semantic category stimulus and require production of as many words beginning with the letter or belonging to the category (e.g. Benton, 1968; Leezak, 1995; Milner, 1964; Newcombe, 1969 cited in Temple, 1997; Thurstone, 1938 cited in Temple 1997; Turner, 1999a). Good performance on the word fluency task can be achieved by self-cueing an appropriate strategy, for example looking around oneself for objects beginning with the key letter. The Uses of Objects paradigm (e.g. Duncker, 1945; Leezak, 1995; Wallach & Kogan, 1966) requires the participant to provide as many possible ways of using an object as they can in a certain time period. As Turner (1999a) notes, responses can be recorded as conventional or imaginative. The Design Fluency Task (Jones-Gotman & Milner, 1977) requires the participant to produce as many different designs as they can, excluding conventional designs. This third fluency task places heavy demands on the individual's ability to generate novel and imaginative responses that are not recalled from a base of stored knowledge.

There are two ways in which performance on fluency tasks can be assessed: the number of responses generated and the relative uniqueness of each response (Wallach & Kogan, 1966). On all these fluency tasks the number of varied responses provided is taken as a measure of response generation. A small number of responses would indicate poor generation; a large number of responses would indicate good generation. Categorising responses as imaginative, conventional, perseverative (direct repetition of a previous response), redundant (a response so similar as to be considered not varied from a previous response) or incorrect (a response that breaks task rules) facilitates interpretation of the level of generativity skill displayed by the participant (Turner, 1999a).

Few studies have been concerned with the development of generative skill in young children. An early study of children aged about 10 years old reported that children did show creativity (original authors' term), and this was not related to IQ level (Wallach & Kogan, 1966). However, the paucity of developmental work on this skill may, in part, be due to the reliance on fluency tasks to measure generation; fluency tasks have proven resistant to downward extension for children. In particular, word fluency tasks are clearly inappropriate for preverbal children and are likely to be a measure of vocabulary size in other children. Tests of imaginative fluency are more promising for youngsters. Play settings provide an excellent opportunity to test the imaginative generativity of a child. One might wish to assess the number of diverse ways a child uses a toy during one play session. Varying the target toy and carefully coding the acts of play would enable different levels of generation to be tapped. Consider, for example, the generation of a highly imaginative act with a non-functional object as opposed to the generation of a typical action in response to a functional object (see Jarrold, 1997 for a similar approach to play in young children).

Development in autistic spectrum disorders

High-functioning individuals with autism (Minshew et al, 1992; Rumsey & Hamburger, 1988; Turner, 1999a) and low-functioning individuals with autism (Turner, 1999a) have demonstrated reduced letter and category performance on word fluency tasks. However, other studies failed to report these deficits in high-functioning individuals (Minshew, Goldstein, & Seigel, 1995) or in lower-functioning individuals (Boucher, 1988; Scott & Baron-Cohen, 1996) with autism. Interestingly, despite finding no impairment in standard word fluency tasks, Boucher's study found that individuals with autism were substantially poorer on a task that provided no cues for strategy use ("provide as many words as you can think of"). This suggests that the ability to self-generate a strategy with minimal support is more difficult for individuals with autism than cued strategy generation and that the severity of self-generation impairment differentiates them from comparison groups.

An impairment of strategy generation in the absence of clear task structure has also been observed in high- and low-functioning children with autism on

ideational fluency tasks (Turner, 1999a). Children and adults were asked to provide a variety of uses for conventional and non-conventional objects. Two aspects of performance are particularly interesting. Firstly, clinical control participants demonstrated better performance when the object presented was non-conventional, whereas the impaired performance by autistic subjects was unaffected by the type of object. Conventional objects are likely to have certain responses associated with them that are more salient than other responses. Non-conventional objects are less likely to have a salient association to action. It seems that the typically developing children are more susceptible to capture by the standard use of an object, whilst the children with autism are impaired regardless of the inhibitory demands. Secondly, the lower-functioning individuals with autism produced more repetitive errors and the higher-functioning individuals with autism produced more redundant errors than their appropriate comparison group. Both these error types can be considered to represent inflexibility of thought and may suggest that these children struggled to inhibit previous responses. Although one other study failed to report an autism-specific generativity impairment when participants were asked for alternative uses for a brick (Scott & Baron-Cohen, 1996) this may have been because the two groups of children were young, with a verbal mental age of four-and-a-half years, and the comparison group were unexpectedly poor on the task.

Performance on the third type of fluency task, design fluency, is worthy of note. On this task, children are asked to produce as many designs as possible using a set of components (such as a line, a triangle and a circle). Children with autism produced as many designs as comparison groups, however both high- and low-functioning children with autism made more error responses than their comparison groups (Turner, 1999a). Specifically, these two groups of children produced more repetitive and redundant designs than the group of control children. This pattern of response indicates that individuals with autism are able to produce an equivalent number of designs to their peers, but they are more susceptible to 'capture' by a previous design. It seems plausible to propose that the generation of novel responses is more susceptible to interference from previous responses in autism than typical development, and that this

susceptibility may be due to an inhibitory control impairment or an inability to plan a response so that capture by an earlier motor program can be resisted.

1.7 Working Memory

Working memory is a key component of successful task performance but it is often seen as a confounding rather than dependent variable in executive function research. Its key role is maintaining a mental representation on-line during tasks requiring executive control (Goldman-Rakic, 1987; Fuster, 1985; Welsh et al., 1991). In fact, we have already seen how even simple object retrieval tasks make working memory demands of infants (e.g. Diamond et al, 1994). Moreover, in a developmental context, the increasing capacity of working memory may provide an essential foundation for maturing executive skills (e.g. Diamond et al, 1997). In addition to this conceptualisation of working memory as a necessary basis for executive function development, factor analysis has suggested it should be considered an executive skill in its own right (Hughes, 1998a; Welsh et al, 1991).

Typical development

The most common way in which working memory has been explored in young children is through visual search tasks[6]. These are particularly suitable for young and preverbal children because they require very little verbal ability. In the prototypical visual search task the child is presented with a number of containers in each of which is placed a reward. The children are encouraged to find all the rewards as quickly as possible, making as few reaches as possible. Usually the containers are hidden from the sight of the child after each reach. In stationary versions of the task the containers remain in the same relative positions throughout the task whilst in scrambled versions the relative positions are altered between each reach. In the scrambled condition the containers are always distinguishable from one another (by colour, shape or size for example). Retrieving all rewards in the minimum number of reaches requires working memory to remember the containers already checked (or to remember the

[6] The term visual search task is used in the same way as Hughes (1998a,b) to describe a visual working memory task in which containers must be searched.

containers not yet checked) to prevent a repeat search to a container. Remembering which locations or containers have been searched are both useful strategies in the stationary version, whilst in the scrambled version a strategy associated with the distinguishing dimension is best.

Developmental improvements are measured by a reduction in the number of reaches required to retrieve all rewards or by successful performance when the number of rewards and containers is increased. Two studies have looked at the application of visual search tasks with preschool children. One study suggested that three boxes were most suitable for 15- to 30-month-old toddlers and six boxes for 3½ to 7-year-olds (Diamond et al, 1997). A second study used eight boxes with children aged three and four years old (Hughes, 1998a).

Development in autistic spectrum disorders

One study has evaluated the working memory skill of pre-school children with autistic spectrum disorders on visual search tasks with three and six boxes (Griffith et al, 1999). This study reported no evidence of impairment compared to a control groups matched for ability. However, the picture is less clear in older individuals with autism.

Adolescents and adults with high-functioning autism (together these studies have assessed individuals aged between 12 and 40 years old) have been reported to perform both more poorly than (Bennetto et al, 1996; Minshew et al, 1999; Minshew et al, 1992; Mottron, Morasse & Belleville, 2001; Ozonoff, Pennington, & Rogers, 1991a) and comparably to (Minshew & Goldstein, 1993; Ozonoff & Strayer, 1997; Rumsey & Hamburger, 1988; Russell, Jarrold, & Henry, 1996) comparison groups on working memory tasks. These contrasting results may be explained in terms of an individual's propensity to initiate an appropriate working memory strategy. Bebko and Ricciutti (2000) provide support for this hypothesis by reporting that moderately functioning individuals with autism required suitable structure and support within the task setting to engage in a rehearsal strategy. Since individuals with autistic spectrum disorders have been shown to find it difficult to self-generate appropriate strategies it is

possible that deficits in working memory may reflect a generativity problem rather than a memory capacity problem per se.

From the small amount of research that has looked at working memory as a distinct executive skill it is unclear whether this skill is impaired or spared in autism, or what the developmental trajectory of this skill might be. Clearly further investigation is required with samples of young children re-assessed over a period of time.

1.8 Planning: A Case of Executive Skills Working Together

There is widespread agreement that planning involves the integration of several component executive skills (e.g. Goel & Grafman, 1995; Welsh, 1991; Wellman, Fabricius, & Sophian, 1985; Willatts, 1997). Minimal requirements for successful planning are the ability to "*recall procedures for actions, anticipate their effects, and coordinate them into coherent sequences without information supplied through overt action*" (Willatts, 1997, p.147). In other words, a combination of working memory, generativity, inhibitory control and some mechanism for evaluation of what actual actions are necessary.

> '*[Planning] involves representing a problem, setting a goal, deciding to plan, creating a plan, implementing and monitoring the plan, and then reviewing the outcome*' (Friedman, Scholnick, & Cocking, 1987).

Typical development

Infants show the ability to think beyond the first step of a problem around the age of 10-months (Willatts & Rosie, 1992) and by 18-months show evidence of the generation and evaluation of alternative strategies, of remembering previous decisions, and of strategy monitoring (De Loache, Sugarman, & Brown, 1985; Willatts & Fabricius, 1993 cited in Bauer, Schwade, Saeger Wewerka, & Delaney, 1999). These findings have been based upon tasks where the infants have to overcome one or more obstructions in the pursuit of a desired object. For example, a toy may be resting out-of-reach on a cloth that the infant must pull

towards them. Several obstacles may need to be removed before the infant can pull the cloth. Likewise, work with older children has focused on their ability to sequence a number of steps in order to achieve a specified goal.

The Towers of Hanoi and London (Humes, Welsh, Retzlaff, & Cookson, 1997; Shallice, 1982; Simon, 1975) require the combination of a series of moves to achieve a goal state. The child (or adult) is presented with a number of rings to be transferred from one peg to a goal state in the fewest number of moves possible. The number of rings and pegs, as well as the number of moves required from the initial state to the goal state vary according to the age of the participant and across study. Other variations have included presenting differently sized rings, imposing certain rules that moves must conform to, supplying a cover story and providing a continual representation of the goal state during the task (e.g. Welsh, 1991). Common to all these variations are requirements to generate, evaluate and implement a sequence of steps that often appear to move away from the goal before reaching it.

Children improve on these planning tasks with age. Welsh and colleagues (Welsh, 1991) showed that six to twelve year olds were more proficient than three to five year olds but that both groups employed similar strategies. Adult performance levels were reached by the age of six for three-disk problems and by 12 for four-disk problems (see also Anderson, Anderson & Lajoie, 1996; Lussier, Guerin, Duffresne, & Lassonde, 1998).[7] Importantly, Welsh and colleagues argued that they had provided appropriate structure through a cover story to elicit planning behaviour in the youngest (three-year-old) children, who would otherwise not have demonstrated such planning skill.

The notion that appropriately structured planning tasks tap into the zone of proximal development (Vygotsky, 1978) enabling young children to demonstrate more sophisticated planning ability than they would in a less structured setting has persisted. Neither 21- nor 27-month-old children were able to solve a problem by sequencing three actions (e.g. make a spinning top, make a gong, make a rattle) without support (Bauer et al., 1999). However, when the

[7] Note that Pea (1982) found that children as old as 12 years still did not appreciate the flexible and revisionary nature of planning.

children were shown the goal state performance improved dramatically so that the percentage of trials on which the three necessary actions were produced rose to 45% and the children were more reliable than chance at sequencing these three moves correctly. Removing the planning demands completely (the solution was modelled for the child to imitate) meant that 73% of these very young children could reach the goal. This study neatly demonstrated the problems two-year-old children have with planning a sequence of actions to an unseen goal, but their readiness to sequence actions to a seen goal.

Three-, four- and five-year-old children demonstrate a similar readiness to plan a more familiar task: shopping in a toy grocery store (Hudson & Fivush, 1991). Five-year-olds could plan and execute shopping for breakfast and party goods regardless of how the appropriate goods were set out in the model store. Four-year-olds were in a transitional phase where they could construct a plan but found execution of the plan onerous unless an adult provided feedback and reminders during the task. Three-year-olds were able to shop for either breakfast or party goods (but not both) when provided with feedback and reminders throughout plan construction and execution. This study illustrates a developmental progression of planning skill whereby five-year-olds can manage generation, evaluation, implementation and inhibition of actions simultaneously but younger children require some or all of these competing executive demands to be reduced before they can demonstrate planning. As this study was cross-sectional in design, longitudinal work must be conducted to ascertain whether this developmental pathway is apparent in individual children.

Development in autistic spectrum disorders

The impairments reported in inhibition, generation, working memory and set-shifting skills for individuals with autistic spectrum disorders suggest that, given the composite nature of planning, these individuals will also display planning deficiencies. The available evidence supports this prediction.

In all studies using the Tower of Hanoi or London tasks that this author is aware of, individuals with autism are less efficient in planning a sequence of moves leading to the goal (Bennetto et al., 1996; Hughes et al., 1994; Ozonoff &

Jensen, 1999; Ozonoff & McEvoy, 1994; Ozonoff et al., 1991a; Ozonoff et al., 1994). Furthermore, Prior and Hoffman (1990) showed that 10 to 17 year-olds with autism used maladaptive strategies to solve a maze task and made the same mistakes repeatedly. Hughes and colleagues (1994) demonstrated that planning impairment extends across the ability range in children with autistic spectrum disorders (mean chronological age 13.2 years) and Ozonoff and McEvoy (1994) showed performance in autism remained stable between the ages of 12 and 15 years old whilst the performance of the comparison group improved.

Studying how individuals with autism approach drawing tasks has also shed light on planning skills within this population. A sample of 13-year-olds with autistic spectrum disorders were not impaired in their ability to copy a complex figure, however they exhibited a more piecemeal and less global approach to the task than other children (Prior & Hoffman, 1990). This suggests that an individual with autistic spectrum disorder may be less likely to formulate a complete plan before starting to copy the figure. An incomplete plan may also explain why individuals with autism are more likely to become captured by a previous response when asked to draw novel designs (Turner, 1999a). Similarly, two studies assessed autistic children's ability to generate (Scott & Baron-Cohen, 1996) and complete (Leevers & Harris, 1998) impossible pictures. Combining the results of these experiments it appears that four-year-olds with autism are able to complete an impossible picture (minimal planning demands) but do not have the required planning skills to *generate* an impossible picture.

The current evidence suggests that individuals are impaired in their ability to produce planful behaviour. These studies have not clearly demonstrated whether this stems from a deficit in generating, evaluating or implementing the plan. The evidence presented earlier that shows executive deficits becoming increasingly apparent as the interaction between inhibition and generation is increased. It seems likely, therefore, that planning impairments in autism reflect a difficulty in integrating inhibitory control with response generation. Further investigation regarding very early planning skills in autism is essential; particularly given the small but growing body of evidence that executive impairment is not evident in very young children.

1.9 A Developmental Perspective on Executive Function in Autistic Spectrum Disorders

A large body of research has provided mixed support for executive dysfunction in autism. Whilst a popular conclusion is that individuals with autism have severe and pervasive executive deficits (e.g. Pennington & Ozonoff, 1996) the fine-grained approach to executive function followed here suggests this is too simplistic. The current hypothesis is that early development of basic inhibition and working memory skills may be unimpaired (or only mildly impaired) in young school-aged children with autism, in comparison to age- and ability-matched comparison groups. The evidence for this assertion comes from carefully designed tasks that single out basic skills and the unimpaired performance apparent in very young children with autistic spectrum disorders. However performance on tasks that place multiple executive demands upon the child (planning tasks, for example) is substantially impaired in children with autism.

The work by Griffith and colleagues (1999) that found no significant group differences on tasks of inhibition-and-initiation or working memory in preschoolers with autism would be in line with this proposal. In that study preschoolers did not display the expected executive function deficit in relation to their controls but it is possible that more complex planning tasks that depended upon the integration of several executive function skills would have elicited group differences.

The developmental trajectory of executive skills over time is more difficult to identify. The preceding sections have suggested there is no reliable evidence of inhibitory impairment in preschool children with autism or in adolescents with autism. However studies on children in the intervening age groups have identified inhibitory deficits. Griffith and colleagues (1999) did re-administer the Spatial Reversal task to some of their sample but found no significant change in performance for either children with autism or children with developmental delays between the mean ages of 3-3 and 4-7 years. Therefore this study identified a lack of developmental progression on this task for both groups over one year. One proposal to bring this study together with the body of

work is that children with autism may continue to 'fail to progress' until their adolescent years whilst individuals with developmental delay may 'catch-up' earlier. Clearly it is important to study the development of executive function over time in individual children to further explore both typical and atypical executive function development. The current thesis aims to extend the Griffith work by taking a developmental perspective of several executive skills over the period of one year.

Chapter 2

Evaluating the Executive Function Hypothesis of Autism

The validity of a psychological account of autism can be assessed by its ability to parsimoniously explain autism. A neuropsychological account of autism that claims a primary role in the development of autism must be able to provide an explanation of the communicative, social, and repetitive qualities of the autism behavioural profile, to explain the early onset of this developmental disorder and to demonstrate how the severity of individual symptomatology can vary at the same time as the deficit being universally displayed within autistic spectrum disorders. Furthermore, a single causal deficit is only tenable if the deficit cannot lead to different symptom profiles as seen in other developmental disorders (e.g. Pennington, Rogers, Bennetto, et al, 1997).

These requirements of a neuropsychological account of autism are stringent and recent views have suggested that one single account may not be sufficient to explain the heterogeneity of autism (e.g. Bailey et al, 1996; Happé, 2000; Minshew, Goldstein & Siegal, 1997). However it is also important to thoroughly evaluate each individual theory that may contribute to a multi-disciplinary account of autism.

Over the last 15 to 20 years interest in explaining autism at the psychological level has increased dramatically. In particular, three hypotheses have been constructed to explain cognitive and behavioural aspects of autism: Theory of Mind (ToM), Central Coherence (CC) and Executive Function (EF). Since the current thesis is focussed upon the EF hypothesis of autism this chapter evaluates the hypothesis against the strict criteria outlined above.

2.1 Can Executive Function Explain the Behavioural Presentation of Autistic Spectrum Disorders?

The behavioural characteristics of autism include abnormal social-communication alongside repetitive behaviours. Although the presence of repetitive behaviours is not alone enough for a diagnosis of autism, they are arguably the most distinctive aspects of the autistic behavioural profile. One of the benefits of the executive dysfunction hypothesis of autism is that it seems able to explain the repetitive nature of some behaviour in autism. In fact it provides a more promising account of these behaviours than the Theory of Mind or Central Coherence accounts.

Repetitive behaviour has proved difficult to explain, in part, because both motor and cognitive behaviours can have the same characteristics of inflexibility and restriction. Repeated hand flapping, for example, is inflexible and repetitive at a motor level whereas repeated talking about a very circumscribed topic is limited and repetitive at a cognitive level. Turner (e.g. 1995; 1997) has begun to address the ways in which executive dysfunction might lead to such varied levels of types repetitive behaviour.

Based on data from a substantial sample of children with autism aged between about seven and 18 years of age, Turner looked at the relationship between certain categories of repetitive behaviour and executive function skills. Having developed a theoretical proposal that dysfunction in inhibition might correlate with 'lower' level repetitive behaviours whilst dysfunction in generativity might correlate with 'higher' levels of repetitive behaviour she then found her data supported such a proposal. On this view, a failure to control one's behaviour may lead to an action being repeated over and over again: if one cannot inhibit the response it will remain activated. This might correspond to lower level repetitive behaviours such as stereotyped movements and manipulation of objects. Alternatively, if one cannot generate an alternative response then the behaviour may not be replaced or developed and thus behaviour may take on a repetitive pattern. This might correspond to higher-level repetitive behaviours

such as circumscribed interests or insistence on sameness for routines and rituals (Turner, 1997).

Although Turner is really the only author so far to propose this theory, her data did support the predictions and there is an intuitive feel that such a theory might be plausible. The studies conducted by Turner relied on parental report of repetitive behaviours which may result in subtle biases. Direct observation of repetitive behaviours is very time-consuming and problematic but it is clearly important that the predictions of this theory be assessed in further samples for both reported and observed repetitive behaviours.

Communication in autism is less frequently related to an executive dysfunction account. However, it has been suggested that an impairment in interpreting and responding to feedback may affect linguistic development (Rutter, 1987). This type of account may be particularly well placed to explain the qualitatively unusual aspects of language seen in many individuals with autistic spectrum disorders, including repetitive topics of conversation, as well as the poverty of language seen in others. Anecdotally, it seems that feedback within the family may revolve around repeating words or correcting the child's pronunciation or grammar. A child who is unable to make use of this feedback may be unlikely to develop language in the same manner as a child who can interpret and benefit from the feedback. Alternatively, it has been suggested that, "*conversation requires the ability to integrate diverse knowledge bases*" (Dennis, 1991). Such integration is likely to be related to a child's ability to inhibit and evaluate a number of alternatives; the previous sections have demonstrated that the integration of these skills is impaired in autistic spectrum disorders.

The ToM account provides a particularly clear account of social impairment in autism: a failure to comprehend the mental life of others leads to impoverished social understanding (see Baron-Cohen, Tager-Flusberg & Cohen, 2000). However, an executive function account also addresses the variability in social skill across settings found in individuals with autism. When social situations are structured and predictable, individuals with autism are more likely to exhibit appropriate social behaviour than when faced with an unstructured setting

(Clark & Rutter, 1981; Dadds et al, 1988; Donnellan et al, 1984). A structured setting reduces the executive demands of strategic behaviour generation, evaluation and implementation.

Executive deficits can also provide a plausible explanation of the pattern of impaired and spared play shown in autism. Harris (1991; 1993; 1994) argued the child's growing ability to impose internal executive control on their behaviour explained the developmental trend in pretence away from action determined by the external context. Although Harris' suggestion may help to explain the increasing sophistication of young children's play it lacks specificity. The specific role of generativity in pretend play has been highlighted by the observations that pretend play is not totally absent in autism nor do individuals with autism have specific difficulty inhibiting a salient object affordance in play (Jarrold, 1997; Jarrold et al, 1994; Jarrold et al, 1996). This view is also able to explain why individuals with autism produce fewer spontaneous pretend acts but are unimpaired in their ability to carry out suggested pretend acts (Jarrold et al, 1996; Lewis & Boucher, 1988).

2.2 Is There a Relationship Between Executive Function Impairment and Severity of Autistic Symptomatology?

Direct comparisons between executive function performance and severity of autistic symptomatology are uncommon and the relationship that may or may not exist between the two is unclear. Evidence to suggest there may be a relationship between executive function ability and social skill comes from work with children who experience damage to the prefrontal lobe area: set shifting impairments are reported to relate to measures of empathy and social skills (Grattan & Eslinger, 1989; Grattan & Eslinger, 1992). A review by Hughes (2001) identified the plausible role executive function may play in sociocommunicative skills.

Three studies have looked more directly at the correlational relationship between symptomatology and executive function skill in young children with

autism. Two studies compared joint attention, social interaction and behaviour regulation with performance on the Spatial Reversal task in a combined group of children with autism and their comparison sample. One of these had reported group differences on the executive task for five-year-olds (McEvoy et al, 1993) and the other had not found these differences for four-year-olds (Griffith et al, 1999). Both studies reported a significant relationship between joint attention and perseverations on the Spatial Reversal task; one study also reported a relationship with social interaction (McEvoy et al, 1993) but neither with behaviour regulation. The third study with five-year-olds with autism explored a number of symptom variables with performance on the A-not-B Invisible Displacement task and a delayed non-match to sample task hypothesised to tap the medial temporal lobe (Dawson et al, 1998). Social orienting, immediate imitation, deferred imitation, shared attention, response to distress, symbolic play and Wing classification were all correlated with performance on the delayed non-matching to sample task whilst the A-not-B Invisible Displacement task showed a relationship with immediate imitation only. The lack of substantial relationship between the A-not-B Invisible Displacement task and some aspects of socio-communicative behaviour suggests there may not be a relationship between inhibition-and-implementation and social-communicative behaviours. However, it is possible that this finding may be accounted for either by the particular measure of performance observed on this task or that this task may in fact relate to repetitive behaviours rather than socio-communicative behaviours.

Therefore, there is mixed evidence to suggest a relationship between social skills and tasks thought to measure executive performance. Turner (1997) reported significant associations for older children with autism between certain measures of executive function task performance and parental report of repetitive behaviours. Specifically, she suggests that recurrent perseveration is associated with repetitive movements and circumscribed interests, stuck-in-set perseveration with repetitive use of language and circumscribed interests and generativity with sameness behaviour and circumscribed interests (Turner, 1997). In contrast, the preliminary results from a study looking at the display of repetitive behaviours in high-functioning children and adolescents with autism

and Asperger's syndrome fails to report any associations between the repetitive behavioural categories and performance on the WCST (South, Ozonoff, & McMahon, 2001).

Evidently, further exploration of the relationship between executive function skills and all aspects of autistic symptomatology is needed. In particular, the developmental course of autism may be better understood by a clearer understanding of the nature of this relationship. An initial prediction would be that executive function and symptomatology were related contemporaneously. Beyond that, a predictive relationship between early executive function skill and later symptomatology would also fit with the Executive Function hypothesis of autism. The opposite pattern would serve to argue against executive dysfunction as a primary and causal impairment in autism. Either of these relationships would provide valuable clues for identifying children who may benefit from specific intervention. For example, if symptomatology was found to predict later executive dysfunction, appropriate and early intervention may diminish the difficulties encountered in the executive domain, or provide the child with alternative skills to counter the executive dysfunction. Furthermore, within the broad categories of executive function skill and autistic symptomatology, relationships may exist between specific executive function skills and specific symptomatology variables. A third possibility is that no reliable relationship between executive function and autistic symptomatology is observed at a group level. This finding would also call the executive dysfunction hypothesis of autism into question. In this situation, further exploration of the individual profiles of development may highlight other key variables or specific groups for whom a relationship exists.

2.3 Is Executive Function Impairment Universal within Autistic Spectrum Disorders?

The data presented in earlier sections has already shown that performance on executive function tasks is not always consistent across or within samples (e.g. Adrien et al, 1995; Prior & Hoffman, 1990). Furthermore, careful reading of most studies of executive function reveals a greater spread of scores in autism

samples than comparison samples. Nevertheless, there is evidence to support a claim that executive dysfunction is widespread in autism and related disorders.

One review reported significant deficits on thirteen out of fourteen experimental studies of executive function in a range of autism samples (Pennington & Ozonoff, 1996). Furthermore, individuals with autism demonstrated deficits relative to controls on 25 out of 32 tasks. The average effect size of the group differences across studies and tasks was large (1.0). Executive deficits have also been reported in individuals with Asperger's syndrome (Ozonoff et al, 1991b) and the unaffected siblings of individuals with autism (e.g. Hughes, Plumet, & Leboyer, 1999; Ozonoff, Rogers, Farnham, & Pennington, 1993). Even more compelling is the fact that discriminatory power shown by executive deficits in distinguishing siblings of individuals with autism for siblings of individuals without autism is greater than that shown by theory of mind skill (Ozonoff et al, 1993).

2.4 Is Executive Function Impairment Unique to Autistic Spectrum Disorders?

Executive function skills have also been studied in a large number of developmental disorders; including children with attention deficit hyperactivity disorder (ADHD), conduct disorder, Fragile-X, obsessive compulsive disorder, Tourette's syndrome, children treated for early phenylketonuria, children with Down syndrome or moderate learning disabilities, and sub-clinical hard-to-manage, angry and antisocial children. Initially, it seems that any executive deficit in these populations would form a major criticism of the executive function account of autism: how could one primary impairment result in such varied symptom profiles? However, as this chapter has already demonstrated, executive function is not a unitary construct. Instead several component skills can be identified. The current approach reflects this understanding of the executive function construct by searching for distinctive profiles of executive impairment that may distinguish between developmental disorders.

The meta-analytic review sought to identify specific executive function profiles for autism, attention deficit hyperactivity disorder, conduct disorder and Tourette's syndrome (Pennington & Ozonoff, 1996). The authors argued that ADHD and autism were the two disorders that demonstrated reliable and severe executive deficits and the profiles varied for each disorder. They suggested that autistic spectrum disorders are characterised by intact inhibitory skill and severely impaired cognitive flexibility and working memory whilst ADHD is characterised by deficits in response inhibitory control and planning with relatively spared generation and flexible set shifting skills.

The Tower of Hanoi planning task elicits performance deficits in ADHD (Aman, Roberts, & Pennington, 1998; Pennington, Groisser, & Welsh, 1993; Weyandt & Willis, 1994; for a non-replication see Ozonoff & Jensen, 1999) although the average effect size is half that for autism samples (Pennington & Ozonoff, 1996). Stroop tasks of inhibitory control in children with ADHD have reliably reported deficient performance in comparison to controls (Barkley, Grodzinsky, & DuPaul, 1992; Boucugnani & Jones, 1989; Gorenstein, Mammato, & Sandy, 1989; Grodzinsky & Diamond, 1992; Hopkins, Perlman, Hechtman, & Weiss, 1979; Lavoie & Charlebois, 1994; Lufi et al, 1990; Ozonoff & Jensen, 1999; Reardon & Naglieri, 1992) as have other tasks of motor response control (Aman et al, 1998; Barkley, 1997; Korkman & Pesonen, 1994; Nigg et al, 1998; Mariani & Barkley, 1997; Shue & Douglas, 1992; Trommer, Hoeppner, Lorber & Armstrong, 1988).

In contrast, performance on the WCST is mixed with more than half of the investigations showing no ADHD deficit compared to controls (Barkley et al, 1992; Fischer, Barkley, Edelbrock & Smallish, 1990; Grodzinsky & Diamond, 1992; Loge, Staton, & Beatty, 1990; McGee, Williams, Moffitt, & Anderson, 1989; Ozonoff & Jensen, 1999; Pennington et al., 1993; Weyandt & Willis, 1994; although deficits are demonstrated by Boucugnani & Jones, 1989; Chelune, Ferguson, Koon, & Dickey, 1986; Gorenstein et al, 1989; Shue & Douglas, 1992) and a smaller average effect size than for autism (Pennington & Ozonoff, 1996). Similarly, letter and category fluency does not seem reliably impaired in ADHD (Fischer, Barkley, Edelbrock, & Smallish, 1990; Loge et al,

1990; McGee et al, 1989; Weyandt & Willis, 1994; but Grodzinsky & Diamond, 1992) except when the variable of interest is the number of rule breaks made (Loge et al, 1990). It is notable that children with conduct disorder only display WCST and inhibitory difficulties when comorbid ADHD has not been removed from the clinical sample (Lueger & Gill, 1990; McBurnett, Harris, Swanson, et al, 1993; Moffitt & Henry, 1989; Pennington & Ozonoff, 1996).

The executive skill pattern for Tourette's syndrome is ambiguous. Stroop tasks do not, except for one study (Georgiou, Bradshaw, Phillips, Bradshaw, & Chiu, 1995), identify inhibitory deficits (Channon, Flynn, & Robertson, 1992; Ozonoff & Jensen, 1999; Silverstein, Como, Palumbo, et al, 1995). Similarly, children in this clinical group were unimpaired on all conditions of the Go-NoGo task (Ozonoff et al, 1994). Cognitive flexibility also seems unimpaired on the WCST (Bornstein, 1990; Bornstein, 1991a; Bornstein & Yang, 1991; Harris et al., 1995; Ozonoff & Jensen, 1999; Randolph, Hyde, Gold, et al, 1993; Sutherland, Kolb, Schoel, et al, 1982; Yeates & Bornstein, 1994; but Gladstone, Carter, Schultz, et al, 1993). However, inhibitory difficulties were observed on Luria's Hand Game and on a task requiring the inhibition of 'yes' and 'no' responses (Baron-Cohen, Cross, Crowson, & Robertson, 1994).

PKU children have also demonstrated impairments in planning and organisation (Cowie, 1971; Koff, Boyle & Pueschel, 1977) the flexible application of strategy (Pennington, van Doorninck, McCabe & McCabe, 1985; Welsh et al., 1990), and semantic category fluency (Welsh et al, 1990) that endure over a number of years and relate to the level of phenylalanine (Diamond et al, 1997 but see Griffiths, Tarrini & Robinson, 1997).

Children with Down's syndrome and/or moderate developmental delay have often been included in heterogeneous control groups for studies of children with autism. When they are the focus of a study these children show a tendency to stick to one rule rather than switch (Zelazo et al, 1996), difficulty in disengaging from a stimulus (Kopp, Krakow, & Johnson, 1983), trouble redirecting attention (Cicchetti & Ganiban, 1990) and problems returning to previously inhibited stimuli (Sersen, Astrup, Floistad, & Wortis, 1970). Mildly or moderately

handicapped children have also demonstrated some difficulties with maintaining a response set but not inhibitory control (Hughes et al, 1994; Ozonoff et al, 1991b). These findings suggest that further investigation is warranted: perhaps considering the role of generativity versus inhibitory control in these populations.

There has been very little research concerning executive function skills in children with speech and language delays. However two very recent studies (Bishop & Norbury, a; b, both under review) have contrasted inhibition and generation in four groups aged 6-10 years: children with high functioning autism, non-autistic children with pragmatic language impairment, children with typical specific language impairment and control children of similar age and nonverbal ability. Generativity was measured using two fluency tasks that were shown by Turner (1999) to be sensitive to autistic spectrum disorders. Bishop and Norbury failed to replicate Turner's findings of low response rates in children with autism, but there was a significant correlation between the number of correct responses on the fluency tasks and measures of pragmatic impairment. Other autistic symptoms were unrelated to fluency performance. Inhibition was assessed using two subtests from the Test of Everyday Attention for Children. Although they found evidence of inhibitory deficits, these were neither specific to autism, nor associated with particular aspects of autistic symptomatology. Rather, they appeared to be associated with poor verbal skills. This study suggested that children with specific language delay showed similar inhibitory deficits to children with pragmatic language impairment and autistic spectrum disorder. Moreover it suggested that autistic symptomatology may not be related to inhibition or generation, but that verbal ability may be the key correlate.

The precise profiles of spared and impaired executive skills in developmental disorders have yet to be described fully. Initial indications suggest that distinctive skill patterns may differentiate between disorders, and in this way the discriminant validity problem may be answered because the differences reflected at the behavioural level of these disorders may result from subtly different cognitive impairments. Further research is key to assessing whether or not this is the case: longitudinal work is required to compare the developmental

trajectories of different populations and samples must be recruited with care so that the effects of comorbidity may be established. Furthermore, in light of the evidence that Down's syndrome and moderately handicapped children may also display some executive deficits the recruitment of clearly defined comparison groups is essential.

2.5 Speech and Language Delay as a Comparison Group for Autistic Spectrum Disorders

Research tends to focus on clearcut cases of autistic spectrum disorder who meet full diagnostic criteria. However children who have some features similar to children with autistic spectrum disorder can have considerable potential for clarifying theoretical relationships among cognitive and behavioural processes, and for eliminating some confounding variables. As Bishop and Norbury (under review) write

> "*suppose we are interested in seeing how far autistic symptoms can be attributed to executive deficits, and we do the conventional kind of study in which an autistic group is compared with a control group matched on some index of mental age. If we find a significant group difference, we can conclude that some aspect of autistic symptomatology is associated with executive dysfunction, but we cannot determine which component(s) of the triad of symptoms are responsible for the association. If, however, we were to include a group of children who had impairments in only one or two of the components of the autistic triad, we would be better able to tease apart underlying relationships.*"

Therefore the recruitment of children with speech and language delays as a comparison group for children with autistic spectrum disorders is likely to reduce the possibility that any findings are due to differences in communication skills. This may be particularly true when the samples are very young children for whom the oddities of autistic language may not yet be established.

Typical language acquisition varies substantially across individuals, however speech and language delay can often be identified in children by the age of three years (e.g. Burden, Stott, Forge & Goodyer, 1996; Whitehurst & Fischel, 1994). ICD-10 (World Health Organisation, 1992) diagnostic criteria for a specific developmental language disorder include: performance on standardised language assessments skills in the lowest 3% of the population; a nonverbal IQ that is higher than verbal IQ; and no known neurological, sensory or physical impairment that may directly affect spoken language. Distinctions can be made between receptive and expressive language delays where one or both of these particular aspects of language are specifically impaired.

Whilst the cornerstone of most diagnostic criteria for speech and language delay is an observed discrepancy between verbal and non-verbal ability (American Psychiatric Association, 1994; World Health Organisation, 1992), other authors have argued that children can display all the linguistic hallmarks of a speech and language delay in the absence of a verbal-nonverbal discrepancy (Bishop, Hartley & Weir, 1994; Stark & Tallal, 1981). This argument is possibly most important in very young children when performance on standardised instruments may be least sensitive and most susceptible to disruption. In recruitment of the children for the current study the discrepancy criterion was not adhered to because of these concerns about its validity in preschool children (although lower verbal mental ages than non-verbal mental ages were reported for the speech and language delay group at both time points: see Table 3.1 in Chapter 3).

ICD-10 criteria also state that the occurrence of a pervasive developmental disorder is an exclusion criterion for a diagnosis of speech and language delay (World Health Organisation, 1992) however other authors have argued these diagnoses need not be mutually exclusive. Rapin and colleagues (Rapin & Allen, 1987 cited in Conti-Ramsden & Botting, 1999) proposed that the labels of developmental language disorder and autism should be used to describe impairments in two distinct domains that can occur separately or together. Although these arguments have yet to be resolved, the point highlights the

theoretical links that have been drawn between the two disorders and need to be addressed in light of this thesis.

Links between communicative difficulty and social impairment in children have been reported longitudinally (e.g. Beitchman, Brownlie, Inglis, et al, 1996; Cantwell & Baker, 1991; Rutter & Mawhood, 1991) and concurrently (Beitchman, Hood, Inglis, 1990; Cohen, Menna, Vallance, et al, 1998; Tallal, Dukette, & Curtiss, 1989; Stevens & Bliss, 1995). Three main theoretical arguments have been constructed to explain the links between language and social impairments: a general information processing impairment (Bishop, 1992; Johnston & Ellis Weismer, 1983; Siegel, Lees, Allan & Bolton, 1981), inadequate opportunity for social learning resulting from impoverished language (Rice, Sell & Hadley, 1991), and an underlying impairment in the social cognition domain that leads to concurrent impairment in language and social skills (e.g. Locke, 1993).

To date there is evidence to support each theory and no single explanation of the relationship between social and linguistic impairment has been convincingly presented. One study did try to compare two of theoretical approaches in six-year-old children (Redmond & Rice, 1998). After comparing Childhood Behaviour Checklist scores for 17 speech and language impaired children and 20 age-matched controls they reported that the speech and language impaired children SLI children received ratings in the normal range and more similar to their typically developing peers than to psychiatric populations. Moreover, they argued that significant differences between parental and teacher ratings suggested social problems were a function of social setting rather than the result of an impaired social cognition domain.

Further support for the view that language difficulties do not necessarily stem from early social impairment comes from the observation that children with Down syndrome who do fail to develop language skills do not show deficits in nonverbal, symbolic or social difficulties early in life (Sigman & Ruskin, 1999). These studies support the validity of speech and language delayed children as a group with distinct social skills to those with autistic spectrum disorders.

The valid distinction between the two groups of children is also supported by data comparing the behavioural patterns of autistic spectrum children and speech and language delay children. Loveland and Landry (1988) found that there was a wider range and greater severity of verbal and non-verbal communication in autistic spectrum than in speech and language delayed individuals. In fact, five-year old speech and language delayed children were very similar to two-year old typically developing children (both groups had similar language levels) suggesting that any socio-communicative impairment in speech and language delay may be better described as *delay* as opposed to the *deviant* behaviour seen in autistic spectrum disorders. Similarly, Lord (Lord, 1995; Lord et al, 1993) reported that the ADI-R and ADOS-G could reliably distinguish the two groups at ages as young as two years old. Therefore, it seems valid to view these two groups of children as reliably distinct in the nature of their developmental difficulties.

There were several specific advantages of recruiting speech and language delayed children as a comparison group[8]. First, any subsequently observed group differences in cognitive performance could not be a simple function of experiencing a communication impairment. Second, children with speech and language delay were likely to follow a general cognitive developmental trajectory more similar to that of the children with autistic spectrum disorders than other clinical groups or non-clinical groups might. This made them a better comparison group for this longitudinal study of development over time. Third, the recruitment of two clinical groups meant that any effect of exposure to clinical services or a developmental disorder would be similar for both groups. Fourth, the children with speech and language delay represented a relatively homogeneous clinically important group about whom much is still to be learnt. This study could therefore begin to provide some preliminary information about the cognitive functioning and behavioural symptomatology in very young children with speech and language delay. A final advantage of this comparison group was that they provided a conservative test of the executive dysfunction hypothesis of autism: spurious significant results were less likely to be reported.

[8] Throughout this thesis the SLD group will be referred to as a comparison group rather than a control group to reflect the positive contribution made to the literature by this choice of a relatively homogeneous clinical group.

However, it must be acknowledged that a conservative test may be accompanied by a loss of sensitivity.

2.6 Summary and Aims

Children with autistic spectrum disorders have been shown to experience difficulties planning, generating ideas and switching rapidly from one set of rules to another. However, they do not appear to be impaired in inhibiting one response. Preliminary suggestions indicate that this profile of executive skill may distinguish autism from other developmental disorders. Nonetheless, the executive dysfunction account of autistic spectrum disorders is weakened by the proposition that very young children with autism may not demonstrate executive difficulties and by the small number of investigations looking for a relationship between this cognitive function and its most likely behavioural sequelae - repetitive behaviour – in young children.

Measuring executive function in preschool children has been done (see Chapter 1) but it is difficult. Issues such as the verbal competence of children and the attention span of very young children have a substantial impact on the tasks that can be administered, the procedure followed and the reliability of the data. Despite these practical and methodological concerns it is of fundamental importance to the executive function hypothesis that these skills are assessed in very young children with autistic spectrum disorders. This thesis endeavours to study the executive function performance of very young children with and without probable autistic spectrum disorders. Through cross-sectional and longitudinal assessment of preschoolers referred for autistic spectrum disorders and speech and language delay the thesis aims to a) establish whether very young children with probable autistic spectrum disorders display executive deficits in comparison to a clearly defined control group, b) rigorously assess the question of a relationship between executive function skill and autistic symptomatology and c) build a picture of executive skill development in both groups of children over a 12 month period.

To this end, a group of very young preschoolers who have been referred to speech and language therapists (or health visitors) for complex communication difficulties indicative of autistic spectrum disorders were recruited. The comparison group were a clearly defined group of children who have been referred for speech and language delay without the added complexities indicative of autism. Thorough diagnostic assessments were administered to all children to assist clinical group decisions and to provide detailed symptom information. All children were given a battery of executive function tasks chosen to tap several component skills with the constraints that the tasks need to be appropriate for relatively low developmental abilities and with minimal verbal demands. Children recruited to the project were followed up after 12 months and the diagnostic and executive measures re-administered.

Chapter 3 assesses whether very young children with probable autistic spectrum disorders demonstrate executive dysfunction when compared to children with speech and language difficulties. This comparison is made both on initial recruitment and at follow-up. Additionally, if appropriate, the developmental trajectories of executive function skill in both groups are explored. Chapter 4 focuses on the repetitive behaviours reported for both groups, and the change over time in quantity or severity of these behaviours. Chapter 5 looks for any relationship between repetitive behaviour and executive function skill at both time points. The final chapter provides a summary and discussion of the overall thesis findings.

Chapter 3

Executive Function Skill in Very Young Children with Autistic Spectrum Disorders

One prediction of an executive dysfunction hypothesis of autism, as already discussed in Chapter 2, is that specific executive deficits should onset early in the child's development. If executive difficulties do indeed underlie autistic symptomatology then children who are already displaying autistic behaviours should demonstrate impairment on executive tasks when compared to young children with communication delays who do not display definite autistic symptomatology. This prediction was examined by comparing the executive function performance of a group of young children with autistic spectrum disorders with a group of children with speech and language delay but no clear autistic symptomatology. The performance of both groups was assessed twice, with a 12-month gap. This chapter focuses on group comparisons at Time 1 and Time 2 separately to establish if there is a specific deficit in executive performance in the ASD group at the ages of three and/or four years.

3.1 Recruitment

Literature and clinical experience suggests that parents often report developmental concerns that may indicate autistic spectrum disorders at around two years of age (e.g. Howlin & Moore, 1997). By recruiting a sample of children through speech and language therapists and their colleagues it was hoped to target a very young group of children who were likely to go on to receive a diagnosis of autistic spectrum disorder or speech and language delay.

Reliable diagnoses of autism can be made by 2 or 3 years of age (e.g. Baird, Charman, Baron-Cohen, et al, 2000; Cox et al, 1999; Lord, 1995) but the average age for a diagnosis is 5 years, and 11 years for Asperger's Syndrome

(Howlin & Moore, 1997). Given the young age of children targeted by this project it was likely that many children would not have yet received a clinical diagnostic label. This had a significant impact upon the design of the project and the procedures followed to recruit and group children.

Once Multi-Regional NHS Ethical approval was obtained, clinicians based in ten districts in the north-east of England agreed to be involved with the project[1]. A named research clinician was identified within each district and local ethical approval obtained. This research clinician, and their clinical colleagues, then reviewed current cases and waiting lists to identify children who might be suitable for inclusion in the project. Criteria for inclusion were a chronological age of 30 to 40 months and a referral for communication and/or social difficulties. Exclusion criteria were severe global developmental delay, severe birth complications or other known diagnosable severe medical conditions that might be of aetiological significance.

The clinician responsible for each identified child then described the project to the family (in many cases the research clinician and responsible clinician were in fact the same person). If the family stated they were interested to hear more about the project their name was passed on to the author. Families were provided with full details of the project by the author before written consent for the child's participation was requested[2].

Behavioural information from the Autism Diagnostic Interview - Revised (Le Couteur, Rutter, Lord, et al, 1989; Le Couteur, Rutter & Lord, unpublished; Lord et al, 1994), Autism Diagnostic Observation Schedule - Generic (DiLavore, Lord & Rutter, 1995; Lord, Rutter & Goode, et al, 1989; Lord et al, 2000) were combined with other clinical information to form a best estimate clinical judgement at both time points. The author had been trained in the administration of the ADI-R and ADOS-G and attended regular reliability sessions within a team of researchers and clinicians (inter-judge

[1] The districts involved were Newcastle, Northumberland, North Tyneside, South Tyneside, Sunderland, Durham, South Durham, Middlesborough, Stockton and North Yorkshire

[2] The responsible clinician for the child retained clinical responsibility at all times during the project. Information obtained through the assessments was fed back to that responsible clinician rather than to the family.

agreement over 70% in all sessions). The author and an experienced, expert, clinician reviewed videotapes of the child sessions and agreed upon a best estimate clinical judgement at Time 1 and Time 2. The expert clinician was blind to the Time 1 decision during the Time 2 decision process. Unless otherwise stated, the judgements made at Time 2 are those applied when diagnostic group comparisons are made in this thesis.

Time 1

Fifty-one names were passed to the principal researcher. Of these, five families did not give written consent to be involved in the project. The remaining 46 children (37 male and 9 female) were recruited to the project. At initial recruitment (Time 1) the children were between the ages of 29 and 46 months (mean age = 37.22 months, standard deviation = 4.79).

The socio-economic status of families with children with autism has been the focus of some research over the years. Initially, a higher occurrence of autism was associated with higher socio-economic status (Lotter, 1966; Lotter, 1967). This may have been a result of self-referral bias towards these families. The observation that this association has decreased in line with improved health services and general awareness of autism (Green, Campbell, Hardesty et al., 1984) suggests that the initial observations may have been an artefact. However, there remains an unexplained slight bias towards autism being diagnosed in more affluent families (Wolff, Narayan & Moyes, 1988).

In this study maternal education was taken as a measure of socio-economic status. Seven categories of qualification were formed: no qualifications, basic secondary school qualifications (such as GCSE, CSE, O Level), additional secondary qualifications (GNVQ, NVQ, BTEC), A Levels, Further education, Professional Qualifications (such as City and Guilds), Degree and Postgraduate Certificate of Education. Table 3.1 shows the number of mothers who had obtained qualifications in each category according to the diagnostic category their child belonged to at the second assessment period. Four mothers did not provide this information. Over 90% of both groups achieved a secondary school qualification. Therefore there is no reason to

believe that the families of children with autism were of a higher SES than the children with speech and language delay.

Table 3.1. Highest maternal qualification by diagnostic group

	ASD	SLD	PDD-NOS	Total
No Qualification	2	1	0	3
GCSE/ CSE/ O Level	5	12	4	21
GNVQ/ NVQ/ BTEC	3	0	0	3
A Level	3	2	0	5
Further Education/ College	2	1	1	4
Professional Qualification	0	1	0	1
Degree/ PGCE	3	1	1	5
Not known	2	1	1	4

Time 2

The author attempted to re-contact all families approximately 11 months after the first assessment period. Two families had moved away from the area and three others withdrew from the project when they were re-contacted. The remaining 41 children (32 male, 9 female) were followed up at Time 2 (89% of original sample). At Time 2 the children remaining in the project were within the age range 41 to 57 months (mean age = 48.63 months, standard deviation = 4.79).

Group allocation decisions were made upon the basis of a combination of ADI-R, ADOS-G, and available clinical information. An experienced and expert clinician assisted in this process. The group decisions made at Time 2 were used to form the diagnostic groups reported here[3].

Following this careful grouping procedure, children were allocated to one of three groups: autistic spectrum disorder (ASD), speech and language delay (SLD) or pervasive developmental disorder – not otherwise specified (PDD-

[3] The five children who were seen only at Time 1 were grouped according to the Time 1 decision. Obviously they provided no data for Time 2 analyses and therefore are only included in Time 1 analyses.

NOS). At Time 2, the ASD group contained 17 children (15 boys and 2 girls), the SLD group 17 children (11 boys, 6 girls) and the PDD-NOS group seven children (5 boys, 2 girls). The clinical decisions for the five children seen only at Time 1 were three in the ASD group (3 boys) and 2 in the SLD group (2 boys). To ensure the group comparisons were made between diagnostically clear groups, PDD-NOS children were excluded from all analyses reported in this chapter.

Table 3.2 provides descriptive information for the two groups to be compared in this chapter. Each child was administered three sub-scales of the Mullen Scales of Early Learning (Mullen, 1995) so that verbal and non-verbal ability could be entered as a covariate. Non-verbal mental ability was quantified as the age equivalent scored by the individual child upon the Visual Reception sub-scale and verbal mental age was the mean of Receptive and Expressive age equivalents obtained by each child. If a child was missing receptive or expressive data (no child missed both) the overall group mean was inserted (Clark-Carter, 1997; Howell, 1997). At Time 1 this procedure was applied to three children: two ASD (one missing receptive language and the other expressive language) and one SLD (missing receptive language). At Time 2 receptive and expressive language data were obtained for every child.

Two-tailed parametric t-tests revealed that the two groups had similar chronological ages (CA) and non-verbal mental ages (NVMA) to each other at both time points (CA; Time 1; $t_{(37)} = -1.68$, n.s.; Time 2; $t_{(32)} = -1.12$, n.s.: NVMA; Time 1; $t_{(37)} = 1.79$, n.s.; Time 2; $t_{(32)} = 1.94$, n.s.). However, the groups differed significantly on the verbal mental age (VMA) variable at both times (Time 1; $t_{(37)} = 2.49$, $p<0.05$; Time 2 $t_{(32)} = 3.31$, $p<0.05$). The ADI-R and ADOS-G algorithm scores for each domain were significantly different between groups at both time points (all $p<0.05$). Whilst it is theoretically possible that the two groups of children represented individuals from the same broad spectrum of communication impaired children, the significantly different symptom scores supports the validity of forming two groups on the basis of expert clinical judgement.

Table 3.2 Descriptive characteristics for ASD and SLD groups based upon Time 2 decisions. Age and ability scores are given as mean age equivalent in months (sd); ADI-R and ADOS-G scores are given as mean algorithm score for each domain (sd).

		Time 1		Time 2	
		ASD	SLD	ASD	SLD
	n	20	19	17	17
Chronological Age		038.3 (4.5)	35.7 (4.8)[b]	49.2 (4.3)	47.4 (5.2)[b]
Verbal Mental Age		19.9 (9.5)	26.1 (5.5)[c]	26.6 (9.2)	35.6 (6.5)[c]
Non-verbal Mental Age		26.9 (6.5)	30.7 (6.6)[b]	33.8 (9.9)	39.5 (7.3)[b]
ADI-R[a]	Socialisation	19.1 (6.8)	4.4 (5.0)[c]	20.2 (5.7)	3.1 (2.7)[c]
	Communication (verbal)	12.7 (0.6) n=3	2.8 (2.7)[c] n=8	12.9 (4.6) n=7	3 (2.8)[c] n=12
	Communication (non-verbal)	9.7 (3.1) n=17	4.4 (3.6)[c] n=11	9.6 (3.1) n=10	3.8 (2.4)[c] n=5
	Repetitive	4.9 (1.5)	0.9 (1.0)[c]	5.1 (2.1)	1 (1.2)[c]
ADOS-G[a]	Socialisation	7.4 (3.5)	0.4 (1.2)[c]	9.1 (2.3)	0.8 (1.4)[c]
	Communication	4.8 (2.3)	0.7 (0.9)[c]	5.3 (2.4)	1.9 (1.4)[c]
	Repetitive	3.1 (1.4)	0.3 (0.6)[c]	3.2 (1.3)	0.9 (1.0)[c]

[a] Higher scores reflect greater abnormality of behaviour
[b] $p>0.05$
[c] $p<0.05$

3.2 Executive Function Tasks

Designing tasks that measure a single cognitive skill in isolation is very complex and probably impossible. Tasks requiring any kind of response implicate motor or verbal response mechanisms by definition in addition to any other executive skill the task may recruit. Furthermore, the introduction to executive function skills in Chapter 2 has also demonstrated the close inter-relation between executive skills such as inhibition, generativity, working memory and planning.

A multitude of tasks have been used in the literature to assess many of these skills and some of these tasks (such as Go-NoGo and Stroop) seem to have gained general acceptance as tasks of inhibitory control. However many of these tasks require verbal skills more advanced than could be expected of the children in the current study. In particular, the fluency tasks of generativity and many planning tasks can place substantial verbal demands upon children. The executive function tasks that exist in the literature for young or lower ability children are predominantly measures of inhibition-and-implementation or working memory. Developing non-verbal generativity tasks has proven exceptionally difficult since even instructions such as "produce as many responses as possible without repeating previously produced responses" require a substantial level of comprehension skill. For this reason tasks of generativity were unable to be developed for the current study.

At Time 1, when the children were three years of age, the tasks employed in the current study focused on inhibition-and-implementation and working memory. By administering tasks frequently used in literature with young children this meant the current study would be suitable for comparison with the existing knowledge of typical and atypical executive function development. When the children were a year older two further tasks were developed to tap attentional set-shifting and planning at Time 2. These two tasks were an evolution of tasks used in the existing literature.

3.2.1 Time 1

Four tasks were presented. The tasks were chosen to be age and ability appropriate. The tasks were piloted with normally developing children in nursery (mean age 31 months). No ceiling or floor effects were found on the tasks.

A-not-B Task (e.g. Diamond et al., 1997; Diamond, 1985). This task has been frequently used to assess inhibition of a previously rewarded response and working memory in infants and young children. The participant watches as a reward is hidden and then a few seconds later must retrieve it.

Materials:

Two identical red boxes (15x15x15cm) were placed in front of the child. Each box had one side that opened on a hinge; the box was always presented with this side towards the child so that the 'door' opened down towards the table. A small toy (e.g. train, frog, car) was hidden in one of the boxes. A white metal tray was placed in front of the boxes (45x30cm) as a screen. A small cloth (15x15cm) was used to cover the toys in pre-testing.

Procedure:

The pre-testing phase began with the child being asked to choose a toy to play with. This toy was then placed on the table and partially hidden and the child was asked to retrieve it. If the child successfully retrieved the toy they were allowed to play with it for a brief period. The toy was then placed on the table and fully covered and the child was asked to retrieve it again. Pre-testing was repeated a maximum of four times. If the child was unsuccessful on the pre-test phase the task was discontinued. If the child was successful once on the pre-test phase, the test phase began.

Two identical boxes were placed in front of the child, one to either side of the child's centre line. The toy that the child had chosen was placed in one of the boxes in view of the child (the first side of hiding was counterbalanced across children). Both boxes were closed simultaneously and a screen placed in front of the boxes for approximately five seconds. The examiner maintained the child's attention during this short period by counting with an excited facial expression. The screen was removed and the child asked to retrieve the toy. As soon as the child opened one box the examiner held the other box. If the child found the toy the examiner said *"Well done!!"* and the child was allowed to play with the toy for a short time. If the child picked the incorrect box they were not able to play with the toy and the experimenter said *"Oh no... [pause]. I've found it! See if [name] can find it! Try again!"* The toy was hidden in the same box until the child had successfully retrieved the toy on two consecutive trials. Then the task was repeated with the toy placed in the other box (a reversal). The reversal process was repeated a maximum of four

times. The experimenter worked to maintain the child's attention for four reversals (e.g. by allowing the child to choose a new toy) but some children became bored or refused to continue before they had attempted four reversals.

Scoring:

Firstly, the percentage of reversal trials on which each child successfully retrieved the toy was calculated (number of successful reversal trials over number of reversal trials attempted). If a child incorrectly repeated a search to the previous box this was classed a perseverative error. The percentage of perseverations (number of perseverative errors made over number of opportunities to perseverate incorrectly) was calculated since the pattern of responding was different for each child. As a measure of continued failure and inability to change performance on the basis of feedback, the longest run of perseverative responses made by each child was also recorded.

A-not-B Invisible Displacement Task (e.g. Diamond et al., 1997). This task was devised by Piaget (1954) as the following step in his object permanence series after the A-not-B Task. The materials and procedure for this task are similar in many ways to the A-not-B Task.

Materials:

Two identical silver boxes (15x15x15cm) were used that opened in the same manner as the boxes in the A-not-B Task. The same metal tray and selection of toys were available as in the A-not-B Task.

Procedure:

The procedure for this task is the same in many ways as that for the A-not-B Task. The main difference is that the child watches a toy being hidden in a centrally located box which is then moved to one side. When the screen is placed in front of the box the other identical box is placed to other side of the child. Once more the child is asked to retrieve the toy.

As before, the child was encouraged to choose a toy to play with. In the pre-testing phase the toy is placed in one central box and then moved to one side (side of presentation counterbalanced across participants). The screen is placed in front of the box whilst the examiner counts to five before it is removed and the child asked to retrieve the toy. This is then repeated to the other side. Note that only one box is present during the pre-test session. Pre-testing was repeated a maximum of twice. If the child was successful to both sides testing began, otherwise the task was terminated.

On each trial the chosen toy was placed in a centrally located box, the box closed and then moved to one side (the first trial was always to the same side as the final pre-test move). A screen was placed in front of the box and, whilst the examiner counted to five, a second identical box was placed to other side of the child. The screen was removed and the child asked to retrieve the toy. As in the A-not-B Task, the child was allowed to play with the toy when he successfully found it but not when unsuccessful. When the child successfully retrieved the toy on two consecutive reaches, the toy was placed in the other box and the task repeated (a *reversal*). The process was then repeated to a maximum of four reversals but, as before, not all children attempted four reversals.

Scoring:

This task was scored in the same ways as the A-not-B Task.

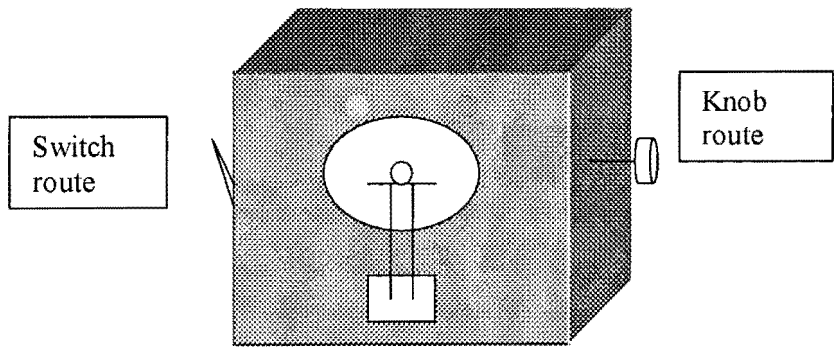
Detour Reaching Box (Hughes & Russell, 1993; Hughes, 1993). This task required the child to inhibit the prepotent response to reach directly towards the marble and to implement a rule that they had been shown.

Materials:

A box was constructed following the descriptions in the original paper by Hughes and Russell (1993). The box was 30x30x30 cm and made of aluminium. In its front was a centrally located, circular hole, 15cm in

diameter, cut in a perspex square. Inside the box was a platform upon which a marble rested. On either side of the front opening were photoelectric cells which generated an infrared beam. If this beam was broken, a trap door automatically operated and the marble fell from view. Two lights (yellow and green) were positioned on the front of the box. When the green light was lit, turning a knob on the right hand side of the box would project the marble down a chute and into a catch tray at the front of the box. When the yellow light was lit, the knob no longer operated. At this stage, the operation of a switch on the left hand side of the box disengaged the infrared beam and the participant could reach through the front opening to retrieve the marble (see Figure 3.1 for a diagram of the apparatus).

Figure 3.1 Detour Reach Task apparatus (Hughes & Russell, 1993)



Procedure:

The child was presented with the apparatus and the marble pointed out to them. They were encouraged to reach in for the marble. On reaching for the marble, it fell from view and the experimenter said "*Oh no! It's gone. I'll show you how to get it*". The experimenter reset the apparatus and demonstrated the knob route for the child. When the marble fell into the catch tray the experimenter applauded and encouraged the child to handle the marble. The apparatus was then reset and the child encouraged to retrieve the marble: "*Can [name] get the ball?*". If the child successfully retrieved the marble, praise was given, the marble replaced and the child encouraged to

repeat the task. This was repeated until the child had achieved a criterial run of three consecutive correct responses, or 15 trials had been administered.

If the child made an incorrect response on the task the examiner said "*Oh no! Let's try again*!". The apparatus was reset and the child given a verbal prompt: "*Remember the knob*". If the child continued to fail the prompt increased to a verbal prompt accompanied by a point, and finally to a demonstration of the successful route. A successful response at any stage meant the next time a prompt was required the cycle of verbal, verbal and visual, demonstration would begin again.

If the child achieved a criterial run on the knob route, the rule was changed. The child was shown the yellow light was now illuminated and told the rule had changed. They were encouraged to try the knob route so they were clear it no longer worked. Then the experimenter demonstrated the switch route with the instructions "*Switch the switch, then reach in*". After the demonstration the apparatus was reset and the child encouraged to retrieve the marble. As on the knob route, the child was encouraged to retrieve the marble and praised if they were successful. The same prompting strategy was invoked if required. The switch-reach route was administered until a criterial run of three consecutively correct reaches was achieved or 15 trials had been administered.

Scoring:

Children were classed as passing or failing this task. Making three consecutive correct retrievals of the marble was classed as a pass. The number of reaches required to reach a criterial run was recorded for the passers. Correct responses following a prompt were not included in a criterial run.

Errors made by each participant were also recorded. For the knob route, the total number of errors, the longest run of errors and the number of direct reach errors were recorded. On the switch-reach route four categories of error types were formed: failure to inhibit direct reach, perseveration to knob rule, confusion between the two rules, and incomplete or unsuccessful attempt. These categories were mutually exclusive. Table 3.3 describes the error types

included in each category. Total number of errors and longest run of errors were also recorded. The longest run of errors was not reported in this thesis due to the difficulty in interpreting this variable: a run of errors could be formed of a variety of errors and therefore could not be interpreted as perseveration to any particular response.

Table 3.3 Error types on the switch-reach route of the Detour Reach Task.

Error Category	**Error description**
Failure to inhibit direct reach	Direct reach error
Perseveration to Knob route	Knob and no reach
Confusion between the two routes	Knob-reach, switch-knob-reach, switch-knob-no reach, knob-switch-reach, knob-switch-no reach, reach-switch
Incomplete or Unsuccessful attempt	Switch-only and repeated switching so that a the subsequent direct reach triggered the trapdoor

Three Boxes Task (e.g. Diamond et al., 1997). This task is a working memory task requiring participants to keep a track of which boxes they have already opened. Two versions of the task were administered, one in which the hiding locations remained stationary between reaches and the other where they were scrambled after every reach.

Materials:

Three differently coloured plastic cups with a height of 9 cm were baited with Smarties, chocolate buttons, crisps or raisins depending on child preference and dietary restrictions. A different selection of cup colours was presented for each version of the task. A white metal tray (45x30 cm) was used as a screen.

Procedure – Stationary version:

Three differently coloured plastic cups were placed in a straight line in front of the child. The cups were positioned so that the rims of adjacent cups were touching in order to maintain a constant distance between the cups. The child watched as the experimenter placed one reward under each cup. A white screen was then placed in front of the cups whilst the experimenter counted to five. The screen was removed and the child encouraged to search one cup for a reward. As soon as the child searched one cup the experimenter held the other cups to prevent the child also searching them. After each search the cups were again hidden by the screen before the child could search again (regardless of whether the child had successfully found a reward). In the stationary version the cups remained in the same relative locations throughout the task. The task continued until the child had retrieved all three rewards, had lost interest or 15 trials had been administered.

Procedure - Scrambled version:

The procedure was exactly the same for this task except that the experimenter moved the relative locations of the cups around while the screen blocked the child's view between each reach (regardless of whether the reach had been successful or unsuccessful).

The order in which the two versions were presented was counterbalanced across participants; each child received both the stationary and the scrambled versions of the task.

Scoring:

For both versions of the task an efficiency ratio was calculated where the number of rewards retrieved was divided by the number of reaches made. The optimal efficiency ratio was 1. The errors made by each child were also recorded. The longest run of perseverative reaches to a location or cup was recorded. For the scrambled version the longest run of perseverative responses to location and cup were recorded separately; for the stationary version location and cup were indistinguishable.

3.2.2 Time 2

The following tasks were administered exactly as in Time 1: A-not-B with Invisible Displacement Task and the Detour Reaching Box. The Three Boxes Task became the Six Boxes Task. All these tasks were scored in the same manner as at Time 1.

The novel tasks for this time period were piloted in nurseries with typically developing children of approximately the same expected ability level as the sample. This pilot work confirmed that the tasks would not have been appropriate for administration at Time 1 and found no floor or ceiling effects.

Sorting Task This task was developed to broaden the range of executive skills assessed in the study by tapping attentional set-shifting skills. It was influenced by card sorting tasks already used with typically developing and Down syndrome individuals (e.g. Hughes, 1998a; Zelazo et al, 1996).

There are several key points that make this task a development of previous work. In Hughes' work children had to identify which cards teddy would 'like' or 'not like' according to the rule. The present task eliminated this extra character from the task and swapped the cards for toy bricks: the child's job would now be to post the brick in the correct hole according to a given rule. The present task also made the possible sorting rules explicit by labelling the dimensions and demonstrating correct sorts. This was different to Hughes' work but the same as Zelazo's. Unlike Zelazo et al, the children were trained directly before each rule-test to diminish working memory demands. There was also a total prop shift between the two rules. This total shift meant the child must make both an intra-dimensional and extra-dimensional shift to be successful and eliminated the possibility of perseveration to a specific colour, shape, or brick Finally, like Zelazo and colleagues, the current task included a third stage when the rule explicitly switched from trial to trial to assess flexible shifting, but the task differs from Zelazo in that the rule was not repeated before each trial as this was felt too strong a prompt.

Materials:

Two wooden boxes (30x26x16cm) each had two square holes cut on the centre line (each hole was 8x8 cm with 7cm between holes). Fabric was attached to the box so that the shape of the hole was not visible; a slit was cut in the fabric to allow objects to pass through the hole into the box. On each box a coloured brick shape was glued above each hole (See Figure 3.2). On one box these shapes were a red cylinder (diameter 3cm, length 6.5cm) and a blue rectangular cuboid (3x3x7 cm). On the other box the shapes were a yellow pyramid (height 6.5 cm, base 2.5 cm square) and a green square cube (3x3x3 cm). These prototypical shapes remained in place throughout the task. Associated with each box were fourteen coloured bricks (red and blue cylinders and red and blue rectangular cuboids for the first; yellow and green pyramids and yellow and green square cubes for the second).

Figure 3.2 Shape Sorting Task apparatus (note no shape information about holes provided)

Procedure:

The participant was asked to post coloured bricks into the appropriate holes to demonstrate colour and shape sorting. The presentation orders of sorting rule and box were counterbalanced across children. The experimenter explained the first sorting rule with verbal and gestural instructions: "*This is a circle,*

this is a square. Circles go in here, squares go in here." Or "*This is yellow, this is green ....etc*".

Training involved four trials when each of the brick types associated with the box were given to the child for sorting. On these trials the examiner pointed out the salient feature: e.g. "*it's a circle, where do circles go?*" Children who failed one or more of the training trials were given a second opportunity to pass the training trials. The task was terminated for children who failed the second training trials; testing trials were administered to children who passed the training trials.

A maximum of 10 testing trials were administered when the experimenter handed the child one block at a time for them to post in the appropriate hole (brick order was random). No support or feedback was given to the child on these trials. If the child made two consecutive incorrect sorts they were verbally reminded of the rule: e.g. "*Remember, circles go in here and squares in here*". If the child made five consecutive correct sorts the second sorting rule was administered.

The first box was placed to one side of the child and the second box positioned in front of the child. The second sorting rule was presented in the same way as the first (training and testing). If the child made five consecutive correct sorts on this rule, the third and final rule was administered.

For the third rule, both boxes were placed in front of the child. The child was reminded of the sorting rule associated with each box. The experimenter said "*Now we're going to play the [first rule presented] game. Where does this go?*" and presented the child with a brick for them to sort according to the first rule. Bricks and boxes retained their association with a specific rule throughout the task. After one trial, the experimenter moved to the second box and said "*Now we're going to play the [second rule] game. Where does this go?*" The sorting rule and box was alternated after each sort. A maximum of 14 trials were administered in this way (i.e. seven to each rule). Note that no further training was provided at this final stage.

Scoring:

The number of correct sorts made for each rule was noted. A strict criterion of five consecutively correct responses was required to 'pass' a sorting rule. This ensured that a passer must have made at least one non-identical sort (in each set four blocks were exact matches and six were non-identical matches). Children were classified as passing or failing Rules 1, 2 and 3 (rule switching). For children who failed, whether they failed at training or testing phases of the rules was recorded.

The number and type of errors made on each sorting rule was recorded. On Rule 2 and Stage 3, errors were recorded as a perseverative error to the previous rule or as unclassified (of no discernible sorting pattern – e.g. a yellow triangle in the green square hole).

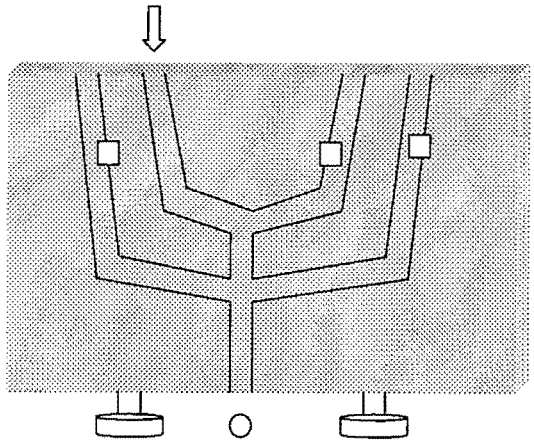
Marbles Task This task was designed to measure elementary planning skills in young children. The child was required to drop a marble down the only available route that would result in the marble exiting the apparatus. Bruce Hood (1995) used a task involving Perspex tubes and simple problem solving. On his task the child had to use a tool to push from one end of the tube to dislodge the toy. If they pushed from the correct end the toy came out but if they pushed from the other end it didn't. The current task drew from Hood's work but was designed to resemble the marble routes commonly given as children's toys. There is also scope with the current task to make it more complex as the child develops.

Materials:

A piece of wood 42x25x3 cm was mounted on two feet so that it was 2 cm off the ground. Four routes were cut through the wood (see Figure 3.3), each of which could be blocked by one of four moveable wooden blocks. Each route was sufficiently large that a marble could be dropped all the way through when no wooden block was in place. A piece of Perspex was attached to either side of the wood so the routes were visible. One piece of Perspex was

attached on hinges so the experimenter could open it to reposition the wooden blocks.

Figure 3.3 The Marbles Task apparatus (white squares represent the wooden blocks)



Procedure:

Pre-testing began when the participant was presented the apparatus with no blocks in position. The experimenter encouraged the child to drop a marble down each route and to observe that it came out the bottom of the apparatus. The apparatus was then removed and wooden blocks placed in all four routes. The experimenter pointed the blocks out to the child and encouraged them to drop the marble in each possible route. This was to ensure the child understood the blocks were not permeable. Then the experimenter removed one of the blocks (the route that was unblocked was randomly determined). The child was encouraged to drop the marble down the clear route so it exited at the bottom. If this was correctly done, the child received an ink stamp on a certificate and testing began.

A maximum of twelve trials were administered. On each trial the apparatus was hidden whilst the experimenter manipulated which route was clear by moving the wooden blocks according to a predetermined randomised order (with the proviso that the first trial was never the same route as the final pretest trial). The apparatus was then placed in front of the child and they were encouraged to drop the marble through the clear route. The experimenter

gave no assistance at this stage. The child had a certificate with 12 spaces on which an ink stamp was placed for each successful solution of the problem. Every time the child erred, the examiner said "*Oh no, it's stuck*" and pointed out the correct route. The trial was not re-administered following the examiner's comments. However, if the child made two consecutive errors the second of these trials was repeated. The examiner then helped the child to make a correct response. This was not marked as correct, but the child received a stamp to reinforce the response. After this, the remaining trials continued.

Scoring:

The percentage of correct trials was calculated for each child. All errors were recorded but in terms of the executive dysfunction hypothesis the errors of interest were an incorrect choice of the same route they had just successfully chosen (an incorrect perseveration) and the longest run of these incorrect perseverations. The longest run of errors regardless of route was also noted for each child.

3.3 Procedure

Each assessment period (Time 1 and Time 2) comprised two child sessions and one parent/caregiver session. Child sessions lasted 1 to 1½ hours and caregiver sessions between 1½ and 3 hours. The ADOS-G was administered in one child session and the Executive Function battery in the other. Administration of the Mullen Early Learning Scales was begun in the first child session and concluded in the second child session. A caregiver session involved the ADI-R; the RBQ was left with the caregiver following this session. To guard against systematic order effects, no fixed order of sessions was followed. Although the ADI-R and ADOS-G were administered at both time points only data from Time 2 was used in forming the diagnostic categories (with the exception of five children who did not provide this data at Time 2).

3.3.1 Time 1

The relationship built between author and family, and the time spent in the family home, meant it was impractical for the author to remain blind to the nature of the child's difficulties. However, the ADOS-G was administered before the ADI-R in half of the cases. For these children, the author only received very brief referral information before she administered the direct diagnostic assessment. The ADI-R was conducted before either child session in 35% (16/46) of the cases.

The author administered all interviews and assessments with three exceptions. Two children were administered the ADOS-G by an experienced clinician with whom the author had established reliability. One ADI-R was administered by an experienced clinician as part of her training on the interview schedule in the month before the family were recruited to the project. The family were unwilling to repeat the interview with the author and therefore she reviewed and coded the videotape of the training interview.

All interviews and assessments were conducted in the family home with two exceptions. One parental interview took place in a Child and Adolescent Service familiar to the mother, and one child was living (with her parents) at her grandmother's house due to the family home being refurbished. Interviews were always carried out with at least one parent of the child. The majority of parental interviews were conducted with the mother alone (28/46). A further 14 interviews were conducted with both parents present. In one case the primary care-giving role fell to the father and he was the respondent in the interview. In two cases the child's mother and grandmother were the respondents.

All three assessments were arranged to be within as short a period as possible. Seventy percent (32/46) of the families were seen all three times within one month (mean = 27 days). The remaining families, with one exception, were seen within two months. One child recruited to the project completely refused to co-operate with the author on any of four visits she made to the family home. Parental session data was available for this child. Unfortunately the

family moved away from the North-East after Time 1 assessment and could not be followed up at Time 2. Therefore no direct observation data was available for this boy.

The order of presentation of executive function tasks was partially randomised. The author ensured that the final task presented did vary across children, however she used her judgment to decide which task order would maintain each individual child's attention and motivation. The position in which each task was presented varied across children. For example, the A-not-B Invisible Displacement was the first task administered to 12 children and the final task presented to 16 children. There is no evidence from the data collected that the children who were administered the task first performed better than those who attempted it last. It cannot, therefore, be claimed that performance on this task was a function of when it was presented to the child.

Half the children received were administered the A-not-B Task with the toy hidden in the right hand box first, and half to the left. The same was true of the A-not-B Invisible Displacement Task. Half the children were administered the stationary version of the Three Boxes Task before the scrambled version, and half the opposite order.

3.3.2 Time 2

The child and parent sessions were constructed in the same way as Time 1. The three sessions were conducted over as brief a period as possible (mean = 15 days). The assessment period was greater than one month for only one child (35 days).

The mean gap between Time 1 and Time 2 assessments was 365+3 days. Two children were seen 14-15 months after the first assessment. In one case this was due to maternal pregnancy and the other maternal illness. One child was seen only 11 months after time 1 assessment. This child had been recruited to another research project that also used the ADI-R and ADOS-G. Rather than overburden the family it was agreed that the data from these instruments

would be shared between the projects. (The author had established reliability with the researcher for the other project who carried out these assessments).

The principal researcher conducted all interviews and assessments at Time 2 (with the one exception described above). To reduce experimenter bias that may have been introduced by the same author seeing the families and children at both testing periods a number of precautions were taken. No reference was made to the Time 1 ADI-R before or during the Time 2 ADI-R. The ADI-R was also administered in full at Time 2; family history was rechecked and all behavioural questions were re-administered. It was not possible to conduct the ADI-R with two families who remained with the project at Time 2.

The majority of respondents at Time 2 were mothers (27/39). A further eight ADI-R were carried out with both parents present, two interviews were conducted with fathers only. Nearly all the children were seen for at least one direct assessment session before the parents were interviewed (38/41). The ADOS-G was administered in the first session for 71% of these children.

3.4 Statistical Analysis

This chapter compares group performance on key executive function task variables. The executive function variables were chosen to reflect overall performance and rates of perseverative errors in line with the current literature.

As performance on executive function tasks may be associated with age and/or ability in younger and less able samples, preliminary analyses were conducted to explore the relationship between performance on each of the dependent variables and CA, VMA and NVMA[4]. Given that the VMA of the ASD group was significantly lower than that of the SLD group, it was particularly important to ensure that any differences between the groups were not attributable to differences in verbal ability. These analyses revealed

[4] Parametric correlations were conducted since the variables did not violate the assumptions of parametric analysis.

surprisingly few significant findings (only 10 out of 99 correlations were significant) with CA and VMA showing a significant pattern of association with performance for just one task each and NVMA showing significant associations with performance across four tasks. Given that VMA and NVMA were also significantly inter-correlated (Time 1: $r=0.82$, $n=39$, $p<0.001$; Time 2: $r=0.61$, $n=34$, $p<0.001$), it was not appropriate to enter all three terms as covariates in an analysis of covariance (ANCOVA). Using covariates which are themselves correlated not only adds nothing to the adjustment of the dependent variable, but also subtracts degrees of freedom from the error term while not removing commensurate sums of squares for errors (Tabachnick & Fidell, 1996). Instead, ANCOVA was only used when correlational analyses had shown age or ability to be significantly associated with the dependent variable in question. Where more than one index was significantly associated with a single performance variable (Marbles Task only), the variable with the greatest correlation with the dependent variable was entered as the covariate (following Tabachnick & Fidell, 1996). This approach served both to ensure that the assumptions of ANCOVA were not violated and to maximise the statistical power of each comparison. Where there was no association between the dependent variable in question and age, verbal mental age or non-verbal mental age, univariate analysis of variance (ANOVA) was undertaken.

Performance on the Sorting Task was classified into four categories and therefore was categorical data. Similarly, the number of children passing the Detour Reach routes was categorical data. These variables were therefore analysed with chi-square or likelihood ratio methods (depending on the size of the expected values). All continuous dependent variables were examined a priori to assess whether they met the assumptions of parametric tests. On two occasions the sample sizes available for a particular task were so small that parametric analysis was inappropriate (Detour Reach Switch-Reach Route and A-not-B Invisible Displacement Task at Time 1). On these occasions non-parametric Mann-Whitney U Tests were undertaken. All other dependent variables did not appear to violate the parametric assumptions.

A priori power analyses were conducted before the participants were recruited. An average effect size of 1.0 has been reported for individuals with ASD on executive function measures (Pennington & Ozonoff, 1996). On this basis, samples of 17 were required for a power of 0.8 with a significance level of 0.05 (Clark-Carter, 1997).

3.5 Time 1 Results

Key variables were selected for each task on an a priori basis. For each task a measure of overall performance was selected. As executive deficits are expected to lead to specific classes of errors, additional measures of errors and, in particular, perseverative errors were analysed.

3.5.1 A-not-B Task

Fourteen children in the ASD group and 16 in the SLD group attempted this task. All other children refused to cooperate on the task. The key variables were percentage of reversals correct, percentage of perseverative errors and longest run of perseverative errors. Descriptive information for these variables is presented in Table 3.4. None of these variables were significantly correlated with age, verbal mental age or non-verbal mental age.

Table 3.4 A-not-B Task: Key variables

	ASD		**SLD**	
	Mean	**(SD)**	**Mean**	**(SD)**
	n=14		n=16	
% reversals correct	71.43	(30.26)	78.13	(28.20)
% perseverative errors	26.05	(23.20)	17.28	(19.79)
Longest run of perseverative errors	1.33	(1.29)	1.0	(1.03)

Performance on this task was good, with both groups averaging over 70% of the reversal trials correct. Whilst this may reflect a ceiling effect on the task, it is important to note that the wide standard deviations demonstrate some children were not performing at ceiling. Univariate analyses of variance revealed no significant effect of group for the percentage of reversals correct ($F_{(1,28)} = 0.39$, n.s.), the percent of perseverative errors made ($F_{(1,28)} = 1.25$, n.s.) or the longest run of perseverative errors ($F_{(1,28)} = 1.03$, n.s.)

3.5.2 A-not-B Invisible Displacement Task

Nine ASD and 13 SLD children attempted this task. However, two ASD and four SLD children failed to attempt any reversal trials. Since the reversal trials are an integral part of the task, these children were excluded from the analysis leaving nine children in the SLD group and seven in the ASD group. Preliminary analyses indicated a significant association between NVMA and two of the three dependent variables (percent reversals correct $r=0.52$, $n=16$, $p<0.05$; percent of perseverative errors $r= -0.71$, $n=16$, $p<0.05$). The longest run of perseverative errors produced showed no significant pattern of association with age or ability. Due to the small number of children in each group, non-parametric analyses were conducted.

The key variables were the same as for the A-not-B Displacement Task. Group means and standard deviations for each variable are presented in Table 3.5. Mean performance on the reversal trials of this task was at or below chance levels for both groups. Although the mean percentage of reversals correct for the SLD group was almost double that of the ASD group, the within-group variation in performance on reversal trials was substantial for both groups. A Mann-Whitney U test revealed no statistically significant difference between the groups on this measure ($U=21.0$, $z= -1.17$, n.s.). Both groups continued to perseverate on around half of the possible opportunities; again there was wide within-group variation and no significant group difference ($U=33.0$, $z= -0.99$, n.s). Comparing the groups on the longest run of perseverative errors produced also revealed no statistically significant difference ($U=32.5$, $z= -1.79$, $p=0.07$).

Table 3.5 A-not-B Invisible Displacement task: key variables

	ASD		SLD	
	Mean	**(SD)**	**Mean**	**(SD)**
	n=7		n=9	
% reversals correct	26.19	(33.13)	51.85	(43.66)
% perseverative errors	54.33	(26.08)	41.53	(34.79)
Longest run of perseverative errors	2.56	(1.24)	1.54	(1.39)

3.5.3 Detour Reach Task

Knob Route

Sixteen ASD and 18 SLD children attempted this task. Nine ASD and fourteen SLD children passed by achieving a criterial run of three consecutively correct responses. Table 3.6 includes descriptive characteristics for the Knob Route performance variables. Preliminary analyses indicated no significant correlation between any of the performance measures and age, verbal mental age or non-verbal mental age.

A Chi-square analysis showed no significant group difference between the proportion of passers and failers (χ^2=1.79, df=1, n.s). An ANOVA revealed no effect of group on the number of trials taken to reach criterion ($F_{(1,21)}$ = 0.04, n.s.). The mean number of errors was small for both groups. An ANOVA revealed no effect of group on the number of errors made ($F_{(1,32)}$ = 2.29, n.s.). Therefore, performance on this part of the task was good and there was no indication that children in either group found it difficult to inhibit the prepotent response to reach directly towards the marble.

Switch-Reach Route

Two ASD children and three SLD children who had passed the Knob route refused to continue with this task. This left seven ASD children and 11 SLD children who had passed the knob route and attempted the switch-reach route. Table 3.6 provides group means and standard deviation for the Switch-Reach Route dependent variables. Preliminary analyses revealed no significant correlation between any of the dependent measures from this task and measures of age and ability.

Three children in each group passed this more complex route. Chi-square analysis revealed no significant effect of group ($\chi^2_{(1)} = 0.47$, n.s.) on passing the task. Given the small number of children who attempted this task, non-parametric Mann-Whitney U Tests were conducted on the remaining variables of interest. A Mann-Whitney U Test revealed no significant effect of group upon the number of trials required to reach criterion ($U=32.5$, $z= -0.65$, n.s.).

Perseveration on this task can be measured by the child's propensity to repeat the knob rule, or by their tendency to make direct reach errors. No child repeated the action required for the knob rule, however responses involving the knob, switch and a reach were fairly common. This seemed to reflect some level of confusion rather than an inhibitory control problem. These confusion errors were significantly more common in the ASD group ($U=16.0$, $z = -2.06$, $p<0.05$).

Table 3.6 Detour Reach Task, Knob and Switch-Reach Routes: Key variables

	ASD		SLD	
	Mean	**(SD)**	**Mean**	**(SD)**
Knob Route	n=18		N=16	
Pass (n)	9		14	
Number of trials to criterion	4.22	(1.72)	4.07	(1.82)
Errors				
Total errors	4.88	(5.07)	2.61	(3.62)
Switch-Reach Route	n=7		N=11	
Pass (n)	3		3	
Number of trials to criterion	7.0	(3.61)	7.33	(4.51)
Errors				
Confusion*	**3.71**	(2.21)	**1.55**	(1.37)
Direct Reach*	**2.67**	(1.75)	**6.18**	(3.25)
Total errors	6.43	(3.21)	8.0	(2.72)

* $p<0.05$

Direct reach errors also occurred regularly. Given the low rate of these errors observed for the knob route, this increase is likely to indicate a failure to inhibit the prepotent response under increased task demands. Non-parametric analysis revealed a significant effect of group on the number of direct reach errors made ($U=14.0$, $z= -2.23$, $p<0.05$). Group mean values (Table 3.6) suggested that the SLD group made direct reach errors more frequently than the ASD group.

Overall error rates did not differ significantly across groups ($U=28.0$, $z = -0.96$, n.s.). The differences between the performances of the two groups on this task were subtle and neither group showed a tendency towards continued perseveration.

3.5.4 Three Boxes Tasks

Stationary version

Seventeen children in the ASD group and 19 children in the SLD group attempted this task. Table 3.7 presents descriptive information for each group and each of the dependent variables. None of these variables were significantly related to age or mental age. An efficiency ratio (number of rewards retrieved/number of reaches made) was calculated as a measure of the efficient use of working memory. A score of 1 indicated that no errors had been made during the task: each search had been to a previously unchecked box. Overall performance for both groups was high and may represent a ceiling effect. An ANOVA revealed no significant difference between the groups on this measure ($F_{(1,34)} = 0.47$, n.s.).

Only seven ASD and six SLD children made any errors on this task. Therefore there was no benefit in further exploring error responses on this task.

Table 3.7 Stationary Version of the Three Boxes task: key variables

	ASD		**SLD**	
	Mean	**(SD)**	**Mean**	**(SD)**
	n=17		n=19	
Efficiency ratio	0.89	(0.22)	0.89	(0.18)

Scrambled version

Fifteen ASD and 19 SLD children attempted this task. Two ASD children who had attempted the stationary version became too disinterested in the task to attempt the scrambled version. Table 3.8 presents group means and standard deviations for each of the dependent variables. None of these variables were found to be significantly related to age of mental age.

Efficiency on the scrambled version was not as high as for the stationary version, but did not differ across groups ($F_{(1,32)} = 0.01$, n.s.). Twelve ASD and 15 SLD children made at least one error on this task. ANOVA revealed no effect of group on number of errors made ($F_{(1,32)} = 0.59$, n.s.). Because the mean numbers of error were low for both groups, there was no opportunity to analyse perseverative errors. This suggests that perseveration to location or colour is not a key feature of the children's performance despite an increase in the number of children making errors in this version of the task.

Table 3.8 Scrambled version of the Three Boxes task: key variables

	ASD		**SLD**	
	Mean	**(SD)**	**Mean**	**(SD)**
	n=15		n=19	
Efficiency ratio	0.65	(0.22)	0.66	(0.24)
Errors	1.73	(1.33)	2.21	(2.10)

3.5.5 Time 1 Summary

Across three of the four tasks administered at Time 1, the performance of the ASD and SLD groups was indistinguishable. There was no significant difference between the groups in overall performance or susceptibility to perseveration for the A-not-B task, the A-not-B Invisible Displacement task or the Three Boxes task. Performance of both groups on the A-not-B task and the stationary version of the Three Boxes task was so good that ceiling effects may have been observed. However the large standard deviations observed for both groups demonstrate that even on these tasks there was considerable inter-individual variation in performance.

The only task to elicit any evidence of group differences was the switch-reach route of the Detour Reach Task. Children with speech and language delay

made significantly more direct reach errors than children with autistic spectrum disorder whilst children in the autism group made more errors reflecting confusion between the rules.

The number of children for whom satisfactory data were elicited on these tasks was small. Therefore, it would be inappropriate to use these data points for longitudinal analysis of executive function development. However the current data does suggest that the two groups of children performed at comparable levels on the executive function tasks.

3.6 Time 2 Results

3.6.1 A-not-B Invisible Displacement Task

Twelve ASD and 15 SLD children attempted this task. Table 3.9 presents the mean (and SD) for each group for percentage of reversals correct, percentage of perseverative errors and the longest run of perseverative errors. There was no significant correlation between age, verbal mental age or non-verbal mental age and any of these variables.

Performance was similar to that observed at Time 1, with the percentage of reversal trials at or below chance levels for both groups. ANOVA showed no significant group differences for percentage of reversals correct ($F_{(1,25)}$ = 3.23, p=0.08). As at Time 1, apparent differences in the mean of the two groups are outweighed by very substantial variability in the performance of individual children.

The percentage of trials on which each child perseverated incorrectly showed similarly wide variation across individuals. There was no difference between the groups on this measure ($F_{(1,25)}$ = 3.02, n.s.). However, at this assessment time, the difference between the two groups' mean longest run of perseverative errors was significant ($F_{(1,25)}$ = 6.57, $p<0.05$). The mean longest run of perseverative errors was greater for the ASD group than the SLD group. This finding echoes the almost significant difference between the two groups on this variable at Time 1.

Table 3.9 A-not-B Invisible Displacement Task: key variables

	ASD		SLD	
	M	**(SD)**	**M**	**(SD)**
	n=12		n=15	
% reversals correct	25.0	(39.89)	52.78	(39.92)
% perseverations	50.94	(30.54)	32.62	(24.34)
Longest run of perseverative errors*	**2.58**	(1.0)	**1.6**	(0.99)

* $p<0.05$

3.6.2 Detour Reach Task

Knob route

Seventeen ASD and 16 SLD children attempted this task. Table 3.10 presents the key variables for this task. Two children in each group failed to achieve a criterial run; all other children were successful on this task. Chi-square analysis revealed no effect of group on passing or failing the task ($\chi^2_{(1)} = 0.00$, ns).

The number of trials required to reach criterion was significantly associated with non-verbal mental age ($r=0.65$, $n=29$, $p<0.05$), but not chronological age or verbal mental age. ANCOVA was used to compare the number of trials required to reach criterion for each group whilst covarying for the effects of NVMA. This analysis revealed a significant effect of NVMA ($F_{(1,26)} = 18.05$, $p<0.001$) but no effect of group ($F_{(1,26)} = 0.1$, n.s.). There were too few errors for analysis.

Switch-reach route

Fifteen ASD and thirteen SLD children continued to this task, of these five ASD and 8 SLD achieved a criterial run (Table 3.10). A chi-square analysis revealed no effect of group on classification as a passer or failer ($\chi^2_{(1)} = 2.23$, n.s.). For this route, chronological age and mental age were not significantly correlated with the number of trials to criterion. ANOVA also showed no difference between the groups on this measure ($F_{(1,11)} = 0.01$, n.s.).

In the same way as was described at Time 1, inhibition can be measured through the tendency to perseverate to the knob route instead of the switch-reach route, or by the tendency to make direct reach errors. At this assessment period, a few children did repeat the knob route rather than implement the switch-reach rule. However the frequency of this response was very low with only three children making one knob route response each and therefore this error type was excluded from analyses. More children made a confusion response (n=16) but the mean confusion response per group was still low (Table 3.10), and lower than Time 1. Therefore these types of error did not seem to be an important feature of performance at this age.

Direct reach errors were more common than knob or confusion responses at this time. Preliminary analyses showed no significant association between chronological age, verbal mental age or non-verbal mental age and the number of direct reach errors or total errors produced. However, ANOVA revealed a significant effect of group for the number of direct reach errors with the ASD group producing significantly more of these errors than the SLD group ($F_{(1,26)} = 5.90$, $p<0.05$). Similarly, the ASD groups were observed to produce significantly higher error totals than the SLD group ($F_{(1,26)} = 5.82$, $p<0.05$). This pattern is in contrast to the results at Time 1, where the SLD group made more direct reach errors than the ASD group.

Table 3.10 Detour Reach Task: key variables

	ASD		SLD	
	Mean	**(SD)**	**Mean**	**(SD)**
Knob Route	n=17		n=16	
Pass (n)	15		14	
Number of trials to criterion	3.73	(1.44)	4.21	(2.19)
Errors				
Total errors	1.71	(3.33)	2.31	(4.05)
Switch-Reach Route	n=15		n=13	
Pass (n)	5		8	
Number of trials to criterion	5.8	(3.11)	5.75	(2.19)
Errors				
Direct Reach*	**4.6**	(3.16)	**2.0**	(2.38)
Confusion	1.73	(1.75)	1.00	(1.29)
Total *	**6.47**	(3.89)	**3.15**	(3.29)

* $p<0.05$

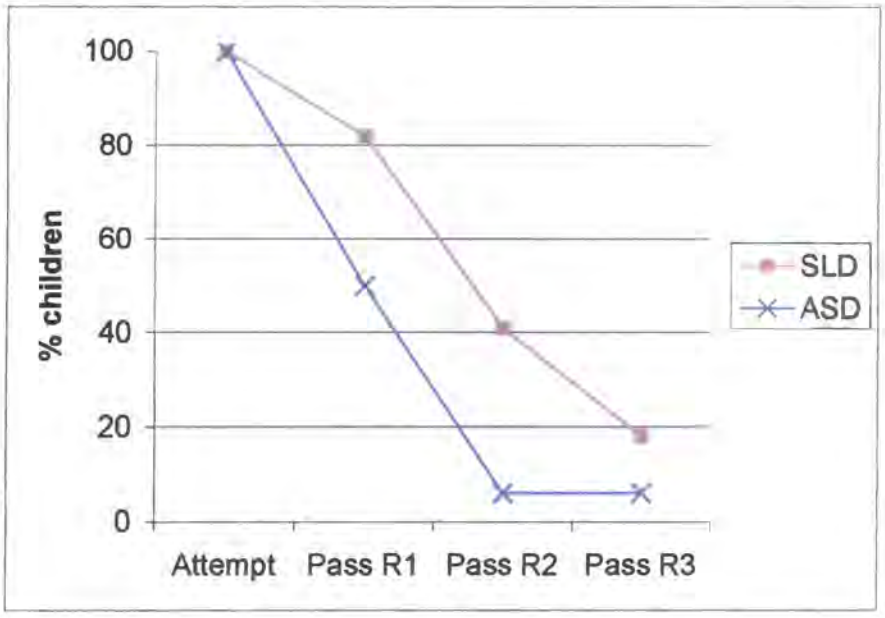
3.6.3 Sorting Task

Sixteen ASD and 17 SLD children attempted this task. Children were grouped according to the highest level of rule they passed. The four categories were: fail Rule 1, pass Rule 1 and fail Rule 2, pass Rule 2 and fail Rule 3, and pass Rule 3. A child in the first category failed the training or testing phase of

Rule 1; a child in the second category had passed the testing phase of the first rule but could not switch to the second rule; a child in the third category had passed the testing phase of both rules but could not shift from one to another in the final phase of the task; and a child in the fourth category was successful at every stage of the task. All children who were administered this task had demonstrated the ability to sort by both shape and colour during the Mullen Scales of Early Learning. Figure 3.4 provides a graphical representation of the performance of the two groups. The percentage of each group remaining at every stage is displayed.

Figure 3.4 illustrates that both groups of children followed a similar trajectory, although a greater percentage of the SLD group passed each rule. Since the expected frequencies for half the cells were below five, a likelihood ratio method for contingency tables was applied to assess whether the two groups performed significantly differently on this task. This analysis revealed differences in the proportion of each group passing each Rule ($2(i)_{(3)} = 8.92$, $p<0.05$). The performance of the ASD group was particularly poor. Almost half (44%) of these children failed to pass the test phase of the first sorting rule they were shown. Of the seven children who did learn the first rule, only one child was able to pass the testing phase of the second rule; this child went on to be successful on all phases of the task. In contrast, over 80% of the SLD children (82%) passed the test phase of the first rule. Seven of these 14 children were also successful on the second rule and three (18%) continued to be successful on all phases of the task.

Figure 3.4 Graph showing the percentage of children remaining at each stage of the Sorting Task by Group



It is possible to look more closely at the performance of the children by checking whether they failed at the training or testing phase of a rule (Table 3.11). As we have already noted, more ASD than SLD children failed Rule 1. Within each group, roughly equal numbers were unsuccessful on the training and testing phases. Some children were unable to sort by the first rule even with verbal prompts and others could not continue to sort correctly when that support was removed. In contrast, the majority of children (86% in both groups) who failed Rule 2 did so on the training phase of the task. This suggests that they were unable to switch from Rule 1 to Rule 2 even when Rule 2 was demonstrated and they were given verbal support. Both groups displayed severe difficulties in switching from Rule 1 to Rule 2.

Table 3.11 Sorting Task: Failure on training or testing?

	ASD			SLD		
	Training	Testing	Total	Training	Testing	Total
	n	n	N	n	n	N
Fail R1	4	4	**8**	2	1	**3**
Fail R2	6	1	**7**	6	1	**7**

Although the order of rule presentation was counterbalanced across children it is important to consider whether the one rule was associated with more failures than the other. Five of the six ASD (83%) and four of the six SLD (67%) who failed training for Rule 2 had sorted by colour successfully as Rule 1 but could not switch to shape sorting. This implies that shape sorting is more difficult for these children than colour sorting. In support of this, eight of the eleven children (73%) who failed Rule 1 had been administered the shape rule. However all children had previously demonstrated the ability to shape sort, 17 children successfully sorted by shape at Rule 1, and 50% of children who passed Rule 2 were attempting the shape rule. So there is no clear evidence to suggest that shape sorting is in fact more difficult than colour sorting for these children, but there is a suggestion that shifting from colour to shape sorting may be more difficult than the alternative direction.

Error analysis can provide information about the underlying reasons for failure on Rule 2. Two error categories were formed: perseverative responses to Rule 1 and unclassified responses where no sorting rule was followed. A child who continues to sort by Rule 1 during Rule 2 might be demonstrating a stuck-in-set cognitive deficit, whereas a child who has a mixture of error types might be demonstrating a more general attentional problem with the task or difficulty with Rule 2 itself.

Ten children attempted the Rule 2 testing phase (two ASD and eight SLD children). Perseverative errors were not a regular feature of any of the children's responses, therefore it would be inappropriate to conclude that these children were perseverative on this task. Interestingly, several parents of children in both groups spontaneously commented during the training phase of Rule 2 that their child could usually sort by the rule but they seemed to be '*stuck with the [first] rule*' but the data does not bear this observation out. Further refinement of the task, perhaps by presenting both training sessions first and then administer the testing sessions like Zelazo and colleagues (Zelazo et al, 1996), will help our understanding of the set shifting skills of these very young children.

In summary, both groups displayed difficulty in passing Rule 1 despite having previously shown successful sorting by that rule. Beyond that, the ASD group demonstrated a significant deficiency in switching from Rule 1 to Rule 2. However perseverative errors were not common for either group. The ASD set-shifting difficulty was present even though the task was structured so that the rule change was clearly identified, the second rule was demonstrated immediately before training began, and training with verbal prompts occurred directly before testing.

3.6.4 Six Boxes Task

Stationary version

Fifteen ASD and 17 SLD children attempted this task. Table 3.12 presents descriptive information for the variables of interest in this task. Non-verbal mental age was significantly associated with efficiency ratio ($r=0.36$, $n=32$, $p<0.05$) and the number of incorrect reaches produced ($r=-0.38$, $p<0.05$). These variables were not significantly correlated with either chronological age or verbal mental age and the longest run of perseverative errors to a specific box/location was not significantly associated with any age or ability variable.

The efficiency ratios for both groups were 0.77. An ANCOVA comparing the efficiency ratios of both groups whilst covarying for the effect of NVMA revealed a significant effect of NVMA ($F_{(1,29)}$ =5.01, $p<0.05$), but no significant effect of group ($F_{(1,29)}$ = 0.48, n.s.). Similarly, ANCOVA for the number of incorrect reaches obtained a significant effect of NVMA ($F_{(1,29)}$ = 5.01, $p<0.05$), but no effect of group ($F_{(1,29)}$ = 0.12, n.s.).

The mean lengths of a run of perseverative reaches to a specific box/location were small indicating continued perseveration was not common in either sample. Twenty children made at least one perseverative reach to a particular box/location. Thirteen of these were from the SLD group and seven from the ASD group. Chi-square analysis reported that there was no reliable difference in group membership for those individuals who made a run of one or more and who did not make a run ($\chi^2_{(1)}$ = 3.02, n.s.).

Table 3.12 Stationary version of the Six Boxes Task: key variables

	ASD		**SLD**	
	Mean	**(SD)**	**Mean**	**(SD)**
	n=15		n=17	
Efficiency ratio	0.77	(0.26)	0.77	(0.17)
Number incorrect reaches	4.38	(2.97)	2.69	(1.8)
Longest run of perseverative errors to box/location	1.63	(1.19)	1.15	(0.38)

Scrambled version

Sixteen ASD and 17 SLD children attempted this task. Table 3.13 presents the means and standard deviations for the key dependent variables. No significant correlations between chronological age or mental age and performance on this task were observed. The efficiency ratios for both groups were lower than their performance on the stationary task, but they were not different from each other ($F_{(1,31)}$ = 0.00, n.s.). Nor were any effects of group observed for number of incorrect reaches made ($F_{(1,31)}$ = 0.09, n.s.).

This task allows perseverative responding to location to be distinguished from perseveration to colour (the distinctive dimension for each box). Considering first perseveration to location, 13 children made no perseverative response of this nature. Of these children, 10 were SLD and three were ASD. Twenty children (13 ASD and 7 SLD) made at least one perseverative error to a previously searched location. Chi-square analysis revealed a significant association between group membership and making a run of perseverative responses to a location ($\chi^2_{(1)} = 5.53$, $p<0.05$): children in the ASD group were more likely to make a perseverative error to a location than SLD children. However, the most common length of consecutive perseverations to location was one; this is reflected in the mean longest length of perseverations (Table 3.13). Although there was a significant group difference between the group means ($F_{(1,31)} = 4.79$, $p<0.05$) this is difficult to interpret as evidence for perseverative responding since the mean lengths were so short. Whilst a greater number of ASD children made one perseverative response to location, they did not tend to become stuck with this response. This limits the interpretation of these findings as evidence for executive function impairment.

The pattern of data for perseverative errors to a previously searched box was similar. Twenty children (8 ASD and 11 SLD) made at least one perseverative error to a specific box. No association between group and presence or absence of a run of perseverative responses to a box was identified through chi-square analysis ($\chi^2_{(1)} = 0.25$, n.s.). Once again the most common length of consecutive perseverative errors to a particular box was one; the low mean values (Table 3.13) show how brief these runs really were for both groups ($F_{(1,31)} = 0.97$, n.s.).

Table 3.13 Scrambled version of the Six Boxes task: key variables

	ASD		SLD	
	Mean	**(SD)**	**Mean**	**(SD)**
	n=16		n=17	
Efficiency ratio	0.59	(0.14)	0.59	(0.17)
Number incorrect reaches	4.44	(2.22)	5.0	(2.63)
Longest run of perseverative errors to a specific location*	**1.06**	(0.68)	**0.56**	(0.73)
Longest run of perseverative errors to a specific box	0.63	(0.62)	0.94	(0.85)

* $p<0.05$

3.6.5 Marbles Task

All 34 (17 ASD, 17 SLD) children attempted this task. Performance on this task was good with the mean percentage of correct trials being over 70% for both groups (Table 3.14). This may represent a ceiling effect in both groups, although the large standard deviations demonstrated some children did not perform at ceiling. Verbal and non-verbal mental age correlated significantly with the percentage of trials correct (VMA: $r= 0.4$, $n=34$, $p<0.05$; NVMA: $r=0.4$, $n=34$, $p<0.05$), and the total number of errors produced (VMA: $r= -0.36$, $n=34$, $p<0.05$; NVMA: $r= -0.35$, $n=34$, $p<0.05$). Verbal mental age, but not non-verbal mental age, was significantly associated with the number of perseverative errors ($r= -0.37$, $n=34$, $p<0.05$). Chronological age was not significantly associated with any of the dependent variables for this task.

Analysis of covariance was used to compare the performance of the ASD and SLD groups on the percentage of trials completed correctly. Given that verbal mental age and non-verbal mental age were significantly correlated with each other, only the variable with the highest correlation with the dependent

variable (VMA) was entered into the ANCOVA procedure (see section 3.4). This analysis revealed a significant effect of VMA ($F_{(1,31)} = 6.97$, $p<0.001$), but no significant effect of group ($F_{(1,31)} = 1.0$, n.s.). This suggests that when mental age was taken into account, both groups were equally able to select the correct route for the marble. Similarly, an ANCOVA for the total number of errors on this task revealed a significant effect of VMA ($F_{(1,31)} = 6.13$, $p<0.05$), but no significant effect of group ($F_{(1,31)} = 1.42$, n.s.).

A repeated response to the same route led to a perseverative error on most occasions. Thirteen children made no perseverative errors of this type whilst the remaining 21 children made between one and seven perseverative responses. Most children made a run of no more than one response of this nature, but 10 (4 ASD, 6 SLD) children made runs of two, three or four perseverative responses. Chi-square analysis revealed no association between group membership and the presence or absence of a run of perseverative responses ($\chi^2_{(1)} = 0.02$, n.s). Similarly, when verbal mental age was entered as a covariate, analysis of covariance for the number of perseverative errors revealed a significant effect of VMA ($F_{(1,31)} = 7.13$, $p=0.01$), but no significant effect of group ($F_{(1,31)} = 2.03$, n.s.). Since the mean numbers of perseverative errors were less than 2 for both groups there was no reason to explore the longest run of this error type. By comparing the group mean values for perseverative responses and for incorrect responses we can see that perseverative responses were not a major type of error response for either sample: in fact perseveration seems to occur on only one-third or thereabouts of incorrect responses.

Table 3.14 Marbles Task: key variables

	ASD		SLD	
	Mean	(SD)	Mean	(SD)
	n=17		n=17	
% correct	71.84	(29.07)	74.84	(19.63)
Number of errors	3.06	(2.97)	3.00	(2.37)
Number of perseverative errors	1.71	(1.99)	1.76	(2.14)

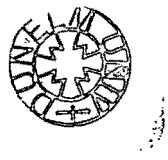
3.6.6 Time 2 Summary

Four out of five tasks revealed some indication of performance differences between the groups. However, these differences tended to be found on only one error variable per task.

Neither group performed above chance on the reversal trials of the A-not-B Invisible Displacement Task; the groups were not significantly different on this measure. However, the children with autistic spectrum disorder did make significantly longer runs of perseverative errors on this task. The switch-reach route of the Detour Reach Task also elicited group differences on error variables with children in the autism group making more errors and specifically, more direct reach errors than the children in the speech and language delay group.

Performance on the Sorting task was poor for both groups, but there was evidence that the children with autism were reliably worse than the children with speech and language delay. Despite this finding, there was no evidence that the children in the autism group were responding in a more perseverative manner than the speech delayed group (although the numbers of children for whom errors could be examined for this behaviour were very limited).

The scrambled version of the Six Boxes Task produced significant group differences in the longest perseverative run to location, but the mean longest



run for the autism group was only about one and therefore cannot be interpreted to represent a perseverative problem. Finally, children in both groups demonstrated high levels of success on the Marbles task with no group differences on any measure.

In sum, comparing the performance of the ASD and SLD groups at four years of age shows some hints of executive dysfunction in the autism group. There was a tendency for the ASD subjects to show greater perseverative responding on some, but not all, of the tasks. The ASD group were also found to be less successful than the SLD group on the Sorting Task, which was designed to assess attentional set-shifting. However, the ASD group was not globally impaired on any task. The ASD and SLD groups achieved scores that were similar or identical on almost all measures of overall performance.

3.7 Executive Function Performance Over Time

Three executive function tasks were included in the test battery at both time points to provide continuity of assessment and enable direct measures of change. The A-not-B Invisible Displacement and the Detour Reach tasks were administered in an identical fashion at both time points. The Boxes Tasks were administered with three boxes at Time 1 and six boxes at Time 2. The sensitivity to change over time of these tasks is largely unknown although the A-not-B Invisible Displacement Task has been used in a longitudinal study of infants and toddlers with PKU (Diamond et al, 1997). The tasks used to measure change were chosen for their appropriateness for the developmental ages at both time points and the Boxes task was made more demanding at Time 2 to take into account developmental progress.

One of the complexities of this type and quantity of data collection is ensuring that all the data is present and of good quality. Every attempt was made to ensure that both these criteria were met during this project. However, inevitably, some of the data are missing due to child fatigue or non-compliance. Unfortunately, the sample sizes at Time 1 were too small to

allow formal analysis of development over time. Therefore this section presents impressions of the patterns of change for individual children who provided data at both time points to demonstrate the variability in performance change in the sample.

A-not-B Invisible Displacement Task

Eleven children attempted the A-not-B Invisible Displacement task at both Time 1 and Time 2 (ASD=4, SLD=7). The percent of reversal trials correct at both time points was directly compared as a measure of inhibitory control. Figure 3.5 provides visual representation of the patterns of development for each individual child from Time 1 to Time 2. Each line represents one case and the numbers on each column represent the percentage of reversal trials correct for each child at both time points.

Figure 3.5 Visual representation of change over time in percentage of reversals correct on A-not-B Invisible Displacement Task.

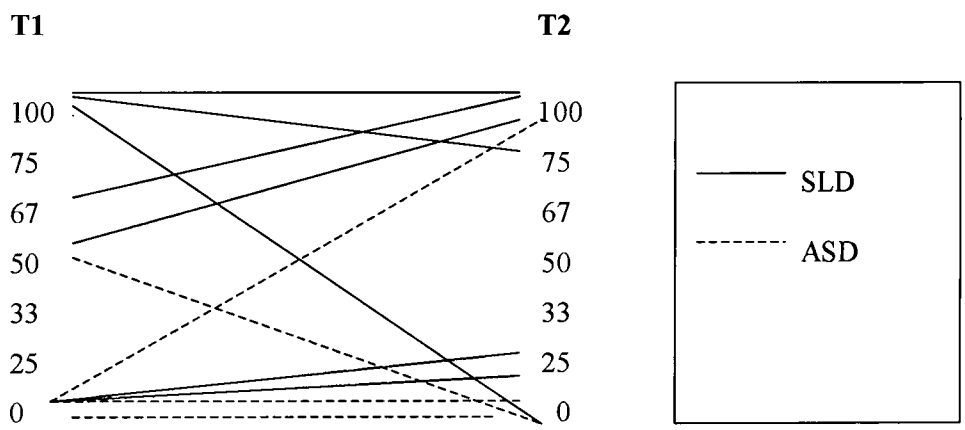


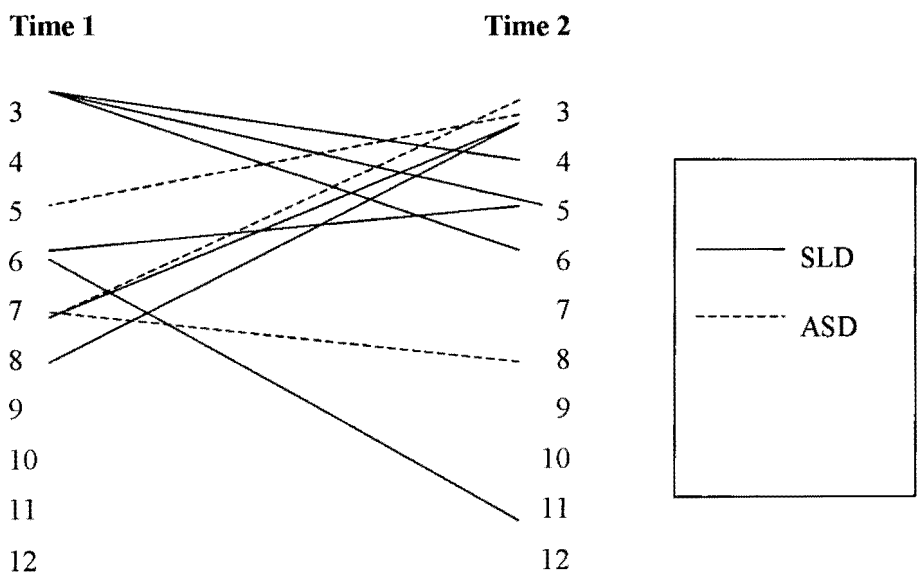
Figure 3.5 shows there is no simple picture of development on this task for these children. Only one child with ASD actually performed better on this task when they were a year older, and this child improved dramatically from 0% to 100%. Although the majority of SLD children did make some improvement over time, none made such a dramatic leap in performance whilst one SLD child (but no ASD) worsened from 100% at Time 1 to 0% at Time 2.

Detour Reach Task

Twenty-nine children attempted the knob route of the Detour Reach task at both time points (ASD=14, SLD=15). Twelve SLD and 7 ASD children passed the knob route at both time points. Nine of these children passed the task in the minimum three trials at both time points (ASD=4, SLD=5). Figure 3.6 provides a visual representation of the change in performance for individual children, excluding the nine who were at ceiling at both times. The two columns represent the number of trials required to pass the task and each line represents one child.

The switch-reach route was only administered to children who had made a criterial run and therefore the numbers of children attempting this task at both times were notably smaller. Fourteen children did, however, attempt the switch-reach route at both time points (ASD=6, SLD=8). Only one child achieved a criterial run on the switch-reach route at both Time 1 and Time 2. This child was a member of the ASD group and passed the switch-reach route in 5 trials at Time 2 as opposed to 8 at Time 1.

Figure 3.6 Visual representation of change over time in number of trials required to pass the knob route at Time 1 and Time 2, excluding those children performing at ceiling on both occasions (maximum number of trials = 15, minimum number of trials = 3).



Three and Six Boxes Tasks

The visual search tasks were administered with three boxes at Time 1 and six at Time 2. As the number of reaches required to retrieve all boxes cannot be directly compared, efficiency ratios allow for consistent and comparable assessment of performance. Performance was classed as "improved" if the efficiency ratio was greater at Time 2 than at Time 1, "unchanged" if the efficiency ratios were identical at both time points and "worsened" if the efficiency ratio was lower at Time 2.

Thirty children (ASD=13, SLD=17) children attempted the stationary versions of the task at both Time 1 and Time 2. The majority of both groups actually worsened over the year. This could reflect a ceiling effect on the Three Boxes task and/or the increased difficulty of the Six Boxes task. Interestingly all four of the ASD children who improved their performance at Time 2 were at ceiling during the second period of testing whereas only two of the three SLD improvers reached ceiling.

Twenty-nine children (ASD=12, SLD=17) attempted the scrambled version of the task at both time points. Once more, a drop off in performance was common within each group. All four SLD children who scored at ceiling at Time 1 performed less efficiently on the Six Boxes task along with six other SLD children. Similarly, two of the six ASD children whose performance dropped fell from ceiling levels. Despite there being six improvers in each group, only one (SLD) child performed at ceiling on the Six Boxes task. Because there were a larger number of children who attempted these tasks at both time points, a graphical representation of individual children was too confusing. Table 3.15 displays the number of children whose performance improved, remained unchanged or worsened on the Box tasks by group.

Table 3.15 Number of children in each category of performance on the stationary and scrambled Boxes Task.

		ASD	SLD
Stationary	Improved	4	3
	Unchanged	4	4
	Worsened	5	10
		13	**17**
Scrambled	Improved	6	6
	Unchanged	0	1
	Worsened	6	10
		12	**17**

Summary

Although the small sample sizes preclude statistical analysis and highlights a need for caution, it can be seen that the direction of change in performance on executive function tasks varies within groups: children improved, remained unchanged and worsened regardless of group. The current data provided no reason to believe that larger samples would produce evidence of reliable developmental progression of executive function in these groups of very young ASD and SLD children. This was further supported by identifying children whose performance changed most dramatically over the year on each task. Many individual children were examined in detail but no consistent pattern was found. Performance change in one direction on an executive function task did not reliably associate with performance change on another executive function, or with change in ability as measured by the Mullen Scales of Early Learning (Mullen, 1995). Four case studies are presented in Appendix IV to further illustrate these conclusions.

3.8 General Discussion

Inhibition of one response and implementation of another, inhibition of attentional set, working memory and planning skills are all aspects of executive function (see Chapter 2 for more detail) that were targeted by this study. These skills were tapped when the groups were, on average, three years old and again when they were four years old.

3.8.1 Do Very Young ASD Children Perform More Poorly on Executive Function Tasks than SLD Children?

The overall finding in the current study was that out of a total of nine tasks administered, only three tasks (A-not-B Invisible Displacement Task, Switch-reach route of the Detour Reach Task and the Sorting Task) elicited any reliable evidence that these very young children with autistic spectrum disorder were performing more poorly than children with speech and language delay. Furthermore, these differences were only observed when the children were, on average, four years old and not when they were three years of age. The tasks that did elicit this group difference were those thought to address the inhibitory skill of these young children. As such, the current findings call into question the prediction that executive function impairment should onset early in a young autistic child's development. Surprisingly, one finding actually ran counter to the predictions of an executive dysfunction hypothesis of autism.

At Time 1, children with autism made significantly fewer direct reach errors than children with speech and language delay when attempting the switch-reach route of the Detour Reach Task. A direct reach error on this task has been interpreted as a sign of inhibitory failure (Hughes & Russell, 1993). On this view, the SLD children appear to have greater difficulty in inhibiting their responses than ASD children. But the performance of these same children on the knob route task had suggested an unimpaired ability to inhibit the direct reach response per se. SLD children had been able to inhibit a direct reach and implement the correct knob response when this was the total demand of the task. The switch-reach response required the child to inhibit a direct reach, and implement a switch response followed by a direct reach response. Failure

to perform any of these actions resulted in an error. More specifically, a difficulty in sequencing the two responses correctly would lead to errors that resembled a failure to inhibit a prepotent response (direct reach). There are two possible explanations for the performance of the SLD children. Firstly, their inhibitory control is less effective when faced with the increased demands of a more complex task. Secondly, the direct reach errors may represent a sequencing problem rather than a more clear-cut inhibition of response problem. Further research is required to elicit the root cause of the direct reach error but it is notable that one mother spontaneously commented that her son was having difficulty sequencing the two responses. However, this pattern of results was not replicated at Time 2. In fact at the second time point the ASD group made significantly higher rates of direct reach errors than the SLD group.

The only hint of an autistic spectrum deficit at Time 1 was in terms of a greater number of confusion responses on the switch-reach route of the Detour Reach Task. These responses involving the knob, the switch and a reach were suggested to represent confusion between the knob rule and the switch rule. This could be interpreted as a type of perseveration where the knob route section of the task cannot be overridden completely by the new switch-reach response. However the perseveration in this confusion response is of a different quality to the perseveration that would be demonstrated by a knob-only response (a response that was not noted in this sample). Moreover, this finding of increased rates of confusion errors in the ASD group was not replicated at Time 2.

At the average age of four most task variables still produced no significant differences. However, more evidence was present to suggest an inhibitory impairment in children with autism. The ASD group showed a tendency towards increased perseveration on both the A-not-B Invisible Displacement Task and the scrambled version of the Six Box Task, producing significantly longer runs of perseverative errors on both tasks. However, it should be noted that the mean values for both groups were still relatively low so that

perseverative responding in ASD group was neither markedly widespread nor pervasive.

The set-shifting required by the Sorting Task proved taxing for both groups. Most children failed to inhibit the first rule and to pass the second rule and the majority of failers, from both groups, were unable to pass the training phase of Rule 2. This suggests a profound inability to shift from one set to the next for these children. Moreover, children with autism were less successful on the task than the children with speech and language delay. This reflects the findings of Hughes et al (1994) who noted significant difficulty in making the intra-dimensional and extra-dimensional shifts for individuals with autism when compared to other clinical groups. However the reasons for this difficulty in the current sample are unclear because perseverative responding was not a common feature of either group and all the children who were administered this task had demonstrated competent sorting according to shape and colour during their ability assessment.

Performance on the Marbles task was unexpectedly good for both groups. Furthermore, the two groups were indistinguishable on all the key variables of this task. If this task does measure planning (see section 6.2.3 for a discussion of this issue), the current study provides no evidence that the ASD group are less efficient at planning in a structured experimental setting than the SLD group.

In conclusion, there is no evidence at either time point to suggest working memory or planning impairments in the children with autistic spectrum disorder. Evidence for impairments in inhibitory control was equivocal. Only one variable at Time 1 reported a group difference where children with autism performed less successfully than children with speech and language delay. At Time 2 there was more evidence to suggest that the very young children with autism displayed inhibitory control impairments when compared to very young children with speech and language delay. However, these impairments were not of the magnitude or the pervasive nature that would have been

predicted by the existing literature in older children with autistic spectrum disorders (e.g. Pennington & Ozonoff, 1996).

3.8.2 Methodological Issues

Before interpreting the findings of this study, there are several methodological issues that must be addressed. First, the tasks administered were predominantly measures of inhibitory control and therefore did not represent the range of executive function skills studied in older and more able individuals. At Time 1 it was particularly difficult to develop tasks that might tackle a variety of executive function skills. The importance of keeping the verbal demands of the task to a minimum precludes the administration of many generation and planning tasks that have been described in Chapter 2. Therefore, three of the four tasks administered at this age focused upon inhibitory control. This is commonly observed in published studies of executive function in preschool children (e.g. Griffith et al, 1999; McEvoy et al, 1993; Hughes, 1998a,b; Diamond et al, 1997). Perhaps performance differences might have been observed if non-verbal generativity or planning tasks appropriate for the low ability of these children could have been administered to these children. At Time 2 two tasks were developed specifically to assess more advanced inhibition and set shifting skills and elementary planning skills. In this way the range of executive skills assessed in the study were broadened as far as possible (section 6.4 deals with this in more detail).

It must also be acknowledged that ceiling effects were observed on two of the tasks at Time 1. The A-not-B task proved easy for most children in this study, as did the stationary version of the Three Boxes Task. This may have influenced the findings, however the standard deviation indicated that performance was not at ceiling for some children. It is notable that, even though the children with autistic spectrum disorder were less able than the children with speech and language delay, they did not perform more poorly on these tasks. There was no clear evidence for ceiling effects at Time 2 (the Marbles Task is a possible exception).

The same experimenter assessed all children in the study in their own homes at both time points. This ensured continuity of experience for the children and consistency across children in that they were all assessed in their home environment. The fact that all the children were assessed in a familiar environment may have played a role in the relatively good performance of the children in the study. Nevertheless, the wide standard deviations on the tasks indicated variable within-group performance that supported the appropriateness of the tasks administered.

A third methodological question is the diagnostic status of these very young children. This issue will be dealt with in more detail in section 6.2.2 but there is good reason to believe that the rigorous grouping procedure that was followed in this study was valid.

Finally, it is possible that the power of the statistical analyses was not sufficient to observe differences between the two groups in question. This issue will also be discussed in more detail in section 6.2.1 but analyses were undertaken in a planned and cautious way to maximise power as far as possible. The fact that the current findings replicate the pattern of results reported by Griffith et al (1999) supports the idea that the current findings are valid and reliable. Furthermore, the small number of children providing data at Time 1 meant that longitudinal analyses of executive function development over time were inappropriate with this dataset. However, the development of a small number of individual children was reviewed and this suggested that executive function did not develop in a reliable way over the period of one year for either group. To further explore the longitudinal development of executive function it would be necessary to have greater sample sizes and power.

3.8.3 How do the Current Findings Relate to the Existing Executive Function Literature in Autism?

The literature with older children and adults with autism generally reports impairment in set-shifting tasks such as the WCST (e.g. Bennetto et al, 1996; Ozonoff & Jensen, 1999; Ozonoff & McEvoy, 1994; Rumsey & Hamburger,

1988; Szatmari et al, 1990) but not on inhibition-and-implementation tasks (e.g. Ozonoff et al, 1994; Ozonoff & Jensen, 1999; Ozonoff & Strayer, 1997; Russell et al, 1999). In conjunction with a recent study assessing four-year-olds with autism (Griffith et al, 1999), the current study extends this pattern to very young children with autistic spectrum disorder.

Griffith and colleagues (1999) administered a battery of inhibition-and-implementation and working memory tasks executive function tests to a sample of young children with autism and a mean chronological age of 4:2 years. The non-verbal ability of these children was 2:10 and the verbal ability 1:10. The comparison group had a variety of developmental delays. Out of seven tasks (A-not-B, A-not-B Invisible Displacement, Spatial Reversal, Three Boxes stationary and scrambled, and Six Boxes stationary and scrambled) they reported only one significant group difference. In fact, this sole group difference reported that children with autism had *superior* performance to that of the control group on the Spatial Reversal Task.

The current study also provides some indication of superior performance for children with autism in that, at Time 1, children with autism made fewer direct reach errors on the switch-reach route of the Detour Reach Task. This particular difference was reported when the children were one year younger than the Griffith sample and may be a result of the small sample size since, by the time they reached they age of four, this effect was reversed so that children with autism did make more direct reach errors than the speech and language delayed comparison group.

The inclusion of a task of shifting between cognitive sets and a task designed to measure early planning skills extended the Griffith et al work. In this way, skills that have been frequently reported as deficient in older individuals with autism (e.g. Bennetto et al, 1996; Hughes et al, 1994; Hughes, 2001; Ozonoff & Jensen, 1999; Ozonoff & McEvoy, 1994; Ozonoff et al, 1991a; Ozonoff et al, 1994; Rumsey & Hamburger, 1988; Szatmari et al, 1990) were explored in very young children with autism. The findings were complicated by the fact that so many children had trouble with Rule 1 on the Sorting Task (despite

previous success on a sorting task in the ability assessment), but there was evidence to support the existing literature that children with autism perform poorly on set-shifting tasks. In contrast, the children with autism showed no impairment on the task designed to assess planning skill. If this task did successfully assess early planning skills (see section 6.2.3) then the current study does not replicate the findings of planning impairment in older individuals.

So, combined with Griffith et al, there seems to be growing evidence that neither very young children with autism nor much older children and adults with autism demonstrate inhibition-and-implementation and working memory impairments. However, there is some evidence that early school-age children (roughly 4 to 7 years of age) with autism do display inhibition-and-implementation (Hughes & Russell, 1993; Russell et al, 1991). One possible explanation for this discrepancy stems from a subset of the Griffith sample which was reassessed on the Spatial Reversal Task at the age of five years. Group differences between children with autism and children with developmental delays were still non-significant, however they noted a trend towards the control group performing fewer perseverative errors with time. They argued

> "*the deficit appears autism-specific at later ages due to an improvement in the executive functioning of young children with developmental delays*" (Griffith et al, 1999, p.827).

A developmental explanation of this nature may provide a way in which to bring together studies showing unimpaired and impaired inhibition-and-implementation skills in autism. It is possible that the *developmental trajectories* of these skills may differ between children with autism and other children. Autistic spectrum disorder is a developmental disorder and therefore a developmental explanation seems eminently appropriate. Unfortunately, the current data set could not further explore this possibility due to the small number of children who provided data at Time 1. But looking at individual children suggests that developmental changes vary for each child and are not

a function of diagnostic grouping. Clearly, studies of specific executive skill development over time are necessary to investigate this possibility. One particular complexity of this type of work is the need to generate tasks that can accommodate a wide range of developmental levels and ages in order to assess the development of a specific executive function skill over a long period of time.

Marked heterogeneity for individuals with autism is commonly reported at a cognitive level (e.g. Adrien et al, 1995; Griffith et al, 1999; Pennington & Ozonoff, 1996) and at a clinical or behavioural level. Wide standard deviations are reported in the current sample. In terms of studies of very young children with autism, these wide standard deviations may have reduced the likelihood of finding reliable group differences in executive function performance. At the same time, they also signal individual variation between children. This is one important point the current thesis has highlighted and should be further explored.

3.8.4 How do the Current Findings Relate to the Existing Literature for Typically Developing Children?

One final issue that must be considered in relation to the current chapter is the relative executive skill of the current sample. It is plausible that the similarity in executive performance of these two groups reflects an executive impairment in *both* samples. Since it was not possible to recruit another comparison sample of typically developing children to this study, the following section attempts to relate the current findings to four published studies looking at typically developing children with similar ages or ability levels. Comparisons across studies are by their nature inexact but it does provide an approximate evaluation of the developmental appropriateness of the performance of the current SLD and ASD samples. In fact, it appears that both the groups in the current study could be said to perform at levels roughly equivalent to their verbal and non-verbal ability.

The A-not-B Invisible Displacement Task was administered by Diamond and colleagues (1997) to a group of typically developing children at three-month

intervals from the age of 15 months until they reached 30 months. These children were recruited as a comparison group for PKU children who were the main focus of the monograph. Twenty-seven month old children in the Diamond study got an average of about 45% reversals correct and 30-month-olds got about 55% of reversal trials correct. The current SLD group were correct on about 52% of reversals at both time points. At Time 1 this meant the SLD sample were performing at a level that might be considered appropriate for their non-verbal and verbal mental age. However, by Time 2 a lack of improvement on the task meant they were now performing slightly less well than would be expected by their ability level. The same pattern holds for the ASD sample, although their 26% of reversals correct at Time 1 was nearer to the 30% correct of 24 month-old infants (this reflects the lower ability levels of the ASD group).

The Detour Reach Task was administered to two samples of typically developing preschoolers. The first of these had an average age of 3 years 8 months (Hughes & Russell, 1993) and the second an average age of 3 years and 11 months on recruitment (Hughes 1998a, b). Although the typical children were not assessed for ability, their chronological age was in excess of the verbal and non-verbal ability levels of the current samples even at Time 2. As might be expected, the Hughes papers reported a higher proportion of success on both the knob and switch-reach routes than was found in the current study. Over 90% of 3:8 year-olds passed both knob and switch-reach routes whereas the pass rate for the switch-reach route at Time 2 was 33% for the ASD group and 62% for the SLD group. Interestingly, the children in the current study who were successful on either route passed as quickly as or quicker than typically developing 3:11 year-olds (Hughes, 1998a,b; see Table 3.15).

Table 3.16 Mean number of trials to criterion for the Detour Reach Task for the current sample and a sample of typically developing children.

	ASD		SLD		Hughes (1998)
	T1	T2	T1	T2	Age 3:11
Knob Route	4.2	3.7	4.1	4.2	3.5
Switch-Reach Route	7.0	5.8	7.33	5.8	7.0

Unfortunately, the Detour Reach Task has not been used with children of younger developmental ages. This comparison would be more appropriate given the developmental delay of the clinical populations. At this stage the interpretation of the current performance of the ASD and SLD groups must be limited to the fact that fewer of them were successful than typically developing children with similar chronological ages, but those who were succeeded as quickly as their chronological equivalents.

The final task of inhibitory control for which there is comparative data is the Sorting Task. Hughes (1998a,b) administered a similar set-shifting task in which the child was required to identify which cards were a teddy bear's favourite on the basis of experimenter feedback. The rules applied were shape and colour, and the rule shift was signalled by a change of stimuli. Unlike the current study, training was not provided nor was the child explicitly told either rule. Hughes reported that a mean number of 1.5 rules were solved by her 3:11 year-old sample. Whilst noting the procedural variations, it is interesting that the SLD children in the current thesis solved a mean number of 1.4 rules when they were four-years-old with a non-verbal ability of just over three-years-old. This suggests that the SLD sample were performing at a level equivalent to their chronological age, and in advance of their developmental age. In contrast the ASD children were less successful.

Turning to the working memory tasks, the Three and Six Boxes tasks were administered by Diamond and colleagues (1997) in their longitudinal study. Performance on the tasks was reported as the mean number of reaches required to retrieve all the rewards; not the efficiency ratios calculated in the current study. By dividing the number of rewards boxes by the mean number of reaches required to retrieve the rewards an estimate of the efficiency ratio was calculated for the Diamond study. It is this estimate with which the current samples are compared.

The efficiency estimate for 30-month-old typically developing children on the Three Boxes stationary task was lower than those obtained by the current samples (0.75 relative to 0.89 for both the ASD and SLD groups). Therefore the current ASD and SLD groups performed at or above their non-verbal ability level and definitely above their verbal ability levels on the stationary version. The Diamond sample found the scrambled version easier than the stationary version, whilst the ASD and SLD children showed the opposite profile. In fact, on this occasion the samples were performing at about the level of 24 month-old children in the Diamond study. This performance level was below their non-verbal ability but above their verbal ability.

Diamond and colleagues have administered the Six Boxes Task to typically developing 42-month-old children. The performance of the ASD and SLD groups for the stationary version of the task was similar to the performance of 42 month-old controls. This equated to a performance slightly below their chronological age but above their ability age equivalents. Performance on the scrambled version was slightly less efficient than that of the 42 month-olds (efficiency estimate about 0.7 compared to 0.6 for the current samples). This is unsurprising given the non-verbal and verbal ability developmental delay in the ASD and SLD samples. Thus, on tasks of working memory there is no evidence of an ASD or SLD impairment relative to typically developing children of equivalent mental age. Both groups appear to perform at levels suitable for, or in excess of, their developmental age.

Taken together, where suitable comparisons are available children with ASD and SLD appear to perform at levels appropriate for their developmental age on tasks of inhibitory control and working memory. This pattern of findings presents a challenge for the executive function hypothesis. Specifically, it queries the prediction that executive function deficits are an early emerging feature of the cognitive profile of children with autism. A second prediction of the executive function hypothesis is that the severity of autistic symptomatology be systematically related to executive dysfunction. Chapter 5 explores the possible relationship between severity of repetitive behaviour and executive function performance at both Time 1 and Time 2. Firstly, Chapter 4 considers the repetitive behaviour displayed by both groups at Time 1 and Time 2.

Chapter 4

Repetitive Behaviour

4.1 Repetitive Behaviour in Young Children: A Brief Introduction

Repetitive behaviour is an umbrella term used to refer to a class of behaviours that are linked by qualities of repetition, rigidity, invariance and inappropriateness (Turner, 1999b). It is commonly accepted amongst parents that young children go through phases where much of the behaviour and play is characterised by these qualities. However, the presence and developmental trajectory of repetitive behaviour in young typically developing children has been sorely neglected in academic circles. Repetitive behaviours have elicited more interest in certain clinical populations such as Obsessive Compulsive Disorder, Tourette's syndrome. Unfortunately, the existing work in these populations is focused upon older children and adults and there has been little rigorous exploration of repetitive behaviour in autism until recently. Given the hypotheses of the current thesis the focus of this section is on the current knowledge about repetitive behaviour in young children.

One longitudinal study reported that 11% to 17% of typically developing infants aged 9, 12 and 18 months exhibited self-injurious behaviour (Shentoub & Soulairac cited in Tröster, 1994). After the age of two this behaviour dropped markedly and by the ages of five and six years it had disappeared. Similarly, developmental change in the quantity of repetitive behaviour has been observed in cross-sectional studies of children aged between one and six years. The Childhood Routines Inventory (CRI) is a questionnaire that identifies two types of repetitive behaviour: "Just Right" phenomena and "Repetitive Behaviours" (Evans, Leckman, Carter, et al, 1997). Just Right phenomena include a preference to have things done in a particular way, lining up of objects and an insistence on certain belongings

being in certain places. Repetitive Behaviour phenomena include a preference for the same household activities every day, acting out the same thing in pretend play and repeating certain actions over and over again. Both categories of behaviour were reported more frequently in two-, three- and four-year-olds than in one- and six-year-olds (Evans et al, 1997).

Evans and Gray (2000) used the CRI to compare the repetitive behaviour of typically developing children and children with Down syndrome and reported no differences in the overall number of behaviours. Both groups displayed more repetitive behaviour at younger ages (2 to 4:11 years) than at older ages (5 to 11 years) with younger typically developing children also displaying more repetitive behaviour than the more able Down syndrome individuals. Together these studies suggest that repetitive behaviour varies as a function of development and may play an adaptive role in the development of typical preschool children.

Repetitive behaviour has long been associated with autism. Kanner himself (1943) noted an obsessive insistence on sameness of behaviour, activity, and routine which he suggested to be core symptoms of autistic spectrum disorders. The epidemiological study of Wing and Gould (1979) also confirmed that stereotyped movements and repetitive patterns of activity co-occurred with social abnormalities of the autistic type. Furthermore, current diagnostic criteria (DSM-IV and ISD-10) require the presence of repetitive behaviour for a diagnosis of autistic spectrum disorder.

A broad distinction between lower-level behaviours and higher-level behaviours can be made. Motor responses fall in the low-level category whilst more complex cognitive behaviours such as insistence on sameness can be considered higher-level behaviours. Lower-level stereotyped movements and self-injurious behaviours have been reported in learning disabled populations of children (Freeman, Ritvo, Schroth et al, 1981; Murphy, Hall, Oliver & Kissi-Debra, 1999; Smith & Van Houten, 1996; Turner, 1997 for a review) and schizophrenia (McKenna, Thornton & Turner, 1998). However, there is some evidence to suggest that individuals with autism have significantly

greater severity ratings for compulsions, stereotypy and self-injury than other individuals with mental retardation (Bodfish, Symons, Parker & Lewis, 2000; Lord, 1995). Similarly, higher-level repetitive behaviours may be particularly characteristic of autism spectrum disorder (Frith, 1989; Wing & Gould, 1979). Although the rate of circumscribed interests (one example of a high-level behaviour) reported in autism and control samples vary considerably (Szatmari, Bartolucci & Bremner, 1989; Tantam, 1991) there is some evidence that routines and ritualistic behaviours are more marked in individuals with autism relative to age and ability matched control subjects (Bartak & Rutter, 1976; Lord & Pickles, 1996).

Categorising repetitive behaviours as lower- or higher-level is useful but must be used cautiously to avoid missing distinct forms of repetitive behaviour (Turner, 1997). Based upon an extensive review of the literature on spontaneously occurring repetitive behaviour in normal and clinical populations, Turner (Turner, 1997; Turner, 1995) has outlined a classificatory system which groups together, as members of a single class, behaviours that are consistently similar in their form, content or common presentation. She identified 11 classes of behaviour: tardive kynesia, tics, stereotyped movements, self-injury, stereotyped manipulation of objects, abnormal object attachments and preoccupations, insistence on sameness of environment, rigid adherence to routines and rituals, repetitive use of language, circumscribed interests and obsessions and compulsions.

Following this theoretical work, she constructed the Repetitive Behaviour Interview (1995) which yielded four summary scores giving an index of the display of repetitive behaviours in each of four categories: (1) repetitive movements (including stereotyped movements and stereotyped manipulation of objects); (2) insistence on sameness behaviour (including insistence on the sameness of environment and insistence upon specific routines and rituals); (3) repetitive use of language; and (4) circumscribed interests. Table 4.1 outlines these categories. Two groups of autistic individuals (22 high-functioning children and adults with autism who had a verbal IQ of above 75

Table 4.1 An outline of Repetitive Behaviour Interview categories (adapted from Turner, 1997)

Label	Definition	Typical Example
Repetitive Movements	Stereotyped movements: apparently voluntary rhythmic movements of the body which are repeated in an invariant manner, and are inappropriate to the current context	Hand flapping; body rocking; finger flicking
	Stereotyped manipulation of objects: Topographically invariant manipulation of objects repeated in a manner that is inappropriate given the nature, and usual function, of the object	Spinning objects; repetitively examining a toy; lining objects up in rows
Insistence on sameness behaviour	Insistence on sameness of environment: An insistence on one or more minor features of the environment remaining unchanged despite no obvious, or logical, basis. Attempts at change are met with marked resistance	Insisting that the curtains remain open or closed, or that ornaments remain in certain positions; insistence on always playing the same record; insistence on wearing the same T-shirt.
	Rigid adherence to routines and rituals: Any routine or ritual which characterized by total invariance and inflexibility, and which is adhered to in every relevant situation	Insisting on dressing in the same highly stereotyped fashion; insisting on buying a newspaper on every trip to the shop, regardless of whether or not one has previously been purchased (even though the child may have no interest in reading the newspaper himself)
Repetitive use of language	Any phrase or linguistic device which is either (1) copied from other sources, or (2) is presumed to be self-generated but is used repeatedly across different times and situations in an inappropriate manner	Immediate or delayed echolalia; repetitive use of the same phrases or questions; verbal rituals
Circumscribed interests	The repetitive and all-absorbing pursuit, or discussion, of one narrowly circumscribed topic or activity	Reading maps and talking about different countries and their flags on an daily or hourly basis (although the child may show no interest in seeing films of these countries on television)

and 22 learning-disabled individuals with autism and a verbal IQ below 75) and two groups of age-, sex-, and ability-matched comparison subjects from outpatient psychiatric departments and special schools were studied. The groups of individuals with autistic spectrum disorders were reported to display more repetitive behaviour than the comparison group and there was little effect of age and ability on this difference.

This work suggests that individuals with autism demonstrate repetitive behaviours that are maladaptive in content and function. Furthermore it suggests that age has little effect on the difference in repetitive behaviour between autistic and non-autistic individuals. Therefore, it would seem likely that very young children with autistic spectrum disorders would also be reported to show greater repetitive behaviour than their non-autistic counterparts. A questionnaire version of the Repetitive Behaviour Interview was administered to the carers involved in the current study to test this hypothesis. The longitudinal design of this project also provided the opportunity to consider the development of repetitive behaviours in both children with autistic spectrum disorders and speech and language delays. This prospect is particularly interesting given the occurrence of repetitive behaviours in young typically developing children of similar ages to those recruited to this project.

4.2 Cross-sectional Analyses

4.2.1 Participants

The participants were the same as those described in Chapter 3. The recruitment and grouping procedure was detailed in section 3.1.

4.2.2 Assessment

The Repetitive Behaviour Questionnaire (Turner, unpublished) is a form of the Repetitive Behaviour Interview (Turner, 1995). It assesses the severity, nature and frequency of repetitive behaviours displayed by the child. The RBQ consists of 33 questions designed to tap specific types of repetitive

behaviour (see Appendix I for a copy of the questionnaire). The respondent is asked to rate each behaviour type either for frequency or severity, depending on the particular behaviour. In general, each question has three or four response options that can be recoded as 0, 1, 2 or 3 to reflect the reported frequency or severity of the behaviour. Four questions differ from this pattern. One of these questions refers to circumscribed interests. Informants are given four response options to the question '*Does he/she have any interests or preoccupations which you would describe as overly keen, obsessional, or unusual in any way?*'. The first three responses describe increasingly strong obsessional interests. However the fourth option is the qualitatively different response that the child has no particular interests they pursue spontaneously. In addition, there are also three open-ended questions in the RBQ that do not conform to the scoring procedure described above.

The ADI-R (Le Couteur et al, 1989; Le Couteur et al, unpublished; Lord et al, 1994) and ADOS-G (Lord et al, 2000; Lord, Rutter, DiLavore & Risi, 1999) were also administered to each child and provide observed and reported information on repetitive behaviours. However, these tools were used as part of the grouping/diagnostic procedure and so it is inappropriate to use them as outcome measures in the current study.

4.2.3 Procedure

The Repetitive Behaviour Questionnaire was left with the parents/caregivers after the ADI-R had been conducted. The completed questionnaire was either picked up on a subsequent visit to assess the child, or returned by post. The RBQ was verbally explained to each parent and written instructions attached to the questionnaire. Parents were told they could contact the experimenter should they have any questions. The RBQ was administered at both Time 1 and Time 2 assessment periods.

Scores for individual questions were summed to provide indexes of repetitive movements, insistence on sameness behaviour, repetitive use of language, resistance to change and circumscribed interests according to the methods developed by Turner (1995) for the Repetitive Behaviour Interview.

4.2.4 Statistical Analysis

Individual items for the RBQ produced ordinal scores, with three or four levels. At this level non-parametric analyses are required. However, when these individual items are summed according the categories defined by Turner, the number of levels increases sufficiently to permit the use of parametric analyses (Clark-Carter, 1997). The maximum possible score for repetitive movement was 33, for sameness behaviour was 26, for repetitive use of language was 9 and for resistance to change was 6.

The construction of the circumscribed interests variable means it is not suitable for standard analyses. Only one question contributed to this variable and the maximum score was three. However the values 0, 1 and 2 represent increasingly strong circumscribed interests but the value 3 represents no interests at all. Given this qualitative change in the scale, scores of value 3 are eliminated before group means are calculated and because of the small range of possible values no further statistical analyses on this variable were carried out.

Since certain types of repetitive behaviour are thought to be associated with ability (see Turner, 1997 for review) ability was entered as a covariate where necessary (see section 3.5 for more detailed explanation of this procedure in relation to the executive function data). Preliminary analyses were undertaken to explore the relationship between categories of repetitive behaviour as measured by the RBQ and chronological age (CA), verbal mental age (VMA) and non-verbal mental age (NVMA). This revealed that, at Time 1, CA was significantly associated with resistance to change on the RBQ ($r=0.45$, $n=34$, $p<0.01$). There were no significant associations between age or ability and RBQ variables at Time 2. Therefore univariate analyses of variance were undertaken except for resistance to change at Time 1.

4.2.5 Time 1 Results

Twenty-nine children categorised as ASD or SLD provided information on the RBQ at Time 1, 12 in the SLD group and 17 ASD. Table 4.2 displays the mean scores for each group and behaviour category.

Univariate analysis of variance show a significant group difference between mean scores for repetitive movements ($F_{(1,27)} = 16.69$, $p<0.01$) and insistence on sameness behaviour ($F_{(1,27)} = 7.69$, $p<0.05$). For both these variables, the mean score is significantly higher for the ASD group than the SLD group indicating the ASD children are reported to have more severe or frequent behaviours of this type. However there was no significant group difference on repetitive use of language ($F_{(1,27)} = 2.72$, n.s.). Nor was there any significant group difference in resistance to change when chronological age was entered as a covariate ($F_{(1,25)} = 3.49$, n.s.). Although no statistical analysis was undertaken, the mean scores reflect higher levels of reported circumscribed interests in the ASD group than the SLD group. This is particularly interesting given the young age of the children and the complex presentation of circumscribed interests.

Table 4.2 Time 1 Mean scores for each repetitive behaviour category

	ASD		**SLD**	
	Mean	**(SD)**	**Mean**	**(SD)**
	N=17		N=12	
Repetitive Movements*	**8.29**	4.52	**2.17**	3.01
Insistence on Sameness Behaviour*	**7.76**	5.21	**3.25**	2.49
Repetitive Use of Language	1.71†	1.65	0.83	0.94
Resistance to Change	1.44‡	1.75	0.08	0.29
Circumscribed Interests	1	0.43	0.27	0.47

* $p<0.05$; †n=16; ‡ n=13

4.2.6 Time 2 Results

Twenty-six children provided RBQ data, 13 in each group. Table 4.3 displays the mean group scores for each category of behaviour. Analyses of variance revealed that repetitive movements ($F_{(1,24)} = 11.46$, $p<0.05$), insistence on sameness behaviour ($F_{(1,24)} = 7.05$, $p<0.05$) and resistance to change ($F_{(1,24)} = 7.88$, $p<0.0$) were significantly different. The differences all reflect more severe or frequent behaviours in the ASD group than the SLD group. In contrast, repetitive use of language was not significantly different ($F_{(1,24)} = 3.46$, n.s.). Once again the mean score for circumscribed interests was higher for the ASD group than the SLD group, although it is not possible to say if this difference is statisticall significant.

Table 4.3 Time 2 Mean scores for each repetitive behaviour category

	ASD		**SLD**	
	Mean	**(SD)**	**Mean**	**(SD)**
	N=13		N=13	
Repetitive Movements*	**5.92**	4.15	**1.69**	1.75
Sameness Behaviour*	**8.23**	5.83	**3.38**	3.04
Repetitive Use of Language	2.46	1.13	1.38	3.09
Resistance to Change*	**1.54**†	1.45	**0.31**‡	0.63
Circumscribed Interests	1.08	0.67	0.18	0.4

* $p<0.05$; † n=12; ‡ n=11

4.3 Change Over time

The presentation of repetitive behaviour is known to alter as a function of age in typically developing young children (Evans et al, 1997; Shentoub & Soulairac cited in Tröster, 1994). However, the change over time of repetitive behaviours in very young children with autistic spectrum disorders has not been assessed. The longitudinal design of the current project allows an initial exploration of the development of repetitive behaviours in very young children with autistic spectrum disorders and speech and language delay between the ages of three and four years. If the development of repetitive behaviours is consistent within the two groups a comparison of the trajectories across group will be possible. This would provide information about whether the developmental trajectories of these two groups were convergent or divergent. Although it would only be with reference to repetitive behaviours, such analysis may lead to further understanding of the development of both groups over time.

The ability of the Repetitive Behaviour Questionnaire to measure change in symptomatology over time has not been explored in previous research. However the interview has been conducted with children and adults of varying ages and showed no signs of being more suited to one age group than another. Furthermore, the questions are designed to capture current behaviours and therefore scores for each time point should be independent of one another.

4.3.1 Participants

For some children, completed RBQ data was available for both time points. Nine SLD children provided data for each RBQ category at Time 1 and at Time 2. Thirteen ASD children provided data for each RBQ category at both times (only 12 for resistance to change category).

4.3.2 Group Analysis

Measures of symptom change were calculated by subtracting the score reported on each RBQ variable at Time 1 from the relevant score at Time 2 for each child. A negative score reflected a *decrease* in recorded symptomatology over time whilst a positive score reflected an *increase* over time. For example a child who scored 15 for repetitive movements at Time 1 and 12 at Time 2 received a symptom change score of –3, representing a decrease in symptom severity. This produced distributions of change for each category of repetitive behaviour.[1]

With the exception of the repetitive use of language variable, the change distributions were approximately normal with modes of 0. The repetitive use of language change variable had a mode of 2 reflecting a tendency for communication to be recorded as increasingly abnormal at Time 2 compared to Time 1. That is to say that the children recruited to this study were described, in general, as more communicatively repetitive at four-years-old than at three-years-old. This suggests that both children with autism and children with speech and language delay display more deviant communication at the age of four than at three. This finding sits well with the knowledge that it becomes increasingly easy to identify communicative deviancy that is distinct from the variation seen in typical development (e.g. Le Couteur et al, 1989; Mawhood et al, 2000).

Table 4.4 displays the mean and standard deviation of the symptom change scores for each group for each category of repetitive behaviour. Independent samples t-tests indicated there were no statistically significant group differences in the change scores for any repetitive behaviour category (repetitive movements $t_{(20)}=0.55$, n.s.; insistence on sameness behaviour $t_{(20)}=0.31$, n.s.; repetitive use of language $t_{(20)}==-0.37$, n.s.; resistance to change $t_{(19)}=0.62$, n.s.).

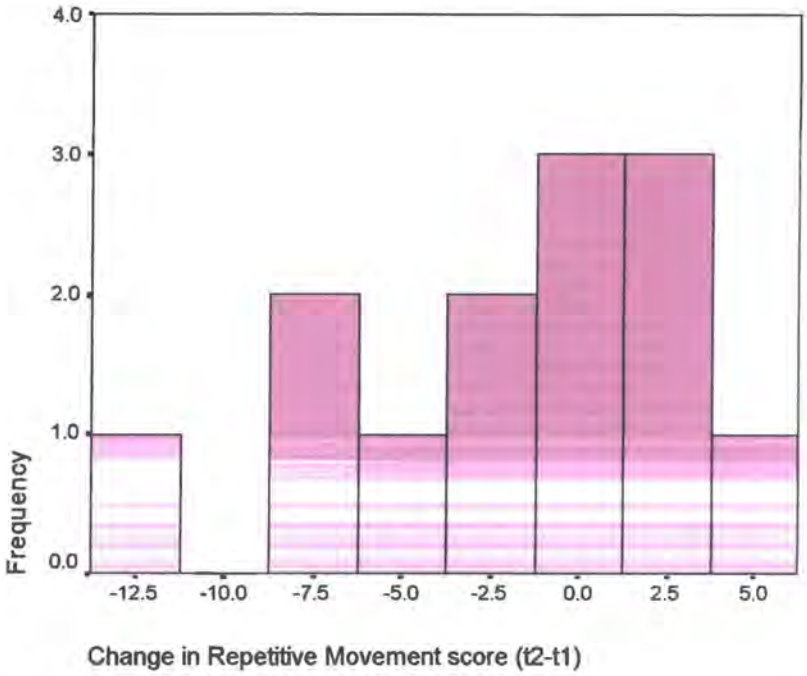
[1]Circumscribed Interests were not analysed since this class of behaviour had a qualitatively different coding scheme.

Table 4.4 Descriptives for Change in Symptomatology scores over time, by group (positive scores reflect greater symptom severity at Time 2; negative scores reflect reduced symptom severity at Time 2)

	ASD			SLD		
RBQ	**N**	**Mean**	**(SD)**	**N**	**Mean**	**(SD)**
Repetitive Movements	13	-1.77	(4.75)	9	-0.78	(3.15)
Insistence on Sameness Behaviour	13	-0.69	(5.44)	9	-0.11	(1.96)
Repetitive Use of Language	13	0.46	(1.56)	9	0.22	(1.39)
Resistance to Change	12	-0.17	(2.29)	9	0.33	(0.87)

It can be seen from Table 4.4 that all the mean values are close to zero (maximum mean value is -1.77) whilst the standard deviations are quite wide, particularly in the ASD group. The variation is likely to be a key reason for the lack of significant group differences, but it also shows that there is no consistent pattern of change in one direction when taken as a group. In fact, this data demonstrates clearly that individual children within each group can have quite distinct change profiles of their own: some children show increases in symptomatology whilst others show decreases in symptomatology. Figure 4.1 presents the ASD group change scores for repetitive movements. This histogram clearly shows change occurring in both directions and can be considered an example of the pattern for the other repetitive variables and groups.

Figure 4.1 Histogram of the change scores for repetitive movements in the ASD group.



Clearly some children in the ASD group do show substantial change in their repetitive movement score over the year. Considering only the central tendencies of the group conceals this pattern of change. Interestingly this change occurs both in the direction of decreased and increased severity. An increase in scores over time indicates increased symptom severity. To explore this further, children were categorised into three groups for each category of repetitive behaviour. Children whose symptomatology score increased over the year were classed as "increasers", children whose score decreased were classed as "decreasers" and those whose score remained the same were classed as "maintainers". Table 4.5 displays the number of children in each group for each symptom score.

Table 4.5 Number of children classed as increasers (I), maintainers (M) or decreasers (D) for each of the symptom variables

RBQ	ASD I	ASD M	ASD D	ASD Total	SLD I	SLD M	SLD D	SLD Total
Repetitive Movements	5	0	8	**13**	1	7	1	**9**
Insistence on Sameness Behaviour	4	2	7	**13**	3	2	4	**9**
Repetitive Use of Language	7	1	5	**13**	3	4	2	**9**
Resistance to Change	3	4	5	**12**	3	5	1	**9**

The important message from Table 4.5 is that symptom scores increased and decreased for children in each group on each repetitive variable. Small expected frequencies make a chi-square analysis inappropriate, however there appears to be no reliable pattern of difference between groups on each variable. The exception to this statement may be repetitive movements where it is interesting to see that no ASD child maintained the same score whereas most of the SLD children did. Whilst there is no directional pattern this does suggest that the presentation of repetitive movements in ASD children is less stable between the ages of 3 and 4 years than SLD children. However, the child who made the change of the greatest magnitude on the repetitive movements variable was an ASD child and the second greatest magnitude of change was for an SLD child: both children showed substantial decreases in reported repetitive movements.

To explore whether individual children were likely to change in the same direction in more than one class of repetitive behaviour, the children with greatest change scores on each variable were identified. The repetitive movement and insistence on sameness variables were used to identify 'changers' since these variables had the widest range of possible scores. For children in the SLD group, substantial change in one class of behaviour did

not relate to substantial change in any other class of behaviour. In fact the child who made the greatest change in each class of behaviour made negligible or no change in the other three categories. In contrast, of the six ASD children who made the greatest change on the repetitive movements variable, four were in the six greatest changers on both the insistence on sameness and resistance to change variables in the same direction of change. However two of these children were noted for increasing symptomatology and two for decreasing symptomatology[2].

In summary, group analysis of symptomatology change over one year failed to produce systematic patterns of change. The central tendencies of each domain were around zero for both groups whilst the wide standard deviations indicated substantial individual variation in the magnitude and direction of change. The lack of overall group trajectories and the within-group variation meant there was no evidence that between-group comparisons would reveal distinctive trajectories and therefore further calculations were not attempted. However it is interesting to note that the ASD children who made substantial change in one class of repetitive behaviour were likely to make substantial change in the same direction in the other classes.

4.4 General Discussion

At the mean age of three years old the two groups had reliably different scores of reported behaviours for repetitive movements and insistence on sameness behaviour. For both these variables, the ASD children were reported to have more severe or frequent behaviours of each type. These findings fit with the conclusions of Turner (1995) that children with ASD were more likely to be reported to display repetitive behaviours than age- and ability-matched comparison groups. It further supports a reliable quantitative distinction in behavioural terms between the two groups of children recruited to this study.

[2] One of these had been engaged upon an intensive home intervention programme that started after the first assessment.

However there was no significant group difference in resistance to change when chronological age was taken into account. It appears that chronological age may be more important in relation to resistance to change than diagnostic status, or ability. Turner (1995) herself argued that resistance to change was a distinct category of behaviour that may not belong to the same taxonomy as the other classes of repetitive behaviour. Only two questions contributed to the resistance to change score: 'how does he/she react if any changes are made to his/her surroundings at home?' and 'how does he/she react if his/her daily routine is changed?' This variable, then, differs from insistence on sameness in that it captures a more reactive behaviour as opposed to an active desire to maintain environments or repeat behaviours.

Similarly, there was no significant group difference in scores for repetitive use of language. This variable was based upon the answers to three questions focussed upon the repetitive use of words or conversation topics. The children assessed in the current sample had low levels of verbal skill: none formed complete sentences and most did not have phrase speech. Parents found answering these questions difficult given the limited linguistic skill of their child and also often commented "don't all children repeat things when they're learning to speak?". Given these limitations it is possible that the repetitive use of language variable is inappropriate for children with very limited language skill.

At the mean age of four years, repetitive movements and insistence on sameness still elicited significant group differences. In addition resistance to change was reported to be significantly different for the two groups. These group differences all reflected more severe or frequent repetitive behaviour in the ASD group than the SLD group and provide further backing for the earlier conclusions that repetitive behaviour can be useful in distinguishing children with autistic spectrum disorders from children with speech and language delay.

Once more, repetitive use of language did not differentiate the two groups. Since resistance to change does elicit significant group differences at Time 2

based upon two questions it seems as if the non-significant findings at Time 1 and Time 2 cannot simply be a result of the variables concerned having a small number of contributing questions and therefore a small range of possible scores. From the current data set it is impossible to say for certain whether this is because repetitive use of language remains inappropriate for both groups of children or whether ASD and SLD children both display repetitive language. However, the mean scores recorded suggest the latter conclusion may be valid.

The analysis of change in scores for each repetitive behaviour category for each child elicited no evidence of systematic within group change. Individual children showed substantial change in their scores from Time 1 to Time 2 but these changes occurred in the directions of increasing and decreasing severity of repetitive behaviour. However, careful analysis of the data highlighted a tendency for those children with autistic spectrum disorders who made the greatest magnitude of change to be the same across the different classes of behaviour. Although these large changes happened in both directions, for each child concerned (four in total) the change was in the same direction. Therefore it appears that children with autistic spectrum disorders may be less stable in their repetitive behaviours between the ages of three and four than their speech and language delayed counterparts. Future research would benefit from attempting to replicate these findings with larger sample sizes. If these 'unstable' subgroups are still identified they may provide a useful way for subgrouping children with autistic spectrum disorders and assessing whether the heterogeneity of cognitive and social functioning is associated with these subgroups. The following chapter examines whether executive function performance is associated with repetitive behaviour in this group of very young children.

Chapter 5

Symptomatology and Executive Function Skill

Autistic symptomatology is commonly divided into three categories of behaviour: social, communicative and repetitive behaviour. A complete explanation of autism should be able to explain the nature and severity of behaviours in each of these domains. In this way, a key prediction of the executive dysfunction hypothesis of autism is that the severity of executive impairment should be related to the severity of symptomatology displayed by the individual (e.g. Pennington et al, 1997; Turner, 1997; Turner, 1999b). Similarly, the severity of symptomatology should be related to executive function performance.

The theoretical link between executive function performance and symptomatology has been made most clearly for repetitive behaviours, but some studies have begun to consider the relationship between executive function and social behaviour in autism (e.g. Dawson et al, 1998; Griffith et al, 1999; McEvoy et al, 1993). However, these studies have produced varied and inconclusive findings. To my knowledge, there have not been any studies looking at the relationship between executive function performance and communication skills, although plausible links have been suggested (Hughes, 2001). Clearly the relationship between executive function skills and social and communication behaviours also requires further exploration.

The current chapter considers the current literature for the relationship between executive function performance and autistic symptomatology. The literature is not extensive but, where possible, the focus will be on the

relationship between symptomatology and executive function skill in very young children.

5.1 Repetitive Behaviour and Executive Function Performance

Repetitive behaviours may be the direct consequence of a failure to generate or inhibit volitional behaviour. An individual who has impairments in generation of responses is likely to find the production of alternative and variable behaviours or interests profoundly difficult. A child with inhibitory problems might struggle to stop an ongoing behaviour or activity. Both of these pathways could lead to a pattern of highly repetitive and invariant behaviour (Turner, 1997). These predicted associations are supported by at least three features of repetitive behaviour in autism, as summarised by Hughes (2001).

> *'First, repetitive behaviour is seen at many levels and in many forms; this pervasiveness suggests a general breakdown in the systems that control behaviour (Ridley, 1994). Second, a primary impairment in executive control could explain why such behaviours are so prevalent, pervasive, and persistent in autism. Third, an executive account would predict that reducing the demands for internal control of behaviour by increasing structure of the environment would lead to reduced stereotypy. This prediction is supported by the results of several independent studies [(Clark & Rutter, 1981; Dadds et al, 1988; Shopler & Olley, 1982)].'* (Hughes, 2001, p. 267)

Expanding upon the proposed pathways from inhibition or generation failure to repetitive behaviour, Turner (1997) argues that different classes of perseverative error would be related to different classes of repetitive behaviour. On this view, perseveration of a previous motor response would be associated with low-level stereotyped movements whilst perseveration to a previous cognitive set would be associated with higher classes of repetitive behaviour. Following an extensive study of 22 children and adults with high-

functioning autism (VIQ>75), 22 children and adults with low-functioning autism (VIQ<75), and two appropriate control groups for whom detailed executive function performance and repetitive behaviour data was collected, Turner reported that there was a definite pattern of association between excessive repetition of one response and parental report of stereotyped movements in the group with autism (Turner, 1995; Turner, 1997). This relationship held for both ability levels despite the HFA group failing to display an inhibitory deficit in comparison to the control group. The HFA group also displayed a relationship between immediate repetition of a response and circumscribed interests. Individuals who showed greater difficulty in moving attention away from the first dimension of the ID/ED task were more likely to display repetitive use of language and circumscribed interests. Considering the relationship between repetitive behaviour and generativity impairment, a negative association between the number of novel responses produced on a task of ideational fluency and an insistence on sameness and circumscribed interests was reported.

Although Turner makes a persuasive argument for specific links between certain classes of repetitive behaviour and types of executive dysfunction, these findings have yet to be replicated. A recent study by South, Ozonoff and McMahon (2001) observed no relationship between the number of perseverative errors on a set shifting task (the WCST) and repetitive behaviours in high-functioning individuals with autism or Asperger's Syndrome. Clearly, further work is necessary to address this discrepancy.

The present chapter explores the relationship between executive function and repetitive behaviour in preschoolers with autistic spectrum disorder and speech and language delay. This is important for the understanding of the role of executive dysfunction in the developmental progression of autism. To explore this question, detailed information concerning repetitive behaviours was systematically collected on recruitment and at follow-up 12 months later. Executive function tasks designed to assess inhibitory control, working memory and planning were also administered to these children. Correlations were calculated between variables representing overall performance and error

responses on the executive function tasks and summary scores of repetitive symptoms.

5.2 Participants

All children who were recruited to the project were available for these analyses since the interest in the dimensional aspects of symptomatology rather than categorical aspects meant none were excluded because of diagnostic uncertainty.

5.3 Materials and Procedure

The executive function measures were administered as described in Chapter 3. The Repetitive Behaviour Questionnaire was used to assess symptomatology (see Chapter 4 for further details). The RBQ provides summary scores for repetitive movements, insistence on sameness behaviour, repetitive use of language, circumscribed interests[1] and resistance to change, permitting a closer examination of the relationship between types of perseveration and classes of repetitive behaviour (Turner, 1997).

5.4 Statistical Analyses

Several considerations were taken into account in deciding the statistical approach to this question. As explained in Section 4.2.4 the use of the summary scores from the RBQ enabled the use of parametric analyses (Clark-Carter, 1997). Preliminary analyses had revealed that, at Time 1, CA was significantly associated with Resistance to Change on the RBQ ($r=0.45$, $n=34$, $p<0.01$) and that there were 10 significant associations between CA, VMA or NVMA and executive function variables and a significant inter-correlation between VMA and NVMA (see Chapter 3, Section 3.4). In order to keep the analyses as consistent as possible and to take age and ability into account, it

[1] The variable for Circumscribed Interests was qualitatively different from the rest of the RBQ variables (see Chapter 4 for more details) and therefore was not included in this analysis.

was decided to enter chronological age and non-verbal ability as covariates. Verbal mental age was not included as a third covariate since this would reduce the degrees of freedom without offering additional information about the child's ability (given the inter-correlation between VMA and NVMA, see section 3.4). Appendices II and III present correlational analyses between key executive function variables and social and communicative behaviours as measured by the ADI-R and ADOS-G and the inter-correlations between these two instruments.

Measures designed to assess overall performance and perseverative responding on the executive function tasks were included in the present analyses. The tasks were designed to tap the following executive function skills: inhibition-and-implementation of responses, inhibition of cognitive set, working memory and planning. Correlational analyses are reported for these executive function variables with repetitive behaviours at Time 1 followed by Time 2.

5.5 Time 1 Results

Fort-six children were recruited at Time 1 and completed RBQs were received for 34 children (a smaller number of children provided both RBQ and executive function data on each task (Table 5.1)).

Table 5.1 Sample sizes available for each executive function task at Time 1.

		RBQ (N=34)
A-not-B Task		27
A-not-B Invisible Task		20
Detour Reach Task	Knob Route	28
	Switch-Reach Route	13
Three boxes	Stationary	32
	Scrambled	30

Four variables measuring perseveration were excluded from analyses at Time 1. The number of errors on either version of the Three Boxes Tasks was low (see Chapter 3). Furthermore, only one child made more than one consecutive perseverative error to a specific box or location on the stationary version of the Three Boxes task. Therefore the number of perseverative errors on these tasks were not entered into correlational analysis. Similarly, the longest run of incorrect responses on the Detour Reach switch-reach route was excluded since this run could comprise a multitude of error types (see Chapter 3) and therefore interpretation of this variable as perseverative responding was difficult.

Table 5.2 presents the correlation coefficients for key executive function variables and four classes of repetitive behaviour identified by the RBQ at Time 1 with chronological age and non-verbal mental age entered as covariates. Four correlations were significant out of a total number of 52 correlations. At the $p<0.05$ level three significant correlations would be expected by chance, therefore these findings may represent chance findings.

The only significant correlations were between variables on the A-not-B tasks. The longest run of incorrect responses reflects a tendency to perseverate to location on both the A-not-B tasks. On the A-not-B Task, this variable was positively correlated with resistance to change ($r=0.48$, $df=23$, $p<0.05$) when chronological and non-verbal mental age was taken into account. The same variable on the A-not-B Invisible Displacement Task was significantly associated with repetitive movements ($r=0.57$, $df=16$, $p=0.01$) and insistence on sameness behaviour ($r=0.55$, $df=16$, $p<0.05$). The positive direction of these relationships indicated that where a child made a longer run of perseverative errors they were reported to display greater repetitive behaviour. The percentage of perseverative errors made on the A-not-B Invisible Displacement Task was also positively associated with insistence on sameness behaviour ($r=0.56$, $df=16$, $p<0.05$). Once again a relationship between increased perseveration and greater repetitive behaviour was reported.

Table 5.2 Partialled correlation coefficients (r) for key variables of executive performance and perseveration and repetitive behaviour summary scores at Time 1.

Task variable	**Repetitive Movements**	**Insistence on Sameness**	**Repetitive use of Language**	**Resistance to Change**
A-not-B Task				
% reversals correct	-0.07	0.09	-0.03	-0.06
% perseverative errors	0.27	0.03	0.20	0.30
Longest perseverative run	0.35	0.19	0.25	**0.48***
A-not-B Invisible Displacement Task				
% reversals correct	-0.53	-0.35	-0.17	-0.38
% perseverative errors	0.34	**0.56***	0.11	0.38
Longest run of perseverative errors	**0.57 ****	**0.55***	0.19	0.31
Detour Reach, knob route				
Trials to criterion	0.27	0.46	0.05	0.29
Longest run errors	0.19	0.02	0.27	0.02
Detour Reach, switch-reach route				
Trials to criterion	0.07	0.28	0.35	0.29
Confusion	-0.22	0.28	0.53	0.38
Direct reach errors	-0.06	-0.52	-0.26	-0.27
Three boxes visual search - stationary version				
Efficiency ratio	-0.12	-0.17	-0.28	0.18
Three boxes visual search – scrambled version				
Efficiency ratio	-0.20	-0.03	0.05	0.11

* $p<0.05$; ** $p<0.01$

5.6 Time 2 Results

Forty-one children were followed up after 12 months, 30 families returned the RBQ.

Table 5.3 Sample sizes available for each executive function task.

		RBQ (N=30)
A-not-B Invisible Task		27
Detour Reach Task	Knob Route	30
	Switch-Reach Route	24
Sorting Task		29
Six boxes	Stationary	29
	Scrambled	30
Marbles		30

Performance on the Sorting Task was measured by categorising the children into four groups: fail Rule 1, pass Rule 1 and fail Rule 2, pass Rule 2 and fail Rule 3, and pass Rule 3. Since the variable was ordinal, with a small number of levels, parametric correlations were inappropriate. Therefore non-parametric correlations[2] were conducted for this task, which meant that chronological age and non-verbal mental age could not be entered as covariates for this task. The correlation coefficients are presented in Table 5.4 alongside the parametric correlations conducted for the other tasks. Moreover, only 10 children attempted the testing phase of Rule 2 and perseverative errors were not common in the responses of these individuals. Therefore, to keep the number of comparisons as low as possible, error variables were not analysed for the Sorting Task.

[2] Kendall's Tau was used because it provides a better estimate of the value that would have been obtained from the population than Spearman's Rho (Howell, 1997).

On the other executive function tasks, the longest run of perseverative responses on any variable was around four, and many children on each variable failed to make a run of perseverative errors. However, there were some children (who varied depending on the task) who did show signs of perseverative responding. Therefore the longest runs of perseverative error response variables were retained in all analyses.

Only three combinations of executive function variable and repetitive variable (out of 68 possible combinations) are related at a significance level of 0.05. Three significant associations would be expected by chance. Therefore there is no reason to believe these significant associations represent a real finding. Table 5.4 presents the partialled correlation coefficients for the executive function variables and the four RBQ category scores with CA and NVMA as covariates.

The A-not-B Invisible Displacement Task produced one significant correlation with repetitive behaviour. The longest run of perseverative errors was positively associated with repetitive use of language ($r=0.70$, $df=23$, $p=0.00$). Longer perseverative runs were associated with greater repetitive language.

No variables on the knob route or the switch-reach route of the Detour Reach Task were significantly associated with any class of repetitive behaviour. Nor did the non-parametric correlational analyses for the Sorting Task reveal any association between the rule reached and repetitive behaviour.

Table 5.4 Partialled (except Sorting Task) correlation coefficients (r) for key variables of executive performance and perseveration and repetitive summary scores at Time 2

Task – variable	**Repetitive Movements**	**Insistence on Sameness**	**Repetitive use of Language**	**Resistance to Change**
A-not-B Invisible Displacement task				
% reversals correct	-0.07	-0.20	0.07	-0.26
% perseverative errors	-0.06	-0.03	0.21	-0.12
Longest run of perseverative errors	0.19	0.05	**0.70****	0.27
Detour Reach Task, knob route				
Trials to criterion	-0.07	0.02	0.12	0.02
Number of errors	-0.14	-0.25	-0.21	-0.16
Detour Reach Task, switch-reach route				
Trials to criterion	0.31	0.17	-0.00	0.18
Direct reach errors	-0.02	-0.02	0.08	0.01
Rule confusion	0.13	-0.04	0.31	0.04
Sorting Task	(*non-parametric*)			
Rule reached (and passed)	-0.25	-0.07	-0.21	-0.18
Six Boxes visual search task – stationary version				
Efficiency ratio	0.14	0.29	0.14	0.30
Longest run of perseverative errors to location/box	-0.09	-0.24	-0.10	-0.27

Table 5.4 cont.

Task – variable	**Repetitive Movements**	**Insistence on Sameness**	**Repetitive use of Language**	**Resistance to Change**
Six Boxes visual search task – scrambled version				
Efficiency ratio	-0.17	-0.23	-0.36	-0.17
Longest run of perseverative errors to a specific location	0.17	0.06	-0.03	0.08
Longest run of perseverative errors to a specific box	0.24	**0.39***	0.21	0.24
Marbles Task				
% trials correct	0.21	**0.39***	0.25	0.30
Number of errors	-0.20	-0.36	-0.25	-0.26
Number of perseverative errors	-0.09	-0.26	-0.22	-0.22

*$p<0.05$; ** $p<0.01$

The longest run of perseverative errors to box on the scrambled version of the Six Boxes Task was significantly associated with an insistence on sameness behaviour ($r=0.39$, $df=26$, $p<0.05$) when chronological and non-verbal mental age was taken into account. The correlation between efficiency ratio on this task and repetitive use of language approached significance ($r= -0.36$, $df=26$, $p=0.06$). There were no significant associations with the stationary version of this task.

Higher percentages of trials correct on the Marbles Task was associated with greater insistence on sameness behaviour when age and ability was taken into account ($r=0.39$, $df=26$, $p<0.05$). The negative relationship between sameness behaviour and the number of errors made approached significance ($r= -0.36$, $df=26$, $p=0.06$).

5.7 Summary

The number of significant associations reported at either Time 1 or Time 2 was equivalent to that which would be expected by chance at the 0.05 level of significance. This suggests that there is no systematic pattern of relationships between repetitive behaviour and executive function task performance in the present sample.

5.8 General Discussion

Correlations between executive function variables and repetitive behaviour symptomatology were calculated at both time points. At each time point a small number of the correlations were significant, however this number was roughly what would be expected by chance. On one hand, these numbers of association may reflect a genuine lack of relationship between the executive skills underlying performance on these tasks and repetitive behaviours. On the other, it is possible that statistical or procedural issues could have contributed to the findings. Section 5.9.1 discusses this second possibility.

5.8.1 Methodological Issues

The RBQ reported that the ASD group were quantitatively more repetitive than the SLD group on all classes of behaviour except repetitive use of language (Chapter 4). There were also qualitative differences in at least some of the repetitive behaviours reported in this group (e.g. some children with autistic spectrum disorder but no children with speech and language delay displayed prototypically autistic repetitive behaviours such as circumscribed interests with smoke detectors or hand flapping). Despite these differences in symptomatology, there was no significant relationship with executive function skill.

However, the RBQ was not developed specifically for use with this very young sample, nor designed to measure behavioural change. These issues are discussed in more detail in sections 6.2.2 and 6.2.4. Also, it is possible that skills untapped by the current study might be related to symptomatology. The

choice of executive function tasks and their importance for the interpretation of the study's findings will be discussed in more detail in the final chapter (Section 6.4).

Given the combined sample size, power should have been sufficient to highlight any relationships between the domains. Additionally, the range of scores obtained from the use of summary domain scores provided a good basis for the analysis. The issue of statistical power will be discussed in relation to the entire study in section 6.1.

5.8.2 How do these findings relate to current literature?

No published work has examined the relationship between executive function and repetitive behaviour in very young children with autism. However, Turner (1997; 1995) reported some evidence to suggest a relationship between repetitive behaviour and autistic symptomatology in older children and adolescents with autism. More specifically, Turner reported associations between perseverative responding and stereotyped movements, impaired attention shifting and repetitive use of language and circumscribed interests, and impaired generation of novel responses and insistence on sameness and circumscribed interests. The present study has failed to replicate Turner's findings, and therefore has suggested that such a relationship may not exist in very young children with autism. Rather, the findings are consistent with the results of a recent study with 61 much older children and adolescents with high-functioning autism and Asperger's syndrome (South et al, 2001). Preliminary results reported no association between the number of perseverative errors on the Wisconsin Card Sorting Task and circumscribed interests, rigid routines/rituals, stereotyped movements or object preoccupations as measured by a version of the RBQ. Once more this calls into question an executive dysfunction hypothesis of autism, which would predict that severity of executive impairment ought to relate to severity of symptomatology.

In contrast the executive function hypothesis, it is possible that a preference for repetitive behaviours reduces opportunities where executive function

skills can be practised and developed. A child who engages in repetitive and invariant actions may remove themselves from situations where the inhibition of one response may be practiced (for example childhood games such as Simon Says or musical statues). The observations that caged animals often display repetitive, invariant and functionless stereotyped behaviour and these behaviours have been shown to correlate with impaired response selection would be consistent with this alternative hypothesis (Garner, 1999; Garner & Mason, 2001).

To conclude, there is mixed evidence concerning the possible relationship between perseveration and repetitive behaviour in autism. Further research in all age groups of individuals with autism is required to elucidate these relationships. This work should include assessment of both inhibitory failure and generation of novel responses to test both the predictions of Turner's hypothesis. These studies are essential before a true evaluation of this prediction of an executive dysfunction hypothesis of autism can be made. In fact, they may lead to a reworking of the executive dysfunction hypothesis to accommodate distinctions between inhibitory control and generation of behaviour.

Chapter 6

Discussion

The final chapter will summarise the experimental findings of the project, discuss their validity in relation to methodological issues, and then consider the implications for theoretical and practical aspects of autistic spectrum disorders.

6.1 Summary of findings

This thesis set out to investigate the validity of the executive dysfunction hypothesis of autism in relation to the development of very young children with autistic spectrum disorders. The executive dysfunction hypothesis seeks to explain the development of autistic symptomatology as a consequence of impaired executive function skill. A strong version of this hypothesis would predict that executive deficits are early emerging, persistent over time and related to the severity of autistic symptomatology. Moreover, early executive function deficits ought to be related to later symptomatology, either in terms of qualitative or quantitative relations. These predictions were evaluated in three different ways: cross-sectional group comparisons, development over time for individual children and correlational analyses between executive function performance and repetitive behaviour.

Few children provided good quality data for the executive tasks at Time 1 and therefore any interpretation of these analyses must be very cautious. Cross-sectional group comparisons revealed very few significant differences in task performance between children with autistic spectrum disorders and children with speech and language delay at Time 1. Moreover, at Time 1, one of these statistically significant differences reported *fewer* errors for the children with autistic spectrum disorders than the children with speech and language delay.

At Time 2 there was some evidence that children with autism made more perseverative responses on certain tasks, but not others, and that they had set-shifting impairments. However, even at this age the groups performed at similar levels to one another on most performance and error variables. The small sample sizes available at Time 1 meant it was not possible to consider statistically the development over time of executive function skill in the current sample. Instead, the performance on executive function tasks was plotted for the small number of children who had attempted the tasks at both time points. Although limited, this process did not highlight any clear developmental trajectories of executive function skill in these samples.

Repetitive behaviours did distinguish between the two groups. The children with autistic spectrum disorders were reported to display greater repetitive movements, insistence on sameness and resistance to change (Time 2 only) than children with speech and language delay. Change over time on these classes of repetitive behaviour was not consistent within- or between-groups. Children belonging to the same group made change in differing directions and the overall group means were around zero. Although there were no clear patterns it was notable that four of the autism group who made substantial change in one class of behaviour also made substantial change in the same direction in other classes. Two of these children showed increased behaviour at Time 2 and the remaining two showed decreased behaviour.

The number of significant associations reported between the repetitive behaviour variables and executive function variables did not exceed the number of significant associations that would be expected by chance at either age. Nor was there any evidence to support Turner's (1997) hypothesis that specific classes of repetitive behaviour were associated with specific types of executive error.

Whilst these findings challenge the executive function hypothesis, it is also possible that the study may have failed to demonstrate differences and relationships that do really exist. Important issues relating this second possibility include the statistical power of the study, the validity of the two

groups of children, the validity of the executive function tasks administered in the study as well as the reliability and sensitivity to change over time of the ADI-R, ADOS-G, RBQ and executive function tasks. These methodological issues will be discussed in more detail in section 6.2. Alternatively, the tasks administered in the current study may not have tapped the specific executive function skills that are impaired in individuals with autism. This issue, and the more general question of fractionation versus integration of executive function, are dealt with in section 6.4.

6.2 Are the current findings valid?

6.2.1 Statistical Power

A priori power calculations based upon the average effect size of 1.0 reported by Pennington and Ozonoff (1996) indicated that groups of 17 would produce a power of 0.8. Both groups in the study had at least 17 members. Unfortunately, not every child attempted every task and therefore some analyses were conducted with smaller sample sizes. For this reason analyses were approached cautiously to maximise power. Preliminary analysis revealed that most dependent variables were not associated with chronological, verbal or non-verbal mental age. The degrees of freedom associated with the analyses are reduced for every variable entered as a covariate. Therefore these variables were only entered as covariates where there was reason to believe they were related to the dependent variables. Furthermore, given the substantial inter-correlation between verbal and non-verbal mental age only one of these variables was ever entered in an analysis. There are two further reasons to believe that the results meaningfully reflect the real pattern. Firstly, the variables analysed were carefully chosen to address specific theoretical questions. Secondly, the large number of comparisons increased the risk of false-positive findings: in this context the small number of significant findings is perhaps even more striking.

Future research would benefit from the recruitment of greater sample sizes. The practical and financial constraints of this type of longitudinal work with relatively rare children makes very large sample sizes prohibitive for one

person and therefore this type of work is a prime candidate for carefully managed multi-site collaborations.

6.2.2 Validity of the Diagnostic Groups

The second issue concerns the validity of the groups compared in the study. The lack of group differences may have resulted because the two groups did not represent distinct groups of children. Whilst some authors have argued that social skills in speech and language delay are impaired in a similar manner to those in autism (e.g. Farmer, 2000) and that the two groups are arbitrarily differentiated by quantitative cut-offs whilst the quality of impairment is similar (e.g. Howlin, Mawhood, & Rutter, 2000; Locke, 1997; Mawhood, Howlin, & Rutter, 2000), there are several reasons to support the distinction between the two groups of young children in the current study.

Two studies have supported the validity of making diagnostic distinctions at very young ages using the ADI-R and ADOS-G in combination with expert clinical judgement (Cox et al, 1999; Lord, 1995). Specifically, the Lord study demonstrated that reliable differentiation between children with autism and those with speech and language delays was possible by the age of two or three years (Lord, 1995). In the current study, the groups differed quantitatively on the summary scores for the socialisation, communication and repetitive behaviour domains on the ADI-R and ADOS-G domains at both times. Moreover, the quality of some behaviour of the children in the two groups was also notably different. For example, several of the children in the autistic spectrum disorders group, but none of the speech and language delay group, were reported to have circumscribed interests or unusual preoccupations of a prototypically autistic nature (see Appendix IV for some case study examples).

Furthermore, by comparing the performance of the current samples with published work of typically developing children, it seems that the two samples were performing at skill levels that would be expected by their mental age. This suggests that neither group was impaired in their executive skill (at least as measured by the tasks administered) in relation to their

developmental level. Therefore the lack of group differences cannot be attributed to an executive deficit in speech and language delayed children.

6.2.3 Validity of the Executive Function Tasks

The third issue addresses the validity of the executive function task measures administered in the study. The tasks administered in this study were thought to tap inhibition-and-implementation (A-not-B Tasks, Detour Reach Task), set-shifting (Sorting Task), working memory (Three and Six Boxes Tasks), and planning (Marble Task). Developing tasks for such young children is very challenging, not least because of the need for non-verbal tasks. However, the tasks of inhibition-and-implementation and working memory administered in this study have already been used in other published studies to measure these skills in young children (e.g. Dawson et al., 1998; Diamond et al, 1997; Griffith et al, 1999; Hughes, 1998a, b; Hughes & Russell, 1993; McEvoy et al, 1993).

Although the procedure for the A-not-B Tasks in the current study follows that used in a number of other studies (e.g. Diamond et al, 1997; Wellman, Cross & Bartsch, 1986), a meta-analysis published since the start of this project has emphasised that the number of correct trials required before a reversal trial may influence the inhibitory control demands made by the task (Marcovitch & Zelazo, 1999). It appears that the greater the number of trials presented before each reversal the more difficult it is for the participant to then inhibit that response and implement the alternative. If a greater number of correct trials had been required in this study it is possible that inhibition-and-implementation deficits would indeed have been observed in very young children with autistic spectrum disorders. However, given the young age of these children, it is important to limit the time demands of the task: it is also possible that the increased length of a task would have resulted in the children attempting fewer reversal trials before they became fatigued.

There is a general consensus in the literature that the A-not-B and Detour Reach tasks tap inhibitory control, that working memory is assessed by the Boxes Tasks, and that tasks on which the Sorting Task was based assess set-

shifting skill (see Chapter 1). In contrast, tasks of planning have not been used in prior studies of early executive function. Therefore the Marbles Task was developed specifically for this study to assess planning at a lower developmental level. This task was very popular with the children in the study, and performance was strikingly good. Interestingly, the performance of a pilot sample of typically developing three-year-olds in nursery was so mixed that the task was nearly eliminated from the battery: we thought the children with developmental delays in this sample would find the task too difficult. The results were quite the contrary. The success of this task may have implications for future task development and the application of similarly captivating tasks as part of therapeutic approaches.

Planning, as defined by Welsh (1991), requires that the child has formulated a series of responses before beginning to act upon the first response. The prototypical task of planning is the Tower of Hanoi, although maze tasks are commonly used with young children in clinical settings (Leezak, 1995). Optimal performance on either of these tasks relies upon the participant evaluating a series of moves before acting. Sub-optimal performance may occur when the child has not pre-evaluated a series of moves before acting. The Marbles Task further simplified the concept of using mazes to assess planning in order to make the task appropriate for these very young children with developmental delays. However, this simplification may have resulted in the planning component being compromised. Optimal performance on the Marbles Task did not require the child to pre-evaluate a series of moves before acting upon one. It was possible for the child to reduce the task to a simple yes/no decision for each possible route. Despite this, the behaviour of some children strongly suggested they were evaluating a number of alternatives before acting upon one. In these cases children could be seen tracing routes with their eyes or fingers before identifying which route was unblocked and then moving the marble to that route.

6.2.4 Assessment over time

For a longitudinal study it is important to use valid and reliable measures that are unlikely to be contaminated by practice effects and are sensitive to change over time. Other studies with pre-schoolers have re-administered some of the current executive function tasks after the period of a year or so: the Detour Reach Task was administered to a group of typically developing children at the ages of four and five years (Hughes, 1998b) and the A-not-B Invisible Displacement Task was administered to toddlers with PKU and comparison groups of typically developing toddlers several times between the ages of 15 and 30 months (Diamond et al, 1997). Neither of these studies reported practice effects ensuing from the re-administration of the tasks. Moreover, the observation that the performance of individual children in the current study sometimes worsened over the year suggests that it is unlikely that practice effects played a role in performance. Turning to sensitivity to change, several children progressed from floor to ceiling performance on the knob or switch-reach routes of the Detour Reach tasks over the year, which may suggest that the task was not sufficiently sensitive to subtle changes in an individual's executive function skill development. However many other children displayed more subtle changes over the year on this and the other tasks. Further work on the reliability over time and the sensitivity to change of executive function tasks is required before these two issues can be resolved.

To the extent that it could be investigated, the current study reported developmental change in executive function skill in both directions: in either group some children improved whilst others worsened. This may simply represent the fact that individuals with autism can be extremely variable in their behaviour, or it may be that development over the period of one year is not informative or reliable. In both cases, longitudinal studies of development over a longer period of time ought to provide more information about the developmental trajectory of executive function skills.

It is important to note that the RBQ was not initially designed to measure change over time, or to be used with such very young children. Several studies have indicated that the ADI-R and ADOS-G are relatively reliable and

valid in very young children over a period of one to three years (Cox et al, 1999; Lord, 1995). However, the ADI-R was designed to assess a child's developmental history up until the time of administration and the ADOS-G to take a snap-shot of the child's behaviour at that specific time. These instruments were used to form the diagnostic groups and were not used to measure change in symptomatology for this study. To my knowledge, no study has looked at change in symptomatology over time as measured by the RBQ in any sample of individuals with autistic spectrum disorders. Whilst this reflects the novel contribution of the current study to the existing literature, it means there is no agreed approach to using this instrument to measure change. However there is no opportunity to compare the current findings to other research, or to evaluate how sensitive the three or four-point scale of behaviour on each question was.

The use of subgroups of individuals who have a diagnosis of autistic spectrum disorders may provide important information upon the nature of executive function skill and its relationship to the experience of being autistic. Although the current data has not elicited a clear candidate variable on which to base the subgroups there are a number of sensible places to start. For instance, children with autism who experience regression may fail to develop their executive skills; or perhaps sub-groups would be best formed according to specific behavioural profiles (e.g. Sevin, Matson, Coe, et al, 1995; Wetherby, Prizant & Hutchinson, 1998). An alternative approach would be to identify groups on the basis of psychologically valid constructs, perhaps even forming these groups from children with speech and language delay, pervasive developmental disorders or developmental delay regardless of clinical diagnosis. Clearly a substantial amount of research is required to explore the most appropriate basis upon which to form sub-groups before the possibility of differential developmental trajectories of executive functioning can be investigated further. It may even be appropriate to use detailed individual case studies rather than group comparisons, as is common in clinical neuropsychology.

6.3 Relating the Findings to the Existing Literature

Having considered the methodological issues concerning the data collected in this project, the findings can be interpreted in relation to the existing literature. In older children and adults with autism, generativity, planning and flexible set-shifting are the areas of most severe deficit, whilst more simple inhibitory skills are relatively unimpaired (Jarrold, 1997; Ozonoff, Pennington, & Rogers, 1991a; Pennington & Ozonoff, 1996; Turner, 1999a). Inhibition-and-implementation skill in children with autistic spectrum disorders, measured by the same tasks as the current study, is reported to be unimpaired four four-year-olds (Griffith et al, 1999) but impaired for five-year-olds (e.g. Adrien et al, 1995; Dawson et al, 1998; Hughes & Russell, 1993; McEvoy et al, 1993). The Three and Six Boxes Tasks did not elicit an autism-specific impairment at the age of four years (Griffith et al, 1999). The current study reflects these findings in so far as there was little reported impairment for three- or four-year-old children with autism on tasks of inhibition-and-implementation and working memory. Signs of impairment on the more complex set-shifting task in four-year-olds echoed the deficits observed in older children's set-shifting skill. However the lack of any planning impairment in the four-year-olds does not fit with the existing data.

Comparisons between the current data and other published studies suggests that three- and four-year-old children with autistic spectrum disorders and speech and language delays display inhibition-and-implementation and working memory skills that can be considered appropriate for their verbal and non-verbal mental age equivalents (see Chapter 3 for details). However, performance on the set-shifting task for both groups was less successful than that of typically developing two-and-a-half-year-olds (Zelazo, Reznick, & Pinon, 1995). This is particularly interesting since the methodological variations applied in the current study to focus the task upon specific set-shifting skills would seem to reduce the working memory and cognitive flexibility demands of the task.

In the original Zelazo study, participants were taught the two sorting rules at the outset of the task. Testing on each rule occurred later, with no reminder of the rules. Children had to hold in mind both rules whilst performing the first rule and then switch to the second rule without the support of a reminder. In the current study, each rule was taught directly before the testing phase of the rule. Working memory demands were therefore reduced and support given to assist the set-shift. However, many of the children in this study were unable to shift from Rule 1 to Rule 2 even with the support provided by the task (despite displaying the ability to sort by colour and shape during an ability assessment). The precise type of support provided by a task is likely to be very important. For example, one study with older individuals with autism reported no autism-specific impairment in a card sorting task but an impairment on an object sorting task (Minshew et al, 1992). The existing studies of set-shifting in young children have used cards (e.g. Hughes, 1998; Zelazo et al, 1995), however the current study used three dimensional blocks that needed to be posted into a box. The impact of this ought to be explored further and considered in relation to young children: the method of presentation of this type of task may be critical to performance. The results from the current study suggest that very young children with autistic spectrum disorders have substantial difficulties with set-shifting that are not alleviated by the supportive method of presentation applied here. These difficulties may reflect a response control failure or a difficulty at the conceptual cognitive shifting level and are in line with the data reporting set-shifting impairment in older children with autistic spectrum disorders.

The lack of a planning impairment in autism, as measured by the Marbles Task, is in contrast to the pervasive difficulties on the Tower of Hanoi tasks reported in older individuals with autistic spectrum disorders (e.g. Pennington & Ozonoff, 1996) and with clinical descriptions of difficulties with the planning demands of everyday life (e.g. organising a shopping trip, cooking, dressing). Given the methodological constraints of the Marbles Task it is clear that further work on the nature of planning skill in young children with autism is required. The development of other non-verbal tasks in which to measure planning behaviour in young children would facilitate this process.

Three published studies have explored the relationship between executive function performance and certain aspects of autistic symptomatology in young children (Dawson et al, 1998; Griffith et al, 1999; McEvoy et al, 1993; see Chapter 5). The Griffith and McEvoy studies reported significant relationships between the number of perseverative errors on a task of inhibition-and-implementation and measures of joint attention and social interaction. In contrast, the Dawson study found an association between the percentage of reversals correct on an A-not-B Task and immediate imitation but no relation with joint attention or several other variables. No published studies have examined the relationship between repetitive behaviour and executive function skill in very young children, but in older children and adolescents the evidence for a relationship is mixed (South et al, 2001; Turner, 1997).

The current study reported no significant pattern of associations between repetitive behaviour and executive function performance, and a preliminary analysis of summary social and communication domain scores also revealed no reliable association with executive function skill. It is possible that more fine-grained analysis between individual aspects of behaviour, such as delayed echolalia, joint attention, or response to name may have produced evidence of a relationship between behaviour at this more specific level and executive function performance. However, the number of comparisons needed by such analysis precluded it from being undertaken in the current study. Future research should seek to clarify the theoretically plausible relationship between repetitive behaviour and executive function through the careful comparison of errors with individual classes of repetitive behaviour in a wide range of individuals with autism. Similarly, the theoretical links between social or communication behaviours and executive function should be refined in a manner that leads to specific predictions about individual aspects of behaviour which could then be tested empirically.

6.4 Implications for Generativity

One possible explanation for the lack of executive impairment in the current study is that the tasks did not tap the specific individual skills that may be impaired in autism. Several different executive function skills were assessed in the study: inhibition-and-implementation, cognitive set-shifting, working memory and planning. As already noted, the current findings were broadly consistent with the existing literature, with the exception of planning skills.

Substantial impairments in generativity for older children and adults with autistic spectrum disorders have been demonstrated in several studies (e.g. Minshew et al, 1992; Rumsey & Hamburger, 1988; Turner, 1999a). The commonly used tasks of word, ideational and design fluency place substantial demands on verbal skill and are therefore inappropriate for use with very young children and children with developmental delays. Because it was not possible to develop a suitable task for the very young children in this study, generativity in these very young children could not be assessed. Therefore, the current study cannot address the question of generativity in very young children with autistic spectrum disorders or speech and language delay.

In fact, the existing literature does not even reveal whether it is appropriate to look for generative skill in typically developing preschoolers. It is possible that generation is a more complex skill that is not present in very early development. Nevertheless, future work would benefit from the development of tasks attempting to measure generativity in younger children. The type of non-verbal task required to tap this skill in very young children may be better drawn from the pretend play literature (e.g Jarrold, 1997; Jarrold et al, 1996). Perhaps providing a number of set objects and encouraging the child to produce as many different acts as possible. One difficulty is making the need for multiple answers clear non-verbally. It is easy to tell a child to produce many varied answers but trying to ensure that the task structure entails this without the need for complicated verbal instructions is difficult. A further difficulty for this approach to generativity in children with autism would be that, unless one can formulate an argument that impoverished play results

solely from a generativity impairment, the production of a limited number of play acts may stem either from impoverished play skills or an impairment in response generation.

6.5 Fractionation vs. Integration of the Executive Function Construct Reviewed

The current study sought to separate out and assess individual executive function skills. This approach follows the fractionation approach to executive function (see Chapter 1). On this approach, the performance of individuals with autistic spectrum disorders ought to have been assessed on each of the possible component executive function skills before the hypothesis can be said to have been rigorously tested. An approach of this nature may lead to a reworking of the executive function hypothesis of autism such that one specific executive function may be the cognitive deficit that leads to the development of certain behavioural characteristics of autism. However, the definition of the individual aspects of executive function and the empirical assessment of each skill is complicated, and may be counter-productive if the key difficulty for children with autism arises when several component executive function skills are required simultaneously.

Several authors have suggested that autism may be associated with a particular difficulty in integrating information from differing sources and manipulating knowledge online (Kanner, 1943; Stuss & Benson, 1987). In the pursuit of tasks that tap only one aspect of executive function in these young children, tasks become simpler. If the dysfunction that is observed later in life is a result of the need to simultaneously manipulate several aspects of executive function whilst in a social setting then the experimental procedures are eliminating the very problem that they are interested in quantifying. Increasingly complex tasks that require the integration of one or more component skills, after assessing them individually, may further delineate the situations in which individuals with autism have executive difficulty.

6.6 Executive Dysfunction: Delay or Deviance?

The mounting evidence that very young children with autism do display appropriate executive skills has implications for the nature of the deficit observed later in life. Specifically, do young children with autistic spectrum disorders grow into an executive disorder and, if so, why? This section discusses the possibilities of delayed or deviant executive function development. Section 6.7 considers how the experience of growing up with autism may adversely affect the development of executive function skill.

If executive function skill is delayed in autism, we might expect the skills to be delayed from an early age. In this case, very young children with autism would demonstrate executive skills at levels appropriate to younger or less able children. However, if children grow into an executive deficit then the developmental trajectory of these skills may be deviant. In this case, it is possible that early executive skills would be present. There is evidence for both the accounts of delay or deviance in the executive function performance of individuals with autism.

The executive delay account is supported by the fact that children with autism have been shown to benefit from increased task structure (Ciesielski & Harris, 1997) in a similar way to typically developing young children (e.g. Bauer, Schwade, Saeger Wewerka, & Delaney, 1999; Hughes, 1998a; Zelazo, Burack, Benedetto, & Frye, 1996), suggesting the underlying developmental path may be similar in both groups. Also, there is data suggesting that very young children with autism have delayed maturation of the frontal cortex (Zilbovicius et al, 1995). However in the current study, three- and four-year-olds with autism seem to perform at levels appropriate for their ability; suggesting early skills, at least, are not delayed. Rather, the development of executive skills in autism may be deviant in nature.

For the deviance account, it is notable that the pervasive executive difficulty in autism may lie in the flexible generation of ideas (Turner, 1995) rather than inhibiting action schemes. If understanding and the generation of ideas come on-line before inhibitory control in typical development (Zelazo & Reznick,

1991; Zelazo, Reznick & Pinon, 1995; Zelazo, Reznick, & Spinazzola, 1998), and if inhibitory control is unimpaired whilst generativity is impaired in autism this suggests executive deviance rather than delay. Such a situation would reflect the deviant quality of socio-communicative behaviour and play skills in autism (Le Couteur et al, 1989; Le Couteur et al, unpublished). It may also address the concern about whether an executive function deficit is unique to autism. There is evidence that executive function skills are delayed in Attention Deficit Hyperactivity Disorder (Oosterlaan & Sargeant, 1998). If the development of executive skill in autism does turn out to be deviant rather than delayed this may provide a distinction between the types of executive disorder that might play a role in the development of these distinct psychopathologies. More work must be addressed specifically to this issue before it can be discussed further.

6.7 How Might Development as a Child with Autism Influence Executive Function Development?

The deviance account of executive function development in autism leads us to a proposal put forward by Griffith and colleagues (Griffith et al, 1999) and Ozonoff and McEvoy (1994). These authors suggested that the executive function skill of comparison groups might improve at a greater rate than that of children with autism. In fact, Griffith suggested that the performance of individuals with autism remained stationary over time. It is therefore important to consider how the experience of growing up autistic may impact upon the development of executive skills.

A fundamental difference between the experiences of a child with autism and a child without autism is the role of social interaction in their development. Social interaction involves the integration of a large amount of information from varying sources, the ability to shift cognitive set to understand the possible reasons for another's behaviour, and the ability to stop oneself from making inappropriate comments or actions. Experience within a social world can have a substantial impact upon self-regulatory ability (Luria, 1961) or on

higher-level cognitive development (Vygotsky, 1978). Therefore it is possible that early impairment in social skills contributes to subsequent executive dysfunction in autistic spectrum disorders. Perhaps the child with autism who removes himself from early social interaction such as joint attention deprives himself of the opportunity to practice and hone these executive skills and therefore fails to make the developmental advances of those involved in social experience.

Likewise, play may have an influential role. Pretend play may require the executive skill to generate an alternative use for an object and to inhibit the action most commonly associated with that object; social play requires the ability to inhibit a response when it is another person's 'turn'. On this view, a lack of interest in play skills may lead to many missed opportunities to refine these skills.

A repetitive behavioural profile may similarly reduce opportunities where executive function skills can be practised and developed (see section 5.7.2). A child who has one interest that he pursues to the exclusion of all others is likely to deprive himself of varied opportunities to extend his executive skill. It is also possible that lower-level repetitive behaviours such as repetitive movements may inhibit the development of flexible cognition. Recent animal studies have supported the suggestion that motor stereotypies may impact upon cognitive skill (Garner, 1999; Garner & Mason, unpublished).

For the children in the current study there was little evidence of concurrent association between repetitive behaviour and executive skill. This would not support the argument that autistic symptomatology leads to executive dysfunction. It is possible, though, that the measures of symptomatology were too general or that it is precisely the experience from the ages of three or four years of age that impacts upon the development of skills that are deficient in older children with autism. Further longitudinal study of the relationship between specific repetitive, social and communication behaviours and cognitive skills is needed in typically developing children and those with developmental disorders. Following the future development of the individuals

involved in this study could provide fascinating information about the long-term outcome of these two groups of children. In time, this might begin to bridge the gap between recent findings in very young children with autism and the prevailing literature with older children and adults with autistic spectrum disorders.

6.8 Implications for Intervention Approaches for Autistic Spectrum Disorders and Speech and Language Delay

6.8.1 Autistic Spectrum Disorders

The executive function account of autism proposes that poor regulation of behaviour may underlie the behavioural aspects of autism. This view would predict that the use of cues and structure to minimise the self-regulation requirements of a task would assist the child. Although intervention approaches such as TEACCH and LOVAAS have developed independently from this psychological hypothesis of autism, they attempt to structure the individual's environment in order to ameliorate the behavioural aspects of autism (e.g. TEACCH: Schopler, Brehm, Kinsbourne, & Reichler, 1971; Schopler, Mesibov, & Hearsey, 1995; Schopler, Mesibov, Shigley, & Bashford, 1984; LOVAAS: Lovaas, 1981). Nevertheless, the current study's lack of support for an impairment in behavioural regulation of very young children with autism does not diminish the role of early intervention.

It is generally accepted that early intervention in autism can be a valuable and effective tool in assisting children with autism and their families by increasing the child's chances of living successfully and happily, possibly attending mainstream schooling and reducing the longer term demands upon clinical services (e.g. Jordan, et al., 1998; Rimland, 1994a). Executive function skills are essential to successful functioning in everyday life. Self-care skills such as dressing or brushing your teeth rely on simple planning abilities (e.g. underwear must be put on before trousers; toothpaste must be placed on brush before scrubbing teeth). The same is true of more advanced skills required by older individuals (e.g. planning a route to the cinema and altering that route if

necessary). Clearly, facilitation of executive function skills can have a substantial impact upon the degree of independence an individual with autism might experience and upon their quality of life throughout their life span.

The current study joins a growing body of evidence that very young children with autistic spectrum disorders do have some early executive function skills. These skills, which include inhibition-and-implementation and working memory, may underlie more complex skills (see Chapter 1). Therefore, if the early skills can be built upon, it may be possible to improve the executive function performance of older children and adults with autism. TEACCH and LOVAAS programs emphasise the importance of breaking down behaviours into a larger number of small steps that are easy to assimilate for the young autistic child. These small steps are often repeated many times and the child given multiple opportunities to practice these skills (Connor, 1998). These techniques may be particularly useful for the development of executive function skills. Since the children already demonstrate some of these small steps (such as basic inhibitory control) these could be reinforced and practiced in many ways. The HANEN intervention approach for young children with autism (Sussman, 1999) focuses upon simple, naturally occurring, social routines which are built on develop socio-communicative skills. Given the relationship between precursors to theory of mind and executive function it is possible that this type of approach may additionally benefit the child's executive function development.

However, one particular problem for children with autism is generalising skills learnt in one setting to another situation (e.g. Hadwin et al, 1997; Ozonoff & Miller, 1995). Perhaps by presenting the simple steps in a variety of situations and contexts, the child will begin to see how the same action can be used in many different settings and in response to different task demands. A recent paper has shown that experience with one type of executive task can be generalised to facilitate performance on another task (Dowsett & Livesey, 2000). In this study, typically developing three-year-olds who were given experience with a variation of the Wisconsin Card Sorting task demonstrated improved inhibitory control on a variation of the Go/No-Go discrimination

task. The implication that experiential learning can be transferred from one executive function task to another is encouraging for those trying to develop intervention packages. Whilst the study concentrated upon typically developing children, it is possible that the same principles could be applied for children with autistic spectrum disorders and/or speech and language delay.

The verbal difficulties of many children with autism emphasise the importance of demonstrating actions and settings in a visual manner and allowing the child hands-on experience of the particular skill. A visual presentation of correct and incorrect responses to a task may enable the child to selectively imitate the correct action (as Want and Harris (2001) demonstrated with tool use in typically developing three-year-olds). In the current study, training for the Marbles task demonstrated both correct and incorrect actions visually: performance on this task was unexpectedly good for both groups of children.

Whilst small steps may form the fundamental aspects of executive skills, it is important that the child is taught how to link them together to facilitate more sophisticated skills. This is likely to require careful structuring of the tasks (e.g. Dawson & Osterling, 1997). Klahr (1978) discussed how task instructions, cues and feedback imposed constraints on strategy construction and execution that could be seen in the child's performance. The work of Welsh (Welsh, 1991) has shown how a typically developing child can benefit from Tower of Hanoi problems being presented in increasing order of difficulty: they appear to apply learning from a simpler problem to the next problem. Support such as goal-state information also assists planning in young typically developing children (Bauer et al, 1999; Hudson & Fivush, 1991).

The supportive structuring of tasks resembles Vygotsky's theoretical zone-of-proximal-development construct (Vygotsky, 1978). The zone represents a skill level that is not attainable by the child individually, but can be achieved with some support from an experienced other. When the structure is reduced,

the child ought to have made the developmental and conceptual gains required to maintain the skill they achieved in the zone-of-proximal-development. Simultaneously, a new zone becomes available. In this way we can see continual development facilitated by a dynamic co-operative process. Whilst the social rewards of cooperation may be less motivating for an individual with autism, the concept of high levels of structure initially that are then reduced can be applied to non-social aspects of the task, or perhaps a non-human partner could be used (a robot, a computer or mechanical operations). Moreover, it is possible that as the child sees their developmental progression the social cooperation involved may become more important to them: they have a 'reason' for social interaction.

In summary, it seems reasonable to suggest that an intervention approach involving the practice of simple cognitive or social skills may facilitate executive function development in autism. The basic skills ought to be repeatedly practiced and then built into more complex skills. Structure can assist this process before being reduced so the child can take an increasingly independent role in the execution of more sophisticated executive skills.

6.8.2 Speech and Language Delay

The literature concerning executive function skills in children with speech and language delay is sparse. This makes the interpretation of the current data in terms of existing knowledge impossible. However, the study does suggest that very young children with speech and language delay exhibit executive skills appropriate to their ability. There is a clear case for more research on the developmental trajectories of these young children. Initially it is important to ascertain whether older children with speech and language impairments have executive deficits and the developmental trajectory of this skill. It would also be useful to consider ways in which to distinguish those children who demonstrate speech and language delay when very young but go on to catch-up by their early school years from those who demonstrate continuing problems throughout development. As was discussed for children with autism, it is possible that these subgroups may have different executive function skill capacity. Further research into the developmental path of

executive skill in speech and language delay may also provide valuable information about the possible mediating role of language in both executive function and theory of mind.

The comments about intervention approaches for children with autism hold true for individuals with speech and language delay as well. Although there is no way of knowing at present whether both groups of children follow similar developmental mechanisms, the Vygotskian approach may be particularly beneficial for children with speech and language delay since they do not show the severe social impairments present in autism that might prevent successful cooperation on a task.

6.8.3 A Wider Perspective

This thesis has called a strong version of the executive dysfunction hypothesis of autism into question. However, the hypothesis still has theoretical strengths. The current view in the literature is that no single account of autism, at the psychological or any other level, will be able to explain the entire phenomenon that is the autistic spectrum. In fact, even within specific theories the picture appears to be more dynamic and complex than had first been thought. This means that research must continue apace in numerous different fields. In this way it may prove possible to emphasise the developmental nature of autism: perhaps best described by different theoretical perspectives at different stages of development but always held together by an overarching developmental framework for this fascinating and complex disorder.

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Appendix I

Repetitive Behaviours Questionnaire

Thank you for agreeing to complete this questionnaire. We still understand relatively little about repetitive behaviours. These are behaviours, or habits, which are repeated frequently, or always followed in the same way or at the same time. By completing this questionnaire you are contributing to our understanding of this important class of behaviour.

Although there are several pages of questions, you will find that many can be answered with a quick 'no' response. In this way you should be able to complete the questionnaire quite quickly. However all answers and descriptions of behaviour will be considered individually and fully so the information you are able to provide will be particularly helpful and informative.

In completing this questionnaire please record the behaviour that your son or daughter shows at the moment (that is **over the last three months**). Please describe and rate the most **usual** way he/she displays this behaviour. Each question is followed by a list of alternatives. Please tick the box next to the alternative that best describes the behaviour shown by your son or daughter. Where he/she shows two or more behaviours of the type probed by one question then please describe and code **separately**. The examples given in each question are only a guide to the type of behaviour that can be shown; please describe any other behaviours of the type probed by the question. If your son or daughter shows any behaviour that is not covered by the questionnaire please describe this and provide as much information as you can on additional sheets of paper.

For those items that ask about the frequency with which behaviour is shown, please rate how frequently your son or daughter might display the behaviour over the course of the day if you were watching them **all** day. Think about this either in terms of the number of bouts of this behaviour he or she would show over the course of the entire day, or if it is more appropriate, the number of bouts of this behaviour that might occur in a typical hour.

Please try to complete each question as accurately as you can and try not to leave any question, or any part of a question, unanswered. If you have any questions or comments about this questionnaire, please contact me at the address below.

Once again many thanks for your help, time and interest.

Heather Shearer
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South Road, Durham, DH1 3LE.

1. Does he/she operate light switches, taps, the toilet flush etc. repeatedly when it is not necessary to do so?

☐ **Never** or rarely
☐ **One** or more bouts of this behaviour daily
☐ **15** or more bouts of this behaviour daily (or at least one bout an hour)
☐ **30** or more bouts of this behaviour daily (or at least two bouts an hour)

Please describe this behaviour

Is there any specific time or situation when this behaviour is especially likely to occur?

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2. Does he/she arrange toys or other items in rows or patterns?

☐ **Never** or rarely
☐ **One** or more bouts of this behaviour daily
☐ **15** or more bouts of this behaviour daily (or at least one bout an hour)
☐ **30** or more bouts of this behaviour daily (or at least two bouts an hour)

Please describe this behaviour

Is there any specific time or situation when this behaviour is especially likely to occur?

---

3. Does he/she repetitively fiddle with toys or other items?
For example, does he/she spin, twiddle, bang, tap, twist, flick or wave anything repetitively?

☐ **Never** or rarely
☐ **One** or more bouts of this behaviour daily
☐ **15** or more bouts of this behaviour daily (or at least one bout an hour)
☐ **30** or more bouts of this behaviour daily (or at least two bouts an hour)

Please describe this behaviour

Is there any specific time or situation when this behaviour is especially likely to occur?

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4. Does he/she touch parts of his/her body or clothing repeatedly?
For example, does he/she repeatedly rub his/her legs, pull at the buttons on his/her clothing, or touch his/her ear or elbow etc.?

☐ **Never** or rarely
☐ **One** or more bouts of this behaviour daily
☐ **15** or more bouts of this behaviour daily (or at least one bout an hour)
☐ **30** or more bouts of this behaviour daily (or at least two bouts an hour)

Please describe this behaviour

Is there any specific time or situation when this behaviour is especially likely to occur?

---

5. Is he/she attached to anything in particular?
For example, does he/she carry a teddy, a blanket or a stick etc. around with him/her?

☐ **No** particular attachment to any object
☐ Attachment to an object of the sort commonly used as a comforter (e.g. teddy, blanket etc.)
☐ Attachment to **unusual** object (e.g. stick, glove etc.)

Please describe this behaviour

---

6. Does he/she obsessively collect or hoard items of any sort?

☐ **No** obsessive, or unusually keen, collecting or hoarding
☐ Very keen collector of **usual** items (e.g. stamps, football cards etc.)
☐ Very keen collector of **unusual** or odd items (e.g. leaflets, jar lids, sticks etc.)

Please describe this behaviour

---

7. Does he/she spin him/herself around and around?

☐ **Never** or rarely
☐ **One** or more bouts of this behaviour daily
☐ **15** or more bouts of this behaviour daily (or at least one bout an hour)
☐ **30** or more bouts of this behaviour daily (or at least two bouts an hour)

Is there any specific time or situation when this behaviour is especially likely to occur?

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8. Does he/she rock backwards and forwards, or side to side, either when sitting or when standing?

☐ **Never** or rarely
☐ **One** or more bouts of this behaviour daily
☐ **15** or more bouts of this behaviour daily (or at least one bout an hour)
☐ **30** or more bouts of this behaviour daily (or at least two bouts an hour)

Is there any specific time or situation when this behaviour is especially likely to occur?

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9. Does he/she bang his/her head? Does he/she do this repetitively?

☐ **Never** or rarely
☐ **One** or more bouts of this behaviour daily
☐ **15** or more bouts of this behaviour daily (or at least one bout an hour)
☐ **30** or more bouts of this behaviour daily (or at least two bouts an hour)

Is there any specific time or situation when this behaviour is especially likely to occur?

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10. Does he/she pace or move around repetitively?
For example, does he/she walk to and fro across a room, or around the house or garden repetitively?

☐ **Never** or rarely
☐ **One** or more bouts of this behaviour daily
☐ **15** or more bouts of this behaviour daily (or at least one bout an hour)
☐ **30** or more bouts of this behaviour daily (or at least two bouts an hour)

Please describe this behaviour

Is there any specific time or situation when this behaviour is especially likely to occur?

---

11 . Does he/she make repetitive hand and/or finger movements?
For example, does he/she repetitively wave, flick, flap or twiddle his/her hands or fingers repetitively?

☐ **Never** or rarely
☐ **One** or more bouts of this behaviour daily
☐ **15** or more bouts of this behaviour daily (or at least one bout an hour)
☐ **30** or more bouts of this behaviour daily (or at least two bouts an hour)

Please describe this behaviour

Is there any specific time or situation when this behaviour is especially likely to occur?

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12. Does he/she make other repetitive body movements?
For example, does he/she repeatedly clasp his/her hands, tap his/her feet, swing his/her legs or jump etc.?

❒ **Never** or rarely
❒ **One** or more bouts of this behaviour daily
❒ **15** or more bouts of this behaviour daily (or at least one bout an hour)
❒ **30** or more bouts of this behaviour daily (or at least two bouts an hour)

Please describe this behaviour

Is there any specific time or situation when this behaviour is especially likely to occur?

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13. Does he/she ever injure him/herself?
For example, does he/she bite, scratch, knock or pick at his/herself? Does he/she do this repeatedly?

❒ **Never** or rarely
❒ **One** or more bouts of this behaviour daily
❒ **15** or more bouts of this behaviour daily (or at least one bout an hour)
❒ **30** or more bouts of this behaviour daily (or at least two bouts an hour)

Please describe this behaviour

Is there any specific time or situation when this behaviour is especially likely to occur?

---

14. Does he/she insist on things about the house staying the same?
For example, does he/she insist on furniture staying in the same place, or curtains being open or closed etc.?

❒ **No**
❒ **Mild** problem which does not effect others
❒ **Serious** problem which effects others on a regular basis

Please describe this behaviour

15. Does he/she insist on other items being put out, kept or stored in the same way?
For example, does he/she like ornaments, toys or cassette tapes kept in the same places or positions?

❒ **No**
❒ **Mild** problem which does not effect others
❒ **Serious** problem which effects others on a regular basis

Please describe this behaviour

16. Does he/she play the same music, game or video, or read the same book repeatedly?

❒ **Never** or rarely
❒ **Regular** feature of behaviour, but will tolerate alternatives when necessary
❒ **Highly regular** and **highly rigid** feature of behaviour. Will not tolerate any alternatives.

Please describe this behaviour briefly

17. Does he/she insist on using the same objects or items in any other situation?
For example, does he/she insist on using the same chair, plate, bed linen or door?
(DO NOT count any insistence on using the same mug or cup)

❒ **Never** or rarely
❒ **Regular** feature of behaviour, but will tolerate alternatives when necessary
❒ **Highly regular** and **highly rigid** feature of behaviour. Will not tolerate any alternatives.

Please describe this behaviour

18. Does he/she insist on wearing the same clothes or refuse to wear new clothes?

❒ **Never** or rarely
❒ **Regular** feature of behaviour, but will tolerate alternatives when necessary
❒ **Highly regular** and **highly rigid** feature of behaviour. Will not tolerate any alternatives.

Please describe this behaviour

19. Does he/she insist that certain items of clothing must always be worn, or worn in the same situation or in the same way?
For example, does he/she insist on always wearing a vest, or wearing a hat to the shops, or always buttoning a shirt to the collar?

❒ **Never** or rarely
❒ **Regular** feature of behaviour, but will tolerate alternatives when necessary
❒ **Highly regular** and **highly rigid** feature of behaviour. Will not tolerate any alternatives.

Please describe this behaviour

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20. Does he/she insist on eating the same foods, or a very small range of foods, at every meal?

❒ **Never** or rarely
❒ **Regular** feature of behaviour, but will tolerate alternatives when necessary
❒ **Highly regular** and **highly rigid** feature of behaviour. Will not tolerate any alternatives.

Please describe this behaviour

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21. Does he/she insist on moving or travelling by the same route?
For example, does he/she insist on taking the same route when moving about the house, going for a walk, or travelling in the car?

❒ **Never** or rarely
❒ **Regular** feature of behaviour, but will tolerate alternatives when necessary
❒ **Highly regular** and **highly rigid** feature of behaviour. Will not tolerate any alternatives.

Please describe this behaviour

---

22. How does he/she react if any changes are made to his/her surroundings at home?
For example, if you move the furniture, or rearrange the way that certain items are stored or organised?

❒ May comment on, or notice, the change but shows no negative reaction
❒ Accepts the change, but shows some degree of anxiety or mildly negative reaction
❒ Will accept the change, but shows extreme anxiety or strong negative reaction (e.g. tantrum)
❒ Will **not** accept the change. Persistently attempts to rearrange the items

---

23. Are there any aspects of routine that he/she insists must remain the same?
For example, does he/she insist on always bathing before breakfast, on going to the shops every afternoon, or on watching a video after every meal?

❒ **No**
❒ **Mild** problem which does not effect others
❒ **Serious** problem which effects others on a regular basis

Please describe this routine

24. Does he/she make rituals out of everyday activities such as eating, dressing, getting in the car, walking up stairs etc.?

❒ **No**
❒ **Mild** problem which does not effect others
❒ **Serious** problem which effects others on a regular basis

Please describe this activity and ritual(s)

25. Does he/she have any rituals that are linked to particular occasions or places?
For example, does he/she have specific rituals for the supermarket, the Doctor's surgery or a relative's house?

❒ **No**
❒ **Mild** problem which does not effect others
❒ **Serious** problem which effects others on a regular basis

Please describe this ritual(s)

26. How does he/she react if his/her daily routine is changed?

❒ May comment on, or notice, the change but shows no negative reaction
❒ Accepts the change, but shows some degree of anxiety or mildly negative reaction
❒ Will accept the change, but shows extreme anxiety or strong negative reaction (e.g. tantrum)
❒ Will **not** accept any change to routine

27. Does he/she 'echo' or repeat what other people say?

❒ **Never** or rarely
❒ **One** or more bouts of this behaviour daily
❒ **15** or more bouts of this behaviour daily (or at least one bout an hour)
❒ **30** or more bouts of this behaviour daily (or at least two bouts an hour)

Is there any specific time or situation when this behaviour is especially likely to occur?

28. Does he/she say the same things, or make the same noises, repeatedly?
For example, does he/she say the same word repeatedly or other sounds such as hums or growls or clicking noises? Or does he/she use the same 'stock phrases' frequently?

❒ **Never** or rarely
❒ **One** or more bouts of this behaviour daily
❒ **15** or more bouts of this behaviour daily (or at least one bout an hour)
❒ **30** or more bouts of this behaviour daily (or at least two bouts an hour)

Please describe this behaviour

Is there any specific time or situation when this behaviour is especially likely to occur?

29. Does he/she talk about the same topic over and over again?

❒ **Never** or rarely
❒ **One** or more bouts of this behaviour daily
❒ **15** or more bouts of this behaviour daily (or at least one bout an hour)
❒ **30** or more bouts of this behaviour daily (or at least two bouts an hour)

Please describe this behaviour

Is there any specific time or situation when this behaviour is especially likely to occur?

30. Does he/she have any interests or hobbies? Please describe these briefly.

In particular, does he/she have any interests or preoccupations which you would describe as overly keen, obsessional, or unusual in any way?
Please describe any such interests in as much detail as you can.

In summary would you say that he/she has:

☐ a varied pattern of interests which he /she will pursue spontaneously and without prompting

☐ one or more obsessional interests, but also other usual interests which he/she will pursue spontaneously and without prompting

☐ only obsessional interests which he/she will pursue spontaneously

☐ has no particular interests or hobbies that he/she will pursue spontaneously

[NB DO NOT include watching TV as an interest or hobby]

31. What was the earliest repetitive activity that you remember your son or daughter showing?

How old was he/she when this began?

32. Of all the behaviours in this questionnaire that your son or daughter engage in, which one would you say is the most marked or the most noticeable?

33. Of all the behaviours in this questionnaire that your son or daughter engage in, which one would you say causes the greatest problem in day-to-day life?

Thank you for completing this questionnaire

Appendix II

Executive Function Skill and Social and Communication Behaviours

EF and Socialisation Variables at Time 1

Correlation coefficients with chronological age and non-verbal mental age partialled out.

Task variable	ADI-R Socialisation	ADOS-G Socialisation
A-not-B Task		
% reversals correct	-0.06	-0.19
% perseverative errors	0.24	0.08
Longest run perseverative errors	0.13	-0.04
A-not-B Invisible Displacement Task		
% reversals correct	-0.30	-0.06
% perseverative errors	0.25	0.21
Longest run perseverative errors	**0.46***	**0.50***
Detour Reach, knob route		
Trials to criterion	-0.13	0.30
Longest run errors	0.14	**0.42***
Detour Reach, switch-reach route		
Trials to criterion	0.20	0.00
Direct reach errors	**-0.47***	**-0.54***
Confusion errors	0.05	0.13
Three boxes visual search - stationary version		
Efficiency ratio	0.08	-0.06
Three boxes visual search – scrambled version		
Efficiency ratio	-0.05	0.01

* $p<0.05$; ** $p<0.01$

EF and Socialisation Variables at Time 2

Correlation coefficients with chronological age and non-verbal mental age partialled out (except Sorting Task which is analysed non-parametrically)

Task – variable	ADI-R Socialisation	ADOS-G Socialisation
A-not-B Invisible Displacement task		
% reversals correct	-0.11	-0.24
% perseverative errors	0.04	0.07
Longest run perseverative errors	0.19	0.33
Detour Reach Task, knob route		
Trials to criterion	-0.03	-0.01
Number of errors	-0.09	0.06
Detour Reach Task, switch-reach route		
Trials to criterion	0.01	-0.02
Direct reach errors	0.29	**0.37***
Rule confusion	-0.28	-0.30
Sorting Task	(*non-parametric*)	
Rule reached (and passed)	-0.24	**-0.29***
Six Boxes visual search task – stationary version		
Efficiency ratio	0.10	0.03
Longest run of perseverative errors to location/box	-0.11	0.00
Six Boxes visual search task – scrambled version		
Efficiency ratio	0.10	0.03
Longest run of perseverative errors to a specific location	0.28	**0.38***
Longest run of perseverative errors to a specific box	-0.08	-0.14
Marbles Task		
% trials correct	0.12	0.12
Number of errors	-0.10	-0.13
Number of perseverative errors	-0.03	-0.09

* $p<0.05$; ** $p<0.01$

EF and Communication Variables at Time 1

Correlation coefficients with chronological age and non-verbal mental age partialled out

Task variable	**ADI-R Comm. verbal**	**ADI-R Comm. non-verbal**	**ADOS-G Comm.**
A-not-B Task			
% reversals correct	-0.16	-0.30	-0.29
% perseverative errors	0.47	0.40	0.27
Longest perseverative run	0.26	0.39	0.10
A-not-B Invisible Displacement Task			
% reversals correct	-0.28	0.07	-0.08
% perseverative errors	0.58	-0.37	0.19
Longest run perseverative errors	0.47	0.42	**0.48***
Detour Reach, knob route			
Trials to criterion	-0.07	-0.44	0.22
Longest run errors	0.06	0.25	**0.38***
Detour Reach, switch-reach route			
Trials to criterion	0.30	0.14	-0.14
Confusion	-0.53	-0.43	**-0.48***
Direct reach errors	-0.89	-0.14	-0.05
Three boxes visual search - stationary version			
Efficiency ratio	-0.41	-0.09	-0.21
Three boxes visual search – scrambled version			
Efficiency ratio	-0.31	-0.03	-0.04

* $p<0.05$; ** $p<0.01$

EF and Communication Variables at Time 2

Correlation coefficients with chronological age and non-verbal mental age partialled out (except Sorting Task which is analysed non-parametrically)

Task – variable	**ADI-R Comm. Verbal**	**ADI-R Comm. Non-verbal†**	**ADOS-G Comm.**
A-not-B Invisible Displacement task			
% reversals correct	**-0.57***	**-0.59**	-0.22
% perseverative errors	-0.00	**0.78****	-0.11
Longest run perseverative errors	0.27	**0.74****	0.26
Detour Reach Task, knob route			
Trials to criterion	0.18	**-0.63***	0.18
Number of errors	0.19	-0.15	0.07
Detour Reach Task, switch-reach route			
Trials to criterion	0.13	-0.40	-0.26
Direct reach errors	0.12	-0.19	0.29
Rule confusion	-0.10	**0.76***	-0.16
Sorting Task	(*non-parametric*)		
Rule reached (and passed)	-0.21	-0.23	-0.11
Six Boxes visual search task – stationary version			
Efficiency ratio	0.34	0.23	0.21
Longest run of perseverative errors to location/box	-0.28	-0.05	-0.23
Six Boxes visual search task – scrambled version			
Efficiency ratio	0.19	-0.01	-0.13
Longest run of perseverative errors to a specific location	0.18	0.36	**0.38***
Longest run of perseverative errors to a specific box	-0.13	0.24	0.00
Marbles Task			
% trials correct	0.36	0.04	0.22
Number of errors	-0.36	-0.05	-0.26
Number of perseverative errors	-0.19	-0.11	-0.14

† Very few children who were non-verbal attempted the A-not-B Invisible Displacement at Time 2 (n=9). This may have had an undue impact on the statistical significance since graphical representations do not reflect these significant findings. * $p<0.05$; ** $p<0.01$

Appendix III

Inter-instrument correlations between behavioural domains

Time 1

	ADI-R	Socialisation	Communication verbal	Communication non-verbal	Repetitive
ADOS	Socialisation	**0.71****			
	Communication		**0.78****	**0.59****	
	Repetitive				**0.54****

** p<0.01

Time 2

	ADI-R	Socialisation	Communication verbal	Communication non-verbal	Repetitive
ADOS	Socialisation	**0.78****			
	Communication		**0.57****	0.20	
	Repetitive				**0.44****

** p<0.01

Appendix IV

Four Case Studies

Case 1

Summary

Case 1 shows an apparent developmental discrepancy whereby inhibitory skills worsened over the year whilst working memory improved. The quantitative measures of his repetitive behaviour reduced over the year, whilst the qualitative aspects became more distinctive. The parental account of symptomatology decreased over the year whilst the direct observation scores increased.

Symptomatology

This child had a diagnosis of autism spectrum disorder and significant repetitive behaviours. The following table contains diagnostic data for each behavioural domain.

		Time 1	Time 2
	Chronological age	3-6	4-7
	Verbal mental age	3-0	3-9
	Non-verbal mental age	3-4	4-6
ADI-R	Socialisation	13*	8*
	Communication (verbal)	13*	10
	Repetitive	8*	7*
ADOS-G	Module	1	2
	Socialisation	6†	9*
	Communication	3†	5*
	Repetitive	3	3
RBQ	Repetitive Movements	11	4
	Sameness Behaviour	11	6
	Repetitive Use of Language	7	3
	Circumscribed Interests	1	2
	Resistance to Change	3	1

* above cut-off for autism spectrum disorder; † above cut-off for pervasive developmental disorder

At Time 1 he had a strong interest in numbers, a fondness for routine that pervaded home life and also rituals associated with specific locations or times. He was fascinated with air conditioning units and smoke detectors at Time 2. On one occasion his mother gave him the home video recorder and he filmed the air-conditioning unit.

Executive Function

At Time 1, he successful on the knob route of the Detour Reach Task but refused to attempt the switch-reach route. On the A-not-B Invisible Displacement Task he made perseverative errors on half of the reversal trials he attempted. His efficiency ratio on the stationary version of the Three Boxes Task was 0.6, and on the scrambled version was 0.43.

At Time 2, he was again successful on the knob route of the Detour Reach Task. This time he attempted the switch-reach route but had difficulty in successfully sequencing his responses: he made many, varied, errors before making a criterial run. He only attempted one reversal trial on the A-not-B Invisible Displacement Task at this time point, on which he perseverated to the previous response. His efficiency ratio on the stationary version of the Six Boxes Task was 1.0, and on the scrambled version was 0.71.

Case 2

Summary

Case 2 showed a mixed pattern of both skill and difficulty with inhibition-and-implementation skills. He showed some evidence of improvement in inhibition-and-implementation and working memory skill over the year. In contrast, his particularly marked preference for sameness behaviour remained relatively consistent over the year.

Sypmtomatology

Case 2 had a diagnosis of autism spectrum disorder with marked repetitive behaviours. The following table contains diagnostic data for each behavioural domain.

		Time 1	Time 2
	Chronological age	2-9	3-9
	Verbal mental age	3-0	3-5
	Non-verbal mental age	2-9	3-1
ADI-R	Socialisation	26*	26*
	Communication	12* (non-verbal)	19* (verbal)
	Repetitive	6*	9*
ADOS-G	Module	1	2
	Socialisation	4†	5†
	Communication	2†	2
	Repetitive	3	4
RBQ	Repetitive Movements	9	2
	Sameness Behaviour	15	12
	Repetitive Use of Language	4	1
	Circumscribed Interests	1	2
	Resistance to Change	6	2

* above cut-off for autism spectrum disorder; † above cut-off for pervasive developmental disorder

At Time 1, he demonstrated several repetitive behaviours: lining up of anything (e.g. trains, food, cups) in perfectly straight lines, repetitive fiddling with car wheels and light switches and some spinning of himself. At Time 2 several other repetitive behaviours were noted. He had a marked interest in trains: he could identify all 80

30. Does he/she have any interests or hobbies? Please describe these briefly.

In particular, does he/she have any interests or preoccupations which you would describe as overly keen, obsessional, or unusual in any way?
Please describe any such interests in as much detail as you can.

In summary would you say that he/she has:

☐ a varied pattern of interests which he /she will pursue spontaneously and without prompting

☐ one or more obsessional interests, but also other usual interests which he/she will pursue spontaneously and without prompting

☐ only obsessional interests which he/she will pursue spontaneously

☐ has no particular interests or hobbies that he/she will pursue spontaneously

[NB DO NOT include watching TV as an interest or hobby]

31. What was the earliest repetitive activity that you remember your son or daughter showing?

How old was he/she when this began?

32. Of all the behaviours in this questionnaire that your son or daughter engage in, which one would you say is the most marked or the most noticeable?

33. Of all the behaviours in this questionnaire that your son or daughter engage in, which one would you say causes the greatest problem in day-to-day life?

Thank you for completing this questionnaire

Thomas the Tank Engine characters, he made hand and arm movements by his side as if he were a train and he would read anything to do with trains, including price lists! He also preferred items to be kept in the same place, to be read the same story at bedtime and insisted on a complex evening routine that took several hours.

Executive Function

At Time 1, he was successful on the knob route of the Detour Reach Task but failed the switch-reach route. He attempted two reversal trials on the A-not-B Invisible Displacement task and made a perseverative error on both trials. His efficiency ratio on the stationary version of the Three Boxes Task was 1.0, and on the scrambled version was 0.75.

At Time 2, he was successful on both the knob and switch-reach routes of the Detour Reach Task. He only attempted one reversal trial on the A-not-B Invisible Displacement Task and made a perseverative error on this trial. His efficiency ratio on the stationary version of the Six Boxes Task was 1.0, and on the scrambled version was 0.67. He was able to shift flexibly between rules on the Sorting Task.

Case 3

Summary

Case 3 demonstrated a pervasive difficulty in inhibition-and-implementation skills. However his social, communication and repetitive symptom scores were low and he did not meet diagnostic criteria for autism spectrum disorder.

Symptomatology

Here was a child who did not meet the criteria for autism spectrum disorder but had significant executive function difficulties. Some repetitive behaviours and social and communicative difficulties were noted for this boy. However, they were less intense and distinctive than other children who received a diagnosis of autism spectrum disorder. Unfortunately, no RBQ data was received for Time 2. The following table contains diagnostic data for each behavioural domain.

		Time 1	Time 2
	Chronological age	3-8	4-9
	Verbal mental age	2-2	2-9
	Non-verbal mental age	2-2	2-8
ADI-R	Socialisation	1	6
	Communication (verbal)	4	5
	Repetitive	2	3*
ADOS-G	Module	1	2
	Socialisation	0	1
	Communication	1	3†
	Repetitive	2	2
RBQ	Repetitive Movements	3	n/a
	Sameness Behaviour	0	n/a
	Repetitive Use of Language	1	n/a
	Circumscribed Interests	1	n/a
	Resistance to Change	0	n/a

* above cut-off for autism spectrum disorder; † above cut-off for pervasive developmental disorder

Executive Function

At Time 1, he was successful on the knob route of the Detour Reach Task but was unable to correctly sequence the actions for the switch-reach route. He attempted one reversal trial of the A-not-B Invisible Displacement Task and made a perseverative error. His efficiency ratio on the stationary version of the Three Boxes Task was 1.0, and on the scrambled version was 0.6.

At Time 2, he was unable to sequence the two responses required for the switch-reach route of the Detour Reach Task. He was also incorrect on the only reversal trial of the A-not-B Invisible Displacement Task that he attempted. His efficiency ratio on the stationary version of the Six Boxes Task was 0.75, and on the scrambled version was 0.55.

Case 4

Summary

Case 4 showed relatively good (and improving) inhibition-and-implementation skills despite having very low ability levels and a definite diagnosis of autism spectrum disorder.

Symptomatology

This child received a diagnosis of autism spectrum disorder at Time 2. He had received monthly injections of secretin between Time 1 and Time 2. The following table presents diagnostic data for each behavioural domain.

		Time 1	Time 2
	Chronological age	3-9	4-9
	Verbal mental age	1-2	1-2
	Non-verbal mental age	1-9	2-10
ADI-R	Socialisation	12*	17*
	Communication (non-verbal)	2	9*
	Repetitive	6*	3*
ADOS-G	Module	1	1
	Socialisation	10*	10*
	Communication	5*	4*
	Repetitive	2	4
RBQ	Repetitive Movements	4	0
	Sameness Behaviour	13	7
	Repetitive Use of Language	3	3
	Circumscribed Interests	1	1
	Resistance to Change	3	1

* above cut-off for autism spectrum disorder; † above cut-off for pervasive developmental disorder

He demonstrated sensory interests in the experimenter's hands by touching and looking at them for a long time (Time 2). His mother also reported that he liked routines to

remain consistent and objects to be in specific places in the home. She also said that repetitive movements such as hand flapping and self-injury had been pervasive at Time 1 but were less frequent at Time 2.

Executive Function

At Time 1, he was successful on the knob route of the Detour Reach Task but could not achieve a criterial run on the switch-reach route. He did not attempt the A-not-B Invisible Displacement Task but was correct on every reversal trial on the A-not-B Task. His efficiency ratio on the stationary version of the Three Boxes Task was 1.0, and on the scrambled version was 0.67.

At Time 2, he was successful on both the knob route and the switch-reach route of the Detour Reach Task. He was also correct on two out four reversal trials on the A-not-B Invisible Displacement Task. His efficiency ratio on the stationary version of the Six Boxes Task was 1.0, and on the scrambled version was 0.55.