# MEASURING AUTISM SPECTRUM DISORDER:

# ASSOCIATED FEATURES AND DIAGNOSTIC CRITERIA

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# CONTENTS

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# LIST OF PUBLICATIONS

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*Note:* The content of Chapter 5 closely resembles that of Kent, Carrington et al (2013).

# LIST OF PRESENTATIONS

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Kent, R. G., Gould, J., Wing, L., LeCouteur, A., & Leekam, S. R. (November 2011). What role does atypical sensory processing play in the core features of ASD? Talk given at Cardiff Psychology Postgraduate Conference. Wales, UK.

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# THESIS BACKGROUND

The work in this thesis was made possible by a CASE 1+3 studentship grant from the Economic and Social Research Council in collaboration with the National Autistic Society from 2009-2013. The CASE studentship programme enables an applied project to be carried out, that will lead to results that are relevant to a non-academic organisation. In this case the purpose of this particular research programme was to develop and promote research use of the Diagnostic Interview for Social and Communication Disorders (DISCO) in collaboration with the National Autistic Society. Until now the DISCO has been predominantly used as a diagnostic tool for autism spectrum disorders and although some research has been published, the DISCO is mostly used for clinical purposes only. For this thesis a secondary database of diagnostic information was analysed. Data from 200 of these cases were collected by clinicians Dr. L. Wing and Dr. J. Gould during clinical assessments at the National Autistic Society's Lorna Wing Centre in Bromley. The remaining 82 cases were part of a non-clinical research study conducted at the University of Kent. Primary data using questionnaires adapted from the DISCO along with a further sample of 115 individuals collected by collaborators were obtained over the course of the research programme.

The work of the CASE studentship was to address questions at the leading edge of the field in the hope of demonstrating the distinctive contribution of the DISCO for research purposes beyond that provided by other instruments. Previously these data had been collected for purposes other than the research questions proposed in the thesis. The current work involved a process of data screening and preparation beyond the focus of this thesis which was completed before analyses were run.

Over the course of the research programme for the thesis the proposed changes to the international classification systems, specifically; the criteria for Autism Spectrum Disorder were proposed (2011) and subsequently published by DSM-5 (2013). The spectrum approach uniquely offered by the DISCO provided the perfect opportunity to respond to these changes and contribute to knowledge about the change in criteria from DSM-IV to DSM-5 and this work was conducted as part of this PhD. This had several effects on the current thesis. The work already conducted to that point had centred on items in the DISCO that were not specifically diagnostic (associated items) in comparison with the current diagnostic criteria at that time which were specified by DSM-IV and ICD-10. This resulted in two relatively distinct sections of the thesis one on the DSM-5 diagnosis the other on “associated” features. In addition, the attention on DSM-5, which is only the focus of one empirical chapter but conducted across an 18 month period, added to the limitations for collecting further primary data that could have been collected to strengthen the validity of findings across the thesis. The limitations of conducting a series of analyses on the same datasets are considered throughout the thesis and acknowledgements are made about the generalizability of these findings without further follow up.

# THESIS SUMMARY

The overall aim of this thesis is to use the distinctive "spectrum" approach uniquely offered by the Diagnostic Interview for Social and Communication Disorders (DISCO) to explore the different descriptions of Autism Spectrum Disorder across four parts of the thesis. Part 1 introduces the concept of an "autism spectrum" and evaluates how this is continually developing. In Part 2, the focus is on the associated behaviours of ASD, that is, the behaviours not used to make a diagnosis. Sensory behaviours were found to be related to the core features of ASD, to maladaptive behaviours and were found to mediate the relationship between maladaptive behaviours and the core features of ASD, implicating sensory behaviours as having a particularly influential role in the behavioural manifestation of ASD. Further work addressed the lack of research on adults and found high-functioning adults with ASD reported significantly more sensory behaviours than IQ matched typically developing adults. Part 3 explored the measurement characteristics of two definitions of ASD: Wing and Gould (1979) and the new Diagnostic and Statistical Manual for Mental Disorders (DSM-5). A new diagnostic algorithm for DSM-5 criteria using DISCO data was designed and tested. Results showed that when the DSM-5 behaviours were mapped accurately onto a tool that is capable of measuring the spectrum, the DSM-5 criteria were found to have both good sensitivity and specificity. An advantage of the DISCO is the range of behaviours included. Comparison with Wing and Gould's measure revealed a strong overlap in spectrum concepts and one single element "quality of social interaction" had excellent diagnostic accuracy alone. Finally, in Part 4 the findings from Part 2 and 3 are brought together and discussed and a revised definition of the "autism spectrum" is proposed with regards to a continuum of reciprocal social interaction, which proposes that the pattern of behaviours seen in ASD need to be considered in combination.

# LIST OF TABLES

# LIST OF FIGURES

# PART 1: MEASURING AUTISM SPECTRUM DISORDER USING THE DIAGNOSTIC INTERVIEW FOR SOCIAL AND COMMUNICATION DISORDERS: AN INTRODUCTION

*"Nature never draws a line without smudging it."*

Wing (2005, p. 202).

## 1 Introduction: concepts of Autism Spectrum Disorder

The understanding of Autism Spectrum Disorder (ASD) is constantly evolving. In a broad sense the term is used to represent a cluster of behaviours characterised by qualitative impairments in social interaction, communication and repetitive behaviours. However, the diagnosis of ASD relies on the behavioural characteristics of individuals rather than biological, genetic or neuro-imaging measures; therefore, changes in diagnostic criteria can have substantial impacts on who meets criteria for a clinical diagnosis and how samples are defined in research. The overall aim of this thesis is to use the distinctive "spectrum" approach uniquely offered by the Diagnostic Interview for Social and Communication Disorders (DISCO, Wing, Leekam, Libby, Gould, & Larcombe, 2002) to explore the different descriptions of Autism Spectrum Disorder. As part of this overall goal, focus will be given to developing new measurement scales of behaviour relevant both for use in diagnosis as well as for use in describing the additional needs of individuals with ASD.

The first part of the thesis will aim to review the history of the term ASD and how our concept of "the spectrum" is continually developing. It will describe the findings from across the literature in ASD and the introduction of the international classification systems that have resulted in autism being conceptualised as Autism Spectrum Disorder. The first route to a conceptualisation of a spectrum focuses on the ongoing research using the concept of Autism Spectrum that started with Kanner and evolved from the work of Lorna Wing and Judith Gould in the 1970s; the second route details the course of the international classification systems of ICD and DSM and the modification to the diagnostic criteria in the past thirty years including changes to the Diagnostic and Statistical Manual for Mental Disorders (DSM-5, American Psychological Association) in 2013. A final section provides an overview of the thesis and summarises the questions addressed in the rest of the chapters.

## 1.1 Section 1: Concepts

### *1.1.1.1 The early history of Autism*

The first systematic recording of the behavioural symptoms of autism was made by Leo Kanner (1943), a specialist in children's psychiatric services. He described a condition, which he believed had not been recorded until this point that was based on similar characteristics seen across 11 children (8 boys; 3 girls, 2-11 years of age). He recorded individuals, abnormal from birth or within 2½ years of age, with impairments in social aloofness and indifference to others, resistance to change in their pattern of activities, impaired communication, mute or deviant language use and also a fascination and unusual dexterity of manipulation of objects.

Kanner (1943) concluded that children had a fundamental impairment in being able to relate themselves to other people or objects in the typical way, which was present from the beginning of life. The continuing clinical work of Kanner resulted in the development of specific criteria for Early Infantile Autism, a psychobiological disorder that occurs within the first two years of life and is characterised by a profound lack of affective contact as well as resistance to change in elaborate repetitive routines (Kanner & Eisenberg, 1956).

Asperger (1944) also made similar observations of children at the same time although his writings were not translated and available in English until much later (e.g.Frith, 1991; I. Gillberg & Gillberg, 1989; Wing, 1981). Asperger detailed a slightly milder form of behaviour than was described by Kanner, while working in Austria. The individuals he observed had socially odd or egocentric behaviour, long-winded and repetitive speech or odd intonation, deficits in non-verbal communication, restricted interests, repetitive routines and behaviour, poor motor coordination and a marked lack of common sense. Asperger (1944) believed the condition to not be recognised in infancy and usually not before the third year of life or later.

### *1.1.2 Conceptualising Autism as a spectrum: clinical reports and literature*

The proposal that autistic symptoms can be described as a continuum or a spectrum was first proposed by Wing and Gould (1979) who conducted an epidemiological study of all children under fifteen years old in Camberwell, London. The purpose of the study was to identify any children with autism or related features by collecting information from parents and teachers using a clinical interview: the Handicaps, Behaviour and Skills schedule (HBS; Wing & Gould, 1978). The HBS, an earlier and much simpler version of the DISCO, was a semi-structured interview used to systematically collect information on individual's clinical behaviours and level of development.

At the time, few studies had conducted epidemiological surveys of early childhood autism in a geographically defined population. The first was conducted by Lotter (1966) in Middlesex in which

all children aged, 8, 9 or 10 years old were screened (N=~78,000) for the main clinical features described by Kanner (1943). Postal questionnaires to teachers or professionals screened individuals for social aloofness, repetitive/ritualistic behaviour, abnormalities of speech and stereotyped movements and 135 children were selected for further assessment. Lotter (1966) found 15 children who had marked behaviours for both of Kanner's original criteria, social aloofness and resistance to change, and 20 with these characteristics but without both behaviours in a severe form. This gave a prevalence of early infantile autism of 4.5 per 10,000 children. However, a third group of children were also identified (although excluded from the main conclusions) who had some autistic features but did not score high enough to meet Kanner and Lotter's inclusion criteria.

In the Camberwell (total population 155,000) study by Wing and Gould (1979), researchers sought out any child below 15 years of age (N=35,000) who was known to educational, health or social services in the London Borough and had one or more of the features described by Kanner, or any child whose behaviour appeared to be peculiar, odd or strange for any reason according to teachers or professionals (taking into account the child's level of development). Screening procedures included identifying individuals with "absence or impairment of social interaction, especially with peers; absence or impairment of development of verbal and nonverbal language; repetitive stereotyped activities of any kind" (page 13; Wing & Gould, 1979). In addition, all children recorded as having a severe intellectual impairment were included. This in turn provided a comparison group of individuals who had severe intellectual impairment but none of the features described by Kanner (1943). This identified 132 individuals, all of whom had a learning disability for full assessment using the Handicap and Behaviour Skills Schedule.

Two approaches were used to attempt to classify individuals with and without the behaviours described above with the best accuracy. First the authors attempted to identify named syndromes such as those of Kanner (1943) and Asperger (1944). The authors identified a small proportion of children who met the criteria for early infantile autism; 17 individuals met the two criteria specified by Kanner and Eisenberg (1956), profound lack of affective contact and resistance to change in elaborate repetitive routines, and only seven individuals met all criteria described by Kanner (1943). In addition, only a few individuals were identified who matched the criteria described by Asperger (1944/1991), although this is also likely due to the sampling bias of individuals in special education. Overall, a large proportion of individuals identified in the study had a range of behavioural features from across descriptions.

The second approach to classification used by Wing and Gould (1979) was to use the current behaviours elicited by the HBS schedule. The authors were able to categorise individuals into two groups using the quality of social interaction score on the HBS: those who had an intellectual

disability but took pleasure and had interest in social interaction (appropriate for their mental age); and those who had inappropriate social interaction. The individuals with a social impairment were almost always found to have impairments in their communication skills both in terms of their non-verbal communication and the reciprocal two-way nature of their interaction. In addition, these children were found to have impairments in symbolic/imaginative activities and these groups of individuals were also characterised by having a limited pattern of activities, usually rigid and repetitive rather than creative and flexible. The authors referred to this collection of behaviours as the "triad of impairments." The triad consisted of impairments in social interaction, social communication and imagination which in turn were associated with a repetitive or restricted pattern of activities. Later the triad extended the description of "imagination" to be confined to "social imagination".

Wing and Gould (1979) described these individuals along a scale, in which individuals could vary widely across any area of the triad; these impairments were conceptualised as independent dimensions along which individuals could score. Wing and Gould did not explicitly define what the definition of a dimension was, however, they described such dimensions, and the continuum as a whole, as ranging from least to most autistic and therefore, in this sense the dimensions referred to the severity of the behaviour, for example, an individual may have severe impairments in interacting with their peer group on the social interaction dimension but relatively less severe behaviours for the repetitive behaviour dimension. The atypical social interaction style (the social dimension) was described as ranging in severity from "aloofness or indifference" to others, such that communication was limited to obtaining needs to "passive" acceptance with no spontaneous social behaviours or "active but odd" social approaches, in which individuals attempted to engage with everyone (friends, family and strangers) but in a limited form such as asking everyone the same set of repetitive questions (see Wing, 1988). Communication impairments could vary in severity from no attempts to communicate, communicating needs only to a lack of reciprocal communication such as asking repetitive questions or engaging in monologues. Imagination could also be absent, limited to play copied from others to having spontaneous imaginative play that is repetitive or cannot be modified by others. Furthermore; restricted and repetitive behaviour can range in severity from simple motor stereotypies, to organising objects, insistence of sameness or circumscribed interests (e.g. calendars or train timetables).

Abilities in other domains could also vary greatly across individuals presenting with the triad of behaviours. This is best presented by (Wing, 1991a, p. 127) who claimed it was possible for any combination of skills and difficulties to occur together and that there was an "uneven profile" of ability in individuals with autism and the triad of impairments:

> "It was possible for a child to be aloof, mute, unable to dress himself without help, but to be adept at jigsaw puzzles, making the most unlikely objects spin and climbing and balancing in the most unlikely places. Another child would be aloof sometimes and passive at others, with poor expressive speech, but slightly better comprehension, unable to read, but good at working with numbers, unable to cross the road safely on his own but capable of cleaning and tidying the house without supervision. Yet another would be active but odd in social interaction, with good grammar and a large vocabulary, but repetitive in speech and poor in comprehension, able to read fluently with little understanding, hopeless at arithmetic, ill coordinated in most gross motor skills but able to dismantle and put together again any spring-wound clock he could lay his hand on." (Wing, 1991a, p. 127).

The authors also found that individuals with a social impairment and the rest of the triad of impairments were more likely than the socially appropriate individuals to present with behavioural problems (maladaptive behaviours) such as temper tantrums, wandering, screaming, random aggression and destruction, self-injury and aimlessly creating chaos as measured by the HBS. They described these behaviours as particularly unpredictable, bizarre and unfamiliar, although problem behaviours were also seen in the socially appropriate children they tended to occur in a social context.

When individuals included in the epidemiological study were followed up in adolescence and early adulthood, further evidence was found that Kanner's criteria were not a discrete disorder (Shah, 1986; Wing, 1988). In all cases the individuals still had impairments across the triad of behaviours found in their childhood. It was noted that around 20% of the individuals changed in their pattern of interaction, in most cases changing from aloof or passive in childhood to passive or active but odd in later years. In a minority of cases, only a subtle impairment could be found but this was still impacting on individuals social and work ability. However, some individuals who met criteria according to Kanner when they were children, less easily fit the criteria as young adults, although they were sill impaired across the triad. This further emphasised the difficulties in differentiating specific syndromes from others.

The key development from the work of Wing and Gould (1979) and subsequent theorising (Wing, 1996, 1997) was that impairments can be seen across a triad of behaviours that vary dramatically in the severity of the skills or behaviour. This continuum relates to Kanner's autism and Asperger's descriptions as well as to the descriptions adopted by DSM-III and DSM-IV (see below) but is also wider than these specific descriptions. "The various clinical pictures of autism and related disorders depend upon the combinations of different impairment, which vary in severity

independently of each other, though they interact to produce the overt behaviour pattern" (Wing, 1991b, p. 111). Wing (1991a) described this continuum as, "at the high end", integrating into "unconventional "or "peculiar but normal behaviour". It was suggested that the decision on whether an individual meets a clinical diagnosis should be decided by whether the individual is limited by their impairments and whether the individual requires support. Furthermore, Wing (1991a) stated that although definitions or cut-offs are required in research, these should not impact on the clinical needs of the individual.

#### *1.1.2.1 Summary*

The epidemiological study conducted by Wing and Gould (1979) was helpful in identifying the prevalence as well as the boundaries of the clinical descriptions of autism. It has been contended that individuals with ASD better fit the concept of a continuum than separate, definable categories. Wing (1988, 1991) argues that the findings from such work are best explained by conceptualising autism as a continuum of impairments. It is argued that the central focus of the continuum is that of an intrinsic impairment of reciprocal social interaction but it also covers other psychological functions such as communication, imagination and the resultant repetitive behaviours (Wing, 1988). Wing's view is not of a continuum that only ranges from least to most 'autistic' but allows any combination of the triad of behaviours to be present, with each aspect of the triad varying along a continuum. This continuum approach would therefore facilitate a diagnosis that meets individual needs, allowing assessment of where the individuals impairments are for each area of the triad such that one individual may have severe social interaction problems but infrequent repetitive routines.

Wing (1991) maintains that while the triad of impairments form the focus of behaviour and are most important to capture, an individual's clinical profile is also impacted by a range of additional behaviours and developmental skills such as non-verbal communication, language, motor co-ordination, visuo-spatial skills and that any combination of these behaviours can occur to produce the individual's clinical picture. This highlights the need to better understand the additional dimensions of untypical behaviours such as sensory processing deficits or anxiety that occur with high prevalence in ASD and the role they may play in the behavioural manifestation on the autism continuum.

This first conceptualisation of a spectrum has some restrictions. The conceptualisation proposed in the paper (Wing & Gould, 1979) is a view largely based on clinical experience and in many ways it is not specific enough to test directly. Greater specificity of the number or types of behaviours that would be classified as socially impaired is required as well as a definition of the

boundaries between a clinical disorder along the spectrum in order to test the proposed classifications. This is addressed to some extent with the publication of the DISCO for which the authors designed a Wing and Gould spectrum algorithm (see Chapter 6). However, it is not clear from the work so far if the continuum described here reflects just a severity scale or the range of clinical conditions that present with impairments in social interaction. More work is needed to assess whether the classification based on social impairment can be used to determine either intervention strategies such as educational needs, management techniques and prognosis and in addition if it is related to specific physiological, biological or psychological impairments or functioning. In addition, whether the triad of impairments are found together in additional samples will provide further evidence for this model. The point made by Wing and Gould (1979) was that any system of classification should be based on the full range of conditions that have impairments in social interaction. This continuum approach was not, however, the view originally adopted by the international classification systems.

### *1.1.3 The path to a spectrum: the International Classification Systems*

At a similar time that Wing and Gould (1979) were conceptualising autism as a continuum or spectrum disorder, the term "autism" was first introduced into the Diagnostic and Statistical Manual for Mental Disorders (DSM-III, American Psychiatric Association, APA, 1980) as a categorical label describing a specific disorder. Before this time the DSM-I had largely overlooked the descriptions made by Kanner (1943) although children demonstrating autistic-like symptoms would have been classified as "Childhood Schizophrenic" (APA, 1952) which was also true of DSM-II; the descriptions for childhood schizophrenia include "autistic, atypical, and withdrawn behaviour" (APA, 1968, p.35). This reflected the view in the 1950s and 60s that autism was a form of childhood schizophrenia likely caused by the confusion of terms used by Kanner "infantile autism" and a psychiatrist Eugen Bleuler who used "autism" and "autistic" in 1911 to describe the aspect of schizophrenia in which a person withdraws from outside world into himself (Sicile-Kira, 2004).

The work of Michael Rutter (Rutter (1968), 1972), 1978)) was influential in differentiating what he termed childhood autism from schizophrenia by detailing that individuals with autism in comparison to those with schizophrenia presented with a stable course of impairments, without hallucinations and/or delusions, that autism could be comorbid with intellectual disability and was more common in males than females and finally that the developmental course was different; autism peaked in infancy and schizophrenia in adolescence. Rutter's (1978) review of Kanner's descriptions and following research emphasised a disorder termed "Childhood Autism" with an onset before 30 months and a focus on the importance of social impairments, language deviance

and atypical behaviours for a child's developmental level. These clinical descriptions with the original "Early Infantile Autism" descriptions of Kanner (1943; Kanner & Eisenberg, 1956) helped form the basis of the criteria used within DSM-III and DSM-III-R.

#### *1.1.3.1 DSM-III*

"Infantile Autism" was introduced under the category of Pervasive Developmental Disorders (PDD) in DSM-III (APA, 1980). The category was comprised of categorical diagnoses of residual infantile autism, Childhood Onset Pervasive Developmental Disorder (COPDD), Residual COPDD and Atypical Autism. In order to meet criteria for Infantile Autism, individuals were required to display symptoms before 30 months of age and present with "pervasive lack of responsiveness to other people; gross deficits in language development; if speech is present, peculiar speech patterns such as immediate and delayed echolalia, metaphorical language, pronominal reversal; and bizarre responses to various aspects of the environment e.g. resistance to change, peculiar interest in or attachment to animate or inanimate objects" (APA, 1980, p. 89).

The approach adopted by DSM-III was monothetic; individuals had to meet all specified criteria. However, revisions of DSM-III-R adopted a polythetic approach (APA, 1987) and terminology was also changed from Infantile Autism to Autistic Disorder, which reflected a change in criteria that allowed individuals to still meet the description as they aged. In addition, diagnoses of COPDD were removed from the criteria and PDD-Not Otherwise Specified (PDD-NOS) was added. In this version, 16 diagnostic criteria were presented which individuals receiving a diagnosis of Autistic Disorder had to meet eight.

#### *1.1.3.2 The introduction of sub-groups*

The descriptions of individuals seen by Asperger (1944) were not recognised until Lorna Wing described the clinical account of such individuals (Wing, 1981) and until Asperger's descriptions were translated into English in 1991 (Frith, 1991). Wing (1981) described the clinical features, course, differential diagnosis and management of Asperger's syndrome based on the original cases that he presented as well as 34 cases (5-25 years) seen by Lorna Wing herself. Wing (1981) added to the behavioural criteria specified by Asperger (1944), which were argued to become apparent through appropriate questioning about the developmental history of the individuals. These additions were: a lack of interest and pleasure usually seen in infants for company of others; in general a lack of the intense urge to communicate such as babble, laughter, smiles and movement; and that imaginative pretend play is not present or limited in individuals who fit Asperger's clinical descriptions. These descriptions of the condition, referred to as Asperger's

Syndrome rather than his term "autistic psychopathy" were readily adopted into the international classification systems, although the condition and specific requirements had not been previously validated beyond clinical description.

### *1.1.3.3 DSM-IV*

The categorical approach of DSM III-R was maintained across DSM-IV (1994) and the next version of text revisions (DSM-IV-TR[1], APA, 2000), which at the commencement of this PhD was the edition currently in use. In these criteria, Asperger Syndrome was added as a separate diagnostic category; the label of PDD now referred to five categorical diagnoses of Autistic Disorder, Asperger's disorder, PDD-NOS, Childhood Disintegrative Disorder (CDD) and Rett disorder. The diagnostic criteria are based on impairments in social interaction, communication and restricted or repetitive behaviours. The full criteria for Autistic Disorder can be seen in Box 1. Autistic Disorder is characterised in DSM-IV-TR as qualitative impairments in social interaction; qualitative impairments in communication; and restricted repetitive and stereotyped patterns of behaviour, interests and activities. Each impairment domain had four sub-criteria; individuals are required to have impairments for six of the criteria, at least two from the social interaction domain and one each from the communication and restrictive and repetitive behaviour domains. In addition, individuals are required to present with such behaviours before the age of three years.

In order to meet the DSM-IV-TR criteria for a diagnosis of Asperger's Disorder, individuals must have impairments in two of three domains required for Autistic Disorder (see box 1): qualitative impairment in social interaction and restricted repetitive & stereotyped patterns of behaviour, interests and activities. Individuals need at least two impairments from the social domain and one from the repetitive behaviour domain. In order to distinguish between Autistic Disorder and Asperger's Disorder, individuals cannot present with any significant delays in developmental milestones such as cognitive or adaptive skills or curiosity about the environment and they must also not have any clinically significant delay in language. In other words, individuals must have used single words by the age of two years old and phrase speech by three years of age. Finally, these impairments must cause clinically significant impairments in social, occupational, or other important areas of functioning.

[1] The text revisions to DSM-IV (DSM-IV-TR) did not have any impact of the diagnostic criteria for PDD.

**Box 1: Diagnostic Criteria for 299.00 Autistic Disorder**

A. Six or more items from (1), (2), and (3), with at least two from (1), and one each from (2) and (3):

1. Qualitative impairment in social interaction, as manifested by at least two of the following:
    a) marked impairment in the use of multiple nonverbal behaviours such as eye-to-eye gaze, facial expression, body postures, and gestures to regulate social interaction
    b) failure to develop peer relationships appropriate to developmental level
    c) a lack of spontaneous seeking to share enjoyment, interests, or achievements with other people (e.g., by a lack of showing, bringing, or pointing out objects of interest)
    d) lack of social or emotional reciprocity
2. Qualitative impairments in communication as manifested by at least one of the following:
    a) delay in, or total lack of, the development of spoken language (not accompanied by an attempt to compensate through alternative modes of communication such as gesture or mime)
    b) in individuals with adequate speech, marked impairment in the ability to initiate or sustain a conversation with others
    c) stereotyped and repetitive use of language or idiosyncratic language
    d) lack of varied, spontaneous make-believe play or social imitative play appropriate to developmental level
3. Restricted repetitive and stereotyped patterns of behaviour, interests, and activities, as manifested by at least one of the following:
    a) encompassing preoccupation with one or more stereotyped and restricted patterns of interest that is abnormal either in intensity or focus
    b) apparently inflexible adherence to specific, non-functional routines or rituals
    c) stereotyped and repetitive motor manners (e.g., hand or finger flapping or twisting, or complex whole-body movements)
    d) persistent preoccupation with parts of objects

B. Delays or abnormal functioning in at least one of the following areas, with onset prior to age 3 years: (1) social interaction, (2) language as used in social communication, or (3) symbolic or imaginative play.

C. The disturbance is not better accounted for by Rett's Disorder or Childhood Disintegrative Disorder.

Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS) is given as a diagnostic label to individuals with impairments in social, communication and repetitive or restricted behaviours, who do not meet full criteria for other PDDs or schizophrenia, schizotypal personality disorder, or avoidant personality disorder. For example, individuals who met behavioural criteria for Autistic Disorder but not the age of onset criteria or those with not enough (less than six) impairments across the criteria would receive a diagnosis of PDD-NOS.

Childhood Disintegrative Disorder is the fourth categorical diagnosis under the umbrella term of PDD. The diagnostic criteria states that individuals must present with a period of typical development of social interaction, adaptive skill and communication for at least the first two years of life, which is followed by regression (a loss of skills) in a least two areas of development including language, social skills, toileting skills, play or motor skills. In addition, the individuals must have

impairments in two of the three diagnostic behaviours for PDD. Finally, a diagnosis of Rett's Disorder is given if an individual has normal development in the first five months of life but a slowing of head growth from 5-48 months of age, loss of manual dexterity, early regression in social skills, deficits in co-ordinated movement and severe deficits in language. The aetiology is known: it is caused by the deletion or mutation of an X linked gene. Individuals must not meet criteria for Rett's disorder in order to be diagnosed with other PDD disorders. The move to the DSM-5, however, subsumes all of these sub-groups under the single broad category of Autism Spectrum Disorder (ASD). The specific changes are detailed in Section 1.1.5 below.

#### *1.1.3.4 ICD-10*

Another international classification system is the International Classification of Diseases (ICD), which is currently in its 10th version (ICD-10, World Health Organisation, WHO, 1994). The journey across criteria specified by the World Health Organisation (WHO) and the International Classification of Disease (ICD) was relatively consistent with the DSM pathway described above. "Infantile autism" first appeared in ICD-8 (WHO, 1967) as a sub-type of schizophrenia, and then under the heading "behaviour disorders of childhood" in ICD-9 (WHO, 1977) and eventually as a Pervasive Developmental Disorder (ICD-10, WHO, 1992), which closely matched with the DSM-IV.

A conscious effort was made to align the two classification systems for the DSM-IV and ICD-10 revisions (Volkmar, Reichow, & McPartland, 2012) and both contain criteria for categorical diagnoses of Pervasive Developmental Disorders. These two systems differ in their approach to diagnosis across research and clinical practice, the DSM-IV criteria were designed to be used for both purposes whereas the ICD-10 criteria provided two sets of criteria for these uses. Overall DSM-IV also has the capacity to allow comorbidity more than ICD-10 but is still restrictive (Volkmar et al., 2012). Despite these design differences, criteria for Autistic Disorder (DSM-IV) and Childhood Autism (ICD-10) are effectively identical and this was confirmed in a field trial of DSM-IV criteria over 21 sites and nearly 1000 cases (Volkmar et al., 1994). The criteria for additional diagnoses are also similar but again differ in diagnostic label: Asperger Syndrome rather than Asperger's Disorder. Furthermore, instead of the term PPD-NOS for DSM-IV-TR to capture individuals who do not meet the specific requirements of other PDDs, this concept is covered by three diagnoses in ICD-10: Atypical Autism (in age, symptomatology or both); other Pervasive Developmental Disorders; and Pervasive Developmental Disorders, unspecified. Overall, the two classification systems gather the same behavioural information and apply the same criteria to categorical disorders. There was a clear advantage of the convergence of diagnostic criteria across both DSM-IV and ICD-10 as this led to a magnitude of new research being conducted on both autism, as categorised as Autistic Disorder

or Childhood Autism, as well as Asperger Syndrome, indicated by 75 peer reviewed papers from 1944-1994, to over 1000 from 1994-2012, Volkmar et al. (2012). This is of clear benefit to advancing knowledge in the field of autism. In addition, this abundance of research resulted in the development of a range of diagnostic tools that could be used to confirm or aid in the diagnostic process of individuals taking part in research as well as in clinical settings, which are reviewed in Appendix 1.

### *1.1.4 Evaluating the international classification systems*

There are important limitations to the classification system descriptions of Autism as summarised above. Firstly, these are based on the view of Kanner, Asperger and clinicians in the field who hypothesised that groups of symptoms which they observed could be conceptualised as a specific disorder. However, as Wing et al. (2002) point out, the pathology, aetiology or causal dysfunctions of these disorders have not been confirmed using impartial criteria. The research that attempts this is essentially circular as clinical opinion is used rather than objective measures (see Volkmar, Cicchetti, Bregman, & Cohen, 1992).

In addition, the growth of research since the alignment of international classification systems DSM-IV and ICD-10 has also resulted in the testing of the accuracy of DSM-IV-TR and ICD-10 criteria and the categorical approach adopted by them both has not been well validated in the literature. The following section reviews the difficulties found when applying the current diagnostic criteria to individuals in research and clinical practice and evaluates the conceptualisation of autism as a spectrum. As discussed later in the chapter, the findings from empirical studies suggest that Asperger Syndrome cannot be reliably differentiated from autism using the ICD-10/DSM-IV criteria.

In all the studies described below the term 'autism' will be used to define either Childhood Autism (ICD-10) or Autistic Disorder (DSM-IV), and 'Asperger Syndrome' will be used to cover both definitions (DSM-IV, Asperger's Disorder), since as mentioned the international classification systems for DSM-IV and ICD-10 are equivalent (Volkmar et al., 1994).

#### *1.1.4.1 A problem with sub-groups: Statistical analyses*

One example of a study used to inform the debate about sub-groups applied a statistical approach of cluster analysis to search for sub-groups from the "bottom up" across a large amount of data from individuals with ASD (Prior et al., 1998). One hundred and thirty five individuals (3-21 years old) were assessed in clinics in UK and Australia; these individuals covered a range of diagnostic categories of PDD-NOS, high functioning Autism and Asperger Syndrome. All the individuals were high functioning with a verbal mental age of at least three years old. In order to

start from such a bottom up approach, a large amount of data was collected on these individuals. Information on background history as well as diagnostic behaviours such as those described in the DSM-IV and Wing and Gould's (1979) triad of behaviours were collected using the Autism Spectrum Disorders Checklist (Rapin, 1996) which had been based on an earlier version of the DISCO and HBS. In addition, developmental history variables such as problems in pregnancy, age of onset, family history and health problems were collected as well as the individual's current vocabulary comprehension.

The cluster analysis revealed three clusters: A, B and C. These clusters did not map onto the diagnostic sub-groups that the individuals had been assigned to in their clinical diagnoses. Of the individuals with high-functioning autism, 50% were found in Cluster A but the other half were spread across Clusters B and C and although the majority of Asperger cases were found to fit Cluster B, 30% were also found in Cluster A. Analyses were conducted comparing the "core autism features" across the three statistically devised clusters. It was found that individuals differed in the severity of the impairments rather than having qualitatively different patterns of behaviour. The children assigned to Cluster C had the least behaviours across all three domains, individuals in Clusters B and C tended to attempt to communicate their ideas and were more aware of social relationships than the Cluster A children. However, when specific symptoms were explored it was the severity of the same behaviours that differentiated groups for example social impairments were found across domains but whereas children in Domain A were socially isolated, children in Cluster B attempted social approaches but were unsuccessful (see Leekam, 2007). Interestingly, there were also a number of symptoms that did not differ across Clusters A or B (e.g. peer relationships and how to greet others) and some that did not differ across any of the individuals included in the study (e.g. impaired eye contact, response to others' emotions and non-verbal communication).

Further investigations were made about how these clusters differed on language ability as well as whether a delay had been present in language acquisition, which is diagnostically discriminative between Autism and Asperger Syndrome according to DSM-IV. The assignment to clusters was not predicted by the presence of a language delay, however, current language ability was extremely important; with children in Cluster A having lower verbal ability than Clusters B and C. This study was influential in applying a novel way of categorising individuals. Although it found that individuals could be categorised into one of three groups, these groups did not fit the categories proposed by the DSM-IV and were distinguished by the severity of their behaviours and their language ability. The authors suggested that it is best to understand these categories within a spectrum concept of autistic behaviour rather than discrete categories and that the severity of the

spectrum is based upon cognitive or current language impairment rather than qualitatively different autistic symptoms.

### *1.1.4.2 A problem with sub-groups: comparing "high functioning" autism and Asperger Syndrome*

There are three main arguments which claim that Asperger Syndrome and autism cannot be distinguished from each other in DSM-IV or ICD-10. The first argument is based on evidence that individuals with a clinical diagnosis of Asperger Syndrome, actually meet the diagnostic criteria for Autism (Autistic Disorder). This contradicts the DSM requirements that when the diagnosis of Autistic Disorder should be ruled out before an Asperger Syndrome diagnosis is given. The second argument makes the case that the criteria for speech delay that is used in DSM-IV to differentiate between Autistic Disorder and Asperger Syndrome is not reliable. Finally, it has been proposed that age and IQ may provide better explanations of differences seen across symptoms in individuals with ASD. These arguments and the supporting empirical studies are presented below.

#### *1.1.4.2.1 Individuals with Asperger syndrome meet criteria for Autism*

The primary argument against DSM-IV Asperger Syndrome being a unique diagnostic entity is that in empirical studies it has been found that most children with a clinical diagnosis of Asperger Syndrome actually meet criteria for autism. This was most surprising when the original case descriptions of the children made by Asperger (1944) were analysed according to the DSM-IV criteria. It was found that all cases met criteria for Autism rather than Asperger Syndrome (Miller & Ozonoff, 1997). In addition, in a sample of 68 preschool children with PDDs, including Autism and 23 with a clinical diagnosis of Asperger Syndrome, only 1 individual met DSM-IV criteria for Asperger Syndrome (Eisenmajer et al., 1996).

Mayes, Calhoun, and Crites (2001) investigated which DSM-IV criteria were met by 157 children with clinical diagnoses of both Autism and Asperger Syndrome. They found that independent reviews of these children, which applied the DSM-IV criteria, showed that all individuals met criteria for autism. That is all of the individuals, including those with a clinical diagnosis of Asperger Syndrome, presented with impairments in social interaction, restricted and repetitive behaviours but also had impairments in communication, that were not required in Asperger Syndrome. The authors concluded that this was not surprising, given that if an individual has problems with social interaction and a restricted pattern of behaviour they are likely to use repetitive language and have difficulty maintaining a reciprocal conversation.

It has been argued by Mayes and Calhoun (2003) that Asperger's original descriptions are different from DSM-IV criteria for Asperger Syndrome and instead are extremely similar to those presented in the DSM-IV criteria for Autistic Disorder. Asperger (1944) did not provide specific diagnostic criteria for his clinical description but made reference to inappropriate use of speech, reversal of pronouns and odd intonation. This is stark contrast to the criteria in DSM-IV in which individuals have to have impairments in social interaction and restricted or repetitive behaviours but not communication impairments. Also, two of his cases he recorded as being delayed in developing speech which again contrasts with the DSM-IV criteria.

In an earlier study, Leekam, Libby, Wing, Gould, and Gillberg (2000) also examined which ICD-10 criteria would be met by individuals who might qualify for either Autism or Asperger Syndrome diagnoses. In this study, they compared individuals who would meet ICD-10/DSM-IV criteria for Asperger Syndrome versus another measure of Asperger Syndrome, which more closely resembled the clinical descriptions made by Asperger. These criteria were defined by Ehlers and Gillberg (1993) and differed from ICD-10 as no constraints were made about early language development; individuals could meet Gillberg's criteria even if they had delayed language onset or language levels below their mental age. In addition, in order to meet Gillberg's criteria individuals must have at least three symptoms for communication impairments such as odd tone of voice, long winded speech, interprets language literally. In addition, the criteria specify six areas of impairment – language peculiarities, social interaction, narrow interests, repetitive routines and motor clumsiness.

The Leekam et al., study examined data from 200 individuals referred for a diagnosis at a tertiary diagnostic clinic where data on developmental history as well as current functioning was collected using the DISCO (Wing et al., 2002). Of the 200 cases only three individuals met ICD-10 criteria for Asperger Syndrome and in addition these three cases also met criteria for Childhood or Atypical Autism. In contrast 45.5% of the sample met criteria for Asperger Syndrome according the Gillberg's criteria (Ehlers & Gillberg, 1993). The cognitive and language abilities of the individuals meeting these criteria were variable, although two-thirds had age-appropriate cognitive and language ability, and one third had levels below their chronological age, 50% of the individuals had a delay in language and 25% had a low IQ. Leekam et al. (2000) found that these individuals differed from the rest of the sample on their level of repetitive behaviour, speech and language, motor clumsiness and non-verbal communication (although these behaviour were also seen across the other individuals) but they did not significantly differ on social impairment and narrow interests as measured by the DISCO. This work highlights that although the criteria better reflecting Asperger's

original descriptions is likely to identify more individuals, there is still a large degree of overlap with a diagnosis of autism, reflecting that the current system of sub-groups is insufficient.

#### *1.1.4.2.2 Differences in individuals presenting with or without a speech delay*

Another argument against the distinction of Autism and Asperger Syndrome in the diagnostic criteria concerns the presence or absence of early speech delay. This criterion which is intended to distinguish these two diagnoses has not been shown to be informative. Eisenmajer et al. (1998) found that the number of autistic symptoms was not significantly related to the presence of early language delay. Analyses were run comparing a group of 46 language delayed and 62 normal language onset individuals with pervasive developmental disorders (mean 11 years old) on ICD-10 and DSM-IV criteria as well as developmental history, receptive language and retrospective information on behaviour before the age of 6 years old. It was found that the presence of a language delay predicted more autistic symptoms when young (<6 years) but not at an older age. Nearing adolescence the only feature that early language delay predicted was social reciprocity although these individuals were also more likely to experience motor delays and impairments in receptive language skills. This led Eisenmajer et al. (1998) to propose that language delay is a feature of general developmental delay and it is this overall delay that is predictive of early symptoms of autism but that this had less effect as the children grew older. In high functioning individuals, early language delay did not seem to be reliable in predicting autistic symptomatology and therefore the distinction of Autism versus Asperger Syndrome in older children, was likely due to the large variability in clinical pattern over time. In addition, in the study described above, in the 135 individuals, early history characteristics including language delay did not predict to which cluster individuals were assigned (Prior et al., 1998).

Mayes and Calhoun (2001) found that 47 children with normal intelligence and a diagnosis of either Autism of Asperger syndrome (23 children with speech delay and 24 without) did not differ on any of the 71 symptoms measured according to the Checklist for Autism in Young Children (Mayes & Calhoun, 1999), even on the language items. By the time of testing (mean age of 6 years old) all individuals had developed language and had at least average verbal ability; the verbal IQ of individuals with a speech delay (verbal IQ = 95.4) was equivalent to that of individuals without a delay (IQ = 94.5) although most children in both groups had problems initiating and sustaining conversations and all but one had atypical speech patterns. In conclusion, it is claimed that the absence of significant language delay for a diagnosis of Asperger Syndrome is neither empirically or clinically justified.

#### *1.1.4.2.3 The role of age and IQ in determining sub-groups*

Finally, a further argument against the diagnostic sub-groups found in DSM-IV and ICD-10 is that autism symptoms have been shown to differ according to individuals' age and IQ. Mayes and Calhoun (2001) found that in a sample of 157 children with ASD, the 100 with IQs below 80 had significantly more symptoms and more problems in social interaction than those with an IQ above 80 but when IQ and age were statistically controlled for, there were no differences between groups. Furthermore, in the cluster analysis study, it was also found that symptom differences were explained by differences in age and IQ rather than diagnosis according to DSM-IV (Prior et al., 1998). The findings suggest that individuals vary in their symptom presentation and that individuals with a lower IQ may have more severe impairments.

Even though individuals can be distinguished by current ability level, this has not been shown to reliably discriminate between individuals with Kanner's Autism and Asperger Syndrome. Differences in the clinical descriptions argue that individuals described by Kanner (1943) developed speech late or not at all and do not use language to communicate whereas Asperger's cases were described as attempting to communicate but in a one sided manner (see Wing, 1991b). In addition, the criteria applied to Asperger Syndrome by the ICD-10 and DSM-IV focus on normal or high intelligence in these individuals. However, findings in the research reviewed above have found that this requirement is not consistent across the two diagnoses. For example, some individuals meeting criteria for Gillberg's Asperger Syndrome had language below their chronological age or IQs below 70 (Leekam et al., 2000).

#### *1.1.4.2.4 Assigning ICD-10/DSM-IV categorical diagnoses*

It has been found that professionals do not use ICD-10 or DSM-IV-TR criteria; instead they are basing their categorical diagnoses on research and case studies (Eisenmajer et al., 1996). In Eisenmajer's study, parent interviews were conducted for individuals with high functioning Autistic Disorder (n=48) and Asperger Syndrome (n=69) and logistic regressions were used to identify which behaviours clinicians used to assign individuals to clinical diagnostic groups. Very few clinical differences were found between the two diagnostic sub-groups; all of the individuals, regardless of whether their clinical diagnosis was of Autistic Disorder or Asperger Syndrome met criteria for Autistic Disorder; all of the individuals presented with some type of communication impairment. Forty three percent of the individuals with a clinical diagnosis of Asperger Syndrome had a language delay; clinicians did not use delay in onset of language as an exclusion criterion but the Asperger group were less cognitively delayed than the autism group. Furthermore, a multi-site study of the DSM-IV criteria and the ADI-R in 2,102 individuals revealed that even though the data collected from

standardised diagnostic instruments was good, clinical diagnoses were variable and different locations were found to give greater importance to additional factors such as verbal IQ or language level (Lord et al., 2012) in making a differential categorical diagnosis.

### *1.1.4.3 Reported differences between Autistic Disorder and Asperger Syndrome*

It is important to acknowledge, that there is also substantial literature that has found behaviour differences between individuals with a clinical diagnosis of Autism compared to Asperger Syndrome, these distinctions refer to differences in a range of areas such as core behaviour, IQ profiles, cognitive level, language ability, motor function, comorbidities, social cognitive tasks such as false belief and theory of mind, epidemiology and outcome. Tsai and Ghaziuddin (2013) argue that in their review of 125 studies (1994-2013) comparing autism and Asperger Syndrome: 30 concluded that both conditions were similar but 95 were reported to show qualitative and quantitative differences between the sub-groups. However these differences were defined as "significant or near significant level of differences" and no distinction was made between those with "significant" or "near significant" differences to assess how many statistically defined differences there were.

In the Tsai and Ghaziuddin (2013) review only six studies were identified that compared low functioning autism to Asperger Syndrome and the review claimed that five of these studies reported significant differences between the groups. However, as presented above, the severity of autism symptoms have been shown to differ across ability level (e.g. Mayes & Calhoun, 2001) and therefore the differences seen between the two groups can also be explained by IQ and furthermore, four (Lotspeich et al., 2004; Nordahl et al., 2007; Schumann et al., 2004; Scott, Schumann, Goodlin-Jones, & Amaral, 2009) of the six studies (Balconi, Amenta, & Ferrari, 2012; Larsen & Mouridsen, 1997) were conducted by the same group of researchers. Evidence for quantitative or qualitative differences between low functioning autism and Asperger Syndrome is limited.

Furthermore, the review claims that out of the 37 studies that compared PDD-NOS to autistic disorder, nine studies found no differences but 28 studies did report differences between these groups. However, only seven of these studies reported differences on the diagnostic features of ASD, for example, social skills (Anderson, Oti, Lord, & Welch, 2009; Fodstad, Matson, Hess, & Neal, 2009; Njardvik, Matson, & Cherry, 1999; Wilkins & Matson, 2009), repetitive behaviours (Matson, Dempsey, & Fodstad, 2009) clinical characteristics (Walker et al., 2004) and core features (Mandy, Charman, Gilmour, & Skuse, 2011; Matson, Dempsey, Lovullo, & Wilkins, 2008). However, given a spectrum of disorder, significant differences in severity for the core features are likely and individuals with PDD-NOS may just have milder behaviours. The remaining 21 studies reported

differences on associated variables such as problem behaviour (e.g. Snow & Lecavalier, 2011) which are not predicted to be identical across individuals with ASD. These differences may provide insight for future sub-typing of the spectrum but are limited in arguing for a distinction between PDD-NOS and autistic disorder.

Additional review studies have found no differences between Autistic Disorder and Asperger Syndrome on a range of clinical, cognitive and associated characteristics in: 26 studies (Howlin, 2003); 14 studies (Myer & Minshew, 2002); 41 studies (Macintosh & Dissanayake, 2004); 16 studies (Sanders, 2009). The review by Tsai and Ghaziuddin (2013) only identified two other review studies that concluded Asperger Syndrome was distinct from Autism (Kugler, 1998; Matson & Wilkins, 2008).

#### *1.1.4.4 Conclusion*

The evidence presented above has led Szatmari, Archer, Fisman, Streiner, and Wilson (1995, p. 1669) to claim that giving a diagnosis of Asperger Syndrome according to the diagnostic classification systems is "virtually impossible." Research has found that early speech delay is not predictive of later autism and that professionals are either not assessing full symptom profile or adapting the diagnostic criteria, as individuals with a clinical diagnosis of Asperger Syndrome are also meeting criteria for Autism. Although, some differences are reported in the literature between groups, which must not be ignored as they may provide useful insights into aetiology or management; what can be concluded from the review above is that individuals are not being assigned to these groups according to the criteria specified by DSM-IV and ICD-10 and therefore, revisions to these classification systems need to reflect this. In addition, it is argued that language delay is difficult to measure retrospectively and therefore it is not reliable to distinguish groups on this variable (Lohr & Tanguay, 2013).

Before the publication of both ICD-10 and the DSM-IV professionals were already arguing for the removal of the term PDD and replacement with ASD. Baird et al. (1991) argues that the introduction of Pervasive Developmental Disorders into the diagnostic criteria was a mistake as there is too much distinction between the categorical labels, for example, parents were less likely to participate in early intervention and seek services if a child was given a PDD-NOS rather than an autism label. In the same year, Happé and Frith (1991) claimed that:

"PDD is uninformative in that is does not describe the nature of the impairments in the disorders it covers. Yet it is the nature of the disorder – the triad of communication, socialisation and imaginative impairments – that is the most homogeneous in this group (Wing, 1988). What is at

least consistent across the patients covered by the "PDD" label is the severity of the impairment – yet this is all that the term "PDD" specifies." page 1167.

Happé and Frith (1991) claim that the term "pervasive" is misleading for two reasons, 1) that this term can be interpreted in many ways, such as brain, cognition or behaviour and 2) that impairments of the autism continuum do not always affect all abilities at all levels. They argue, for example, pervasive abnormalities have not been found across the brain in individuals with autism; individuals may have exceptionally good rote memories at the cognitive level and in some individuals daily living skills remain unimpaired.

Mayes and Calhoun (2003) summarise this issue by asking "Is the DSM always correct?" And their answer is "No". If it were, "we would not have DSM-I, II, III, III-R, IV, and IV-TR. The DSM is a diagnostic system that is being revised continuously. The DSM proposes diagnostic categories, which then must be validated or modified according to the results of empirical research (page, 20)." This research, therefore, has led to the proposal of an autism spectrum in DSM-5 (APA, 2013).

### *1.1.5 The DSM-5*

The DSM-5 committee recognised the evidence presented above that categorical sub-group distinctions are not supported and decided there was enough evidence to replace PDD with a single broad category of Autism Spectrum Disorder (ASD) which subsumes the previous categorical diagnoses (see Swedo et al., 2012). This single category of ASD would fit with the current terminology and concepts of Autism (e.g. Wing and Gould, 1979; Happé and Frith, 1991). The criteria for DSM-5 ASD can be seen in Chapter 5. The section below described the changes that distinguish criteria for DSM-5 from DSM-IV-TR PDD.

Five main changes were proposed within the DSM-5 draft criteria, which are detailed in Part 3 and summarised here. First all categorical sub-diagnoses within DSM-IV-TR PDD would be subsumed under a single concept of ASD. The second change was that the social and communication domains from DSM-IV-TR were to be combined into one domain. The third change is that the DSM-5 repetitive behaviour criteria will include criteria on the presence of hyper or hypo sensitivity to sensory input or the sensory environment. Fourthly, the criterion for early childhood onset includes the caveat that although symptoms must be present in early childhood, they may not become fully manifest until social demands exceed limited capacities, to allow all of the previous categorical distinctions to fall under the umbrella diagnosis term. Finally, the requirement for significant severity (i.e. negative impact on daily functioning) is more explicitly stated in the revised guidelines.

### *1.1.5.1 Concerns about the DSM-5*

With the release of the draft criteria in 2011 and official release of DSM-5 ASD in 2013, there were many publications raising concerns about the proposed changes. The main worry was the removal of the Asperger Syndrome label. This caused a negative reaction among individuals with Asperger Syndrome who identified strongly with the diagnosis or perceive it be an advantage to them and endorse it as separate to autism and this is also a view held by professionals. A survey of 547 health and education professionals who were asked their views about the DSM-5 change demonstrated that 50% of respondents were opposed to the changes, 22% supported and 28% were uncertain and a main reason for this was the increased stigma attached to an "autism" rather than "PDD" or "Asperger" label (Kite, Gullifer, & Tyson, 2013).

Wing, Gould, and Gillberg (2011) raised a number of concerns with the first draft of the DSM-5 ASD criteria. Firstly, they identified that the imagination aspect of the original triad that they proposed is still widely overlooked in the new criteria, they argue that a lack of imagination leads individuals to be unable to foresee the consequences of their actions and this has a huge impact on the individuals social actions and on family or caregivers of the individual and should therefore be given more attention in the diagnostic criteria. Secondly, Wing et al. (2011) argue that more details should be included in the new criteria about diagnosis in early childhood. Diagnosis in infancy is important for early intervention and therefore better outcome but the DSM-5 has not focussed on this and Wing et al. (2011) argue that symptoms such as odd patterns of behaviour, lack or dislike of social interaction, problems in pre-speech communication and limited or specific interests, as identified as being present in early in individuals with ASD (e.g. Barbaro & Dissanayake, 2009), must be included to enable early diagnoses to be made. Thirdly, they argue that the criteria for onset in early childhood may need further elaboration, although this is extended to include individuals whose problems may not be apparent until social demands exceed their ability, some individuals being referred as adolescents or adults may not have an informer who knew them in early childhood or even over their childhood and therefore clear instructions are required for these cases. This may be particularly important for females who have been shown to miss a diagnosis of autism for this reason as they are not referred until adolescence (Kopp, Beckung, & Gillberg, 2010).

The DSM-IV system for autism allowed over 2000 combinations of criteria that could be met in order to receive a diagnosis, for example, only six criteria had to be met but they could be selected across 16 criteria, 4 for each impairment (social interaction, communication, repetitive behaviours), the only restrictions were at least two from social interaction, and one each from the other two impairment categories. The arrangement of domain and sub-domains in DSM-5, however, means only a small amount of combinations are possible, which could lead to a narrowing of the

criteria (Rutter, 2011; Volkmar & Reichow, 2013; Volkmar et al., 2012). There are concerns across professionals that the narrower criteria for DSM-5 ASD may make it harder for an individual with a DSM-IV diagnosis to meet criteria, especially individuals with Asperger Syndrome or PDD-NOS. In addition, this may impact on the access to services especially in countries such as America that rely on diagnostic labels to receive support from insurance companies (Lord & Jones, 2012). The DSM-5 committee argued that although some cases may no longer meet criteria, they are likely to be identified as having "social communication disorder" instead (Swedo et al., 2012), which is a new disorder in the DSM-5 that has similar behaviours to Domain A of DSM-5 ASD without the repetitive Domain B behaviours. However, this disorder is newly introduced and has no research background. In addition, the needs of an individual with this diagnosis have not been assessed so some individuals could lose the support they would have had with a previous DSM-IV categorical diagnosis.

The changes to DSM-5 will invariably make comparability across research using the different diagnostic systems difficult because individuals are recruited and compared according to their diagnostic label, this will be even more problematic if some DSM-IV defined individuals may now meet "social and communication disorder" (Vivanti et al., 2013). This raises the importance of using standardised instruments and even more, as total or severity scores across these instruments will be comparable regardless of diagnostic systems. The main concern is whether these changes will affect the ability to reliably track prevalence rates (Vivanti et al., 2013) or the issues in following up individuals in longitudinal or epidemiological samples (Volkmar & Reichow, 2013). The focus of Part 3 of this thesis is on more accurately measuring the behaviours described by DSM-5 (APA, 2013) using the DISCO.

#### *1.1.5.2 DSM-5 Summary*

For research, the advantage of the DSM-5 conceptualisation of ASD over the conceptualisation made by Wing and Gould (1979) is that specific criteria are proposed. These set of behaviours, examples and criteria that specify the number of impairments required can be directly applied to the behavioural profile of an individual or their clinical information. This allows specific questions to be tested such as whether individuals who previously met criteria for a categorical diagnosis of PDD in DSM-IV-TR also meet DSM-5 ASD criteria. The research conducted so far on the draft and official release of DSM-5 ASD criteria is the focus of the literature review in Part 3, Chapter 5. However, the existing research on DSM-5 ASD consistently shows that the criteria are more specific than the previous diagnostic criteria; DSM-5 ASD does not identify many clinical comparison individuals as having autism but does find that some individuals with ASD, especially high functioning

individuals or those with Asperger syndrome or atypical autism (or PDD-NOS), are likely to be missed according the DSM-5 ASD (e.g. Matson, Belva, Horovitz, Kozlowski, & Bamburg, 2012; McPartland, Reichow, & Volkmar, 2012; Taheri & Perry, 2012). There are some inherent limitations of the work conducted so far, namely, the use of checklists or research tools designed around the DSM-IV criteria, which may, therefore, not provide enough information to accurately map the behaviours descried in DSM-5 (see Part 3).

### *1.1.6 What is a spectrum?*

This introduction has described two conceptualisations of the autism spectrum, according to the DSM-5 and the work of Lorna Wing. DSM-5 describes a disorder with persistent deficits in social communication and social interaction across multiple contexts and restricted, repetitive patterns of behaviour, interests or activities that present in the early developmental period and cause clinically significant impairments in current functioning. Lorna Wing defines the autistic spectrum as a range of behaviours, which have in common life-long effects and the presentation of the triad of impaired social interaction, communication, and imagination, associated with a rigid, repetitive pattern of behaviour. In her description this spectrum is wider than the descriptions of Kanner and Asperger and the categorical disorders described in DSM-IV and ICD-10 but include these within the spectrum. Onset is usually at birth or during the first three years of life, but problems can begin later in childhood. The triad can be recognised at all levels of intelligence and can occur alone or together with any other physical or psychological disorder (Wing, 1997).

Lai, Lombardo, Chakrabarti, and Baron-Cohen (2013) argue that it is important to define what we mean by the autism "spectrum" in order to ensure consistency across research studies and to progress knowledge. They described the three definitions of the autism spectrum:

> "1. Spectrum can refer to the dimensional nature of the cardinal features of autism within the clinical population
>
> 2. Spectrum can also refer to the continuity between the general population and the clinical population.
>
> 3. Spectrum can also refer to subgroups. It has been suggested that "the autisms" may be a useful concept to reflect the substantial heterogeneity within the autism spectrum. (p. 2)."

Both the DSM-5 and Wing and Gould's conceptualisation of Autism Spectrum Disorder most closely fits the first description of a spectrum, as described above, as they define symptoms present in individuals who should receive a clinical diagnosis. In both descriptions it is the combination of the behaviours that results in individuals meeting criteria for a "disorder" (the centre of the figures below). For DSM-5 (Figure 1-1) this is the presence of social-communication impairments and

restricted or repetitive behaviours. For Wing and Gould (Figure 1-2) this is the presence of the triad of impairments in social interaction, communication and imagination associated with a repetitive or restricted pattern of behaviours.

The three definitions (Lai et al., 2013), however, are not mutually exclusive, indeed Wing (1991a) describes the mild end of the spectrum to merge into "normal eccentricity", which highlights the link between the clinical and typical range of ASD behaviours as per the second definition of the spectrum. The arrows on the figures below represent the severity of the behaviour, this varies across all individuals presenting with any behaviour but also across individuals that present with all behavioural criterion and are defined as having ASD.

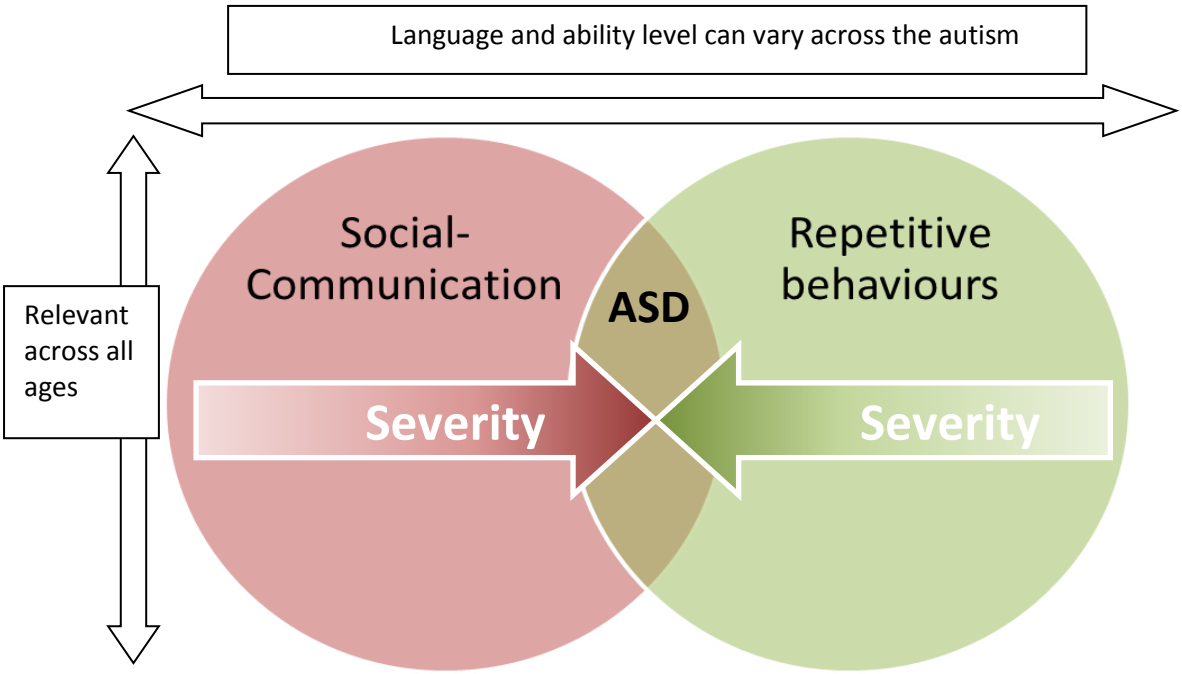



**Figure 1-1: Figure showing Autism Spectrum Disorder as defined by DSM-5 (APA, 2013)**

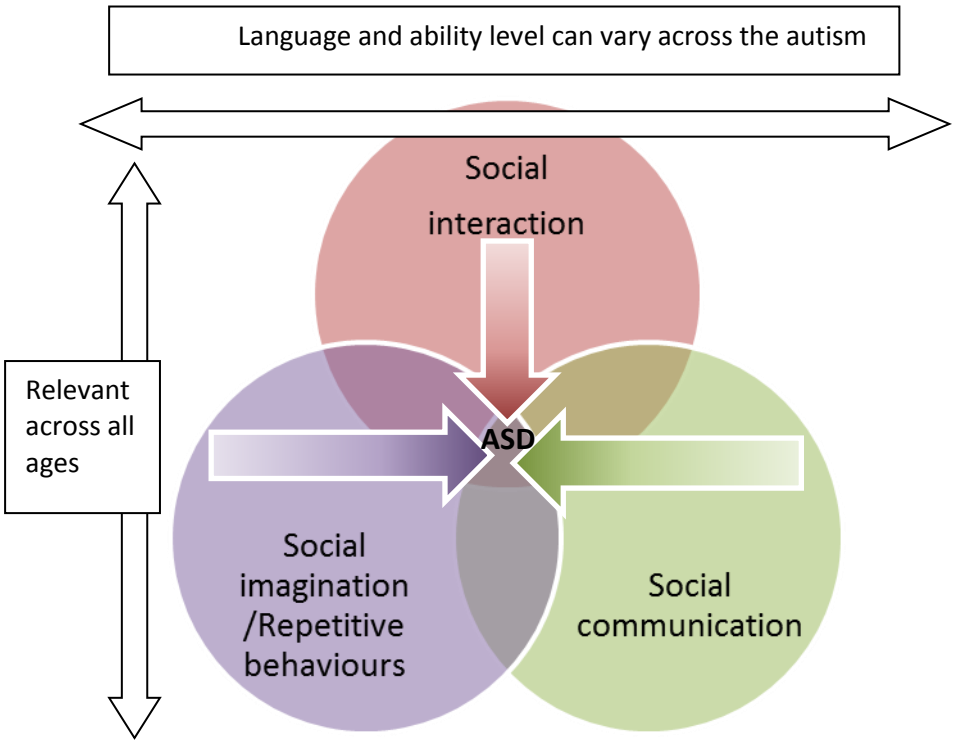



**Figure 1-2: Figure showing Autism Spectrum Disorder as defined by Wing and Gould.**

### *1.1.6.1 New sub-typing of the autism spectrum*

There has recently been much discussion about the need to further divide the spectrum into subgroups, which fits with the third definition of the spectrum above (Lai et al., 2013). By collapsing previous categorical approaches and adopting the term "spectrum" a diagnosis of ASD according to DSM-5 will result in a large amount of heterogeneity under the same label. Although, this may be beneficial in clinical practice for example, to allow all individuals to access services, it is difficult to search for causes of such a large range of behaviours present in ASD. As summarised by Amaral (2011, p. 8) "given the incredible heterogeneity of this disorder, understanding that one size will never fit all is a reasonable perspective to frame all future findings." It is argued that sub-groups will aid in the understanding of the aetiology of ASD at both the genetic and neurophysiological level (Szatmari, 1999). The term "the autisms" has been adopted by some researchers (e.g.Coleman & Gillberg, 2012; Geschwind & Levitt, 2007) and in order to be able to best understand the biology of "the autisms" both the similarities and differences need to be assessed (Lai et al., 2013).

The idea of this new set of subtyping is that rather than being constrained by existing categories, it is possible to search for patterns across behaviours or performance on tests, and attempt to link these patterns to either clinical patterns seen in other conditions or neuropathology (Leekam, 2007). This will hopefully, improve the research on the underlying cause of autism but may

also be of use in clinical practice where there is a trend to move toward individualised treatment. The DSM-5 committee has considered this to some extent by including "specifiers" in the diagnostic criteria. Therefore, individuals can have a diagnosis of ASD with or without an intellectual impairment or a language impairment and ASD can be associated with a known medical or genetic condition, another neurodevelopmental or behavioural disorder or with catatonia. Furthermore, individuals are rated on the level of severity of the ASD symptoms across three levels from requiring support to requiring very substantial support (APA, 2013). For example, an individual, rather than getting a categorical diagnosis of Asperger Syndrome would be described instead as "ASD with good language skills and high intelligence, requiring support for his social and communication and requiring very substantial support for his repetitive behaviour" (Vivanti et al., 2013, page 259). Sub-types are also proposed with Wing and Gould's concept of a spectrum as described in section 1.1.6.3 below.

### *1.1.6.2 Previous research on sub-typing*

An example of work that has already been examined for potential sub-types is individuals' language ability. For example, it has been found in a longitudinal study of individuals with ASD, followed up every two years (Time 1, 4-6 years of age), that the presence of Specific Language Impairment (SLI) at Time 2 was the best predictor of functional outcome in adolescence at Times 3, 4 and 5 (Bennett et al., 2008). Furthermore, individuals with ASD who also met criteria for SLI showed the same reversed asymmetry in the frontal language area similar to that found in individuals with SLI and no autism (Tager-Flusberg & Joseph, 2003). Evidence of brain structure differences within individuals with ASD is an excellent example of how useful such an approach may be in differentiating individuals across the autism spectrum, however, language ability level is not the only measure on which individuals differ on across the spectrum. With the official introduction of a spectrum approach to autism, it is essential that research continues to explore avenues for sub-typing in order to improve work on aetiology.

Other attempts at sub-typing have looked for links to specific chromosome locations. Liu, Paterson, and Szatmari (2008) attempted to examine six dimensional trait sets, which could be tested across 976 multiplex families in the Autism Genome Project Consortium. They examined scores on two sub-scales form the ADI-R; social reciprocity and repetitive or stereotyped patterns of behaviour as well as delayed onset of first words, delayed onset of first phrases, verbal status and IQ above or below 70. Although, the binary sub phenotypes of IQ and delayed onset of phrases showed some relations with specific chromosomes, no linkage was found across the other trait sets and research has not found strong associations to abnormal chromosomal sites (Waterhouse, 2013).

This research holds promise for best understanding how individuals on the spectrum differ from each other but as described by Rutter (2011, p. 339) "all that can be concluded firmly at the moment is that it is highly likely that there are meaningful subcategories of autism spectrum disorders but that these are not well identified."

### *1.1.6.3 Wing's social sub-types*

Wing (1988) argues that the groups are best defined according to their social interaction style. The aloof group is made up of individuals who are aloof and indifferent to others, especially their age peers. These children are those closely fitting the description of ICD-10 Childhood Autism or DSM-IV-TR Autistic Disorder and some of this group are likely to fit Kanner's criteria for Early Infantile Autism. Wing describes these children as likely having mild to moderate intellectual disability, potentially with higher skill in some areas. Individuals may have delayed or no speech and others may have characteristic language peculiarities such as echolalia and stereotyped speech. In those with more developmentally appropriate language level they do not use this to communicate in a reciprocal manner and individuals may use physical prompts to obtain needs. Play can be non-existent and pattern of activities are usually limited to repetitive routines. Furthermore, Wing proposes that beyond the triad these individuals are likely to present with unusual response to sensory input, problems with sleep, motor stereotypies, poor attention span as well as refusal to eat more than a few types of food.

The passive group is unlikely to be consistently seen in one categorical disorder in particular and individuals may be classed as Childhood Autism, Atypical Autism or Asperger Syndrome (ICD-10). This group are defined by their lack of social approaches but passively accept interaction from others, although the communication and imagination impairments are similar to those in the aloof group, individuals are less upset by changes in routine and their behaviour is often more calm. Some individuals may have average to high intelligence and problems may not be fully manifest until impairments with learning or social interaction appear throughout schooling.

The active but odd group make social approaches to others but these are usually inappropriate, naïve or one-sided. It is the Asperger Syndrome label (ICD-10) and the description by Asperger (1944) that best fit the behaviours seen in these individuals, however, it is only a minority of these cases that show no delay in language before three years of age. Most individuals develop language and good grammar but do not use this in a reciprocal way and it may become very repetitive. These individuals are likely to show play and superficial imagination, although again this is likely repetitive and not shared with others. In terms of repetitive activities, individuals in the active but odd group have fascination with and talk about particular topics e.g. train timetables. Wing

claims, motor co-ordination difficulties are prominent in the active but odd group but unusual responses to sensory stimuli, although they can happen, are less likely in this group. Behavioural difficulties such as temper tantrums and aggression are likely to occur if individuals are not able to engage in the activities they enjoy.

Finally, Wing describes a group of "high-functioning" individuals at the least extreme end of the triad. These individuals, described by some as loners (e.g. Wolff, 1995), present with the most subtle impairments. Individuals tend to have average, high or exceptional cognitive ability and fluent speech but these individuals prefer to be alone, are concerned with their own interests and may lack empathy. Wing describes these individuals as finding structured school life and peer interaction stressful in childhood but most are more successful in adulthood; social interaction rules may be learnt or individuals may remain happier alone. Wing proposes that this group do not tend to present with associated features.

In all groups, impairments in the triad of behaviours are present and this is what defines the spectrum. The question of need and eligibility to receive services is what defines these individuals from the typical population. This classification has received little empirical validation and will be analysed further in Part 3 of the thesis.

#### *1.1.6.4 Associated features*

One area that has been widely overlooked is what role the behaviours that tend to co-occur with the diagnostic symptoms of autism play in the condition. With the move toward a spectrum approach of categorising autism, these additional behaviours could be important in defining sub-groups and need more research focus. A range of secondary or "associated" features have been shown to co-occur with the "core" triad of symptoms such as sensory, motor and behavioural problems including externalising behaviours and anxiety and depression, as well as sleep disturbances and impairments in adaptive functioning. Wing (1996) descriptions of social sub-types include behaviours such as sensory and feeding atypicalities as well as motor co-ordination difficulties. Studies have found these behaviours, in some cases, to be extremely prevalent, for example over 90% individuals have been reported to have at least one sensory processing deficit (Leekam, Nieto, Libby, Wing, & Gould, 2007; Tomchek & Dunn, 2007).

A potential avenue for future research is whether these behaviours are beneficial in sub-grouping individuals on the autism spectrum and whether they have predictive value for future outcome. For example, a key finding in the cluster analysis conducted by Prior et al. (1998) was that they found Clusters A and B differed in the severity of their social and communication behaviours

but they did not differ on a measure of sensory impairments, although both groups differed on sensory impairments compared to Cluster C.

This area of sub-grouping is especially relevant given the focus on comorbid conditions with ASD and that these associated features are likely to overlap with other relevant conditions. Further knowledge about this overlap may aid in the discovery of common neurobiological pathways or genetic connections. For example, problems in attention or hyper-activity are common across both ASD and Attention Deficit Hyper-activity Disorder (ADHD) and these behaviours have been found to co-occur (e.g. Ronald, Simonoff, Kuntsi, Asherson, & Plomin, 2008). Furthermore maladaptive behaviours are seen in ASD as well as disruptive behaviour disorders which have shown variable but substantial overlap with ASD e.g. Conduct Disorder (1-10%) and Oppositional Defiance Disorder (4-27%; Kaat & Lecavalier, 2013). Another example is motor atypicalities in ASD that can also be found in Catatonia (e.g. Wing & Shah, 2000; Wing & Shah, 2006) or Developmental Co-ordination Disorder (e.g. Dewey, Cantell, & Crawford, 2007; Fournier, Hass, Naik, Lodha, & Cauraugh, 2010). It is with the exploration of these behaviours that overlaps with similar clinical conditions are likely to be seen and this could be important for learning about shared aetiology.

However, this area is relatively understudied in comparison to the core features of ASD and in comparison to other symptoms that have been used to sub-type individuals such as language level. Before the work in this area can progress to defining sub-types, it is essential to gain a more systematic understanding of when these behaviours co-occur: whether they are found across the whole spectrum and like social or communication behaviours differ in their severity across the spectrum; whether they are only present in certain types of individuals with ASD, for example more prevalent in higher IQ or younger age; or whether the patterns or numbers of behaviours present are specific to ASD. We need to have a better understanding of the role that associated features play in ASD and only once this is established can we look at using these items to sub-group ASD; "new work on symptoms that co-occur with diagnostic symptoms points to the possibility of behavioural sub-grouping that may be more valuable than previous distinctions between Asperger Syndrome and autism in helping us to understand patterns of associated and risk factors for autism" (Leekam, 2007, page 242).

#### *1.1.6.5 Why is it important to study associated features of ASD?*

There are two main reasons why the study of associated features is important: firstly, for what they may be able to tell us about ASD and secondly for what they can inform us about the individual. The relation between associated behaviours and the core features of ASD are important to consider. Robust relations may indicate shared neurobiological mechanisms are underlying both

areas of functioning, which could provide insights into the aetiology or causal nature of the core behavioural manifestation. For example, it has been proposed that the presence of repetitive behaviours such as motor stereotypies and self-injury have been predicted to provide an intrinsic sensory stimulation to the individual (Lovaas, Newsom, & Hickman, 1987). In addition, relations may indicate similar intervention approaches could be used to manage both types of behaviours or even that additional descriptions of associated behaviours could be added to diagnostic criteria to better characterise the clinical phenotype.

The associated features of ASD also represent the needs of the individuals and exploration of these behaviours allow better understanding of the problems experienced by families and other carers. For example, aggression and temper problems are widely reported in individuals with ASD (e.g. Kaat & Lecavalier, 2013; Matson, 2009) and the management, intervention and care programmes put in place for these individual are likely to be unique from individuals without aggression or problem behaviours but who may present with feeding problems or additional mental health problems such as anxiety or depression. In addition, these behaviours may impact on the behavioural presentation of the core features, for example in ASD the presence of sleep problems have been shown to predict higher levels of autistic symptoms (e.g. Mayes & Calhoun, 2009). Overall, the associated features of ASD are thought about in two distinctive ways: as a causal role (e.g. sensory, see Chapter 4) or as a secondary symptom. This process has meant that associated features are not measured in a consistent way and that the effect of associated features on each other in addition to the impact on the core features of ASD is rarely considered. The measurement of the associated features and work on role they play in ASD is explored in Part 2 of the thesis.

### *1.1.7 Thesis summary*

The definition of a spectrum that is focussed on in the current thesis is of clinical disorder in which the core features of autism have a dimensional nature within the clinical population. In the remaining part of this chapter I provide an outline of the thesis chapters, Chapter 2 follows in which I provide detail on the measurement of ASD using the DISCO. This completes Part 1. The focus of Part 2 will be on the associated symptoms of ASD and how these behaviours interact with the core features of ASD. These core features will be based on behaviours used to make a diagnosis of Pervasive Development Disorder according to the ICD-10 (and DSM-IV-TR). This is because Part 2 was predominately conducted before the release of the draft criteria for DSM-5 ASD (2011). The specific behaviours that best identify who falls into Autism Spectrum Disorder will be the question under investigation in Part 3 and the two definitions of the spectrum described above (DSM-5 and Wing and Gould) will be compared.

### *1.1.7.1 Aims of the thesis*

This thesis will explore how we conceptualise autism spectrum disorder. The aim is to identify which patterns of behaviour best identify individuals that fall within the clinically defined autism spectrum to define who gets a diagnosis and what additional behaviours these individuals may present with.

### *1.1.7.2 Chapter overview*

The empirical work in this thesis is split across two parts. In Part 2 of this thesis, the focus will be on the range of behaviours that are present in individuals with ASD. In Part 3 the focus is on how to define diagnostic criteria that capture individuals who fall within the clinical autism spectrum. Although both Part 2 and 3 are conducted using secondary datasets collected with the DISCO, they do focus on different aspects of the behavioural presentation of ASD.

Over the course of the research programme the proposed changes to the international classification systems, specifically; the criteria for Autism Spectrum Disorder were proposed (2011) and subsequently published by DSM-5 (2013). The spectrum approach uniquely offered by the DISCO provided the perfect opportunity to contribute to knowledge about the change in criteria from DSM-IV to DSM-5 and this work was conducted as part of this PhD. The work already conducted using the DISCO was based on diagnostic criteria specified by DSM-IV and ICD-10 and therefore the decision was made to sub-divide the thesis in two Parts. The DISCO has two key roles in a clinical setting, to aid clinicians in making a diagnosis and to aid them in creating a profile of the individual's strengths and weaknesses across behaviours that may not be diagnostically relevant but important in improving the quality of the individual's life. These roles of the DISCO are reflected in the research aims of the thesis and are explored in Parts 2 and 3 as described below.

#### *1.1.7.2.1.1 Chapter 2:*

Chapter 2 will describe the contents of the Diagnostic Interview for Social and Communication Disorders (DISCO), and how the interview should be conducted and scored. This chapter will focus on how the DISCO was designed and the impact that this has on how the information is collected and can be interpreted. How the DISCO can be used to guide a diagnosis will be covered as well as what additional information the DISCO can gather to aid individual's diagnosis, management and needs. This chapter will also describe the three secondary datasets that will be used throughout the thesis to answer the questions set out below.

#### 1.1.7.2.2 Part 2: Measuring the autism spectrum: Associated features

Part 2 of the thesis focused on the "associated" behaviours present in ASD, that is, behaviours that are not included in the diagnostic algorithms. The reason for including associated behaviours was two-fold. The main aim of Part 2 related to the search for measurement of the boundaries of the 'spectrum' of autism and this was to explore whether one or more associated feature of ASD has a particularly influential role in the behavioural manifestation of ASD. Firstly, from a purely methodological perspective the associated scales from the DISCO had not been tested for reliability of their use in research or as independent scales of behaviours. In order to address the main aim, the first purpose was to assess how well the DISCO measures associated features of ASD and preliminary analyses were used to identify which scales within the DISCO were reliable. The main aim of identifying the role of the associated features was tested across Chapters 3 and 4 using the scales identified as being reliable and ASD specific in the DISCO: the maladaptive behaviour and pattern of activities scales were explored together in Chapter 3 and the sensory behaviour scale in Chapter 4.

##### *1.1.7.2.2.1 Chapter 3:*

*How prevalent are maladaptive behaviours in ASD across age and IQ? How are they related to the core features of ASD? And the other "associated" features of ASD?*

Chapter 3 will focus on the maladaptive behaviours measured by the DISCO. Although, research has identified that individuals with ASD frequently present with maladaptive behaviours the terminology and tools used to measure these behaviours are inconsistent and hardly any work controls for the effects of age and IQ in the presentation of these behaviours. The analyses presented in this chapter show strong associations between maladaptive behaviours and individual's social interaction and repetitive behaviours. However, the work of the thesis finds that both of these relationships are influenced by the presence of sensory behaviours.

##### *1.1.7.2.2.2 Chapter 4:*

*How can we measure sensory behaviours in ASD? How are sensory behaviours related to the core and associated features of ASD?*

The first study in this chapter examines the association between the core and associated features of ASD with the sensory behaviours as measured by the DISCO. Replicating the previous chapter, the social and repetitive behaviours had strong associations with the sensory behaviours. In Study 2 of this chapter, the sensory items from the DISCO are adapted into a self-report questionnaire (the Sensory Preferences Questionnaire, SPQ) and tested in a sample of adults with

high functioning ASD or typical development. This was done for two reasons, firstly to assess the external validity of the DISCO sensory items; the SPQ showed good correlations with total scores on questionnaires already used in the literature. Also, this addressed a gap in the literature on sensory behaviours in adults and using self-report questionnaires. It was found that 94% of high-functioning adults with ASD reported at least one marked sensory behaviour and had significantly higher SPQ scores than IQ matched typically developing individuals.

#### *1.1.7.2.3 Part 3. Measuring Autism Spectrum Disorder: Diagnostic Criteria*

Part 3 focuses on the diagnostic criteria for autism spectrum disorder. A set of empirical studies explore the measurement characteristics of two different operational definitions of Autism Spectrum Disorder- those of DSM-5 (APA, 2012/2013) and Wing & Gould (Wing, 2996; Leekam et al., 2002). The first aim of Part 3 is methodological and the goal is to assess how well the DISCO can measure DSM-5 ASD. In addition, I reflect on the adequacy of the DSM-5 criteria for the diagnosis of Autism Spectrum Disorder, in the light of the earlier debate about its lack of sensitivity and consider how comparisons between DSM-5 and Wing & Gould descriptions and their measurement may help to move forward in our understanding of the autism spectrum.

##### *1.1.7.2.3.1 Chapter 5:*

*Can the DISCO measure DSM-5 ASD? What impact will the changes to DSM-5 have on an individual with a clinical diagnosis according to DSM-IV?*

This chapter focuses on the design and validation of a DSM-5 ASD algorithm for the DISCO. The overall purpose of Chapter 5 was to examine the question of whether individuals who have previously been given a diagnosis of Autism or Asperger Syndrome would be captured when the DSM-5 criteria are run using the new DISCO DSM-5 algorithm. The DISCO is suitable to use to test the DSM-5 ASD criteria on as it was designed to measure the spectrum and it has a large range of behaviours that the DSM-5 criteria can be mapped on to. This is a strong advantage over other tools that were designed around the DSM-IV criteria. Particular attention is also given to the way in which behaviours are mapped on to the criteria as this is inconsistent across instruments being used to measure DSM-5. I designed several alternative new diagnostic algorithms for DSM-5 criteria (APA, 2012) and tested them using DISCO data in order to establish an acceptable algorithm that optimised sensitivity and specificity. It is proposed that the range of behaviours are useful as the DISCO DSM-5 ASD algorithm performs well at identifying individuals with an clinical diagnosis according to DSM-IV and did not identify many individuals in the clinical comparison groups.

#### *1.1.7.2.3.2 Chapter 6:*

*How similar is the DSM-5 conceptualisation of a spectrum to Wing and Gould's original description? How can we measure Wing and Gould's ASD? Does Wing and Gould's ASD capture the categorical diagnoses of DSM-IV?*

Chapter 6 explored another measure of the autism spectrum in the DISCO. Wing and Gould conceptualised autism as a spectrum condition before the introduction of autism to the international classification system and therefore based the design of the DISCO on this conceptualisation. The overall purpose of Chapter 6 was to evaluate and compare the measurement of DSM-5 and Wing & Gould descriptions of ASD using the DISCO. I investigate in detail the psychometric properties of the existing DISCO algorithm for Wing and Gould's ASD. The Wing and Gould ASD algorithm performs well in comparison to clinical diagnosis, other DISCO algorithms and across age and ability level. The good overlap with DSM-5 indicated that the change to a spectrum in the diagnostic criteria is similar to spectrum approach already proposed in the literature. Analysis at the item level looks at why there may be such a strong overlap between DSM-5 and Wing and Gould's ASD, one Wing and Gould item in particular "quality of social interaction" performs extremely well diagnostically and is further explored in Chapter 7.

#### *1.1.7.2.3.3 Chapter 7:*

*What can individuals' quality of social interaction inform us about that individuals' core autism and associated symptoms?*

Chapter 7 is dedicated to analysing a single element within the Wing and Gould algorithm, Wing & Gould's Quality of Social Interaction. This item is rated by the interviewer at the end of the DISCO using information gathered across the whole interview, it is made up of numerous codes that can be used to assign individuals into the binary distinction of "typical" or "impaired" quality of social interaction as well as into Wing's social sub-types of "aloof and indifferent," "passive" or "active but odd". Exploratory analyses in this chapter highlight that the information gathered from the summary item, quality of social interaction, is significantly predicted by the information that the interviewer collects throughout the interview. This judgement of quality made by the interviewer is extremely powerful and the same judgement of other areas of impairments could also be important to consider or to include in such diagnostic assessments.

#### *1.1.7.2.4 Part 4:*

##### *1.1.7.2.4.1 Chapter 8:*

The final chapter will discuss the main findings from across the thesis. The aim will be to review which behaviours are found in the autism spectrum and how to define whether individuals should meet criteria for a spectrum diagnosis according to DSM-5 or Wing and Gould's conceptualisation of the spectrum. A model used to conceptualise ASD, using the results from the current thesis is proposed and highlights the key areas for future research on the autism spectrum. The DISCO will be reviewed for its use in research on both the associated features of ASD as well as measuring the spectrum. Improvements to the tool are suggested in order for the research with the DISCO to be applicable to the wider literature. Finally, the key contributions that this thesis has made to the literature will be reviewed.

# 2 Measuring Autism Spectrum Disorder and using the DISCO for research

The aim of this chapter is to provide a more detailed description of the contents of the DISCO. All the research in this thesis is confined to the DISCO. Full descriptions of other diagnostic interview methods and their comparisons with the DISCO are given in Appendix 1. The focus on this description will be on the items and codes that are investigated in the empirical chapters of the thesis. This chapter will also present three secondary datasets that were collected using the DISCO and are used in the thesis to further explore the reliability of measurement of ASD behaviours using the DISCO as well as answering empirical questions with the DISCO data. Sample 1 provides a group of children with high and low functioning ASD as well as low and high functioning clinical and typical comparison groups. This sample allows the specificity of items or sub-sets of behaviours in individuals with ASD compared to high and low functioning comparison children. Sample 2 is an ideal sample to use to measure the spectrum in this thesis as it consists of individuals with ASD who vary across age and ability level, which allow exploration of patterns of behaviour in ASD across a wide demographic. The addition of Sample 3, which is very similar to the composition of Sample 1, provides an independent validation of the results found in Sample 1. The secondary datasets have all been published before and full demographic and recruitment processes are reported in these publications but summarised here in order to provide information on how these datasets are useful for use in this thesis. Primary data collection was conducted for the analyses presented in Part 2, Chapter 4 and the recruitment process and sample characteristics are presented for that primary dataset separately in that chapter.

## 2.1 The DISCO

The Diagnostic Interview for Social and Communication Disorders (DISCO) was designed for use in clinical practice to assist the clinician to collect information about an individual's broad development and behaviour (Wing, 2003; Wing et al., 2002). It is a semi-structured interview which facilitates clinicians and researchers in the identification of an individual's pattern of diagnostic symptoms, developmental skills, associated capabilities and difficulties. The main attributes of the DISCO as a clinical tool are that it can be used to measure behaviours of individuals of any age; it captures information about an individual's current developmental level as well as any developmental delays; and also covers a broad range of items not only for diagnostically relevant behaviours but also additional behaviours seen in clinical practice such as sensory, emotion, gross and fine motor and maladaptive behaviours. The range of skills permitted by the DISCO allows it to

be used for assessment of needs and can therefore be used for recommendations regarding care, education, occupation and interventions (Leekam, 2013; Leekam, Libby, Wing, Gould, & Taylor, 2002; Wing et al., 2002).

The DISCO is also appropriate for use in research. In addition to the broad coverage of behaviours that allow for a range of research questions to be addressed, diagnostic algorithms have been designed for both of the international classification systems criteria for Pervasive Developmental Disorders and their sub-groups (DSM-III-R, DSM-IV & DSM-IV-TR, American Psychological Association, 1987; 1994, 2000; ICD-10, World Health Organisation, 1993) as well as diagnostic categories proposed in the literature. These include Kanner's criteria for Early Infantile Autism (Kanner & Eisenberg, 1956), Wing and Gould (1979) criteria for Autism Spectrum Disorder and Gillberg's criteria for Asperger Syndrome (Ehlers & Gillberg, 1993; Wing, 1981).

### 2.1.1 History of the DISCO

The DISCO was developed from the Handicaps, Behaviour and Skills (HBS) Schedule (Wing & Gould, 1978), which in turn was created from the Childhood Behaviour Schedule (Wing, 1969; Wing & Wing, 1971). This was designed in order to collect brief information systematically from parents about children's social, language, imaginative, sensory and motor impairments and stereotyped behaviours. The HBS also included items on developmental skills and was designed for use in an epidemiological study of ASD (Wing & Gould, 1979). The HBS was then expanded further to include wider measures of past behaviour from infancy onwards to form the DISCO, which can be used to collect information about individuals of any age and level of ability. The reliability studies published in 2002 used the first version of the DISCO (DISCO 9; Wing et al., 2002; Leekam et al., 2002). Since then the DISCO has had two revisions and the current schedule and algorithms are DISCO 11 (see Maljaars et al., 2011 for DISCO 11 ICD-10 algorithm). The coverage of behaviours and skills has remained consistent across these versions and overall the changes are minimal between DISCO 9 and 11. The sections below about the content and scoring of the DISCO are, therefore, relevant to all DISCO versions.

### 2.1.2 Content of the DISCO

The DISCO measures a very large range of behaviours across eight parts as shown in Table 2-1.

**Table 2-1: Table showing the seven sections from the DISCO and the types of DISCO items used in each section**

| Section | Title of section | Types of items* |
|---|---|---|
| 1 | Family Medical Background and Identifying Information | Record of factual information |
| 2 | Infancy | Untypical behaviour |
| 3 | Developmental Skills | Current Level<br>Developmental Stages<br>Untypical behaviour |
| 4 | Repetitive, stereotyped activities | Untypical behaviour |
| 5 | Emotions | Untypical behaviour |
| 6 | Maladaptive behaviour | Untypical behaviour |
| 7 | Interviewers' Judgement of Quality | Quality of behaviour |
| 8 | Psychiatric Disorders and Forensic Problems | Untypical behaviours |

** The types of items are described below in section 2.1.4.*

The first part "family medical background and identifying information" consists of five specific items and a list of suggestions for information on the family and medical background such as problems in pregnancy, visual or hearing impairments and the structure of the family. The information from this section can be used to rule out or identify causes for the behaviours recorded throughout the interview by confirming medical abnormalities and family history. Part 2 focuses on behaviour in infancy only. Two medical questions of foetal abnormality and microcephaly are useful for diagnosing Rett's syndrome, 28 items are then asked to collect information on behaviour in infancy such as feeding, excessive crying, reciprocation in baby games and oversensitivity to noise.

Part 3 of the DISCO "developmental skills" is the largest part of the manual and collects information on an individual's current level, development, and untypical behaviours across a range of developmental skills: gross motor skills; self-care (toileting, feeding, dressing, hygiene); domestic; independence; communication (receptive, expressive and non-verbal); social interaction (with adults and age peers); social play or leisure activities; imagination; visual-manual and spatial skills; pictures, reading and writing; and cognitive skills and achievements. The three types of information collected (current level, development and untypical behaviours) are described separately in the section "scoring the DISCO" below. This section is particularly important for a diagnosis according to DSM or ICD as it covers impairments in social interaction and communication; in addition, it captures information on an individual's imaginative activities, which is an important part of Wing and Gould's triad of impairments.

The fourth part "repetitive, stereotyped activities" covers a range of repetitive, restricted and sensory behaviours that were previously seen in clinical practice by the authors of the DISCO. Again these items are particularly relevant for making a diagnosis according to the international classification systems which require the presence of restricted or repetitive behaviours or routines.

This part of the DISCO has: 12 stereotyped movements and vocalisation items; 15 items on responses to proximal sensory stimuli; four items on responses to auditory stimuli; five for responses to visual stimuli; 19 items capturing routines and resistance to change; and overall pattern of activities is captured by eight items.

Part 5, Part 6 and Part 8 include additional behavioural items often present in ASD and relevant to other related neurodevelopmental disorders and associated conditions. Part 5 contains nine items measuring emotions such as lack of emotional expressions, laughing for no reason and anxiety. Part 6 is entitled "maladaptive behaviour" and measures behaviours affecting other people such as wandering, personal modesty and difficult or objectionable personal habits (25 items) as well as five items on sleep disturbances for example, difficulty in falling asleep and night terrors. Part 8 measures psychiatric problems and forensic problems but is rated only if these behaviours are relevant in light of the age of the individual (adolescent or adult), and type and severity of their behaviours. The aim of this section is to obtain enough information to decide if an individual may require further investigation for a diagnosis of another psychiatric condition such as schizophrenia, eating disorders or personality disorders. Nine items measure catatonic features, six measure sexual problems, there are 16 items on psychiatric conditions and six items measuring forensic problems. These specific features (Part 8) are not included in the research reported in this thesis.

Finally, Part 7 is unique. This is the one section where interviewers are required to make judgements about the information that they have gathered over the course of the interview, Part 7 focuses on the *quality* of the behaviours and interviewers are asked to consider all the available evidence collected from the DISCO to code an individual's quality of social interaction and in addition in DISCO 11 the individual's quality of social communication; quality of imagination and quality of pattern of activities.

In clinical practice, the use of the DISCO can be adapted. It is suggested that, if possible, the interview should be conducted with one or both parents or with caregivers who have known the individual since infancy and that observation; past reports and information gained from additional informants such as teachers should be used to supplement the DISCO. However, this ideal scenario is not always possible. For adults, in particular there may not be a caregiver who has known the individual since birth or the interview may have to be conducted with the individual themselves and in this case information about past behaviour and developmental delays is omitted and only the current clinical manifestation is measured by the DISCO. Furthermore, data about family history in Part 1 may not be required if the case notes already contain this information or Part 8 (psychiatric conditions) may be left out in children (Leekam, 2013).

### 2.1.3 Using the DISCO interview:

The DISCO is a semi-structured investigator based interview schedule. This means that it is the responsibility of the interviewer to gain enough information from the informant to reliably make a judgment and assign a code to the DISCO item in question. In the clinical use of the DISCO, information from outside of the interview with the parent or caregiver should be used to both help assign codes to relevant items, for example, observing whether the individual uses descriptive gestures or engages in appropriate eye contact when interacting with the clinician and to guide the interviewer through the interview at the appropriate level i.e. not asking about long winded and pedantic language if they know the individual has no speech. It has been suggested that supplementation of clinical information from direct observation of the individual, case notes and information from other informants such as teachers or carers not only improves the data recorded in the DISCO but also in aiding service provision and interventions for the individual (Wing et al., 2002). More recently, the National Institute for Health and Care Excellence (NICE) guidelines for children specify that every assessment should include "assessment (through interaction with and observation of the child or young person) of social and communication skills and behaviours" (NICE, 2011, p. 8)

For the purpose of research, however, it is not always possible for the interviewer to meet the individual whose parent or caregiver they are interviewing. If the researcher is meant to be blind to the diagnostic status of the child, background information should not be available and the child should not be seen but this will require the interviewer to do more probing or ask for more examples from the parent in order to code the DISCO item appropriately. The original research on the reliability and validity of the DISCO was carried out with interviewers blind to the child's diagnosis (Leekam et al., 2002; Maljaars et al., 2009; Wing et al., 2002).

All registered users of the DISCO have completed the two session (5 day) training course conducted by the authors of the DISCO. The interview takes around 2-4 hours to complete by an experienced interviewer.

### 2.1.4 Scoring of the DISCO:

The DISCO contains three types of interview items across a range of skills and behaviours. The table (Table 2-1) presented above demonstrated which sections these types of items are used in. Section 3, is the largest section of the DISCO and measures individuals' current level of development, whether there was a delay in requiring these skills (developmental stages questions) and whether the behaviour is atypical (untypical questions) for each skill e.g. social interaction with peers.

### *2.1.4.1 Current level*

The "current level" items are used to capture the individual's highest level of achievement for that current skill (see the example below). The code for this item may therefore be dependent on the individual's developmental level but the aim is to capture what level of the specified skill the individual has achieved. These items were included in the DISCO by the authors based on their experience of using the HBS (Wing and Gould, 1979). During these interviews the authors reported a clear discrepancy between the current level of development i.e. what the individual is capable of doing such as dressing themselves and how these skills are used in everyday life i.e. whether the child does dress themselves or prefers their caregivers to do it. In order to capture these discrepancies a current level of development is essential to measure (Wing et al., 2002).

The codes for these items are arranged in chronological order according to the average age that the behaviour is expected to be achieved, up to the complete achievement of the specified skill. These recognisable steps in development are assigned a code in numerical order for computer entry. The age that each skill is typically achieved in normal development is next to each code to guide the interviewer. The typical developmental levels (age expected to achieve skill) were attained from standard measures of development available at the time of DISCO design; the majority of information was taken from the Vineland Adaptive Behaviour Scales (Sparrow, Cicchetti, & Balla, 1989) and further supplementary publications (Cooper, Moodley, & Reynell, 1978; Egan, Illingworth, & MacKeith, 1969; Griffiths, 1954; Sheridan, 1973, 1977).

EXAMPLE "Current level" DISCO question

Asking questions – level of language used
*Does A ask any questions?*

| | | |
|---|---|---|
| | 0 | Does not ask questions |
| 2 years | 1 | Asks names of objects and people |
| 3 years | 2 | Asks what where and who |
| 4 years | 3 | Asks why, when and how and meaning of words |
| | -8 | Not enough language |

### *2.1.4.2 Developmental stages*

The second type of DISCO interview items are the "developmental stages" items, which track how these behaviours that have been rated for current level develop and whether there was a delay in the individual achieving specific developmental milestones. For specific skills (walking, toilet training, saying meaningful words and saying meaningful phrases) the actual ages of achieving in months were coded. The majority of developmental items, however, are not concerned with the specific age of achievement but whether the individual was delayed in achieving the skill. These

items ask, for a particular skill, “Was A slow in achieving the following skills? Was A so slow that is caused concern?” These types of items follow one of two coding schemes, coding whether there was a delay, for example:

EXAMPLE 1 “developmental stages” DISCO question

Indicating object or person to share interest (Joint referencing)
*Was A slow in achieving this skill? Was A so slow that is caused concern?*

| | |
|---|---|
| 0 | Marked delay |
| 1 | Some delay |
| 2 | No problem |
| -8 | Too young or physically disabled |
| -9 | Not known |

The other format is to ask whether the specific skill was achieved by a certain age, for example:

EXAMPLE 2 “developmental stages” DISCO question

Asking questions: what, where, who?

| | |
|---|---|
| 0 | Not achieved by three years |
| 2 | Achieved by three years |
| -8 | Too young or physically disabled |
| -9 | Not known |

In both cases the two or three point scales are assigned a numerical code, as shown above (0, 1, or 2) for computer entry. In some cases a particular DISCO item may not be relevant to the individual in question for example, if the child was younger than the typical ages for achieving tying of their shoe laces or using money then a rating of delay in development would not be applicable. Another possibility is that information about the specific behaviour could not be obtained, for example if the parent or caregiver cannot remember the individual’s behaviour in infancy or the time that skills were achieved in early childhood. As can be seen above numerical codes were assigned if the item was not applicable for example “too young or physically disabled” or “not known.”

### *2.1.4.3 Untypical behaviours*

The final type of DISCO items, which are referred to as “untypical behaviours” make up the majority of the DISCO questions. Some of the untypical behaviour items follow on from the current level and developmental stages items and are relevant to that particular behavioural domain i.e. the

current level questions allow the interviewer to first establish if the individual has expressive language and then goes on to ask about language atypicalities, as shown in example 1 below.

EXAMPLE 1 "untypical behaviour" DISCO question

Long winded, pedantic speech
*Is A formal and long winded in speech, does not use colloquial expressions, uses long phrases where short ones would do?*

| | |
|---|---|
| 0 | Marked, frequent |
| 1 | Minor, occasional |
| 2 | No problem |
| -8 | Not enough speech to rate |
| -9 | Not known |

Untypical behaviour items that are not directly related to a specific developmental domain ask about behaviours that individuals with ASD typically present with or behaviours that were observed by the DISCO authors in clinical practice, see example 2 below.

EXAMPLE 2 "untypical behaviour" DISCO question

Clinging to home or familiar places.
*Does A intensely dislike leaving home? (e.g. refuses to go on holiday; will go on holiday only to certain places. The attachment is to the house rather than the inhabitants).*

| | |
|---|---|
| 0 | Marked |
| 1 | Minor |
| 2 | No problem |
| -8 | Too young or physically disabled |
| -9 | Not known |

### *2.1.4.4 Ever and Current*

Each "untypical behaviour" item can be coded for both "ever" and "current". The "ever" code is used when a behaviour is present at some stage in the individual's life, whether or not it is present at the time of rating. The ever period refers to the phase in the life of the individuals when the relevant behaviour was present in its most marked form for example, if the individual used to engage in rocking movements with their body when young but do not do so currently. If an individual scored an Ever=0 and Current=2, then at some time in the past, the individual had that behaviour in a "marked" form but it is no longer present. The "Current" code is used to rate the current manifestation of a relevant behaviour. For example, an adolescent referred for diagnosis may be showing behaviours reflecting high levels of anxiety that were not present when the individual was a child.

This scoring system means that if an untypical behaviour is occurring currently then it must also be rated for the ever code; it is not possible to score a more severe code for the current code than the ever, because if a behaviour is being displayed currently then it has occurred at any point in the individual's life. Thus, it is possible to indicate a changed from marked to minor or absent, but a recent onset of untypical behaviour cannot be indicated.

The untypical behaviours are rated (for both ever and current) on a three point severity scale. A behaviour is rated as "marked" if it occurs daily, when no strategy is in action, or whenever the opportunity arises. The following description from the DISCO manual (Wing et al., 2002) best describes the use of the "marked" code:

> "**Marked (0):** For behaviours that can occur at any time (echolalia, arm flapping, self-spinning) the rating of 0 should be used for any that can be observed every or nearly every day, when the individual is not being involved in some activity that is incompatible with the behaviour.
>
> OR For behaviours that can be diminished or prevented by the carer's strategies (for example, self-injury prevented by protective clothing) the rating of 0 should be used if the strategy has to always be in use.
>
> OR for behaviours that require specific opportunities for their occurrence (for example, running away when walking out with parents, inappropriate reactions to visitors) the rating of 0 should be used if the behaviour occurs always or nearly always when the opportunity arises.
>
> OR for behaviours that occur less frequently but are severe when they do occur (harmful physical aggression, intense long lasting tantrums) rate 0 if the carer's and or others around the individual have to constantly vigilant, or feel uneasy and anxious in case the behaviour occurs, or if the individual's daily life has to be limited because of the effect of the behaviour when it does occur."

The "minor" code can be used when these behaviours are less frequent or severe and the final rating of "no problem" is used when the individual does not display signs of the measured behaviour. Like the developmental stages DISCO items, the untypical behaviour items also have codes for "not known" or not applicable. For example, if the individual did not have any speech then the untypical items on use of language such as "long winded or pedantic speech", would not be able to be rated. Furthermore, if a child was too young to understand specific social rules such as not making embarrassing remarks in public this item could be rated as not applicable. A three-point numerical code assigns individuals to the "marked", "minor" or "no problem" ratings.

### 2.1.5 Reliability and Validity of the DISCO:

Inter-rater reliability scores have been conducted for over 300 items from the DISCO and the majority of items had high inter-rater reliability; kappa coefficients or intra-class correlations were above .75 in 80% of items (Wing et al., 2002). The DISCO also enables algorithm diagnoses according to ICD and DSM. The original ICD-10 Childhood Autism algorithm was based on 88 DISCO-9 items and a set of rules specifying how these items convert into diagnostic outcome (Leekam et al., 2002). This algorithm was originally tested along with the Wing and Gould ASD algorithm in a sample of 36 children with autism, 31 individuals with a non-ASD clinical condition and 15 typically developing children. The items in both algorithms had good inter-rater reliability and the two algorithms were significantly related to individual's clinical diagnoses (Leekam et al., 2002).

The ICD-10 algorithm has also been shown to have good criterion and convergent validity in a study using the Dutch translation of the DISCO-11 (van Berckelaer-Onnes, Noens, & Dijkxhoorn, 2008). Again individuals with autism (n=52), non-ASD intellectual disability (n=26) and typically developing children (n=37) were compared. The ICD-10 DISCO algorithm had good sensitivity (.96) and specificity (.79), although the comparison to clinical diagnosis was better in individuals with average intelligence or a mild intellectual disability than those with moderate to severe intellectual disabilities. The DISCO also showed strong agreement with scores on the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000, k=.69), a structured observation task designed to be used in conjunction with a development history interview (the ADI-R) to also make a diagnosis. However, lower agreement was found with the Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2003, pp., k=.49), which is parent report questionnaire on an individual's social behaviours (Maljaars, Noens, Scholte, & van Berckelaer-Onnes, 2012). The DISCO ICD-10 algorithm was also tested against the ADI-R and showed excellent agreement with it and clinical diagnosis and again the inter-rater reliability of these items were excellent for 70% of the items (Nygren et al., 2009). The DISCO, however, has not been tested in very young children and therefore it is unknown whether additional cut-offs or algorithms may be required like for the ADI-R (see Appendix 2).

### 2.1.6 DISCO algorithm and non-algorithm items:

The DISCO includes a wide range of items including 147 that are used in the diagnostic algorithms (across all algorithms) and over 200 that are used to collect a picture of the clinical pattern and needs of an individual. The DISCO contains algorithms for both the international classification systems (ICD-10/DSM-IV-TR) as well as other diagnostic categories described in the literature (Gillberg's Asperger Syndrome criteria, Kanner and Eisenberg's Early Infantile Autism and

Wing and Gould's criteria for Autism Spectrum Disorder). The algorithm items were selected from across all three types (current level, developmental skills and untypical behaviours) of DISCO items.

The DISCO 9 algorithms for the ICD-10 Childhood Autism (Leekam et al., 2002), Gillberg's Asperger criteria (Leekam et al., 2000) and Wing and Gould's ASD criteria (Leekam et al., 2002) and the DISCO 11 ICD-10 Childhood Autism algorithm (Maljaars et al., 2009) have already been published. However, there are minor changes in the algorithms from DISCO 9 to DISCO 11 in response to research findings from DISCO-9. The secondary data described below have been collected with both DISCO 9 (Sample 1 and 2) and DISCO 11 (Sample 3). The decision was made to use DISCO 11 algorithm items in the current thesis for data collected with either version. A full summary of the changes between the DISCO 9 and DISCO 11 algorithms and the diagnostic accuracy of these algorithms in the DISCO 9 samples are detailed in Appendix 2 but no changes significantly reduced the diagnostic accuracy of any of the algorithms.

In summary, changes that were made between DISCO-9 and DISCO-11 were as follows: five items were removed for DISCO-11 and three items were added. Of the added items, one was already measured by DISCO 9 (other repetitive routines, this item was a non-algorithm item before), one item (copying imaginative activities) was covered within the responses to other DISCO 9 items (imaginative activities and acting out role) and only one item was new and could not be mapped back to DISCO-9 (fascination with violence). Part 2 of the thesis uses the items from the ICD-10 Childhood Autism algorithm as measures of the core features of ASD and in Part 3, the algorithms are tested in more detail.

There was one exception to the good overlap between DISCO 9 and DISCO 11 algorithms. Although, the content covered by both versions of the algorithm were identical, there was a substantial change to the imagination items in the Wing and Gould's ASD algorithm between DISCO 9 and 11. However, both the DISCO 9 (Sample 1 and 2) and the DISCO 11 (Sample 3) datasets contained the items required for the DISCO 9 algorithm and therefore for this one algorithm the DISCO 9 version was retained instead to ensure compatibility across samples.

The non-algorithm items cover impairments or skills not described in the diagnostic criteria but seen in individuals with ASD or relevant to an individual's developmental history or current profile of the individual's strengths and difficulties. Of these additional or non-algorithm items, some DISCO items relate to factual information such as family history, ability level or special skills whereas other behaviours cover "associated" behaviours such as sensory processing, motor behaviours, maladaptive behaviours and daily living skills. Again, these additional DISCO items cover all three DISCO types of items and across all sections of the DISCO. These two sets of items are both used across the thesis. Part 2 will examine how well the DISCO provides a measure of these

associated behaviours as well as exploring their role in ASD in relation to the core features. Items are recoded for analyses differently according to whether it is an algorithm or non-algorithm item and therefore coding of data is discussed in the relevant chapters.

## 2.2 Data used in the thesis:

The majority of the statistical analyses presented in the thesis are from two samples of individuals for whom full DISCO interviews were conducted with a parent or caregiver. Sample 1 contains children with a clinical diagnosis of ASD as well as clinical and typically developing comparison children, this sample is used throughout the thesis in order to look for differences between individuals with ASD and individuals with non-ASD diagnoses as well as to test the accuracy of diagnostic algorithms at identifying the ASD group and excluding the non-ASD group. Sample 2 consists of 200 individuals who were referred to the Lorna Wing Centre for Autism, a tertiary referral service. Throughout this thesis this dataset is used to compare across age and IQ as the range of these variables is large in this sample. Both Sample 1 (Leekam et al., 2002; Wing et al., 2002; Leekam et al., 2007) and Sample 2 (Leekam et al., 2000; Leekam et al., 2007) have been used in previous research with the DISCO. Ethics for both datasets to be analysed at Cardiff University were obtained from the School of Psychology Ethics Committee. In addition, a third sample (Sample 3; Maljaars et al., 2011) was used to validate the newly designed algorithms in Part 3. This was essential as the design of the algorithms was made using Sample 1 and therefore testing on the design sample is limited, an independent dataset improves the reliability and validity of the findings in Sample 1. These three samples will be presented separately below and referred to as Samples 1, 2 and 3 throughout the thesis. Any additional recruitment and participants will be detailed in the relevant chapters.

### *2.2.1 Sample 1:*

#### *2.2.1.1 Participants:*

Participants were the parents of 82 children aged 35-140 months at the time of interview (50 school age children and 32 pre-school) residing in London, or the South East of England. Children's clinical diagnoses at the time of recruitment assigned them to one of four groups, which originally consisted of 15 typically developing children, 19 children with an intellectual disability, 15 children with a language disorders and 33 children with ASD, none of whom were diagnosed by a member of the research team. The recruitment of the clinical comparison children (intellectual disability or language disorder) was made according to their current diagnosis at the time of interview, schools and clinics were asked to exclude children with a diagnosis of Autistic

Disorder/Childhood Autism, sensory impairments or Down's Syndrome. However, no systematic clinical autism assessment was undertaken as this was not possible through the clinics used for recruitment. At telephone follow up two years after data collection, the parents of 22 of the 33 children with ASD and all children with an intellectual disability or language disorder were contacted and their original diagnoses were checked. None of the children with ASD had changed diagnosis but two of the children with intellectual disabilities and one child with language disorder were re-diagnosed with ASD. The updated diagnoses were used for the research reported in Wing et al (2002) and Leekam et al (2002) and also used in the work of this thesis. The characteristics of these groups are presented in Table 2-2.

#### *2.2.1.1.1 ASD group:*

Diagnoses were made according to either the ADI (or ADI-R) at the Maudsley or Guy's hospitals in London (22 children) or the ICD-10 criteria used by local diagnostic centres in Kent (11 children). Four of the 33 original children with ASD had a diagnosis of Asperger Syndrome. All children had received a full assessment and none had any additional diagnoses. Two children originally recruited with an Intellectual Disability and one child with a language impairment were re-diagnosed as having an ASD at two year follow up (2 male; 1 female) and were therefore included in the ASD group for the analyses conducted in this thesis. The 36 children in the ASD group were separated into high (n=18) and low functioning groups (n=18) on the basis of their IQ information at the time of recruitment.

#### *2.2.1.1.2 Intellectual Disability group:*

The original group of 19 children (11 male) who had IQ levels in the moderate to severe range (IQ <70) were recruited through local voluntary organisations and specialist schools for intellectually disabled children. Again, professionals were asked to exclude individuals with a diagnosis of ASD, sensory impairments or Down's syndrome as research has shown that around 10% of individuals with Down's syndrome may also have an undiagnosed ASD (Howlin, Wing & Gould, 1995; Wing & Gould, 1979). Associated conditions were prevalent and consisted of: cerebral palsy (3); microcephaly, (1); calcification in the brain (1); maple syrup urine disease (1); mucopolysaccharide disease (1); attention deficit, hyperactivity disorder (2); eight had no specific aetiology. One child who had a cerebral haemorrhage in infancy was diagnosed as having an ASD at the two year follow up, as was one individual with an unknown chromosomal and metabolic disorder. Seventeen children (10 male) remained in the intellectually disabled group and these individuals were used as the low functioning clinical comparison group for all Sample 1 analyses reported in this thesis.

**Table 2-2: Table showing the chronological ages, non-verbal IQ and language abilities of the children in Sample 1**

| | ASD | | Clinical Comparisons | | TD controls |
|---|---|---|---|---|---|
| | Low IQ | High IQ | Intellectual Disability | Language Impairment | |
| N | 18 | 18 | 17 | 14 | 15 |
| Male:female ratio | 17:1 | 15:3 | 9:8 | 9:5 | 9:6 |
| Mean CA in months | 83.67 | 87.28 | 84.18 | 89.07 | 89.40 |
| Standard deviation | 32.465 | 30.263 | 33.416 | 29.865 | 30.661 |
| Range | 34-133 | 35-131 | 40-140 | 49-136 | 51-135 |
| **Non Verbal IQ** | | | | | |
| Leiter N | 9 | 18 | 9* | 13** | 15 |
| Mean | 62.78 | 100 | 59.56 | 92.23 | 108.6 |
| Sd | 12.80 | 21.53 | 22.51 | 16.84 | 15.71 |
| Bayley N | 9 | | 7 | | |
| Mean | 30.67 | | 34.14 | | |
| Sd | 14.79 | | 10.72 | | |
| **Non-verbal MA** | | | | | |
| Leiter N | 9 | 18 | 9* | 13** | 15 |
| Mean | 65.44 | 85.5 | 53.78 | 82.46 | 96.87 |
| Sd | 10.55 | 33.30 | 16.67 | 34.73 | 33.53 |
| Bayley N | 9 | | 7 | | |
| Mean | 16.11 | | 18.86 | | |
| Sd | 4.91 | | 6.230 | | |
| **Language age (months) – expressive*** ** | | | | | |
| MacArthur N | 8 | 1 | 2 | | |
| Mean | 11.25 | 12 | 10 | | |
| SD | 2.38 | | 2.83 | | |
| Reynell N | 8 | 9 | 13 | 8 | 5 |
| Mean | 28.88 | 44.44 | 38.08 | 54.88 | 61 |
| SD | 7.90 | 20.18 | 13.47 | 18.29 | 12.83 |
| WOLD | 1 | 8 | 1 | 6 | 11 |
| Mean | 75 | 100.5 | 87 | 108 | 119.1 |
| SD | | 36.92 | | 21.88 | 30.07 |
| **Language age (months) – comprehension*** ** | | | | | |
| MacArthur N | 8 | 1 | 2 | | |
| Mean | 9.88 | 16 | 9 | | |
| SD | 3.48 | | 0.00 | | |
| Reynell N | 8 | 9 | 13 | 8 | 5 |
| Mean | 23.38 | 40.78 | 29.15 | 42.38 | 55.6 |
| SD | 9.58 | 18.65 | 16.52 | 21.72 | 18.84 |
| WOLD | 1 | 8 | 1 | 6 | 10 |
| Mean | 81 | 95.63 | 87 | 95 | 115.8 |
| SD | | 30.27 | | 18.65 | 28.22 |

* One child with cerebral palsy could not manipulate Leiter material

**One child with language disorder was not available for testing

***One child with LFA autism would not cooperate with the testing and one child with cerebral palsy could not be tested (intellectual disability group)

#### *2.2.1.1.3 Language Impairment group:*

Individuals with language impairments were recruited as a high functioning clinical comparison group. Fifteen children (10 male) without an autism diagnosis and without severe sensory impairments at time of recruitment were referred by speech and language therapists or recruited through schools for children with specific language disorders in Kent and Sussex. Five children had additional diagnoses of dyspraxia (2), dyspraxia and dyslexia (1), severe auditory memory loss (1) and Laudau-Kleffner syndrome (1). One of the individuals with dyspraxia was diagnosed at follow up with an ASD diagnosis and is therefore excluded from this group. Fourteen participants (9 male) remained in the final language disorder group.

#### *2.2.1.1.4 Typically developing group:*

Parents of 15 typically developing children aged 51-135 months old (9 male) who attended school or nursery in South-East England participated (IQ range 81-138).

### *2.2.1.2 Matching procedures:*

#### *2.2.1.2.1 Age*

Groups were matched on chronological age. When the original groups were recruited fifteen children in each of the five groups (high functioning ASD, low functioning ASD, language disorder and intellectual disability, typically developing) were individually matched for chronological age within six months of another individual in each of the remaining groups. This had left an additional 7 individuals (2 high functioning ASD, 1 low functioning ASD and 4 individuals with intellectual disability) unmatched. With the rearrangement of individuals at follow up, individual matching was disrupted. The LFA group was individually matched within six months of age with the intellectual comparison group for 15/18 individuals and the HFA group with the language impaired comparison group in 13/18 individuals. However, there is no significant difference in age between the ASD and non-ASD individuals ($F(56) = 1.28$, $p=.25$, n.s.) in the sample and specifically there were no significant differences between the high functioning ASD and either the language comparison group ($t(30) = -.17$, $p=.87$) or the typically developing group ($t(31) = -.20$, $p=.84$) and no difference between the lower functioning ASD individuals and those with an intellectual disability ($t(33) = -.05$, $p=.96$).

#### *2.2.1.2.2 Non-verbal IQ*

Matching of individuals for their ability level was complicated as the formal IQ tests were not conducted by researchers until after the DISCO interviews had been conducted to avoid bias during the interviews. The individuals with ASD were assigned according to IQ information available at the time of recruitment to high or low functioning ASD groups based on an IQ of above or below 70. Intellectually disabled individuals were recruited as a low functioning control group and the

language disorder group as a high functioning group. All but two individuals completed either the Leiter International Performance Scale (Leiter, 1979) or the Bayley Scales of Infant Development (Second Edition, Bayley, 1993), one individual in the language impairment group could not be visited by the researchers and one individual in the clinical comparison group had cerebral palsy and could not manipulate the materials. The proposed groupings (above) are maintained for the analyses in the current thesis to allow comparability across previous published work, however, the formal IQ tests scores meant individuals assigned to high or low functioning ASD groups and in the high and low comparison groups scored above or below an IQ of 80 on formal tests.

Formal IQ tests confirmed the expected group differences, an ANOVA revealed a significant effect of diagnostic group on IQ ($F(58) = 2.15$, $p<.05$) across the whole sample but not mental age ($F(58) = 2.17$, $p=.22$). Importantly, follow up analyses revealed the LFA and ID group do not significantly differ (IQ, $T(33)=0.21$, $p=.84$, n.s.; MA, $T(33)=.59$, $p=.56$, n.s.), nor did the HFA and LI (IQ, $T(30)=1.60$, $p=.12$, n.s.; MA, $T(30)=0.72$, $p=.47$, n.s.) or TD groups (IQ, $T(31)=-1.29$, $p=.21$, n.s.; MA, $T(31)=-0.97$, $p=.34$, n.s.) but as expected IQ was significantly higher in the HFA than LFA group (IQ, $T(34)=-7.47$, $p<.001$; MA, $T(34)=-4.45$, $p<.001$) and in the LI than ID group (IQ, $T(29)=-3.90$, $p<.001$; MA, $T(29) = -3.38$, $p<.01$).

### *2.2.2 Design and procedure of Sample 1:*

In the original study (Leekam et al., 2002) the reliability and validity was tested by two researchers who conducted all of the parent interviews using the 9th version of the DISCO. Both researchers were trained for three months in the use of the DISCO by clinicians and DISCO authors (Judith Gould and Lorna Wing) specifically for the data collection. This involved coding of DISCO interviews and conducting at least four parent interviews which were recorded and rated by trainers. The codings of each researcher and trainers resulted in high levels of agreement (see Wing et al., 2002). In this research procedure the interviewers coded the DISCO only on information gained from the parent's interview. One researcher was blind to the diagnosis of all the children in the study and the other blind to all the school aged children (66% of the sample) although the diagnosis of three children was revealed by the parent during the course of the interview. Both researchers were present for all interviews; one researcher interviewed the parents and coded the parents' response and the second researcher only coded the parent's responses. This procedure was counter-balanced so each researcher conducted one half of the interviews each.

### 2.2.3 Sample 2:

#### 2.2.3.1 Participants:

Participants were assessed at the Lorna Wing Centre for Autism (formally, the Centre for Social and Communication Disorders/Elliot House), which is a specialist tertiary referral centre for Autism Spectrum Disorders. The DISCO is conducted as part of the assessment procedure for diagnosis at the centre and the DISCO interviews used in the current sample are those for which the DISCO was completed in full and coded for computer entry. The sample of 200 participants included (167 male; 33 female) ranging from 32 months to 38 years who all received a clinical diagnosis of an ASD according to J. Gould or L. Wing. The age and ability levels of the sample can be seen in Table 2-3. Throughout the current thesis, this dataset is commonly used to compare across age and IQ and has therefore been divided into three main age categories, child adolescent and adult.

**Table 2-3: Table showing chronological ages and non-verbal IQ ranges of children in Sample 2**

| | Children | | Adolescents | | Adults | |
|---|---|---|---|---|---|---|
| | Low IQ (<70) | High IQ (>70) | Low IQ (<70) | High IQ (>70) | Low IQ (<70) | High IQ (>70) |
| N | 44 | 75 | 14 | 20 | 12 | 35 |
| % male, female | 84, 16 | 87, 13 | 71, 29 | 85, 15 | 75, 25 | 83, 17 |
| Mean CA in months | 81.57 | 87.93 | 176.57 | 176.55 | 296.97 | 299.26 |
| Standard deviation | 33.216 | 28.858 | 17.704 | 17.160 | 71.939 | 66.447 |
| Range | 32-139 | 37-142 | 145-206 | 145-208 | 216-450 | 216-456 |
| IQ range | | | | | | |
| Profound 0-19 | 1 | 0 | 1 | 0 | 2 | 0 |
| Severe 20-34 | 4 | 0 | 3 | 0 | 4 | 0 |
| Moderate 35-49 | 20 | 0 | 10 | 0 | 1 | 0 |
| Mild 50-69 | 19 | 0 | 14 | 0 | 5 | 0 |
| Borderline 70-89 | 0 | 31 | 0 | 9 | 0 | 18 |
| Average 90-119 | 0 | 38 | 0 | 9 | 0 | 16 |
| Superior 120+ | 0 | 6 | 0 | 2 | 0 | 1 |
| Leiter | | | | | | |
| N | 12 | 28 | 3 | 0 | 3 | 1 |
| Mean | 55.9166 | 76.5 | 76 | / | 85 | 195 |
| Range | 30-88 | 30-186 | 65-90 | / | 70-95 | / |
| WISC-III-UK – verbal | | | | | | |
| N | 5 | 27 | 7 | 14 | 1 | |
| Mean | 57 | 96.888 | 61 | 97.857 | 46 | |
| Range | 48-70 | 65-153 | 55-72 | 77-120 | / | |
| WPSSI | | | | | | |
| N | 5 | 9 | | | | |
| Mean | 63 | 94.556 | | | | |
| Range | 54-73 | 71-116 | | | | |
| WAIS | | | | | | |
| N | | | | 2 | 2 | 32 |
| Mean | | | | 119 | 71.5 | 92.281 |
| Range | | | | 88-150 | 70-73 | 71-120 |

#### *2.2.3.1.1 Ages and ability levels:*

Table 2-3 presents participants' chronological ages, non-verbal IQ estimate and verbal IQ where available (also see Leekam et al., 2000; 2007). Formal intelligence tests were unable to be conducted for 47 low ability or uncooperative individuals. These individuals' behaviour was observed by the psychologist in structured and unstructured situations at the diagnostic centre and these assessments along with information from the developmental section of the DISCO were used to make an estimate of level of ability. Non-verbal IQ is therefore recorded as an IQ estimate to be comparable for all participants in Table 2-3. The age groups were divided into high and low ability according to this estimate if they had an IQ above or below 70. For the majority (n=153) formal IQ tests and language assessments were carried out, a range of different types of assessment were used across the sample due to the variety of age and ability levels. Intelligence was measured using the Wechsler Intelligence Scale for Children (WISC-III-UK), the Wechsler Preschool and Prima Scale of Intelligence (WPSSI) the Wechsler Adult Intelligence Scale (WASI), the Leiter International Performance Scale or Merill Palmer. These were selected according to age and current level of ability. Formal assessments of language were collected using information from the verbal scale of the Wechsler tests, the British Picture Vocabulary Scale or the Reynell Language Development scales. For those who had formal assessments, the IQ range scores for their results were combined with the IQ estimates for those who had informal assessments (described above) .

There was no significant difference in age between the low and high functioning participants across the whole sample using a Mann-Whitney test ($U(1) = 3985.5$, $z=-1.45$, $p=.15$, n.s.) and there is no significant difference between the age groups in the number of high and low functioning individuals (above or below non-verbal IQ of 70) using a chi-square test ($x^2(2)=2.63$, $p=.27$, n.s.).

### *2.2.3.2 Procedure:*

As described in Leekam et al (2000; 2007) the DISCO was conducted by one of the two clinicians who designed the DISCO and worked at the Lorna Wing Centre for Autism. In the period of time when the 200 DISCO interviews were collected (and all information completed and coded for computer entry) additional individuals were assessed at the centre but not included because there was either no parent or caregiver that could provide an early developmental history or the interview was conducted by another clinician. Individuals were seen in chronological order from the confirmation of a referral and assigned to an available clinician for that assessment date. There was no selection process of inclusion that would introduce bias to the clinical picture of individuals included compared to those excluded.

### 2.2.4 Sample 3:

Sample 3 was used as a validation sample only, to compare results of the algorithms proposed in Part 3. Permission to use the anonymised dataset was given from all professionals involved in the data collection. The full details of recruitment and participant characteristics can be found in Maljaars et al. (2011). All DISCO interviews were conducted using the Dutch translation of the 11th version of the DISCO (van Berckelaer-Onnes et al., 2008).

#### 2.2.4.1 Participants:

This validation sample comprised parents of 115 children with a chronological age of below 12 years old and developmental age below six years from the Netherlands who were recruited into one of three groups: 52 individuals with ASD, 26 individuals with an intellectual disability and 37 typically developing children. Originally, recruitment identified 129 individuals, however, 14 were excluded due to refusal to complete the DISCO or insufficient Dutch language to do so or the clinical diagnosis of the child was uncertain. Children were also excluded if severe sensory or motor problems were reported to avoid coding issues on the DISCO. Professionals at schools or day centres for children with special needs were asked to refer children between 2 and 12 years old with an official diagnosis of ASD or an intellectual disability. Typically developing children were recruited through four ordinary day centres. All schools and centres had been randomly selected from all those in three regions of the Netherlands (Zuid-Holland, Noord-Holland, and Utrecht).

##### 2.2.4.1.1 ASD group

Fifty two children (34-137 months; 43 male) had a clinical diagnosis of autism made by an independent clinician according to DSM-IV criteria for Pervasive Developmental Disorders. The diagnosis was not conducted by any member of the research team and the DISCO was not used for the original clinical diagnosis. Parents provided the diagnostic report to the researchers; the time between diagnosis and study recruitment was between one and seven years. Thirty five children had a co-morbid intellectual disability (15 mild; 12 moderate and 8 severe intellectual disability) and 17 were high ability (no or borderline intellectual disability).

##### 2.2.4.1.2 Intellectual Disability group

Diagnosis of an intellectual disability was again confirmed through clinical reports sent by parents. Some children presented with a known cause of intellectual disability such as Down's syndrome but some had unknown aetiology. None of the children in this group had a diagnosis of ASD. The 26 children (16 male) ranged in age from 48-134 months and in level of intellectual disability (11 mild; 12 moderate; and 3 severe).

#### *2.2.4.1.3 Typically developing group*

This group comprised 37 children (15 male; 22 female) aged 24-49 months. None of the children in this group had an intellectual disability.

### *2.2.4.2 Matching:*

Matching at the individual level was not conducted during the recruitment of individuals for inclusion in the sample or reported in Maljaars et al (2011). Subsequent analyses revealed that there was no significant difference in age between the children in the intellectual disability group and the ASD group ($t(76) = .346$, $p=.730$) but there was a significant difference in chronological age between the typically developing children and the children with ASD ($t(87) = -8.608$, $p<.001$). This is because the typically developing children were recruited to be much younger (mean 37.9 months) than the high functioning ASD group (mean 79.6 months) so that non-verbal mental ages would be comparable across groups (mean ASD=43; ID=38.2; TD=42.5: sd ASD=14.2; ID=12.5; TD=9.2).

### *2.2.4.3 Procedure:*

The interviews were conducted by one of seven pairs of interviewers at the participant's home. As in sample 1, the interviewers were blind to clinical diagnosis and the majority of the time did not see the child before or during the interview. All interviewers were trained on the administration of the DISCO for the purpose of the data collection.

## 2.3 Primary data collection

In addition to the secondary samples above, the work in Part 2, Chapter 4 on sensory behaviours measured in the DISCO, offered the opportunity for primary data collection. The DISCO sensory items under investigation were converted into self-report questions and a sample of high functioning adults with ASD were recruited to complete the questionnaire, along with an IQ matched comparisons group of typically developing adults. The full procedure is detailed in Chapter 4.

Further primary data was constrained for two reasons. Firstly, this PhD was the first conducted solely at the Wales Autism Research Centre and recruitment registers and local connections were not in place for recruitment at the beginning of the research programme. The recruitment for the sensory study conducted in Chapter 4 took over 4 years, this was relevant to both identifying individuals with ASD as well as appropriate age and IQ matched comparison individuals. Some of the restrictions to the recruitment of the sensory study were specific to the design. This was a study of adults and the Adult NHS Diagnostic Network was not set up and operational in Wales until 2013, making recruitment a challenge. Secondly, the shift in focus

throughout the PhD to the DSM-5 diagnostic criteria meant that resources were shifted; although the focus on DSM-5 is limited to one chapter, the work was conducted across an 18 month period. In addition, the secondary data described above provided ideal samples for the design and testing of new diagnostic algorithms, with a sample far larger than would have been possible to recruit during the time period of this thesis. This is especially the case as the research was conducted at a research centre rather than a clinic and the original study had taken 3 years to complete (Leekam et al., 2002). An additional secondary dataset (Sample 3) was used to ensure the design and testing of the sensitivity and specificity of the DISCO DSM-5 algorithm was not solely conducted on the same dataset.

## 2.4 Limitations

### *2.4.1 Limitations of the samples selected*

The great advantage of using secondary datasets in the current thesis is that they provide data on a number of participants that would have been impossible to recruit during the current thesis due to recruitment issues (as described above) and the DISCO requires extensive training and at least 3-4 hours to conduct[2]. However, the use of secondary dataset means that there was no control over how the data were collected and in turn this means that the samples described above have a range of limitations that need to be considered. These limitations will be reflected on throughout the thesis where relevant, and the effects of them minimised where possible.

Firstly, the recruitment of a sample of individuals into an ASD or non ASD groups can bias the results of group comparisons, for example, when testing the diagnostic algorithms in ASD and non-ASD groups. This limitation is true of the data used in the thesis. When the data were collected, individuals in Sample 1 were recruited to have ASD and professionals were asked to refer individuals for the clinical comparison groups who did not have or were not suspected of having social and communication deficits or ASD; a similar process was conducted for the recruitment of individuals in Sample 3. This is a limitation of many studies in the current literature because there is a selection bias that means that only those with "core" autism are selected. This is not representative of the general population or individuals who are referred for a diagnosis and is therefore not ideal for studies aiming to examine the spectrum of autism. The most reliable samples to use instead are individuals who have been recruited through the diagnostic process such that individuals referred for a diagnosis who go on to get a diagnosis of ASD can be compared with individuals who are referred but do not get an ASD diagnosis in order to assess the sensitivity and specificity in the real

[2] I am a fully trained and accredited user of the DISCO after attending a two part course (July & October 2011) with successful completion of pre-course preparation and DISCO interviews between course attendance.

world setting. However, in order to minimise the effect that this limitation may have, all group comparisons were conducted on ASD versus the clinical comparison groups rather than the TD group in order to identify differences that are unique to ASD rather than a more general developmental delay or neurodevelopmental characteristic.

A major limitation of Sample 2 was that not all individuals were tested using formal IQ tests, as they were too low functioning or un-cooperative. In order to include all individuals in the analyses the "IQ estimate" variable was used instead, for which the clinicians estimated an individual's IQ from additional information and assigned that individual a categorical code. This limited the type of analyses that could be run with the measure of IQ as well as losing information about the range of IQs across the sample. However, this IQ variable can be converted into "high" (IQ>70) or "low" (IQ<70) functioning groups and a binary variable can be entered into regression analyses when recoded as "0" and "1" (Field, 2009). This allows a measure of IQ to be controlled for and helps to assess if differences are likely to be found in high or low functioning groups specifically, which is how groups are usually divided in the literature.

Another limitation is that some of the data were collected with DISCO 9 and some with DISCO 11. However, analyses presented in Appendix 2 and outside of this thesis have found that these minimal differences do not affect the data significantly as for the majority of DISCO items, the same information is collected. However, the DISCO 9 algorithms have now been used frequently to research the psychometric properties of the DISCO and more research is needed with DISCO 11 to ensure the findings are up to date. The addition of Sample 3 as a validation dataset across the thesis alleviates the impact of this limitation to some extent.

The final limitation is that the authors that collected some of the data may be inherently biased in their data collection. For example, in Sample 2, the DISCO interviews were conducted by the DISCO authors, Lorna Wing and Judith Gould, therefore, the codings of items that are used in the algorithms are already known. In particular, the small amount of items for the Wing and Gould ASD algorithm may be approached differently by these clinicians as they know the importance that they have placed on them in an algorithm. This limitation could also be relevant, in a less severe way, to researchers collecting data because all professionals trained on the DISCO are trained in the conceptualisation of Wing and Gould's spectrum approach, however, in data collection for both Samples 1 and 3 researchers were blind to an individual's diagnosis. The conclusions drawn from the studies on Wing and Gould's measure of ASD are taken with caution; however, as this measure is specific to the DISCO further work would be required to have an independent measure of such behaviours (see Chapters 6/7).

### 2.4.2 Limitations on the research in the thesis

The same samples are used to run nearly all of the analyses. This limits the generalizability of these findings beyond the datasets used and more generally beyond data that has not been collected using the DISCO. To reduce the effect of this limitation, careful consideration of multiple comparisons are made in each chapter and Bonferonni corrections (dividing the alpha level of significance by the number of comparisons in each analysis) applied in all cases. Furthermore, where possible, effect sizes are also calculated and conclusions are not drawn about tests with small effect sizes in cases these are limited by power (e.g. sample size).

This thesis was also limited in the statistical analyses that could be applied. Firstly, the sample size was limited given the multiple data points for each individual across the DISCO items. Furthermore, the questions under investigation across the thesis and in particular in Part 2 meant that the research questions need to be treated as exploratory and with caution in interpretation. For this reason robust statistical tests were used (e.g. regression) but no strong hypothesis or theoretical lead statistical analyses were adopted. Path analysis, specifically and structural equation modelling more generally should only be conducted when there is a clear hypothesis, or small number of hypothesis within one model, to test. Such techniques should not be adopted at the exploratory stage of research (Everitt & Dunn, 1991). In addition, as mentioned in the sample limitations above, some of the dependent or covariate variables, gender and ability level (high or low) use binary scales of measurement and path analysis with nominal or ordinal measurement with a small number of categories (including dichotomies) is not reliable and cannot be conducted, as different methods cannot be combined in a single path analysis (Bryman & Cramer, 1994; Everitt & Dunn, 1991).

In conclusion, this is the first research to test the effect of DSM-5 ASD using the DISCO (Part 3). In addition, the analyses conducted in Part 2 are also exploratory in their search for relations between core and associated features and therefore the assumptions and deductions made from these analyses are restricted. Hopefully, however, they lay systematic and important groundwork for the replication of these findings in additional datasets collected both with the DISCO and other research tools.

## 2.5 Summary

Part 1 of this thesis introduced the concept of the autism spectrum. This thesis was conducted at a time of change in diagnostic criteria which reflect the introduction of “autism spectrum disorder” into the international classification systems and adopted across research. Part 3 addresses this change directly by designing and testing a new DISCO DSM-5 algorithm and

comparing this to the diagnostic outputs according to clinical diagnoses, previous classification systems as well as another measure of the autism spectrum. Part 2, however, reflects the triad of behaviours of social interaction, communication and repetitive behaviours as categorised in ICD-10 and DSM-IV-TR.

# PART 2: MEASURING AUTISM SPECTRUM DISORDER: ASSOCIATED FEATURES

## Part 2 introduction

In Part 1, the concept of the spectrum of autism was introduced in relation to diagnostic criteria and in relation to the diagnostic interview methods used to measure these criteria. Part 1 also described a set of 'associated features' that are also found alongside the core diagnostic criteria. Associated features (maladaptive behaviours, sleep behaviours, emotion, motor behaviours, daily living skills, pattern of activities and sensory features) are extensively recorded within the DISCO. Information about these features has an important role in aiding the clinician to formulate recommendations for support, management and education. Although they are not core diagnostic criteria, these features are of interest to the work of this thesis as the nature of the 'autism spectrum' is explored. At the time when the work for Part 2 was being completed, the DSM-5 draft criteria had not yet been released and therefore there was a need to explore additional behaviours that might contribute knowledge to a new description of autism spectrum disorder.

Part 2 of the thesis first set out to test whether the selection of items in the DISCO that measure which subsets of associated behaviours such as maladaptive behaviours, sleep behaviours, emotion, motor behaviours etc. actually formed reliable scales. These reliable associated scales were then used to answer a set of empirical questions in Chapters 3 and 4.

## Summary of literature on the role of associated features

Associated features have been described in the autism spectrum since the first publications of Asperger (1944) who described special abilities, difficulty with attention, behaviours problems and unusual sensory responses; these have been implicated in ASD in two main ways. Some associated features that have been found to co-occur with ASD are predicted to influence or compound the difficulties of individuals with ASD. For example, sleep problems are widely reported in ASD (Richdale & Prior, 1995; Schreck & Mulick, 2000; Simonoff et al., 2008) and in 650 children with neuropsychiatric conditions and 135 typically developing children, the children with autism slept significantly less, had more difficultly falling asleep, woke earlier and had more bed wetting than children with typical development, ADHD, anxiety, depression and acquired brain injury (Mayes, Calhoun, Bixler, & Vgontzas, 2009). Mayes and Calhoun (2009) studied 447 children with autism (1-15 years) and found nearly all parents reported sleep problems. They also found that elevated sleep problems were found in individuals with higher levels of autistic symptoms as well as increases in other associated features such as aggression, attention deficit, mood problems and hyperactivity.

Furthermore, in ASD it has been found that children with ASD with more sleep problems also had higher scores on internalising and externalising behaviour and poorer adaptive functioning (Sikora, Johnson, Clemons, & Katz, 2012). Research in the typically developing literature indicates that sleep problems may exacerbate aggression or be caused by the same underlying mechanisms (e.g. Goldman, Richdale, Clemons, & Malow, 2012).

Another example is that aggression and temper problems are also reported in individuals with ASD (Kaat & Lecavalier, 2013; Matson, 2009) and these variables have been shown to influence treatment referral (Matson & Smith, 2008; Mudford et al., 2008), aggressive individuals are five times more likely to be hospitalised (Mandell, 2008) and are also more likely to be in residential placements (McIntyre, Blacher, & Baker, 2002; Tyrer et al., 2006). Such externalising problems in typical development are consistently shown to impact on academic achievement (Galéra, Melchior, Chastang, Bouvard, & Fombonne, 2009) and later problems (Reef, Diamantopoulou, van Meurs, Verhulst, & van der Ende, 2010). Furthermore, the impact of maladaptive behaviours goes beyond the individual and has been shown to increase parental stress in individuals with developmental disorders (Baker, Blacher, Crnic, & Edelbrock, 2002) and individuals with ASD (Davis & Carter, 2008; Lecavalier, Leone, & Wiltz, 2006; Tomanik, Harris, & Hawkins, 2004) and parent mental health problems (Herring et al., 2006) in addition to mothers' perceived negative impact of their child on the family (McIntyre et al., 2002).

For other associated features, however, their proposed role in ASD has been more central with some researchers proposing that sensory or motor behaviours may be causing the core features of ASD. Ornitz and Ritvo (1968), for example, proposed that sensory processing atypicalities may have a causal role in the presentation of repetitive behaviours. They proposed that brainstem abnormalities resulted in states of arousal, which changed from over-excitation to over-inhibition. It was suggested that in order to compensate for these arousal inconsistencies individuals engaged in repetitive and restricted behaviours, which allowed them to create order or predictability in order to manage arousal states. In turn, these sensory and repetitive behaviours impede communication and social interaction, which were viewed as secondary symptoms of the condition.

Others (e.g. Dewey et al., 2007; Leary & Hill, 1996) propose that motor impairments are a core feature of ASD and are linked to social impairments, these theories predict that:

> "a child requires a full movement repertoire of functional actions to engage in social interactions…yet, many children with ASDs exhibit qualitative or quantitative abnormalities in one or more aspects of movement detected as early as infancy…..will result in missed opportunities and reduced engagement with coordinated and agile peers, which, in turn, limits the initiation and maintenance

> of friendships and may contribute to delayed social skills and long term social impairments." (Bhat, Landa, & Galloway, 2011, p. 1123).

The purpose of this brief review was to provide an overview of the way in which associated features are examined. Overall, the associated features of ASD are thought about in two distinctive ways: as a causal role or as a secondary symptom. This process has meant that associated features are not measured in a consistent way and that the effect of associated features on each other in addition to the impact on the core features of ASD is rarely considered.

The alternatives below illustrate the range of different possibilities when considering one associated behaviour, sensory features. The predictions may include:

- Sensory behaviours are related to core features but not other associated features
- Sensory behaviours are related to other associated features but not core features
- Sensory behaviours are related to both the core and associated features
- Sensory behaviours are related to core features when examined alone but not when other associated features controlled for
- Sensory behaviours are related to associated features when examined alone but not when core features controlled for
- The relation between core and/or associated features may be mediated by the core or associated features

These different possibilities may inform us about the presentation of ASD at the behavioural level. One of the aims of the multivariate analyses presented in the next chapters is to identify the role that associated features are playing in relation to the core features of ASD. A second aim is to assess if any associated behaviour has a particularly important or influential role in the behavioural manifestation of ASD.

## Reliability of associated symptoms scales in the DISCO

Before examining the role of associated symptoms in Chapters 3 and 4, the remaining part of this introduction reports the reliability of the associated scales. The DISCO measures a range of items that do not belong to the diagnostic algorithms and instead capture the many associated features of ASD and are used in clinical use of the DISCO to create a clinical picture of the individual's strengths, weaknesses and pattern of behaviours in order to improve the recommendations of needs for the individual. The DISCO covers a large range of such items, however, varying numbers of items are used to capture each section and with the exception of one study on the sensory processing items (Leekam et al., 2007), none of the associated features as measured by the DISCO

have been used in research and none have been directly tested for reliability and validity in research. The first step is to assess the reliability of these scales of behaviours to accurately and comprehensively measure the behaviour in question.

Since there has been virtually no research that has previously examined the psychometric properties or explored any empirical questions from the associated items, this part of the thesis is particularly exploratory and the methods and analyses used reflect this.

Preliminary analyses using Sample 1 assessed the reliability of the scales of associated items as they are arranged in the DISCO (see Table 2-4): Daily living skills, motor behaviours, sensory behaviours, sleep disturbances and emotion behaviours, maladaptive behaviours and pattern of activities. The final two sets of behaviours in this list are commonly thought of as the same concept. Behaviours classed in the DISCO as "pattern of activities" are also found in the descriptions of maladaptive or challenging behaviour in the literature[3]. However, the aim was to test the scales of behaviours specifically as used in the DISCO so these two scales were kept separate.

Ideally, the assignment of associated items to the behaviour scales provided in the DISCO would have made upon or confirmed using clustering or factor analytic techniques. However, even the combined sample numbers used in the current thesis are not statistically robust enough to reliably use these techniques. It is recommended that a minimum for five participant cases per DISCO variable should be used in these analyses, however, due to the large number of items in the DISCO this was not possible (Tabachnick & Fidell, 1989). The associated scales were tested as they are presented in the DISCO.

Associated features were selected to answer further empirical questions if they met two conditions. The first was that they should form a statistically reliable scale and the second was that they should statistically discriminate between ASD and clinical control groups. Scales which only distinguish between individuals with ASD and those with typical development were not included to ensure the behavioural presentation was not just due to a more general neurodevelopmental delay. The resulting analyses allowed further exploration of the role that these associated behaviours play in ASD.

[3] For example, the Child Behaviour Checklist (Achenbach,. 1991) measures both externalising behaviours like those in the DISCO "maladaptive behaviour" scale and behaviours such as hyper-activity like the "pattern of activities" DISCO scale).

**Table 2-4: Table of the associated scales in the DISCO, where they are organised in the DISCO and the type of DISCO items that are included in each scale**

| Associated feature | Part | Section | Types of items/scale |
|---|---|---|---|
| Maladaptive behaviours | Part 6 Behaviour | Section i. Behaviour affecting other people | Untypical behaviours |
| Emotion behaviours | Part 5 Emotions | Section i. Emotions | Untypical behaviours |
| Sensory Behaviours | Part 4 Repetitive stereotyped activities | Section i. Stereotyped movements and vocalisations (1) | Untypical behaviour |
| | | Section ii. Response to proximal sensory stimuli | |
| | | Section iii. Responses to auditory stimuli | |
| | | Section iv. Responses to visual stimuli | |
| | | Section v. Routines and resistance to change (1) | |
| | Part 3 Developmental Skills | Section vi. Self-care – Hygiene | |
| | | Section iv. Self-care - Feeding | |
| Motor Behaviours | Part 3 Developmental Skills | Section ii. Gross motor Skills | Developmental stages |
| | | Section xvi. Visuo-manual and spatial skills | Untypical behaviour |
| Daily Living Skills | Part 3 Developmental Skills | Section iii. Self care – toilet training | Developmental stages |
| | | Section iv. Self-care - Feeding | |
| | | Section v. Self-care – Dressing | Untypical behaviour |
| | | Section vi. Self-care – Hygiene | |
| | | Section vii. Domestic skills | |
| | | Section viii. Independence | |
| Pattern of activities | Part 4 Repetitive stereotyped activities | Section vi. Overall pattern of activities | Untypical behaviours |
| Sleep behaviours | Part 6 Behaviour | Section ii. Sleep disturbance | |

A four stage process was endorsed in which items had to: meet acceptable levels of inter-rater reliability (taken from Wing et al., 2002); be endorsed by at least a minority of individuals with ASD; contribute to the internal consistency of the scale; and correlate with the total scale. Items were only selected for inclusion in the scales of associated behaviours if they meet all the criteria, which is specified in detail in Appendix 3 where the results of these analyses are also presented. These are summarised in Table 2-5.

**Table 2-5: Table showing the DISCO associated scales that met criteria for further exploration in Chapters 3 and 4.**

| Associated Scale | Number of items | Reliable? | ASD different from clinical controls? | Explored in Chapters 3 and 4? |
|---|---|---|---|---|
| Maladaptive behaviours | 16 | √ | √ | √ |
| Emotion behaviours | 4 | X | X | X |
| Sensory behaviours | 22 | √ | √ | √ |
| Motor behaviours | 6 | X | X | X |
| Daily living skills | 23 | √ | X | X |
| Pattern of activities | 6 | √ | √ | √ |
| Sleep behaviours | 4 | X | X | X |

## Results of psychometric analyses:

Four scales were found to be reliable according to all of the criteria above. Details are provided in full in Appendix 3. The items included in the emotion (.697), motor (.719) and sleep (.630) scales did not reach good levels of internal consistency, measured using Cronbach's alpha. This is an indication of how much each behaviour represented the concept being measured and all the items included in the scale were related to each other and the total score. However, the maladaptive behaviours (16 items; .918), pattern of activities (6 items; .807) sensory behaviours (22 items; .859) were all reliable and the total scores for each scale were significantly different from the clinical comparison groups. Although the Daily Living Skills scale was reliable (.956, 23 items) the only significant difference was found between the ASD and TD groups and not with the clinical comparison groups and was therefore not selected to focus on in the following chapters. Regression analyses predicting the reliable scales of maladaptive behaviours and patterns of activities are explored in Chapter 3 and the reliable associated scale of sensory behaviours is explored in Chapter 4.

# 3 Measuring maladaptive behaviours in ASD

The aim of this chapter is to explore the DISCO scales of "maladaptive behaviours" and "pattern of activities" which were identified as being reliable scales at the beginning of Part 2 (see Table 3-1). The first part of the chapter presents a review of the literature related to maladaptive behaviours and instruments previously used to measure these behaviours. The term maladaptive behaviours is used to cover both the maladaptive behaviour and pattern of activities scales from the DISCO as these are described as the same type of behaviour in the literature[4]. All conclusions and predictions referring to maladaptive behaviours are related to both DISCO scales. The second part presents the empirical work. In the study reported in this chapter, I present analyses which explore the presence of maladaptive and activity level behaviours across age, gender and ability level and their relation to both the core features of ASD and the other associated features measured by the DISCO.

The work in this chapter was specifically designed to explore the relative strength of the relations between the core and associated features of ASD, a question that is overlooked in the literature. In general research points to an association between maladaptive and the core features of ASD although very little work has controlled for age, gender and IQ in these relations which have been shown to have inconsistent relations across the literature. Even less work focuses on the relation between maladaptive behaviours and other associated behaviours, however, some correlations have been found. Further work is needed on the relative contribution of the core and associated features in their relation with maladaptive behaviours.

The analyses that were conducted further our knowledge about the predictors of maladaptive behaviours in ASD and are also relevant to the overall aim of Part 2, which is to determine if any one or more associated features have a particularly important role in predicting behaviours in ASD. As mentioned earlier, the empirical work for this part of the thesis was carried out before the release of DSM-5 or the draft DSM-5 criteria and therefore the core features of ASD are measured according to ICD-10's triad of impairments in social interaction, communication and restricted or repetitive behaviours.

[4] For example, the Child Behaviour Checklist (Achenbach,. 1991) measures both externalising behaviours like those in the DISCO "maladaptive behaviour" scale and behaviours such as hyper-activity like the "pattern of activities" DISCO scale).

**Table 3-1: Table listing the DISCO items included in the reliable scales of "maladaptive behaviours" and "pattern of activities"**

| **Maladaptive behaviours** |
|---|
| Wandering |
| Destructiveness |
| Noisiness |
| Physical aggression |
| Behaviour in public places |
| Personal modesty |
| Psychological barriers |
| Approaching strangers |
| Embarrassing remarks in public |
| Interrupting conversations |
| Inappropriate response to others' emotions |
| Difficult or objectionable personal habits |
| Scatters or throws objects around |
| Lack of co-operation |
| Needs constant supervision |
| Demands carer's attention |
| **Pattern of activities** |
| Limited pattern of activities |
| Inability to remain sitting |
| Continual motor restlessness |
| Hyperactivity |
| Fixed, repeated motor stereotypes |
| Excessive repetition of activities |

## 3.1 Literature review

### *3.1.1 Maladaptive behaviours in ASD*

As well as experiencing impairments in social interaction, communication and repetitive behaviours, individuals with ASD are at risk of a range of maladaptive behaviours and difficulties above and beyond the core features. These associated maladaptive behaviours are referred to by a range of names in the literature including problem behaviours, challenging behaviours and behaviours that challenge. Overall these terms cover behaviours of socially unacceptable behaviours, aggression, oppositional conduct, hyperactivity, self-injury and irritability (Anderson, Maye, & Lord, 2011; Howlin, 2007; Lecavalier, 2006; Shattuck et al., 2007; Simonoff et al., 2008). The work of this thesis focuses on these "externalising" behaviours although some studies also include internalising behaviours such as anxiety and depression in their measure of maladaptive behaviour but as the DISCO has an independent scale for emotional disturbances, research on these constructs is not specifically reviewed.

Maladaptive behaviours have been demonstrated to be highly prevalent in individuals with ASD (Hartley, Sikora, & McCoy, 2008; Horovitz, Matson, Rieske, Kozlowski, & Sipes, 2011; Matson, Wilkins, & Macken, 2009) with estimates as high as 94% (Jang, Dixon, Tarbox, & Granpeesheh, 2011). These behaviours are also marked in their severity, which could lead individuals to meet criteria for additional clinical conditions (e.g. Brereton, Tonge, & Einfeld, 2006); research has found 46% of children (3-5 years olds) reach clinical or borderline clinical scores on the Child Behaviour Checklist (CBCL, Achenbach & Edelbrock, 1991) (Eisenhower, Baker, & Blacher, 2005) as do a third of children aged 1.5-5.8 years old (Hartley et al., 2008).

DSM-IV-TR (APA, 2000) defines Attention Deficit Hyper-activity Disorder (ADHD) as a disorder with a persistent pattern of inattention and/or hyperactivity-impulsivity that is more severe and more frequently displayed than in others of the same developmental level. These descriptions have overlaps with the "pattern of activities" scales, which include behaviours such as "inability to remain sitting" and "hyper-activity" as shown in Table 3-1. In addition, the maladaptive behaviour items from the DISCO (Table 3-1) also show similar characteristics as the behaviours described by DSM-IV-TR as diagnostic of Oppositional Defiance Disorder (a pattern of negativistic, hostile and defiant behaviour) such as losing temper, actively defies or refuses to comply with adults' requests or rules and to a lesser extent the behaviours of Conduct Disorder ("A repetitive and persistent pattern of behaviour in which the basic rights of others or major age-appropriate societal norms or rules are violated", DSM-IV-TR, APA, 2000). The content of the following chapter does not focus on these disorders specifically, but the overlap in behavioural characteristics may have important implications.

A review of the literature between 2000-2012 of co-morbid disruptive disorders in ASD found that the prevalence of Oppositional Defiance Disorder (ODD) alongside ASD varied from 3-37% and a 1-10% with Conduct Disorder, the authors estimate that in total one in four individuals with ASD would meet criteria for ODD or CD (Kaat & Lecavalier, 2013). Furthermore, Simonoff et al. (2008) explored the comorbidity of other disorders alongside ASD in a sample of 112 children with ASD (10-14 years old), 70% of children had at least one comorbid disorder, and in particular 29% met criteria for social anxiety, 28% for ADHD and 28% for ODD.

### *3.1.2 Why is it important to study maladaptive behaviours?*

Maladaptive behaviours are important to study in relation to ASD because individuals with Autistic Disorder are reported to have significantly more maladaptive behaviours than individuals with PDD-NOS (Kozlowski & Matson, 2012), other developmental disabilities (McClintock, Hall, & Oliver, 2003), individuals with intellectual disability (Blacher & McIntyre, 2006; Tonge & Einfeld,

2003), individuals with Down syndrome (Capone, Grados, Kaufmann, Bernad-Ripoll, & Jewell, 2005) or typical developing children (Gadow, DeVincent, Pomeroy, & Azizian, 2004). Tantrum behaviours are also a common occurrence in children and adolescents with ASD and such behaviours have been shown to positively correlate with ASD severity score (Matson, 2009) and are more prevalent in children with ASD than children with ADHD or typical development (Tureck, Matson, May, & Turygin, 2012). Aggression is also reported at high levels across individuals with ASD. Around 50% of individuals present with aggression with the level of aggression being highest in young children (Mazurek, Kanne, & Wodka, 2013). Parent reported symptoms indicate that 67% of children with ASD have demonstrated aggression to a caregiver and 47% to non-caregivers (Kanne & Mazurek, 2011). Other studies show that 27% of children with Autistic Disorder have scores in the clinically significant range for externalising behaviours and 22% clinically significant scores for the aggression sub-scale; these scores were related to lower non-verbal intelligence, poor expressive language and adaptive functioning (Hartley et al., 2008).

Problem behaviours significantly impact on an individual's daily living, their family and their social development. In typical development, externalising problems have consistently been found to impact on individuals' academic achievement (Galéra et al., 2009) and later problems (Reef et al., 2010). The impact of maladaptive behaviours goes beyond the individual and has been shown to increase parental stress in individuals with developmental disorders (Baker et al., 2002), individuals with ASD (Davis & Carter, 2008; Lecavalier et al., 2006; Tomanik et al., 2004) and parent mental health problems (Herring et al., 2006) in addition to influencing the mothers' perceived negative impact of their child on the family (McIntyre et al., 2002). Furthermore, it has been shown that maladaptive behaviours cause more stress to parents than the core features of ASD themselves (Hastings et al., 2005; Lecavalier et al., 2006). Maladaptive behaviours also impact on the pressure on teachers (Hastings & Brown, 2002), which interferes with an individual's ability to learn and can also interfere with interventions that causes disruptions in the prognosis of individuals with ASD (Horner, Carr, Strain, Todd, & Reed, 2002; Horner, Diemer, & Brazeau, 1992).

In addition, one of the main reasons for treatment referral for individuals with ASD is the presence of maladaptive behaviours (Matson & Smith, 2008; Mudford et al., 2008). Individuals with ASD who present with aggressive behaviours are five times more likely to be hospitalised than individuals with ASD without severe aggressive behaviour (Mandell, 2008), aggressive individuals or those with high levels of behavioural problems with ASD are also more likely to be in residential placements (McIntyre et al., 2002; Tyrer et al., 2006). Further research is needed to understand better the maladaptive behaviours in ASD and how these progress across age, gender and ability

level in order to improve individuals' independence, daily functioning, family network and social support (quality of life).

### *3.1.3 Maladaptive behaviours and core features of ASD*

Little research has been conducted on predictors of maladaptive behaviours in ASD and their relation with the core features of autism. It is important to understand these relationships in terms of creating the most effective behavioural interventions for ASD as well as improving knowledge about shared behaviours.

Overall maladaptive behaviours (Dominick, Davis, Lainhart, Tager-Flusberg, & Folstein, 2007; Jang et al., 2011; Matson, Wilkins, et al., 2009) and tantrum behaviours (Konst, Matson, & Turygin, 2013; Tureck et al., 2012) are significantly positively correlated with ASD symptom scores and appear to be more prevalent in individuals diagnosed with Autistic Disorder compared to PDD-NOS (Kozlowski & Matson, 2012; Medeiros, Kozlowski, Beighley, Rojahn, & Matson, 2012). In addition, in 141 individuals with severe intellectual disability (of 166 originally assessed) maladaptive behaviours were best predicted 15 years later by poor expressive language, poor quality of social interaction and diagnosis of autism at the time of assessment (Murphy et al., 2005).

One theory for the expression of problem behaviours in children and adolescents is that they are due to an individual's inability to competently express his or her needs and therefore he or she uses problem behaviours to mediate communicative interactions (Carr & Durand, 1985; Day, Horner, & O'Neill, 1994; Durand, 1993). Naturalistic observations in classroom revealed 50% of non-verbal children showed maladaptive behaviours and analysis of videotapes revealed these individuals used these behaviours for requesting and rejecting communicative functions (Chiang, 2008). Furthermore, language and communication delays have been shown to be predictive of disruptive behaviour (Durand & Merges, 2001) and interventions which focus on effective communication strategies reduced maladaptive behaviours in preschool children (Koegel, Koegel, Hurley, & Frea, 1992).

In addition, strong relations between maladaptive and repetitive behaviours have also been proposed. In pre-school children, conduct problems were more common in children with ASD who scored high on three of the four sub-scales of the Repetitive and Restricted Behaviour Scale (33 items, Bourreau, Roux, Gomot, Bonnet-Brilhault, & Barthelemy, 2009): sensorimotor stereotypies; reaction to change; and modulation insufficiency (e.g. aggressiveness towards self and others, need to control the progress of activities, stereotyped emotional manifestations and agitation). Hyperactivity was also related to these three scales as well as "restricted behaviours" (Ghanizadeh & Moeini, 2011). Repetitive and restricted behaviours independently predict maladaptive behaviours

in children and adolescents with severe intellectual disabilities: the presence of high frequency repetitive and restricted behaviours increased the risk of self-injury by sixteen times and increases the risk of having two or more severe maladaptive behaviours by twelve times (Oliver, Petty, Ruddick, & Bacarese-Hamilton, 2012).

### *3.1.4 Maladaptive behaviours and associated features of ASD*

Maladaptive behaviours are also associated with additional co-existing conditions and behaviours in ASD. It has been found that children with ASD with more sleep problems also had higher scores on internalising and externalising behaviour sub-scales of the CBCL and poorer adaptive functioning (Sikora et al., 2012). In addition, maladaptive behaviours measured by the Aberrant Behaviour Checklist (Aman, Singh, Stewart, & Field, 1985) were significantly predicted by medication, sleep problems and anxiety (42% of the variance, Rzepecka, McKenzie, McClure, & Murphy, 2011). Anxiety has also been shown to be related to aggression and tantrums (Quek, Sofronoff, Sheffield, White, & Kelly, 2012). Self-injury, sleep problems and sensory problems have been shown to be the strongest predictors of aggression in a sample of children (Mazurek, Kanne, et al., 2013). Research indicates that sleep problems may exacerbate aggression or be caused by the same underlying mechanisms (Goldman et al., 2011; Goldman et al., 2012).

The small amount of literature on sensory processing and maladaptive behaviours has consistently found sensory behaviours to be related to maladaptive behaviours in ASD (e.g. Baker, Lane, Angley, & Young, 2008). Ninety-five percent of individuals with ASD who scored high on internalising behaviours scored on at least one quadrant of the Sensory Profile, with 81.8% of those scoring high on externalising also scoring on one Sensory Profile quadrant. Specifically, in ASD sensory avoiding and male gender were associated with internalising behaviours and sensory sensitivity with externalising behaviours, in TD internalising behaviour was predicted by sensory avoiding and externalising behaviour by sensory seeking sub-scales of the Sensory Profile (Tseng, Fu, Cermak, Lu, & Shieh, 2011). However, a limitation of the work by Tseng et al is that individuals' autism severity and cognitive functioning were not included in the statistical analyses and these may have an impact.

### *3.1.5 Inconsistencies across the literature*

The review of the literature on maladaptive behaviours reveals a number of contrasting findings; the main discrepancies are found when comparing the basic measurements of age, IQ and gender. This indicates that further research on the additional predictor variables may be limited by the sample of individuals to whom that research is targeted e.g. high versus low functioning

individuals or children versus adolescents and adults. The three core inconsistencies are detailed below:

### *3.1.5.1 Age:*

In typical development aggression is very common in early childhood but decreases such that only a minority of adolescents present with aggressive behaviour (Tremblay, 2010). Cross-sectional research that has included a range of ages across childhood and adolescence in individuals with an intellectual disability or autism have also reported a decrease in symptoms (Shattuck et al., 2007; Tonge & Einfeld, 2000), however, some decreases for example in aggressive behaviour are only from 54.6% in 2-4 year olds to 48.3% of 14-17 year olds, with both levels still being exceptionally high (Mazurek, Kanne, et al., 2013). In addition, other reports claim maladaptive behaviours to remain constant across childhood and adolescence (Matson, Mahan, Hess, Fodstad, & Neal, 2010) or that age is not a significant predictor of frequency or severity of maladaptive behaviours (McTiernan, Leader, Healy, & Mannion, 2011). Little work has examined whether these behaviours are maintained at the same level across adulthood too. The current study was able to control for age across early childhood (pre-school age), childhood, adolescence as well as adulthood as the sample covers ages up to 38 years old.

### *3.1.5.2 Gender:*

Findings on the presence of maladaptive behaviours in ASD has also identified contrary results for the effect of gender. A large proportion of studies have shown high problem behaviours in males with ASD (Baghdadli, Pascal, Grisi, & Aussilloux, 2003; Eisenhower et al., 2005; Hartley et al., 2008; Mayes et al., 2012), which mirrors the effects found in typical development (e.g. Archer, 2004) and intellectual disabilities (McClintock et al., 2003). However, some studies show no effect of gender in ASD (Kanne & Mazurek, 2011; Kozlowski, Matson, & Rieske, 2012; Mazurek, Kanne, et al., 2013; McTiernan et al., 2011; Murphy, Healy, & Leader, 2009; Shattuck et al., 2007) or even that girls present with more maladaptive behaviours (Gadow et al., 2004; Holtmann, Bölte, & Poustka, 2007; Nydén, Hjelmquist, & Gillberg, 2000). Kaat and Lecavalier (2013) discuss the possibility of gender being an "equalizer" for co-morbidity in ASD. In individuals with an intellectual disability, the effect of gender has been shown to be absent when studying additional associated features such as disruptive behavioural disorders (Einfeld et al., 2010). This proposal still needs further exploration. Furthermore in many of the studies above research has explicitly compared males and females on the basis of number of maladaptive behaviours on which they present rather than including gender

into the multivariate analyses, which would identify interactions between gender and the other predictive variables.

### *3.1.5.3 IQ:*

The current study also allows a range of high and low functioning individuals to be measured. Previous research has focussed on either individuals with an intellectual disability (e.g. Chiang, 2008; Murphy et al., 2005; Oliver et al., 2012; Rzepecka et al., 2011) or high functioning (e.g. Bauminger, Solomon, & Rogers, 2010) rather than assessing the whole range and to enable the effect of IQ and a range of adaptive functioning to be controlled for. In the typical literature the occurrence of disruptive and aggressive behaviour is reported to occur at higher levels in individuals with a lower IQ (see reviews: Lahey, Waldman, & McBurnett, 1999; Moffitt, 1993) and similar findings have been shown in ASD: lower IQ predicted severity and frequency of stereotypies and self-injurious behaviours and the frequency of aggression in children (McTiernan et al., 2011) and lower cognitive functioning is a risk factor for maladaptive behaviours (e.g. A. De Bildt, Sytema, Kraijer, Sparrow, & Minderaa, 2005; Dominick et al., 2007; Shattuck et al., 2007). However, in other ASD studies, verbal individuals met more criteria relevant to Oppositional Defiance Disorder (Witwer & Lecavalier, 2010) than did non-verbal individuals and parent reports of individuals with a low IQ have resulted in fewer symptoms of ODD than individuals with IQs above 70 (Gadow et al., 2004). Furthermore, the review by Kaat and Lecavalier (2013) found many studies with no effect of IQ on maladaptive behaviours (cf Gadow, DeVincent, & Drabick, 2008; Kanne & Mazurek, 2011; Mayes, Calhoun, Murray, Ahuja, & Smith, 2011) or their effect was lost when controlling for ASD symptoms or diagnosis (Bauminger et al., 2010). Medeiros et al. (2012) found an interaction between diagnosis and developmental level; individuals with ASD higher developmental level were associated with greater levels of maladaptive behaviour than were individuals with a developmental delay without ASD (e.g. Cerebral Palsy, Down Syndrome) in which higher developmental level was associated with fewer maladaptive behaviours.

One limitation of the research on maladaptive behaviours in ASD is that the measures used to capture these behaviours are also measuring behaviours that are inherent in the diagnostic criteria for ASD. A large amount of data reviewed above used the Strengths and Difficulties Questionnaire (SDQ, Goodman, 1997) or the Child Behaviour Checklist (CBCL, Achenbach & Edelbrock, 1991). However, these measures include core features of ASD such as social interaction behaviours, for example the SDQ items “has at least one good friend,” “picked on or bullied by other children” and “generally liked by other children” are key behaviours used to check for impairments in peer interactions that is a core features of autism (DSM-IV-TR, APA, 1994). Furthermore, the CBCL

has items such as “avoids looking others in the eyes” and “speech problems” as well as behaviours that may have sensory or repetitive behaviour causes such as “smears or plays with bowel movements” or “eats or drinks things that are not food,” which may interfere with correlations between these variables. The Aberrant Behaviour Checklist (Aman et al., 1985) is better validated for maladaptive behaviours, with empirically derived scales of hyperactivity, irritability, inappropriate speech, lethargy/social withdrawal and stereotypic behaviour, however, it still contains some items on social isolation and social reactions (see Kaat, Lecavalier, & Aman, 2013). This overlap is important to consider as relations between the core features or even the associated features such as emotion may be inflated if the measures are capturing similar behaviours. Attempts were made to overcome these limitations in the current study by removing any items that are used in the DISCO diagnostic algorithms for ICD-10 (e.g. impairments in social interaction) from the measure of maladaptive behaviours.

### *3.1.6 Hypotheses*

It is proposed that the core features of ASD are likely to predict a significant proportion of the variance in maladaptive behaviours, as operationalised as both the maladaptive behaviours DISCO scale and the pattern of activities DISCO scale. This is predicted as occurrence of these maladaptive behaviours is high in ASD and previous research has found autism symptom scores to be positively correlated with maladaptive behaviours (Dominick et al., 2007; Jang et al., 2011; Matson, Wilkins, et al., 2009). However, only a minority of studies have investigated the role of all of the three core ASD features in combination e.g. using ADI-R scores which have measures of social, communication and repetitive behaviours (Kanne & Mazurek, 2011) and very few have then controlled for all additional factors such as age, IQ, gender and the interactions between them. Therefore no prediction is made about the strength of core symptom and maladaptive behaviour relations when age, IQ or gender are controlled. Previous research also highlights the relation between maladaptive behaviours and other associated features such as sleep, emotion and sensory behaviours (e.g. Sikora et al., 2012). Therefore, it is also predicted that the other (non-core) associated features will have a significant relation with the presence and severity of maladaptive behaviours. Finally, the literature presented above does not lead to consistent predictions about the independent role of age, IQ or gender in predicting maladaptive behaviours. Significant differences are not predicted for the significant predictor variables of the maladaptive behaviour scale score and the pattern of activities scale score as these behaviours are usually grouped together in other measures in the literature that has been reviewed above. It was predicted that the total score for pattern of activities would be predicted by the same variables as the maladaptive behaviour scale.

## 3.2 Empirical work

### *3.2.1 Aims of the study*

The aim of the empirical work presented in the following section is to further the knowledge of the impact of these characteristics alone and in combination with each other for predicting maladaptive behaviours. No study has assessed the predictive power of both the core and associated features in one model to assess the relative contribution of each set of behaviours. In addition the current study also takes into account the role of age, IQ and gender. The aims of the current study are listed below:

1. To better understand the role of age, gender and ability level in the presentation of maladaptive behaviours
2. To examine the associations between the core features of ASD as measured by ICD-10 and maladaptive behaviours
3. To examine associations between the other associated features of ASD and maladaptive behaviours
4. To explore whether the core ASD features or associated features account for more of the variance in maladaptive behaviours
5. To examine the associations between the core and associated features with scores on the pattern of activities scale to assess if the same relations are found as for the maladaptive behaviour scale.

### *3.2.2 Methods*

#### *3.2.2.1 Participants:*

DISCO data from 200 individuals referred to a tertiary referral centre for social and communication disorders for diagnosis is used as the study sample in this study. Full details of this sample are presented in the Methods chapter (section 2.2.4). This sample was chosen as it contains a large number of individuals, allowing multivariate analyses to be conducted with appropriate power and the individuals included in the sample provide a wide range of ages.

#### *3.2.2.2 Data analysis:*

Multivariate regression analyses were chosen in order to control for the effects of all variables on the relation between each independent variable and the dependent variable (maladaptive behaviours); “All IVs enter into the regression equation at once: each one is assessed as if it had entered the regression after all other IVs had entered. Each IV is evaluated in terms of

what it adds to prediction of the DV that is different from the predictability afforded by all the other IVs" (Tabachnick & Fidell, 1989). Firstly, however, univariate analyses were conducted; that is, correlations between the total maladaptive score and all of the predictor variables (age, IQ, gender, social interaction, communication, RRB, sensory, daily living skills, pattern of activities, emotions, sleep) in order to assess the original relation between these variables.

A series of hierarchical multiple linear regressions were conducted to assess the relation between the core, associated and individual characteristics alone and in combination at predicting the presence of maladaptive behaviour and pattern of activities score in order to address the aims set out above. The assumptions of each multiple regression were tested and any problems were reported in the results section.

### *3.2.2.3 Analysis plan:*

1. Conduct multivariate analyses including interaction terms to better understand the role of age, gender and ability level in the presentation of maladaptive behaviours. Gender, age, IQ as well as the interaction between these variables were entered as predictor variables for a model with maladaptive behaviour score as the dependent variable.
2. Conduct a multiple regression analysis to look for specific associations between the core features of ASD as measured by ICD-10 (predictor variables) and maladaptive behaviours (dependent variable). This approach allows the independent contribution of each core feature to be assessed.
3. Conduct a multiple regression analysis to look for specific associations between the other associated features of ASD (sensory, motor, pattern of activities, emotion, daily living skills, sleep) and maladaptive behaviours.
4. Conduct two hierarchical multiple regression analyses to address whether the core ASD features or associated features account for more of the variance in maladaptive behaviours by in the first model entering the core features and then the associated features and in the second model reversing the order. At this stage, analyses also look for mediating relationships between the core and associated features on predicting maladaptive behaviour score.
5. The same process was conducted to look for significant predictors of pattern of activities score.

### 3.2.2.4 Measures:

#### 3.2.2.4.1 Dependent variables:

There were two dependent variables examined in this study. The first consisted of the total score on the items identified to belong to the maladaptive behaviour scale (16 items) as measured in Part 2 introduction. The second is that score on the pattern of activities scale (6 items), which was also found to be reliable. All individuals who scored "marked" were assigned a score of three, "minor" scores were assigned a score of two and "no problem" scores were converted to one. All maladaptive or pattern of activities items were added together to form the dependent variables, the full scales are reported in Table 3-1.

#### 3.2.2.4.2 Independent variables:

##### 3.2.2.4.2.1 Age, IQ and Gender

Age was measured continuously and both gender and IQ were binary measures. The sample consisted of 33 females and 167 males. Measurement of IQ is detailed in the method section the IQ estimate variable consisted of seven groups, for these analyses this variable was collapsed into a binary "high" (>70) or "low" (<70) IQ groups.

##### 3.2.2.4.2.2 Diagnostic core features of ASD

A full list of the items and their codes for the core features are detailed in Appendix 2 and Leekam et al (2002). Although the DISCO has a large range of items that measure social interaction, communication and repetitive behaviours, only the items used in the ICD-10 diagnostic algorithm were selected to capture social interaction, communication and restricted or repetitive behaviours to enable a measure of core features as described by the international classification systems. The scores for these core behaviours were the total number of items that were rated as "present", the codes that determine if a DISCO item is "present" vary for each item according to the clinical description of ICD-10. This measure of social interaction, includes 3 items (behaviour in public places, personal modesty, psychological barriers) that are selected from the maladaptive section of the DISCO and included in the maladaptive scale in the DISCO. Analyses were conducted with these items removed from both the social and maladaptive measure. Also, for the purpose of the current chapter, the sensory items that are included in the ICD-10 algorithm measure of repetitive behaviour were removed because they are included in the separate sensory scale as part of the analysis, the overlap of sensory behaviours in the diagnostic criteria for repetitive behaviours can be seen in Appendix 2.

#### *3.2.2.4.2.3 Associated features*

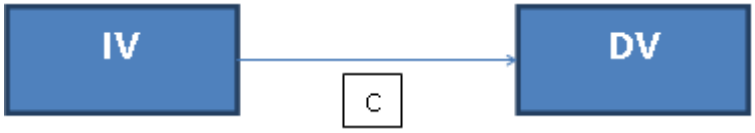
The measures of associated features introduced in the introduction to Part 2 revealed two additional reliable scales; sensory behaviours (22 items) and daily living skills (23 items). For each scale, the total score forms independent predictor variables that were added to the multiple regression with maladaptive total score as the dependent variable. The scales found not to be reliable (motor, emotion, motor, sleep) were converted to binary options of "1" - at least one behaviour is present to a "marked" degree and "0" - no DISCO items present for emotion, motor and sleep behaviours. Multiple regressions can deal with binary variables, but usually if they are coded as 0 or 1 (Field, 2009).

Changes were made to the sensory scale. The literature reviewed above classifies self-injury as maladaptive behaviours, whereas the DISCO scales classify self-injury as a sensory behaviour. In order to prevent confounding these associated two items "self-injury" and "self-stimulation" were removed from the sensory scale.

### *3.2.2.5 Mediation analyses*

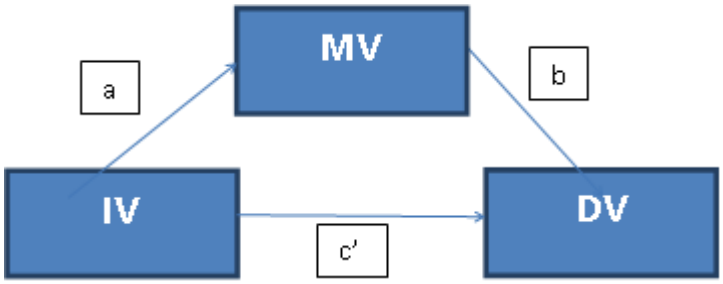
The most commonly used method to test for mediation is the original criteria proposed by Baron and Kenny (1986). They suggest four criteria to test for mediation that can be examined using three regression models as described below:

1. The IV is significantly related to the DV; this is measured by running a simple regression with the IV (e.g. social interaction) predicting the DV (maladaptive behaviours) – Path c.




2. The IV is significantly related to the mediating variable (MV); this is measured by running a simple regression with the IV (e.g. social interaction) predicting the MV (e.g. sensory behaviours) – Path a.
3. The MV is significantly related to the DV; this is measured as the first step in a hierarchical regression model with the MV (e.g. sensory) predicting the DV (maladaptive behaviours) – Path b.
4. When controlling for the effects of the MV on the DV, the effect of the IV on the is no longer significant; this is measured in the second step of a hierarchical regression analysis, where the IV (e.g. social interaction) is entered as a predictor of the DV

(maladaptive behaviours); once the effect of the MV (e.g. sensory) has been controlled for in the first part of the hierarchical regression – Path c'.




However, more contemporary analysts focus on the "indirect model", this is a measure of how well the ab (mediating path) performs compared to the c' or direct effect path (Hayes, 2013; MacKinnon, 2008). Kenny (2014) describes four methods of calculating the indirect effect, however, the test applied here is the bootstrapping method (Preacher & Hayes, 2008). This is selected over the other tests as it not affected by the unrealistic assumption that others tests such as the Sobel test makes about the shape of the sampling distribution of the indirect effect. Instead the bootstrapping test generates 95% bias-corrected bootstrap confidence intervals using z = 1000 bootstrap samples. This is calculated using PROCESS (Hayes, 2012), a computational tool running under SPSS, which was used to directly estimate the strength of the indirect (ab) paths.

Both tests of mediation will be conducted, for any relations that are identified as being relevant to include during the first stage of analyses. The Baron and Kenny approach allows the mediating model to be directly compared to the previous regression analyses and the indirect approach is adopted as a confirmation of the mediating effect following the concerns of modern mediational analysts.

Finally, more complex statistical models were not used here for two reasons: path analysis, specifically and structural equation modelling more generally should only be conducted when there is a clear hypothesis, or small number of hypothesis within one model, to test. Such techniques should not be adopted at the exploratory stage of research (Everitt & Dunn, 1991), which is what is being conducted here. In addition, path analysis with nominal or ordinal measurement with a small number of categories (including dichotomies) is not reliable and cannot be conducted (Bryman & Cramer, 1994; Everitt & Dunn, 1991).

### 3.2.3 Results:

#### 3.2.3.1 Maladaptive behaviours

Table 3-2 shows the associations between all the independent predictor variables and maladaptive behaviours score (dependent variable).

**Table 3-2: Correlation analyses (r) for all predictors and maladaptive "Mal" behaviours (log transformed repetitive behaviours "RB", sensory behaviours "sens" and pattern of activities "pattern" and square root transformed social interaction "SI")**

| | Mal | IQ | Age | Sex | SI | Com | RB | DLS | Sleep | Emotion | Sens | Pattern |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Mal | 1 | -.140* | -.169* | .029 | .331** | .296** | .305** | .294** | .229** | .119 | .492** | .434** |

Note: *p<.05 **p<.01

The initial analyses explored the normality of the included variables. As the Sample was large (200 cases) significant tests for normality may over emphasise the non-normality. Therefore skewness of the variables was examined; skewness values divided by the standard error of skewness indicates whether a distribution is too positively or negatively skewed and therefore non-normal (Field, 2009). Values greater than 1.96 are significant at the p<.05 level of significance, as the sample is large, only values above 2.58 (p<.01) were cause for concern. Examination of these values revealed a number of variables to have non-normal distributions. Tabachnik & Fidell (1989) advise different forms of transformation depending on the extent and direction of the skew, therefore, the final variables to be used in the analyses were square root transformation of social interaction score, log transformed repetitive behaviour, pattern of activities and sensory scores. All other variables had normal distributions in their original format.

Assumptions for multiple regression analyses were tested for all models. The Durbin-Watson statistic did not deviate far from 2 (average = 1.88), the VIF values did not deviate far from 1 (1.02-1.88), the tolerance statistics were well above 0.2 (0.53-.99) and the standardised residuals were normally distributed as tested by Shapiro-Wilk statistics (Field, 2009).

#### *3.2.3.1.1 Aim 1: To examine the role of age, gender and ability level in the presentation of maladaptive behaviours*

Age, IQ and Gender were all entered into a multivariate linear regression, the total maladaptive score was used as the dependent variable. The three variables alone were entered in Block 1 and then the interaction between the variables (three two-interactions and one three way) were entered in Block 2. The interaction terms were created by converting gender and IQ (previously coded as 0,1) into -0.5 or 0.5 and multiplying by each other or with a centred score for age. This was done by standardising the age variable as distance from the mean; all interaction term variables were centred before multiplication as suggested by Aiken and West (1991). All assumptions for multiple regression were met.

**Table 3-3: Hierarchical multiple regression model showing the effects of age, IQ, gender and their interactions as the predictor variables of maladaptive behaviour score.**

| | B | SE B | β |
|---|---|---|---|
| **Step 1** | | | |
| Constant | 31.236 | 1.646 | |
| Gender | .567 | 1.409 | .028 |
| Age | -.012 | .005 | -.153* |
| IQ | -1.901 | 1.101 | -.122 |
| **Step 2** | | | |
| Constant | 28.998 | 1.905 | |
| Gender | 1.297 | 1.429 | .065 |
| Age | -.006 | .007 | -.077 |
| IQ | -.005 | 1.429 | .000 |
| IQ * Gender | -6.436 | 2.858 | -.209* |
| IQ * age | -.019 | .014 | -.122 |
| Age * Gender | -.005 | .014 | -.033 |
| Age * Gender * IQ | -.007 | .028 | -.024 |

*Note:* $R^2$ = .043 for Step one (F(3,196)= 2.935, p<.05), $\Delta R^2$ = .041 (F(4,192) = 2.136, p=.078, n.s.), *p<.05, **p<.01, ***p<.001

The results of the hierarchical multiple regression can be seen in Table 3-3. The first model (age, gender, IQ) explained 4.3% of the variance and the second, with the addition of the interaction terms explained 8.4% (2.8% and 5% adjusted). In Stage 1, age was a significant predictor but the second step also revealed a significant interaction between IQ and gender: as this interaction was significant a graph was designed to demonstrate this relationship. As can be seen in Figure 3-1 (below) in the low functioning group the males have more maladaptive behaviours than females whereas in the high functioning group females appear to have more behaviours than males, although this line is not as steep.

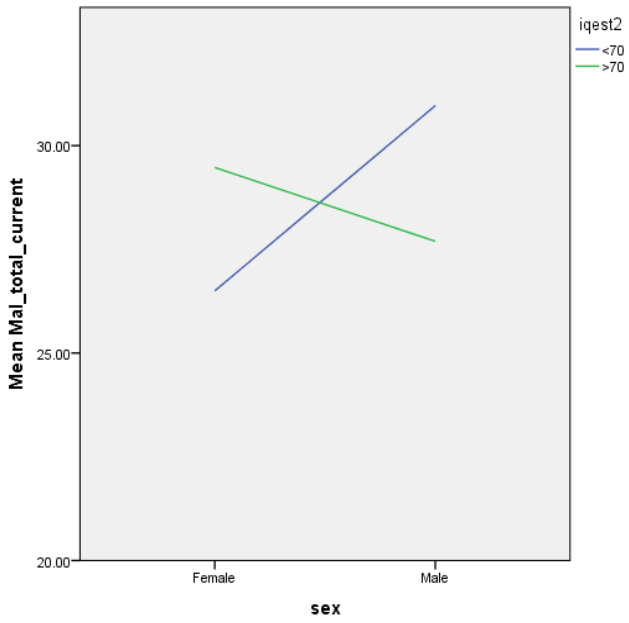



**Figure 3-1: Line graph showing the interaction between IQ and gender on the total maladaptive behaviour score.**

#### *3.2.3.1.2 Aim 2: To examine specific associations between the core features of ASD as measured by ICD-10 and maladaptive behaviours*

The results of the hierarchical multiple regression can be seen in Table 3-4. Both age and the IQ by gender interaction were significant in block one but with the addition of the core features the interaction was no longer a significant predictor and all three of the core features were significant predictors in the model. Model 1 predicted 7.2% (adjusted $R^2$ = 0.052) but the addition of the core features of ASD improved the variance accounted by the Model to 21.3% (adjusted $R^2$ = 18.3%).

**Table 3-4: Hierarchical multiple regression model showing effects of age, IQ, gender and the core ASD features as predictor variables of maladaptive behaviours score.**

| | B | SE B | β |
|---|---|---|---|
| **Step 1** | | | |
| Constant | 29.832 | 1.733 | |
| Gender | 1.091 | 1.426 | .055 |
| Age | -.010 | .005 | -.139 |
| IQ | .221 | 1.429 | .014 |
| IQ * Gender | -7.004 | 2.849 | -.228* |
| **Step 2** | | | |
| Constant | 21.476 | 2.176 | |
| Gender | -.408 | 1.357 | -.021 |
| Age | -.011 | .005 | -.146* |
| IQ | .491 | 1.399 | .032 |
| IQ * Gender | -5.123 | 2.683 | -.167 |
| Social | 1.568 | .677 | .182* |
| RB | 5.076 | 2.088 | .177* |
| Communication | .656 | .276 | .171* |

*Note:* $R^2$ = .072 for Step one ($F(4,185)= 3.571$, $p<.01$), $\Delta R^2$ = .142 ($F(3,182) = 10.926$, $p<.001$), *p<.05, **p<.01, ***p<.001

#### *3.2.3.1.3 Aim 3: To examine specific associations between the other associated features of ASD and maladaptive behaviours*

The results of the hierarchical multiple regression can be seen in Table 3-5. In the model with the associated features as predictors both age and the IQ by gender interaction remained significant predictors of the number of maladaptive behaviours. In addition, daily living skills, sensory behaviours and pattern of activities were strong significant predictors. The second full model explained 35.5% (32.1% adjusted) of the variance in total maladaptive behaviour score.

**Table 3-5: Hierarchical multiple regression showing the effect of age, IQ, gender and the associated features of ASD as predictor variables for maladaptive behaviour score**

| | B | SE B | β |
|---|---|---|---|
| **Step 1** | | | |
| Constant | 29.841 | 1.749 | |
| Gender | 1.138 | 1.420 | .057 |
| Age | -.012 | .005 | -.153* |
| IQ | .106 | 1.421 | .007 |
| IQ * Gender | -6.234 | 2.834 | -.202* |
| **Step 2** | | | |
| Constant | -26.144 | 8.404 | |
| Gender | .447 | 1.250 | .022 |
| Age | -.011 | .005 | -.138* |
| IQ | 2.431 | 1.254 | .156 |
| IQ * Gender | -6.075 | 2.458 | -.197* |
| DLS | .162 | .045 | .229*** |
| Sleep | 1.518 | .958 | .097 |
| Emotion | 1.137 | 1.281 | .054 |
| Motor | .159 | 1.082 | .009 |
| Sensory | 18.925 | 6.403 | .218** |
| Pattern | 20.211 | 5.751 | .257*** |

*Note:* $R^2$ = .066 for Step one ($F(4,195)= 3.454$, $p<.01$), $\Delta R^2$ = .289 ($F(6,189) = 14.091$, $p<.001$), *p<.05, **p<.01, ***p<.001

#### *3.2.3.1.4 Aim 4: To explore whether the core ASD features of associated features amount for more of the variance in maladaptive behaviours*

To explore the relative contribution of the core and associated features of ASD to the prediction of total maladaptive behaviour score, two hierarchical multiple regressions were run. In both models the first block contained the individuals' characteristics (age, IQ, gender and IQ*gender). In the first regression, the core features of ASD were entered in the second block and the associated in the third whereas in the second regression model the associated features were entered first. The results of the final block for both models (all variables entered) can be seen in Table 3-6. In total the model explained 37.8% of the variance in maladaptive behaviours (33.2%

when $R^2$ adjusted). When all variables were entered into the model age, the IQ by gender interaction remained significant, the core features were no longer significant predictors but daily living skills, sensory behaviours and pattern of activities remained predictive. The amount of variance explained by the blocks of each model differed depending on the order that the variables were entered. When the core features were entered first and therefore their predictive value was controlled for, the addition of the associated features significantly improved the variance in maladaptive behaviours explained by the model ($\Delta R^2$ step 3 = .164 ($F(6,176) = 7.751$, $p<.001$).

However, when associated features were entered first, the addition of the core features (once associated features are held constant) did not significantly improve the variance explained by the model ($\Delta R^2$ step 3 = .012 ($F(3,176) = 1.143$, $p=.333$, n.s.). The previous models indicate that the core features do significantly predict maladaptive behaviours when entered separately; this indicates a mediating relationship between core and maladaptive behaviours by the associated features of ASD.

**Table 3-6: Hierarchical multiple regression model showing the effects of age, IQ, gender, the core and associated features of ASD as predictor variables of maladaptive behaviour score.**

| | B | SE B | β |
|---|---|---|---|
| **Step 3** | | | |
| Constant | -22.410 | 9.406 | |
| Gender | .247 | 1.279 | .013 |
| Age | -.010 | .005 | -.129 |
| IQ | 2.049 | 1.316 | .132 |
| IQ * Gender | -6.090 | 2.488 | -.199* |
| Social | -.054 | .680 | -.006 |
| RB | 1.433 | 2.032 | .050 |
| Communication | .419 | .259 | .109 |
| DLS | .143 | .047 | .204** |
| Sleep | 2.112 | .964 | .137* |
| Emotion | 1.059 | 1.316 | .051 |
| Sensory | 17.995 | 7.095 | .209* |
| Pattern | 16.021 | 6.046 | .206** |
| Motor | -.292 | 1.113 | -.017 |

*Note:* $R^2$ = .072 for Step one ($F(4,185)= 3.571$, $p<.01$), $\Delta R^2$ step 2 = .142 ($F(3,182) = 10.926$, $p<.001$), $\Delta R^2$ step 3 = .164 ($F(6,176) = 7.751$, $p<.001$) *p<.05, **p<.01, ***p<.001

#### *3.2.3.1.5 Mediation analyses*

The results above indicate that associated features influence the association between the core features of ASD and maladaptive behaviours. Mediation analyses according to Baron and Kenny's (1986) criteria were run for all associated and core behaviours that were found to be significant predictors. The relation between maladaptive and the core features (social interaction,

communication and repetitive or restricted behaviours) were tested again when significant associated variables (sensory, sleep, daily living skills and pattern of activities) were included as mediating variables. Twelve exploratory models were tested; each core feature mediated by daily living skills, pattern of activities, sleep or sensory behaviours.

The analyses revealed two significant mediation models, with sensory processing mediating the relationship between social interaction and its relations with maladaptive behaviours, and repetitive behaviours and its relations with maladaptive behaviours but the relationship between communication and maladaptive behaviours was not mediated by sensory behaviour. The results of the two significant mediated models are shown in Table 3-7 and Table 3-8 below.

#### *3.2.3.1.5.1 Social interaction, sensory and maladaptive behaviours*

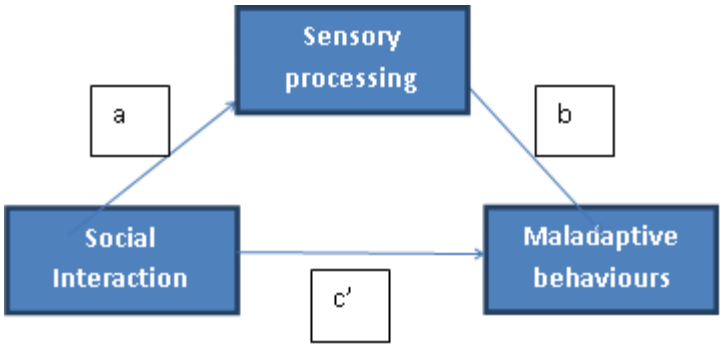



**Table 3-7: Table showing the three regression analyses used to test the mediation analyses of sensory behaviours on relation between social interaction and maladaptive behaviours.**

| | $R^2$ | R2 change | β |
|---|---|---|---|
| **Regression one:** | | | |
| Social interaction predicts maladaptive behaviours | .106*** | | .325 |
| **Regression two:** | | | |
| Social interaction predicts sensory processing | .265*** | | .515 |
| **Regression three:** | | | |
| Step 1: Sensory processing predicts maladaptive behaviours | .224*** | | .416 |
| Step 2: Social predicts maladaptive when controlling for effects of sensory processing | .233 | .009 | .111 |

#### *3.2.3.1.5.2 Repetitive, sensory and maladaptive behaviours*

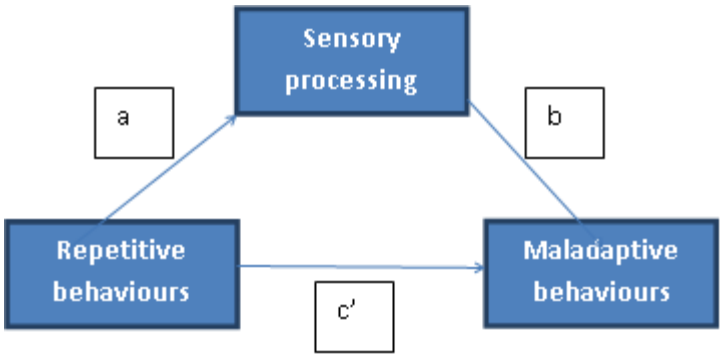



**Table 3-8: Table showing the three regression analyses used to test the mediation analyses of sensory behaviours on relation between repetitive behaviour and maladaptive behaviours.**

| | $R^2$ | R2 change | β |
|---|---|---|---|
| **Regression one:** | | | |
| RB predicts maladaptive behaviours | .082 (.078)*** | | .278 |
| **Regression two:** | | | |
| RB predicts sensory processing | .159 (.154)*** | | .399 |
| **Regression three:** | | | |
| Step 1: Sensory processing predicts maladaptive behaviours | .239*** | | .489 |
| Step 2: RB predicts maladaptive when controlling for effects of sensory processing | .250 | .010 | .109 |

Finally, to confirm whether sensory behaviours mediate the relationship between social interaction or repetitive behaviours and maladaptive behaviours, PROCESS (Hayes 2012), a computational tool running under SPSS, was used to directly estimate the strength of the indirect (ab) paths for the two models. The results (see Table 3-9) show similar results to the mediation analyses presented above, the c path was significant, the direct effect was not but in both cases the indirect effects through the sensory behaviours were significant.

**Table 3-9: Tests of direct and indirect effects of repetitive and social on maladaptive behaviours through sensory behaviours.**

| | Maladaptive behaviours | | |
|---|---|---|---|
| Effect | B | SE | 95% CI |
| Repetitive on Maladaptive (c) | **.010** | **.002** | |
| Direct Effect (c') | .004 | .003 | |
| Indirect Effects* (ab) | | | |
| Sensory | **.0061** | **.0016** | **.0032 -- .0095** |
| Social on Maladaptive (c) | **.039** | **.008** | |
| Direct Effect (c') | .011 | .008 | |
| Indirect Effects* (ab) | | | |
| Sensory | **.0276** | **.0051** | **.0184 -- .0385** |

* Bootstrapped confidence intervals. Effects in bold are significant at p<.001.

### *3.2.3.2 Pattern of activities*

Finally, a multiple linear regression analysis was conducted for pattern of activities. This was conducted to examine whether the two separate DISCO scales, which are usually conceptualised and measured under the same category of maladaptive behaviours in previous literature, have the same relations with core and associated features of ASD. The square root of pattern of activities was used as the dependent variable due to non-normally distributed data as presented at the beginning of the methods section. Again the first hierarchical model only included age, IQ, gender and their interaction terms but as no interaction terms were significant, this step is not shown. The second model incorporated the additional predictor variables (Table 3-10) and this final model predicted 37.5% of the variance in pattern of activities, with age, IQ, social interaction maladaptive and sensory behaviours as significant predictors.

**Table 3-10: Multiple linear regression showing effect of the core and associated features of ASD in addition to age, IQ and gender as predictors of pattern of activities score.**

| | B | SE B | β |
|---|---|---|---|
| **Step 1** | | | |
| Constant | 3.198 | .071 | |
| Age | -.001 | .000 | -.185** |
| Sex | .080 | .060 | .088 |
| IQ | -.207 | .047 | -.293*** |
| **Step 2** | | | |
| Constant | 1.600 | .246 | |
| Age | -.001 | .000 | -.158* |
| Sex | .061 | .054 | .067 |
| IQ | -.096 | .045 | -.136* |
| SI | .014 | .005 | .222** |
| COM | -.007 | .011 | -.044 |
| RB | -.008 | .008 | -.062 |
| DLS | .001 | .002 | .030 |
| Sen | .230 | .045 | .384*** |
| Sleep | .013 | .042 | .018 |
| Emotion | .043 | .058 | .046 |
| Maladaptive | .250 | .045 | .401*** |

*Note:* $R^2$ = .137 for Step one (F(3,196)= 10.400, p<.001), $\Delta R^2$ step 2 = .238 (F(7,189) = 10.267, p<.001), *p<.05, **p<.01, ***p<.001

No mediating models between the core and associated features were found. Specifically, sensory behaviours did not mediate the effect between pattern of activities and core features as firstly repetitive behaviours did not significantly predict pattern of activities and secondly social interaction score remained significant on the direct effect even when sensory behaviours were controlled for. This is shown in Table 3-11 using the modern mediation technique (Preacher & Hayes, 2008)

**Table 3-11: Tests of direct and indirect effects of repetitive and social on pattern of activities through sensory behaviours.**

| | Maladaptive behaviours | | |
|---|---|---|---|
| Effect | B | SE | 95% CI |
| Repetitive on pattern of activities (c) | .493 | .195 | |
| Direct Effect (c') | .135 | .217 | |
| Indirect Effects* (ab) | | | |
| Sensory | **.628** | **.137** | **.380 -- .914** |
| Social on pattern of activites (c) | **.4.150** | **.592** | |
| Direct Effect (c') | **.2.235** | **.650** | |
| Indirect Effects* (ab) | | | |
| Sensory | **1.915** | **.405** | **1.217 – 2.740** |

*Bootstrapped values. Significant effects are shown in bold.

## 3.3 Discussion

### *3.3.1 Findings in comparison to previous literature*

The original correlation matrix showed that in univariate analyses the total maladaptive behaviour score was predicted by age, IQ, daily living skills, sensory behaviours, social interaction, communication, repetitive behaviours and sleep problems. All of these variables have previously been identified in the literature as associated to the presence of maladaptive behaviours, however, this study went on to include these predictors into multivariate models in order to hold all other variables constant and examine the association between each independent variable and the total maladaptive behaviour score. The first set of analyses identified both age and an IQ by gender interaction as being significant predictors with maladaptive behaviours decreasing with age and maladaptive behaviours being more present in low functioning males and high functioning females. When the core features of ASD were added to the analysis the presence of social interaction problems, communication problems and repetitive behaviours all significantly increased the prediction that an individual would also present with maladaptive behaviours. Again, the addition of associated features identified that the presence of maladaptive behaviours significantly increased with the presence of daily living skill problems, sensory and pattern of behaviours as was predicted given previous research that has linked additional behavioural features to the presence of maladaptive behaviours (e.g. Sikora et al., 2012; Tseng et al., 2011; Mazureck et al., 2013).

However, when all the predictor variables were entered into the model simultaneously, only five variables remained significant predictors. This model confirmed the majority of literature suggesting that the presence of maladaptive behaviours decrease with age across typically developing individuals and individuals with ASD (Shattuck et al., 2007; Tonge and Einfield, 2000; Mazurek et al., 2013). The minority of studies that did not report an effect of age on challenging behaviours may well be due to a limitation of age range included in the sample, for example Mctiernan et al (2011) found age was not a significant predictor of frequency or severity of problem behaviours but the sample only included children aged 3-14 years old. This current sample on the other hand covered all of adolescence and adults ranging up to 38 years of age.

The IQ and gender interaction also remained significant when all variables were included, however, the sample used in these analyses has a large gender bias toward males and therefore the interaction with IQ is interpreted with caution and should be replicated in more evenly balanced samples, however, it may have implications in explaining the inconsistent findings across the literature. This supports a large proportion of the recent literature that gender by itself is not predictive of maladaptive behaviours (e.g. Kolowski and Matson 2011; 2012; Mazureck et al., 2013; Shattuck et al., 2007) and a few recent studies claiming that IQ by itself has no predictive value (e.g.

Kanne and Mazurek, 2011; Mayes et al., 2011; Gadow et al., 2008). There is also conflicting findings for IQ across the typical and atypical literature, one explanation for this may therefore be that due to the diagnostic bias in ASD most ASD samples are predominately male and if the typically developing literature samples more girls this could affect the role that IQ may be playing in those analyses. Furthermore, it is predicted that the IQ of the sample may affect the results of gender comparisons, for example, Holtmann et al (2005) claimed females presented with more problem behaviours, however, their sample was IQ matched between males and females but also the average IQ was 88.8 and therefore above the range of learning disability and all their participants (range 70-128) would fall in the high IQ group in the current study.

Future research needs to take into account the age, IQ and gender of the sample participants and look for relationships among these variables as well as their independent contribution and to be careful to not draw conclusions beyond those sampled in their study. Additional findings, support this concern to be cautious as it was found that the effect of IQ disappears when controlling for diagnosis of ASD. This was unable to be controlled for in the current study but highlights that interactions may also exist between diagnostic status and age, IQ, gender and therefore diagnosis of ASD should also be included (Bauminger et al., 2010).

In the final model, in addition to age and an IQ by gender interaction, the significant predictors were all from the associated features category. The presence of sensory behaviours has already been implicated in the presentation of maladaptive behaviours (Baker et al., 2008; Tseng et al., 2011; Duerdan et al., 2012) and this is supported in the current study. However, no research had previously explored whether such associations remain even when core features of ASD are also controlled for, which was found here. This adds a novel finding to the literature that implicates sensory behaviours as important in the presence of maladaptive behaviours. Increased problems in daily living skills also significantly predicted the presence of maladaptive behaviours. This replicates some of the findings indicating that lower adaptive functioning is predictive of the presence of maladaptive behaviours (de Bildt et al., 2005). Interestingly, this variable was not significantly correlated to IQ or communication level, which suggests that the daily living skills of an individual are independently related to the presence of maladaptive behaviours.

Sensory processing behaviours were consistently significantly predictive of maladaptive behaviours and this relationship could be explored further. The analysis conducted here are not causal and therefore longitudinal research would be required to confirm the direction of prediction, however, it would be hypothesised that individuals with a greater number of sensory behaviours are likely to find their everyday surroundings a more harsh and challenging environment and therefore

the individuals under additional pressure and stress from the sensory input are more likely to express this in the form of maladaptive behaviours.

Sensory behaviours were also found to "mediate" the relationship between social and repetitive behaviours and the total maladaptive behaviour scale. When the effect of the relationship between sensory and maladaptive behaviours had been controlled for, the relationship between the core features and maladaptive behaviours was no longer significant when tested in a series of regression analyses. This represents a significant mediating model according to the criteria proposed by Baron and Kenny (1986). However, modern mediation analysts argue that a more accurate test is comparing how well the ab (mediating path) performs compared to the c' or direct effect path (Hayes, 2013) for this reason an SPSS macro (Hayes, 2012) using the bootstrapping method tested the direct and indirect effects of the mediation triangle. The results of these analyses also support a significant mediation model of sensory behaviours impacting on the relation between maladaptive behaviours and the core features of repetitive behaviours and social interaction.

However, a further argument is that mediation should not be tested at all in data of only one time point and it is true that the analyses reported in the current study cannot determine directional or causal effects. With this caution, what can be drawn from these data is that maladaptive, sensory, social and repetitive behaviours are all associated and that sensory behaviours have the potential to influence associations with maladaptive behaviours. This may be in an additive approach such that lack of social understanding can lead to maladaptive behaviours but these are even more likely to occur if the individuals also has sensory behaviours, which may make social interaction harder and therefore lead to more maladaptive behaviours or an indirect approach in which the presence of social interaction impairment leads to the expression of maladaptive behaviours only in individuals with additional sensory behaviours, this would be interesting to test in longitudinal studies. In terms of repetitive behaviour again an additive approach could mean that the presence of both repetitive and sensory behaviours increase the risk of maladaptive behaviours or that repetitive behaviours only lead to maladaptive behaviours when additional sensory demands are placed upon individuals with ASD. Another possibility is that the same mechanism is causing all three behaviours to present in individuals with ASD. Further investigations are needed into what role sensory behaviours are having in the relationship between core and maladaptive behaviours and longitudinal data will need to be collected to achieve this as the direction of such effects is as yet unclear. More complex models, which are likely to require stronger data such as from different time points, could model whether the core features are also impacting on these relations e.g. if both sensory and repetitive behaviours mediate the relation between maladaptive and social behaviours.

It was predicted that the total score for pattern of activities would be predicted by the same variables as the maladaptive behaviour scale. However, this was not the case and different relations with the core and associated features of ASD were found. When all core and associated variables were entered into a model predicting total maladaptive behaviour score, the core features were no longer significant predictors of maladaptive behaviours, instead the associated features of ASD explained an additional proportion of the variance. However, in the complete model predicting the pattern of activities score, with both core and associated features entered as predictor variables, the core feature of social interaction remained a significant predictor of pattern of activities score, along with maladaptive and sensory behaviours and no mediating models were found to be significant.

This research potentially adds to the literature base about the overlap in behaviours of ASD and ADHD and between ASD and Oppositional Defiance Disorder/Conduct Disorder and highlights that sensory and social behaviours appear to be key impairments to examine across conditions. In addition, these findings highlight the importance of considering different types of maladaptive behaviours as the maladaptive behaviour scale in the DISCO and the pattern of activities scale were predicted by different core and associated features. This could have implications for potential explanations behind the presentations of the different types of behaviours and for identifying key areas to target in maladaptive behaviour interventions. Further work should focus on identifying consistent sub-types of maladaptive behaviours, in case different behavioural presentations have different relations to the core features of ASD or different aetiologies.

### *3.3.2 Limitations*

This chapter has added to the literature by using more complex multivariate designs to control for the effects of many variables on the prediction of maladaptive behaviours, however, the data and design of the analyses in this chapter have some limitations. Firstly, the gender balance in the study is extremely biased to males with 83.5% of the sample being male and therefore the results here need to be replicated in samples with a greater proportion of females especially when interactions such as IQ and gender appear to be playing a significant role. In addition, although the analyses presented here controlled for many variables in each model, there are no measures of wider demographics such as parenting stress or household income, which have previously been shown to have significant predictive effects (Bauminger et al., 2010; Kanne & Mazurek, 2012). Finally, the measure of IQ is limited as much of the variance is lost by creating a binary (high versus low functioning) distinction, however, this was done to make the analyses conducted more reliable and to include data from the whole sample.

Another limitation is the apparent overlap of concepts across social interaction and communication and the types of maladaptive behaviours measured in the DISCO. Some maladaptive behaviours were categorised as impairments in social interaction in ICD-10, to control for this the decision was made to remove any overlapping items from the social interaction measure and maladaptive behaviour measures. However, in order to progress work with the DISCO, a larger dataset will be required to apply statistical analyses to group subsets of behaviours to assess if these behaviours can be reliably separated rather than using the predefined groupings in the DISCO. The same applies for the pattern of activities scale and the repetitive behaviours, interestingly, however these two groups of DISCO items were not found to be significantly associated.

### *3.3.3 What have we learnt/Future Directions*

The main issue that this study has demonstrated is the importance of conducting multivariate analyses that are capable of controlling for a multitude of variables and the interactions between them. For example, the presence of sleep problems has previously been implicated in the presence of maladaptive behaviours but does not remain significant when other associated features such as sensory processing are controlled for. Furthermore, the failure to find relations between IQ or gender in predicting maladaptive behaviours may be true when these behaviours are measured separately but in combination these variables may be playing an extremely important role. The DISCO is therefore an excellent tool for this as it measures a range of behaviours and can be used across individuals of all ages and levels of ability.

In addition, what we categorise as a problem or maladaptive behaviour needs to be carefully explored such that these behaviours being measured as maladaptive are independent from social or communicative behaviours in their own right. The core features of ASD may complicate the accurate identification of problem or psychiatric behaviours due to individuals being non-verbal or having a lower developmental level to make it difficult to judge if behaviours are “vindictive” such that the individual understands appropriate norms and is deliberately misusing them (Kaat and Lecavalier, 2013). In addition, these results add to the conclusions of different relationships between variables in the typically developing literature and the ASD literature indicating that maladaptive behaviours and furthermore disruptive behaviour disorders may be different across diagnostic groupings.

Finally, in order for the findings from the DISCO to be applicable to the wider literature on maladaptive or challenging behaviours future studies could examine whether the DISCO items are capturing the same information as the other tools (e.g. Child Behaviour Checklist, Strength and Difficulties Questionnaire and Abberant Behaviour Checklist). The DISCO items may have an advantage in capturing such behaviours in ASD as the items were specifically included if they were

seen in clinical practice, however, this may also prove to be a disadvantage in capturing the range of maladaptive behaviours. In addition, the analyses presented in this chapter raised the concern of categorising maladaptive behaviours as a whole when the DISCO scales of maladaptive behaviours and pattern of activities items showed different relations with the core and associated features of ASD. This means that analyses conducted when the two scales are measured together may be misleading. Factor analytic or cluster analysis techniques could be employed to identify sub-groups of maladaptive behaviours in the DISCO and whether pattern of activities and maladaptive behaviours scales should remain separate.

### *3.3.4 Summary*

The focus of Chapter 3 was on the maladaptive behaviours measured by the DISCO, which included two reliable scales of behaviour: maladaptive behaviours and pattern of activities. Previous research has shown that individuals with ASD frequently present with maladaptive behaviours, however, the terminology and tools used to measure these behaviours are inconsistent and hardly any work controls for the effects of age and IQ in the presentation of these behaviours and little work has controlled for the effect of other associated behaviours. The analyses presented in this chapter found strong associations between maladaptive behaviours and individual's social interaction and repetitive behaviours. However, the exploration of these relationships in hierarchical regressions and mediation analyses found that both the relations between maladaptive behaviours and social interaction and repetitive behaviours are influence by the presence of sensory behaviours. The overall aim of Part 2 of this thesis was to identify if any associated features were playing a particularly important role in the behavioural presentation of ASD. The sensory behaviours are further explored in Chapter 4.

# 4 Chapter 4: What role do sensory behaviours play in ASD?

The aim of this chapter is to explore the DISCO scale of sensory behaviours, one of the reliable 'associated' scales tested earlier and of significance for the results in Chapter 3. Again, the first section of this chapter is dedicated to a comprehensive[5] literature review of the measurement and findings of the research conducted so far on sensory behaviours in ASD. The second part of the chapter presents the empirical work. Two studies are described in this chapter. In Study 4.2.1, I present analyses which explore the presence of sensory behaviours across age, gender and ability level and their relation to both the core features of ASD and the other associated features measured by the DISCO using the analysis method set up in Chapter 3. In Study 4.2.2, primary data are collected using the sensory items from the DISCO converted into a self-report questionnaire, the Sensory Preferences Questionnaire (SPQ). This study provides information about sensory behaviours in high functioning adults with ASD in order to overcome a major limitation in the sensory literature. In addition, this study allows the sensory items from the DISCO to be compared to existing questionnaire methods to explore the external validity of the DISCO sensory scale.

The analyses conducted in this chapter help to address the overall aim of Part 2, which was to identify if any associated behaviours have a particularly important role in the behavioural manifestations of ASD. The findings in Chapter 3, highlighted the role of sensory behaviours in the presentation of maladaptive behaviours as well as having an impact on the relations between repetitive and maladaptive behaviours and between social interaction behaviours and maladaptive behaviours. Relations between the core features of ASD and sensory behaviours are explored directly in Study 4.2.1[6].

## 4.1 Literature review

### *4.1.1 Sensory behaviours in ASD*

Atypical responses to sensory stimuli have been part of the clinical description of ASD since the first publications of Kanner (1943) and Asperger (1944/1991). Furthermore, the sensory aspects of everyday lives are at the forefront of many autobiographical accounts of ASD (O'Neill, 1999; Williams, 1994). For example Temple Grandin described her auditory sensitivity as like "having a

[5] Sensory behaviours/features/atypicalities/processing/modulation/disorder/profile and Autism/ASD/PDD/Aspergers were used as search terms in Web of Knowledge in 2010 and updated in 2013. Although studies focused on one type of sensory behaviour e.g. hyper-responsivity were excluded from the review.

[6] The relation of sensory behaviours and the core features, independent of individual characteristics (age, IQ, gender) and associated features, also formed part of a masters dissertation project which preceded the PhD research (Kent, 2010).

hearing aid with the volume control stuck on 'super loud.' It is like an open microphone that picks up everything. I have two choices: turn the mike on and get deluged with sound, or shut it off. Mother reported that sometimes I acted like I was deaf." (Grandin, 1995, p. 107).

Research has found an extremely high prevalence of sensory abnormalities in individuals with ASD; as high as 95% (Tomchek & Dunn, 2007) and moreover these symptoms do not appear to be limited to one or a selection of modalities but are multi-modal (Adamson, O'Hare, & Graham, 2006; Harrison & Hare, 2004; Kern et al., 2007; Leekam et al., 2007). Significantly higher atypical sensory behaviours have been found in individuals with ASD compared with typically developing children (Baranek, David, Poe, Stone, & Watson, 2006; Ben-Sasson et al., 2008; Dunn, Myles, & Orr, 2002; Kientz & Dunn, 1997; Leekam et al., 2007; Ornitz, Guthrie, & Farley, 1977; Rogers, Hepburn, & Wehner, 2003; Talay-Ongan & Wood, 2000), children with intellectual disabilities (Baranek et al., 2006; Rogers et al., 2003; Wiggins, Robins, Bakeman, & Adamson, 2009), children with Down Syndrome (Carter, Capone, Gray, Cox, & Kaufmann, 2006) and individuals with ADHD (Ermer & Dunn, 1998).

Children investigate and experience the world using sensory input. Parental reports have revealed that parents rated sensory behaviours as one of the first signs that they notice (Baker et al., 2008) and that sensory problems are a major factor affecting their child's social, cognitive and sensorimotor development (Dickie, Baranek, Schultz, Watson, & McComish, 2009; Kranowitz, 2005) and in children with ASD, sensory seeking, under-responsiveness as well as atypical auditory responsiveness have been shown to be related to academic underachievement (Ashburner, Ziviani, & Rodger, 2008). In addition, sensory problems may limit opportunities for children to engage with others during play and therefore limits engagement in new learning experiences (Bundy, Shia, Qi, & Miller, 2007). Some of the key findings in the ASD research are that individuals with ASD are likely to present with sensory abnormalities in more than one modality (Leekam et al., 2007; Baker et al., 2008). Sensory behaviours along with social impairments were identified as atypical in the first year of life using retrospective video analysis (Baranek, 1999) as was found in a case study of an individual who presented with hypersensitivity to touch in the first year (Osterling, Meltzoff and Kuhl, 2000). These findings emphasise how important research into sensory behaviours is as so many individuals experience them.

### *4.1.2 What methods are used to measure sensory behaviours in ASD?*

The majority of research that has been conducted on the sensory symptoms in individuals with ASD has been conducted using parent report measures. In the large majority of cases this has been conducted with the Sensory Profile (Dunn, 1999) or the Short Sensory Profile (McIntosh, Miller,

Shyu, & Dunn, 1999; Tomchek & Dunn, 2007). The Sensory Profile was designed in order to measure the reactions of both children with and without disabilities to frequently occurring sensory input. The full Sensory Profile consists of 125 parent report items which are rated on how frequently the sensory response is seen in their child. It covers eight modalities (Auditory, Visual, Activity level, Taste/smell, Body position, Movement, Touch & Social/emotional) as well as the four "quadrants" proposed by (Dunn (1997), 1999)) and colleagues (Dunn et al., 2002). These quadrants are based on an individual's neurological threshold and the individual's response to sensory input: passive responses to sensory input are categorised as either "low registration" if the individual's neurological threshold is high or "sensory sensitivity" if their threshold is low. Active responses in individuals with a high threshold are classed as "sensory seeking" and active responses in individuals with a low threshold are termed "sensory avoidance." In addition, a number of other parent-report instruments have also been used in the sensory literature such as the Sensory Experiences Questionnaire (Baranek et al., 2006), the Sensory Sensitivity Questionnaire-Revised (Talay-Ongan & Wood, 2000), the Sensory Profile Checklist-Revised (Bogdashina, 2003) and the Sensory Processing Measure (Glennon, Miller-Kuhaneck, Henry, Parham, & Ecker, 2007).

All of these instruments utilise parent's report of individuals' sensory functioning. This has two inherent limitations. Firstly, the majority of research findings are based on research with children and this can severely impact on the implications of these findings for individuals across their lifespan. Secondly, a limitation of parent report data is that it only measures a caregiver's interpretation of an individuals' behaviour and parent/caregiver report has also been found to be discrepant with self-report in other behaviour domains such as emotional and behavioural problems (Achenbach, McConaughy, & Howell, 1987). This is particularly relevant to the consideration of sensory behaviours and how individuals interpret sensory information as this is an extremely personal level and also parents often attach reasons behind their child's behaviour that may be more social than sensory i.e. does not like being in crowds, when instead it may be the noises which are affecting the individuals behaviour. Autobiographical accounts overcome these limitations such as Temple Grandin "the nerve endings on my skin were supersensitive. Stimuli that were insignificant to most people were like Chinese water torture" (Grandin & Scarino, 1996). However, although these accounts are rich in detail they are less useful for research because they stem from the individual's subjective account and only from that individuals' perspective and therefore cannot be generalised to other individuals with ASD, in addition, these accounts tend to be from high functioning individuals which again limits their applicability to individuals with intellectual disability or non-verbal individuals.

Self-report questionnaires are another way of measuring individual's sensory behaviours in ASD. Again the Sensory Profile is the most widely used; the Sensory Profile has been adjusted from parent to self-report for use with adults or adolescents (Adult and Adolescent Sensory Profile, AASP; Brown & Dunn, 2002). Crane, Goddard, and Pring (2009) used the AASP and found 94.4% of individuals with ASD had an extreme sensory behaviour on at least one domain but also found a large amount of variability across the ASD group. This contrasts with many findings using parent reports that suggest that sensory behaviour abate with age (see Section 4.1.3.3 below). This indicates that individuals with ASD may learn to use coping strategies to deal with sensory input even though they are still affected by sensory problems and this may lead parent or caregiver reports to be inaccurate as they can only report what is observable (Robertson, 2012).

The only other questionnaire available for capturing sensory behaviours in a self-report format, at the time of study design, was the Highly Sensitive Persons Scale (HSP; Aron & Aron, 1997). Although research has found the measure to be reliable and have good levels of internal consistency (Aron & Aron, 1997; Liss, Mailloux, & Erchull, 2008) the behaviours asked about in this scale vary widely from behaviours that are relevant to ASD e.g. "I am bothered by intense stimuli, like loud noises or chaotic scenes" to others which are unlikely to be relevant to sensory behaviours in ASD such as "a rich, complex inner life" and this scale has not been validated in the ASD population. Both of the existing self-report questionnaires have two main limitations. Firstly neither were designed to measure sensory behaviours that are present specifically in autism. In addition, both questionnaires contain items that overlap with behaviours used in the diagnosis of autism such as "I like to go to places that have bright lights and that are colourful" or "I touch others when I am talking (for example, I put my hand on their should or shake their hands)" in the AASP and "I make it a high priority to arrange my life to avoid upsetting or overwhelming situations" in the HSP, which limits the conclusions that can be made when either tool is compared to the "core" or diagnostically relevant features of ASD as the correlations may be inflated by both measures containing similar items rather than a relation between distinct behaviours. However, the majority of research in this area has used Dunn's model of Sensory modulation problems and the Sensory Profile, Short Sensory Profile or AASP.

Since I carried out the study design that is reported in Study 2 (below), another sensory behaviour self-report tool has been designed. Robertson and Simmons (2013) developed the Glasgow Sensory Profile. This questionnaire allows measurement of proprioceptive, vestibular, taste and smell modalities separately, which are combined as "taste/smell" and "movement" in the AASP as well as measures of visual, auditory and touch sensitivity. This questionnaire is less dependent on the quadrants proposed by Dunn that may not be relevant to individuals with ASD. Robertson and

Simmons (2013) found scores on this measure were significantly related to individual's scores of the Autism Spectrum Quotient, designed to measure autistic traits in the typical population but did not specifically recruit individuals with ASD into the study.

### *4.1.3 Sensory behaviours relation to core features*

#### *4.1.3.1 Why the relation to core features is important to study:*

The large proportion of research on sensory behaviours focuses on the pattern or number of behaviours in individuals with ASD in comparison to typically developing individuals or other clinical conditions as described in Section 4.1.1. Very little research has focussed on the relation between the core features of ASD and sensory behaviours. This is important to consider for several reasons. Firstly, the sensory system is the system in which individuals acquire information about the world, which in turn allows the individuals to respond and adapt to their environment (cf Hilton et al., 2010), impairments in sensory functioning may therefore have huge impacts on an individual's ability to interact with others. This has been argued as a reason to consider sensory behaviours as a primary impairment in ASD (e.g. Baranek, Parham, & Bodfish, 2005; Iarocci & McDonald, 2006; Rogers & Ozonoff, 2005) and research has found early sensory behaviours in infancy and toddlerhood predict poorer language and social interaction abilities and later ages (DeGangi, Breinbauer, Roosevelt, Porges, & Greenspan, 2000). A strong relation between sensory and social interaction would support this proposal and highlight a key area for further exploration of the aetiology of ASD. Equally, no evidence of such a relation may indicate that the social impairments in ASD are independent of the sensory behaviours present in the majority of individuals with ASD or that sensory may be best categorised as a repetitive behaviour.

Secondly, one possibility is that sensory features have been overlooked because of the strong assumption that they are "just" one type of repetitive behaviour that is seen in ASD. The conceptual overlap between sensory and repetitive behaviours is important to consider. The description of the repetitive and restricted behaviour domain in DSM-IV-TR (APA, 1994) such as "stereotyped and repetitive motor mannerisms" and "persistent preoccupation with parts of objects" allow sensory behaviours to also be captured within the description of Autistic Disorder (DSM-IV-TR, APA, 1994) and such the sensory behaviours are categorised within the non-social aspects of ASD. In addition, the repetitive nature of the behaviours seen in ASD such as motor stereotypies and self-injury have been predicted to provide an intrinsic sensory stimulation to the individual (Lovaas et al., 1987). However, the relation or lack of association between sensory and repetitive behaviours is important for diagnostic criteria, pathogenesis and treatment strategies as described by (Boyd et al., 2010, p. 2):

> Strong relationships (as indicated by a high degree of co-occurrence) may suggest that additional descriptions of sensory symptoms should be added to the diagnostic criteria for autism to better characterize the clinical phenotype. Further, robust linkages may indicate shared neurobiological mechanisms underlie these behavioural features, or that similar intervention approaches could be used to treat both classes of behaviours. In contrast, weak relationships may suggest these behaviours are more distinct than previously thought and that differential treatment approaches and taxonomies are needed, possibly because of dissimilar pathogenesis. (page 2).

Although, sensory behaviours have consistently been found to correlate with overall autism severity (Adamson et al., 2006; Ben-Sasson et al., 2009; Kern et al., 2007), these findings do not specify the contribution of the individual core features of ASD (social interaction, communication and repetitive behaviour) in the relation with sensory behaviour. The research below indicates why it is proposed that sensory features may be related to the core features of ASD, and reviews the limited research on the associations between sensory behaviours and the core features of ASD. No research has investigated the link between each core features of ASD and sensory behaviours independently of the effects of the other core features. This is important to consider given the conceptual overlap with sensory and repetitive behaviours; both social and repetitive behaviours are found in individuals with ASD, therefore, a relation between sensory and social behaviours may only reflect the fact that sensory is related to repetitive behaviours which in turn are related to social impairments in ASD (as they co-occur).

The original theory of Ornitz and Ritvo (1968) proposed that sensory behaviours may have a causal role in the presentation of repetitive behaviours. The argument was that brainstem abnormalities resulted in states of arousal, which changed from over-excitation to over-inhibition. It was suggested that in order to compensate for these arousal inconsistencies, individuals engaged in repetitive and restricted behaviours. These repetitive behaviours allowed them to manage their arousal state by increasing order or predictability. These sensory and repetitive behaviours also impede communication and social interaction, which at that time were considered to be secondary symptoms of the condition. This theory has not received much attention in the ASD literature but would predict that there would be strong associations between sensory and repetitive behaviours and although sensory behaviours may also be associated with social interaction and communication scores, these may be smaller than the effects with repetitive behaviours.

Research on the association between sensory and repetitive behaviours in ASD is limited to only seven studies at the time of writing (2014). In several studies using questionnaires, more repetitive behaviours have been shown to be associated with higher levels of sensory behaviour (Boyd, McBee, Holtzclaw, Baranek, & Bodfish, 2009; Chen, Rodgers, & McConachie, 2009) even when controlling for IQ (Gabriels et al., 2008). Further studies compared the relation with sensory behaviours across the different subtypes of repetitive behaviours (insistence on sameness and motor stereotypies) but these results are inconsistent across the different studies e.g. sensory hyper-responsivity was related to insistence on sameness behaviours but not motor stereotypies (Baranek, Foster, & Berkson, 1997), and hypo- rather than hyper-responsivity has been associated with motor behaviours (Gal, Dyck, & Passmore, 2010), whereas Boyd et al. (2010) found that repetitive behaviours were related to hyper-sensitivity but not sensory seeking or hypo-sensitivity. There are some limitations to using questionnaire methods. Gabriels et al (2008) describes that the most potentially confounding factor is the overlap of items between repetitive and sensory behaviours. One example is the use of the item "touches items and people'" described as sensory seeking in the Short Sensory Profile (McIntosh et al., 1999) but it is rated as a compulsive behaviour on the Repetitive Behaviour Scale – Revised (Bodfish et al., 1999). However, the relation between sensory and repetitive behaviours remained even when all overlapping items were removed from both measures (Boyd et al., 2009).

In Study 4.2.1 overlapping items were removed in order to control for the conceptual overlap in sensory and repetitive behaviours. This further highlights why it is important to hold the effects of the other core features constant in such analyses in order to explore these relations independently. The multivariate design adopted in the current chapter overcomes this overlap in another way by exploring the relations between the core features of ASD, e.g. the repetitive and social aspects as well as their independent and combined associations with the sensory behaviours.

It has been shown that early regulatory disorders of sensory processing in infancy and toddlerhood predict poorer language and social interaction abilities and clinical diagnosis at later ages (DeGangi et al., 2000). In addition, atypical responses to sensory input in the first year of life have been shown to predict a later diagnosis of autism (Baranek, 1999; Osterling & Dawson, 1994); increased mouthing, aversion to touch and decreased visual orienting at 9-12 months distinguished individuals who went on to receive a clinical diagnosis of ASD (Baranek et al., 1999). This suggests that reactions to sensory input at a young age may lay developmental foundations for social communication and language in individuals that go on to receive a diagnosis of ASD. These relations, however, have been even less studied than the association between sensory and repetitive behaviours. However some research has found an association between sensory and social and

communication problems in individuals with ASD. The Social Responsiveness Scale (Constantino & Gruber, 2002) is a 65-item parent or teacher report (15-20 minutes) on an individual's ability to engage in emotionally appropriate reciprocal social interaction. In children with high functioning ASD (N=36) and age-matched controls (N=26) between 6-10 years old, a strong positive relation was found between scores on the SRS and sensory scores across four quadrants for the Sensory Profile (Hilton, Graver, & LaVesser, 2007; Watson et al., 2011) and across different sensory modality scores (Hilton et al., 2010). Sensory Profile modality scores of multisensory responsiveness, proximal senses of oral sensory/olfactory and touch were strongest predictors of SRS score. This is the only study to investigate sensory modalities and social behaviours and was limited to high functioning children between 6-10 years old.

Sensory behaviours measured using another less recognised parent report sensory measure the Japanese Sensory Inventory-Revised (JSI-R; Ota, Tsuchida, & Miyajima, 2002) were also significantly associated with social interaction deficits again measured using the SRS in a group of high functioning children (n=42 ASD and n=42 controls) in Japan (Matsushima & Kato, 2014). The JSI-R is a parent report questionnaire consisting of 147 items in eight sub-categories of vestibular (30 items), tactile (44 items), proprioception (11 items), auditory (15 items), visual (20 items), olfactory (5 items), taste (6 items), and other (16 items). Liss, Saulnier, Fein, and Kinsbourne (2006) also found positive correlations between sensory under-reactivity and sensory seeking on the one hand and social and communication scores from the VABS or Vineland Adaptive Behaviour Scales (Sparrow et al., 1989) on the other. However this association was not found for sensory over-reactivity. Finally, Baker et al. (2008) found specific relationships in 2-8 year old children with ASD with sensory problems and communication and socialisation as measured using the VABS.

In addition to the very limited work in this area, research has been limited to total SRS score or sub-scales from more general measures that are not specific to ASD and measuring the social interaction and communication deficits as described by the international classification systems may provide a more valid measure of the relation between sensory behaviours and the core features of ASD. Examining the relations between sensory and core symptoms of ASD with items that are specific to diagnosis would provide more comprehensible and explicit information about the relationships in ASD. If relations exist then it indicates that the sensory features have the ability to impact on the core features of ASD, which in turn suggest that shared biological mechanisms could underlie the behaviour presentation and that similar intervention programmes could be developed to target both sets of behaviours.

### *4.1.3.2 Relation within associated features*

An additional confound in understanding the association between core and sensory behaviours in ASD that has not been considered is the relation between sensory and the additional associated features of ASD. This is important to identify as it may provide insights into the mechanism by which sensory may be impacting on the core features of ASD. For example, the theory of Ornitz and Ritvo (1968) refers to states of arousal in sensory processing, which may be psychologically distressing and that repetitive behaviours were a way of making the sensory world more predictable. This indicates that the impact of sensory on repetitive behaviour may also be influenced by the level of anxiety seen in the individual; anxiety and other emotional problems are one of the associated features also reported in ASD (e.g. J. A. Kim, Szatmari, Bryson, Streiner, & Wilson, 2000; Sukhodolsky et al., 2008). If analyses do not control for the additional features/behavioural presentation within ASD then important associations may be overlooked by confounding variables.

One strand of research has investigated the role of sensory behaviours in adaptive behaviour. It has been suggested that successful sensory integration is an essential tool in children's development (Bundy et al., 2007; Kranowitz, 2005). Baker et al (2008) found a negative relationship between daily living skills and sensory behaviours; more sensory problems were found in individuals with low daily living skills. Such difficulties have also been shown to be related to high levels of depression and anxiety in children with ASD (Ben-Sasson et al., 2009; Goldsmith, Van Hulle, Arneson, Schreiber, & Gernsbacher, 2006; Mazurek, Vasa, et al., 2013; Pfeiffer, Kinnealey, Reed, & Herzberg, 2005) and furthermore with behavioural difficulties (e.g. Rogers et al., 2003) and maladaptive behaviours (Baker et al., 2008). Not all research shows such significant relationships. Robinson and Magill-Evans (2009) found no significant relationship between scores of the Short Sensory Profile and daily living skills as measured by the Self Care Functional Skills scale of the Paediatric Evaluation of Disability Inventory in young children with ASD. Similar results were found by Jasmin et al. (2009) in that there was no significant difference between total Sensory Profile scores and daily living skills as measured by the Vinelands Adaptive Behaviour Schedule in 35 children with ASD. This area has been widely overlooked in the current literature and may be essential in understanding the patterns of behaviours between core and sensory behaviours as well as how associated behaviours interact with these.

Across all the work assessing the relations between sensory and other behaviours there is a limit to how age and IQ are controlled for. Although some studies have taken the effect of age and IQ on sensory features into account (e.g. Boyd et al., 2010) others have failed to do so which may be influencing the correlational analyses. In addition, many studies do not control for effects of core

features when examining the relationship between sensory and associated behaviours and vice versa. It is imperative to look at the relative power of all variables in analyses to best understand the pattern of behaviours.

#### *4.1.3.3 Effect of age and ability level*

The findings across chronological age and IQ are mixed and these individual characteristics are likely to impact on the associations between core and sensory features if they are not properly understood and controlled for in statistical analyses. In a sample of 3-56 year olds, sensory problems were found to be less frequent in older individuals, there were less differences reported between ASD and TD individuals at an older age (Kern et al., 2006) and correlations between sensory behaviours and ASD severity were found in children but not adolescents or adults (Kern et al., 2007). However, in a sample of children below 14 years old, sensory behaviours were more prevalent in the 10-14 year old group than the 6-9 year old group, which in turn showed more than the 4-5 year old group (Talay-Ongan & Wood, 2000).

Other work concludes that the pattern of sensory symptoms across the lifespan may be dependent on the categorisation according to Dunn (1997, see above). In a meta-analysis of 14 studies, Ben-Sasson et al (2009) found that scores for hyper-sensitivity and sensory seeking increased up to the ages of 6-9 years old and decreased after 9 years old, however, patterns were less consistent for hypo-sensitivity. Differences have also been shown across modalities, with proximal and auditory sensory abnormalities remaining consistently high across a sample of 3-38 year olds, whereas visual symptoms were found to decrease with age (Leekam et al., 2007).

There is also much discrepancy across ability level with some studies finding no effect of Sensory Profile scores and developmental level (Baker et al., 2008; Baranek et al., 2006; Kientz & Dunn, 1997; Lane, Young, Baker, & Angley, 2010; Rogers et al., 2003). However, it has also been shown that sensory behaviours were significantly higher in individuals with lower IQs for scores on hypo-sensitivity, hyper-sensitivity, sensory avoidance but not sensory seeking (Crane et al., 2009). In addition, visual symptoms were also more likely to be found in individuals with a lower IQ (Leekam et al., 2007).

#### *4.1.3.4 Summary of research on Sensory and core features of ASD*

The literature so far on sensory behaviours in ASD has found that sensory behaviours are very frequent in individuals with ASD in children (e.g. Leekam et al., 2007; Tomchek & Dunn, 2007) but this is mixed in adults (Kern et al., 2007; Crane et al., 2009). In terms of what role the sensory behaviours play in the behavioural manifestation of ASD, the strongest finding in the literature on

sensory behaviours is the association between repetitive and sensory behaviour (e.g. Boyd et al., 2009), however, relations have also been found between social and communication symptoms and sensory behaviours (Hilton et al., 2007). Research on the relation between sensory behaviour and core features is limited in that each core feature has been examined independently of the additional core features; Study 4.2.1 aimed to overcome this by examining the core features in combination, this study also examined the effects of age, IQ, gender and associated features to best understand the role of sensory behaviours.

### *4.1.4 Aims of Chapter 4*

This study is split into two studies. The first study explores the relation between sensory behaviours and the core and associated features of ASD and the second study focuses on the measurement of sensory symptoms in ASD using a new self-report measure of sensory behaviours.

The aim of Study 1 is to further explore the relations between sensory behaviours and the core features of ASD using the DISCO. The core features of ASD were taken from the items selected to make a diagnosis according to ICD-10 Childhood Autism DISCO algorithm. This was the first study to assess these associations with all three core features alone and in combination. The role that associated features play in these relations is also considered. This study adds the following novel aspects to the current literature on sensory behaviours, by examining:

- Effects of core features while controlling effects other core features
- Effects of core and associated features in the same model
- Examine association between core/associated and sensory in a model controlling the effects of age, IQ and gender
- The association between sensory and core features using behaviours that are directly relevant to the diagnosis of the core features (algorithm items)
- Effects of age and ability level on sensory behaviours and their relations with behaviours in ASD in a large sample of ages and in high and low ability individuals.

The second study has two objectives. The first objective is to address a clear limitation in the literature of a lack of research with adults and data collected using self-report. In order to do this the DISCO sensory items were converted in a self-report questionnaire and both adults with ASD and typically developing adults were recruited to compare sensory measures. Analyses explore how prevalent these sensory behaviours were in high functioning adults with ASD compared to IQ matched typically developing individuals. The second objective focuses on the need to externally validate the sensory items from the DISCO. This study provides one way to do this. Preliminary

analyses have already found the sensory items to have excellent internal consistency. However, comparisons have not been made between the DISCO sensory items and the existing sensory questionnaire scores that are used in the literature. Therefore, the objective of Study 4.2.2 is to convert the sensory items from the DISCO into a self-report questionnaire and compare the total scores with scores from existing sensory profile measures.

## 4.2 Empirical work

### *4.2.1 Study 4.1.2*

The aim of Study 1 is to explore the relations between sensory behaviours and the core and associated features of ASD across age, gender and ability level using data from the DISCO core and associated scales. The aim is to advance the knowledge about the role of sensory behaviours in the manifestation of core and associated features to help answer the overall aim of Part 2 of whether one set of associated behaviours play a particularly influential role.

The reliable DISCO measure of sensory behaviours consists of 22 items. All items met acceptable levels of inter-rater reliability. Three of the Sensory items "other Sensory problems," "other auditory behaviours," and "other visual behaviours" were not reported in any individuals with ASD were therefore removed from the reliability analysis. The first set of analyses revealed four items with inter-total correlation less than 0.3: mouthing of objects (.161), likes to eat the same foods (.210), dislikes having sticky hands (.170) and dislikes lumpy food (.221) and so these items were removed from the sensory scale. This resulted in a sensory scale of 22 items from the DISCO that can be seen in Table 4-1 (below). This scale had an overall cronbach's alpha value of .859. These items are comprised of four modalities as arranged in the DISCO and in Leekam et al (2007): visual (4 items), auditory (3 items), proximal (10 items) and mixed (5 items). These items do not map consistently onto the four quadrants proposed by Dunn (1999): sensory sensitivity, sensory seeking, sensory avoidance and low registration, for example only one items matches the description for low registration (indifference to pain/heat/cold).

**Table 4-1: Table showing the sensory items included in the reliable sensory scale from the DISCO.**

| Sensory items |
|---|
| **Auditory** |
| Distress caused by sounds |
| Fascination with sounds |
| Acuteness of hearing |
| **Visual** |
| Bright lights and shiny objects |
| Interest in watching things spin |
| Twisting hands or objects near eyes |
| Interest in studying angles |
| **Proximal** |
| Smelling objects or people |
| Touching objects |
| Scratching and tapping surfaces |
| Repetitive, aimless manipulation of objects |
| Being spun round |
| Indifference to pain, heat, cold |
| Reaction to gentle touch |
| Reaction to firm touch |
| Self-spinning |
| Dislikes being washed |
| **Mixed** |
| Smearing |
| Self injury |
| Self stimulation without injury |
| Repetitive destructive activities |
| Over-breathing |

The core features of ASD were measured using the items from the DISCO selected to make a diagnosis according to ICD-10 Childhood Autism DISCO algorithm. It is predicted, in line with previous findings (e.g. Boyd et al., 2009) that sensory behaviours should be strongly related to individuals' repetitive and restricted behaviour score. Associations may also be found between sensory scores and the social and communication scores of individuals with ASD as has previously been found using the SRS (e.g. Hilton et al., 2007). However, it is predicted that these relations will not be as strong as the association between sensory and repetitive behaviours as the theory proposed by Ornitz and Ritvo (1986) place the social and communicative symptoms as secondary. It is predicted that when all core features are explored in a simultaneous model, the repetitive behaviour score will be most predictive of sensory behaviours.

Hypotheses on the relation between other associated features of ASD and sensory behaviours are less clear. Little work has focussed on these associations but the findings suggest sensory behaviours may be related to: adaptive functioning (e.g. Baker et al., 2008) as measured by "daily living skills" in the current study; high levels of anxiety or depression (e.g. Pfeiffer et al., 2005)

as measured by the DISCO emotion scale; and the presence of more maladaptive behaviours (e.g. Miller et al., 2005; Rogers et al., 2003). However, no work has first controlled for the core features of ASD in these relations. The findings across age and IQ are variable although in general sensory behaviours are shown to dissipate with age (Wing, 1996) but no clear hypotheses are made regarding these variables. The aims were as follows:

#### *4.2.1.1 Aims of the study*

- To better understand the role of age, gender and ability level in the presentation of sensory behaviours
- To examine the associations between the core features of ASD as measured by ICD-10 and sensory behaviours
- To examine associations between the other associated features of ASD and sensory behaviours
- To explore whether the core ASD features or associated features account for more of the variance in sensory behaviours

#### *4.2.1.2 Method*

##### *4.2.1.2.1 Participants:*

DISCO data from 200 individuals (Sample 2) referred to a tertiary referral centre for social and communication disorders for diagnosis is used as the study sample in this study. Full details of this sample are presented in the Methods chapter.

##### *4.2.1.2.2 Analysis plan:*

The series of analyses conducted followed the procedure set up in Chapter 3, which allows the relative impact of core and associated features to be tested using regression models. The analysis strategy was to:

- Conduct multivariate analyses including interaction terms to better understand the role of age, gender and ability level in the presentation of sensory behaviours. Gender, age, IQ as well as the interaction between these variables were entered as predictor variables for a model with sensory behaviour score as the dependent variable.
- Conduct a multiple regression analysis to look for specific associations between the core features of ASD as measured by ICD-10 (predictor variables) and sensory behaviours (dependent variable). This approach allows the independent contribution of each core feature to be assessed.

- Conduct a multiple regression analysis to look for specific associations between the other associated features of ASD (maladaptive, motor, pattern of activities, emotion, daily living skills, and sleep) with sensory behaviours.
- Conduct two hierarchical multiple regression analyses to address whether the core ASD features or associated features account for more of the variance in sensory behaviours by in the first model entering the core features and then the associated features and in the second model reversing the order.

#### *4.2.1.2.3 Measures:*

##### *4.2.1.2.3.1 Dependent variables:*

The dependent variable consists of the total score on the items identified to belong to the sensory processing scale (25 items) as described in the previous chapter and shown in Table 4-1. All individuals who scored "marked" were assigned a score of three, "minor" scores were assigned a score of two and "no problem" scores were converted to one. All sensory items were added together to form the dependent variable, the scale reported in Table 4-1.

##### *4.2.1.2.3.2 Independent variables:*

The independent variables of Age, IQ, gender, diagnostic core features of ASD and associated features are identical to those described in Chapter 3, these are reiterated below:

*4.2.1.2.3.2.1 Age, IQ and gender:*

Age was measured continuously and both gender and IQ were binary measures. The sample consisted of 33 females and 167 males. Measurement of IQ is detailed in the method section the IQ estimate variable consisted of seven groups, for these analyses this variable was collapsed into a binary "high" (>70) or "low" (<70) IQ groups.

*4.2.1.2.3.2.2 Diagnostic core features of ASD:*

Items used in the ICD-10 diagnostic algorithm were selected to capture social interaction, communication and restricted or repetitive behaviours to enable a measure of core features as described by the international classification systems. A full list of the items and their codes for the core features are detailed in Appendix 2. As described in Chapter 3, the three overlapping social items in the core social score and the maladaptive behaviour scale were removed from both scales, this was maintained here as both variables were entered into the same models to predict sensory behaviours. In addition, the sensory items that are included in the ICD-10 algorithm measure of repetitive behaviour were removed from the repetitive measure because they are included in the

separate sensory scale as part of the analysis, the sensory behaviours found in the diagnostic criteria for repetitive behaviours can be seen in Appendix 2.

*4.2.1.2.3.2.3 Associated features*

The measures of associated features revealed three additional reliable scales along with the sensory behaviour scale. These measures of daily living skills (23 items), maladaptive behaviours (16 items) and pattern of activities (6 items) are the independent predictor variables that were added to the multiple regression with sensory total score as the dependent variable. The remaining associated behaviours did not form reliable scales and were added to the regression analyses as binary options of "1" - at least one behaviour is present to a "marked" degree and "0" - no DISCO items present for emotion, motor and sleep behaviours.

Multivariate regressions analyses were chosen in order to control for the effects of all variables on the relation between each independent variable and the dependent variable (sensory behaviours). Assumptions of each multiple regression were tested and any problems reported in the results section. Firstly, however, univariate analyses were conducted; correlations between the total maladaptive score and all of the predictor variables (age, IQ, gender, social interaction, communication, RRB, maladaptive, daily living skills, pattern of activities, emotions, sleep) in order to assess the original relation between these variables. Secondly, a series of hierarchical multiple linear regressions were conducted to assess relation between the core, associated and individual characteristics alone and in combination at predicting the presence of sensory behaviour score in order to address the aims set out above.

### *4.2.1.3 Results*

The initial analyses explored the normality of the included variables. As in Chapter 3, exactly the same procedures were followed. Sample 2 was large (200 cases) and as significant tests for normality may over emphasise the non-normality, the skewness of the variables was examined; skewness values divided by the standard error of skewness indicates whether a distribution is too positively or negatively skewed and therefore non-normal (Field, 2009). Values greater than 1.96 are significant at the $p<.05$ level of significance, as the sample is large only value above 2.58 ($p<.01$) were cause for concern. These tests of normality revealed that some of the variables had non-normal distributions. As described in Chapter 3, the variables used in the analyses consist of square root transformation of social interaction score, log transformed repetitive behaviour, pattern of behaviours and sensory scores; all other variables had normal distributions in their original format.

Assumptions for multiple regression analyses were tested for all models. The Durbin-Watson statistic did not deviate far from 2 (average = 1.88), the VIF values did not deviate far from 1

(1.02-1.88), the tolerance statistics were well above 0.2 (0.53-.99) and the standardised residuals were normally distributed as tested by Shapiro-Wilk statistics (Field, 2009).

The first set of analyses entered all of the variables described above into univariate correlation analyses and as can be seen in Table 4-2 all but two of these variables were significantly correlated with the sensory behaviours as measured in the DISCO. The following analyses investigated whether these significant correlations remained in multivariate analyses.

**Table 4-2: Table showing the correlational analyses (r) of all predictor variables and their univariate relations with sensory behaviours in Sample 2**

| | sex | IQ | Age | Com | RB | social |
|---|---|---|---|---|---|---|
| Sensory | -.030 | -.247** | -.154* | .298** | .399** | .516** |

| | DLS | Mal | sleep | emotion | pattern | motor |
|---|---|---|---|---|---|---|
| Sensory | .249** | .579** | .261** | .115 | .529** | .184** |

#### *4.2.1.3.1 Aim 1: To examine the role of age, gender and ability level in the presentation of sensory behaviours*

Age, IQ and Gender were all entered into a multivariate linear regression, the total sensory score was used as the dependent variable. The three variables alone were entered in Block 1 and then the interaction between the variables (three two-interactions and one three way) were entered in Block 2. The interaction terms were created by converting gender and IQ (previously coded as 0,1) into -0.5 or 0.5 and multiplying by each other or with a centred score for age. This was done by standardising the age variable as distance from the mean; all interaction term variables were centred before multiplication as suggested by Aiken and West (1991). All assumptions for multiple regression were met. No interactions were found to be significant and therefore were not presented here. The model with age, IQ and gender predicted 7.9% of the variance in sensory behaviours scores and IQ was the only significant predictor as shown in Table 4-3.

**Table 4-3: Table showing the multiple regression analyses of gender, age and IQ predicting sensory behaviours**

| | B | SE B | β |
|---|---|---|---|
| **Step 1** | | | |
| Constant | 1.565 | .018 | |
| Gender | -.004 | .016 | -.020 |
| Age | .000 | .000 | -.139 |
| IQ | -.039 | .012 | -.227** |

#### *4.2.1.3.2 Aim 2: To look for specific associations between the core features of ASD as measured by ICD-10 and sensory behaviours*

The results of the hierarchical multiple regression can be seen in Table 4-4. IQ was significant in block one and remains a significant predictor when the core features were added, repetitive behaviours and social interaction but not communication scores were also significant predictors. Model 1 predicted 7.9% (adjusted 6.4%) but the addition of the core features of ASD improved the variance accounted by the Model 2 to 37.5% (adjusted 35.5%).

**Table 4-4: Table showing the hierarchical multiple regression analysis results for age, gender, IQ and the core features of ASD as predictor variables of sensory behaviours score.**

| | B | SE B | β |
|---|---|---|---|
| **Step 1** | | | |
| Constant | 1.565 | .018 | |
| Gender | -.004 | .016 | -.020 |
| Age | .000 | .000 | -.139 |
| IQ | -.039 | .012 | -.227** |
| IQ * Gender | | | |
| **Step 2** | | | |
| Constant | 1.433 | .021 | |
| Gender | -.025 | .013 | -.113 |
| Age | .000 | .000 | -.139 |
| IQ | -.025 | .011 | -.142* |
| Com | .004 | .003 | .090 |
| RB | .092 | .021 | .289*** |
| Social | .033 | .007 | .348*** |

*Note:* $R^2$ = .079 for Step one (F(3,186)= 5.339, p<.01), $\Delta R^2$ = .296 (F(3,183) = 28.920, p<.001), *p<.05, **p<.01, ***p<.001

It was hypothesised that the reason communication was no longer a significant predictor of sensory behaviours in a multivariate analysis was the combination of social and communication variables being entered, indeed when social interaction is removed, communication does remain a significant predictor as shown in Table 4-5. However, this model only explains 29% (27% adjusted) of the variance in sensory behaviours compared to 37.5% when social is included.

**Table 4-5: Table showing the hierarchical multiple regression analysis results for age, gender, IQ, repetitive behaviours and communication as predictor variables of sensory behaviours score.**

| | B | SE B | β |
|---|---|---|---|
| **Step 2** | | | |
| Constant | 1.463 | .022 | |
| Gender | -.015 | .014 | -.069 |
| **IQ** | -.045 | .011 | -.259*** |
| Age | .000 | .000 | -.124 |
| **Com** | .007 | .003 | .171** |
| **RB** | .124 | .021 | .390*** |

#### *4.2.1.3.3 Aim 3: To look for specific associations between sensory and the other associated features of ASD: maladaptive, emotion, daily living skills (adaptive behaviour) and pattern of activities*

The results of the hierarchical multiple regression can be seen in Table 4-6. The addition of the associated features improved the variance accounted for by the model to 44.9% (41.8% adjusted). In the second step, IQ was no longer a significant predictor, only maladaptive behaviour and pattern of activities scores were significant predictors.

**Table 4-6: Table showing the hierarchical multiple regression analysis results for age, gender, IQ and the associated features of ASD as predictor variables of sensory behaviours score.**

| | B | SE B | β |
|---|---|---|---|
| **Step 1** | | | |
| Constant | 1.565 | .018 | |
| Gender | -.004 | .016 | -.020 |
| Age | .000 | .000 | -.139 |
| IQ | -.039 | .012 | -.227** |
| IQ * Gender | | | |
| **Step 2** | | | |
| Constant | 1.132 | .056 | |
| Gender | -.015 | .013 | -.070 |
| Age | .000 | .000 | -.008 |
| IQ | -.001 | .011 | -.005 |
| DLS | .001 | .000 | .128 |
| Sleep | .008 | .010 | .048 |
| Emotion | .003 | .013 | .014 |
| Maladaptive | .006 | .001 | .391*** |
| Pattern | .257 | .060 | .298*** |
| Motor | .006 | .011 | .034 |

*Note:* $R^2$ = .079 for Step one (F(3,186)= 5.339, p<.01), $\Delta R^2$ = .370 (F(7,179) = 17.164, p<.001), *p<.05, **p<.01, ***p<.001

#### *4.2.1.3.4 Aim 4: To address whether the core ASD features of associated features account for more of the variance in sensory behaviours by entering all the variables into one analysis to control for the effects of all variables*

To explore the relative contribution of the core and associated features of ASD and their predictive value of total sensory behaviour score, two hierarchical multiple regressions were run. In both models the first block contained the individuals' characteristics (age, IQ and gender). In the first regression, the core features of ASD were entered in the second block and the associated in the third whereas in the second regression model the associated features were entered first. The results of the final block for both models (all variables entered) can be seen in Table 4-7. In total the model explained 54% of the variance in sensory behaviours (50.6% when $R^2$ adjusted). When all variables were entered into the model the previous significant predictors remained: repetitive behaviours, social interaction, behaviours affecting others and pattern of activities. In stage three of both models the addition of either the associated features ($\Delta R^2$ step 3 = .165 ($F(7,176) = 8.994$, $p<.001$) or the core features ($\Delta R^2$ step 3 = .091 ($F(3,176) = 11.593$, $p<.001$) significantly improved the model and highlighted that both aspects were associated with sensory behaviours.

In addition, the inclusion of interaction terms between the core and associated features were added in a fourth step, however, ($\Delta R^2$ step 4 = .033 ($F(7,170) = 1.762$, $p= .098$) these variables did not significantly improve the model fit and no interactions terms were significant.

**Table 4-7: Table showing the final step of the hierarchical multiple regression analysis results for age, gender, IQ, the core and associated features of ASD as predictor variables of sensory behaviours score.**

| | B | SE B | β |
|---|---|---|---|
| **Step 3** | | | |
| Constant | 1.170*** | .052 | |
| Gender | -.022 | .012 | -.102 |
| Age | .000 | .000 | -.004 |
| IQ | -.004 | .010 | -.024 |
| Communication | .004 | .003 | .103 |
| RB | .076 | .019 | .239*** |
| Social | .014 | .007 | .143* |
| DLS | .000 | .000 | .060 |
| Sleep | .009 | .009 | .052 |
| Emotion | -.006 | .012 | -.026 |
| Maladaptive | .005 | .001 | .341*** |
| Pattern | .193 | .057 | .223** |
| Motor | -.007 | .011 | -.037 |

*p<.05, **p<.01, ***p<.001

#### *4.2.1.4 Discussion*

Study 1 explored the presence of sensory behaviours across age, gender and ability level and their relation to both the core features of ASD and the other associated features measured by the DISCO. It was found that in univariate analyses IQ and age are significantly associated with sensory score but in a model with age, IQ and gender, only IQ remains a significant predictor. Furthermore, all three core features are related to sensory behaviours, however, in a model with all three – sensory is not predicted by communication. This is due to the relation with social interaction; in a model where only repetitive behaviours are held constant and social interaction removed communication remains a significant predictor of sensory behaviours but the model with all three core features explains the best amount of variance.

In terms of associated features, daily living skills, maladaptive behaviours, sleep problems, pattern of activities and motor impairments are univariately significantly associated with sensory behaviours, however, in a model only maladaptive behaviours and pattern of activities are significant predictors. Both the core and associated features add significant explanation to the model predicting sensory behaviours and when all variables are included in the model repetitive behaviours, social interaction, maladaptive behaviours and pattern of activities remain significant predictors. As found in Chapter 3, this highlights the importance of considering relations between variables within a multivariate context, the theoretical and clinical implications of these results are presented in general discussion (section 4.3).

The analyses conducted in Study 4.2.1 explored the associations within multivariate models in which the effects of core and associated features can be controlled for. However, the introduction also highlighted the need for further work on sensory behaviours in adults. The DISCO sensory items also have the potential to address these limitations as the content of the DISCO was designed to be relevant across all ages. The second study explores the potential of these items to be converted into a self-report questionnaire to be used in adults with ASD.

### *4.2.2 Study 4.2.2*

Study 2 designed and tested the reliability of a self-report questionnaire using the sensory items from the DISCO. This was done for two reasons. The first aim was to overcome a limitation of the research conducted so far on sensory behaviours in ASD which has been predominately collected using parental report about children's sensory responses; there is a lack of research on adults and very limited work using self-report measures. The second aim is to provide some external validity of

the items used in the DISCO in comparison to tools already used in the literature on sensory behaviours in ASD.

Previous questionnaires have adapted items from the DISCO. Both the Repetitive Behaviour Questionnaire-2 (RBQ-2; Leekam et al., 2007) and the Activities and Play Questionnaire-Revised (APQ-R; Honey et al., 2006) utilised DISCO items and converted them into parent/caregiver-report questionnaire items. Both attempts were successful with 18 items from the RBQ-2 being taken or adapted from the DISCO and resulting in a reliable measure of repetitive behaviours in children (Cronbach's alpha = .85). In the APQ-R, seven sensory DISCO items and seven play DISCO items were adapted and made up part of a reliable tool to measure both Play and Repetitive behaviours with all Cronbach's alpha levels across ASD and TD children above 0.84. No DISCO items have yet been converted into self-report questionnaire measures.

From all reports of the prevalence of sensory behaviours in ASD it is apparent that there is a lack of information regarding sensory behaviours in adults and a reliance on parent reports rather than first person accounts.

#### *4.2.2.1 Development of the questionnaire*

This was conducted in four steps:

1. All the DISCO items identified by Leekam et al (2007) as having sensory properties were selected for inclusion in the questionnaire and presented in the same format as the Adult and Adolescent Sensory Profile.
2. Interviews were conducted with three researchers experienced in ASD and a Parent Consultant at the Wales Autism Research Centre and the questionnaire in the original format was given to two males with a diagnosis of ASD to provide feedback
3. Feedback from the interviews and individuals with ASD was adopted and resulted in the following changes:
    - The layout of the questionnaire was improved; different boxes for different groups of items were used to break up the text and these boxes were spread across pages so individuals were not overwhelmed with text on each page.
    - Items that could cause offence or distress to individuals being asked about their current behaviour were removed. These included all behaviours labelled as "mixed" by Leekam et al (2007): self-injury, self-stimulation, smearing, over-breathing. These items are appropriate in the DISCO as parents or caregivers are asked rather than the individual themselves.

4. The final questionnaire was called the Sensory Preferences Questionnaire (SPQ) and was used in the following study. The items in the final SPQ can be seen below in Table 4-8 and the full layout and formatting can be found in Appendix 4.

**Table 4-8: Table showing the DISCO sensory items converted into self-report format for the Sensory Preferences Questionnaire**

| Number | Code | Question |
|---|---|---|
| 1 | Audist | I am upset by some sounds that do not affect other people (e.g. vacuum cleaners, aeroplanes, fire engines or road drills) |
| 2 | Audfas | I have an unusual interest in some sounds (e.g. bells, water hissing in pipes, records played at the wrong speed) and I spend time listening to these sounds |
| 3 | Hearac | I have unusually acute hearing (e.g. I can hear the jangle of car keys, the rustle of a sweet paper or a quiet sound from a long distance away) |
| 4 | Lights | I am unusually interested in shiny lights (e.g. silver paper, tinsel, patches of sunlight, street lights or lights on a motorway) |
| 5 | Twisth | I get unusually excited at seeing things spin |
| 6 | Spinvis | I twist or flick my hands or objects near to my eyes |
| 7 | Angles | I like to look at objects from many different angles for no obvious reason or I examine objects such as lines of toy trains, by eyeing them closely on their level, perhaps kneeling down or lying on the floor to do this) |
| 8 | Touch | I have an unusual interest in the feel of surfaces (e.g. fur, velvet, hair or smooth or rough surfaces) |
| 9 | Scratch | I scratch or tap on different surfaces in order to feel the sensation. |
| 10 | Gentle | I react negatively if gently touched (e.g. if someone softly touches my arm or shoulder) |
| 11 | Firm | I react negatively to being held firmly or tightly (e.g. if someone hugs me). |
| 12 | Manip | I flick things like pieces of string or sticks, tap two objects together or roll pieces of cotton in my fingers because I like the sensation. |
| 13 | Wash | I dislike or resist being washed, having my hair washed, my nails cut or my hair cut. |
| 14 | Sticky | I am bothered by having dirty or sticky hands or dislike handling Play-Doh, sand, glue or other messy materials |
| 15 | Smell | I tend to explore people or objects by smelling them |
| 16 | Faddy | I have very unusual food fads (e.g. I will only eat marmite sandwiches or I will insist on one brand of drink only) |
| 17 | Mouth | I tend to put inappropriate objects in my mouth or I swallow inedible objects (e.g. cigarette ends, small pieces of metal or paper) |
| 18 | Lump | I refuse to eat food that is lumpy or hard to chew |
| 19 | Spun | I enjoy being spun round or going on roundabouts, more than other people my age |
| 20 | Spin | I like spinning round or running round in circles, more than other people my age |
| 21 | Pain | I DO NOT react to unpleasant or painful sensations (e.g. cuts and bruises, toothache, sore throat or broken bones) |

As described in the introduction, at the time of study design there were two instruments that adopt a self-report format, which the SPQ could be compared with. The Adult and Adolescent Sensory Profile (AASP) was chosen over the Highly Sensitive Person (HSP) scale as it was more relevant to sensory behaviours rather than the wider social and environment nature of the HSP scale. At the time of study design (2009), the Glasgow Sensory Profile (GSP) had not been published, however, it was designed according to the sensory behaviours reported in ASD and was therefore the most relevant to compare the SPQ to so an additional sample of typically developing adults were recruited at a later date to compare the information gathered from the GSP and the SPQ.

In addition, the Autism Spectrum Quotient (AQ: Baron-Cohen, Wheelwright, Skinner, Martin, & Clubley, 2001), a short, self-administered tool which determines the degree to which someone has traits similar to those of individuals on the autism spectrum was also given to the same participants as it allows measurement of whether sensory responses on the SPQ are related to individual's social functioning. Both the Glasgow Sensory Profile and the AASP have been shown to have significant correlations with the AQ and if the SPQ is also significantly correlated it would add to the convergent validity of the tools. In addition the AQ was included in the study to confirm an individual's group membership of ASD or TD using the number of traits present (see section 4.2.2.2.1.2 below). The comparative group analyses assess the frequency and severity of sensory behaviours in high functioning adults with ASD in comparison to typically developing adults.

This study utilised two samples of individuals. The final assessments given to the two samples are summarised in Table 4-9. Sample A was recruited by the author for this study to test the reliability of the SPQ as well as to compare diagnostic groups on their sensory preferences. However, the questionnaire measures were collected as part of a wider study to investigate the neurobiology of sensory symptoms using brain imaging techniques ("tactile sensitivity in ASD" McGonigle & Leekam, funded by the Waterloo Foundation). Participants were first recruited for the questionnaire study and referred on to the brain imaging project if they were interested and met the criteria (right handed males with no current or history of epilepsy). Participants recruited in the current questionnaire study were therefore limited to individuals capable of taking part in both studies if interested and therefore limited to high-functioning males above the age of 18 years. Adjustments could have been made if individuals not meeting these criteria wanted to take part in the questionnaire study but were not targeted in recruitment. Secondary data were also used to validate the SPQ from a study, also carried out at the Wales Autism Research Centre by a final year undergraduate student supervised by Prof Leekam (Catherine Walters). Data collected in this project (2012-2013) came from a sample of typically developing undergraduates and this allowed comparison of the Sensory items from the DISCO with the Glasgow Sensory Profile in typically

developing individuals (Sample B). Both studies were given ethics approval from Cardiff University, School of Psychology ethics committee. The method and results for the two samples are presented separately below.

**Table 4-9: Table showing the measures given to participants in Samples A and B of Study 4.2.2**

| Sample A | Sample B |
|---|---|
| SPQ | SPQ |
| AASP | GSP |
| WASI | |
| AQ | |

### *4.2.2.2 Sample A: Comparing the SPQ to the AASP and AQ and examining group differences on the SPQ.*

#### *4.2.2.2.1 Methods*

##### *4.2.2.2.1.1 Participants:*

All participants were male, over the age of 18 and capable of providing their own consent to participate. Thirty participants with ASD were recruited and 22 typically developing adults.

##### *4.2.2.2.1.2 Recruitment methods:*

###### *4.2.2.2.1.2.1 Individuals with an ASD:*

Contacts were made with specialist colleges and schools, however, the majority of ASD participants were recruited through student support services at universities in South Wales and parent support groups in Cardiff and surrounding local areas. Individuals in the ASD group were recruited if they had a clinical diagnosis of Autism of Asperger Syndrome and were asked to confirm this on the consent form, however, no other independent measure of an individual's ASD symptoms (such as a diagnostic interview or observation schedule) were used to confirm this diagnosis. This was due to the protocols devised for the wider study. The Autism Spectrum Quotient (AQ; Baron-Cohen et al., 2001) was included as both a measure of individuals' everyday functioning but also to act as a broad indication of whether individuals were correctly classified in the right group.

###### *4.2.2.2.1.2.2 Typically developing individuals*

Participants for the neuroimaging study (n=15) were recruited through the university. Emails were sent to the Cardiff University Brain Imaging Centre (CUBRIC) database to participants signed up to hear about brain imaging studies being run and individuals were also recruited through events held by the Wales Autism Research Centre and email lists. An additional seven participants

were recruited for the questionnaire study only to act as an additional comparison with the ASD participants, these individuals were recruited through the Cardiff Electronic Management System for recruitment of undergraduate students or individuals signed up to take part in research in the School of Psychology and posters were displayed in the department. The figure below demonstrates the participants recruited in both groups.

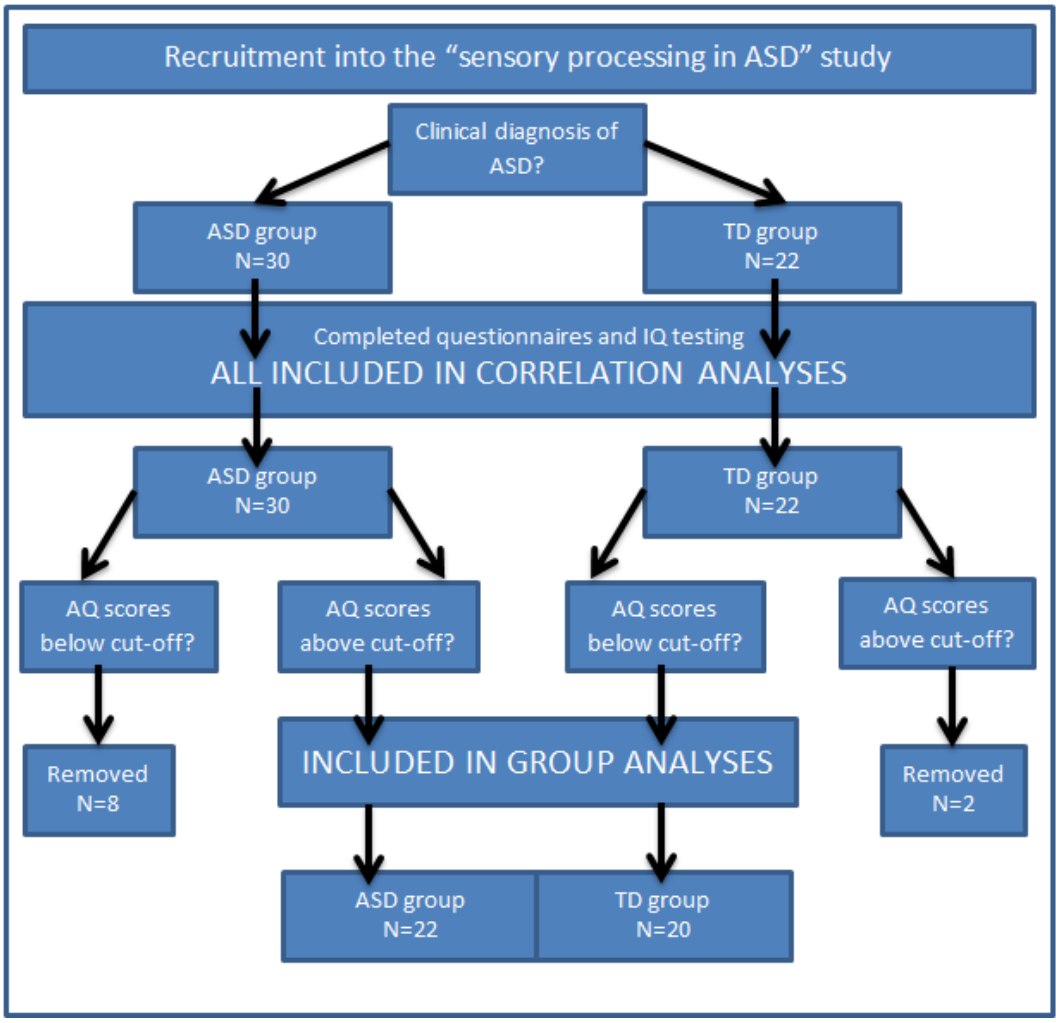



**Figure 4-1: Flowchart showing the recruitment of individuals into Sample 1 that was used to test the Sensory Preferences Questionnaire**

#### *4.2.2.2.1.3 Measures*

##### *4.2.2.2.1.3.1 WASI:*

The Wechsler Abbreviated Scale of Intelligence (Wechsler, 1999) based on the Wechsler Adult Intelligence Scale–Third Edition (Wechsler, 2008) was used in order to ensure IQ was matched across the ASD and TD groups in the study as well as to ensure individuals had a sufficient level of understanding to have completed the written self-report questionnaires reliably. The WASI was

designed as a reliable short form version of the adult intelligence scales and consists of four subtest, two verbal and two performance which allows IQ scores to be calculated for total, verbal and performance IQ based on the age of the individual (according to typically developing norms; Wechsler, 1999). Verbal IQ was assessed using the Vocabulary subtest, which scored individuals definitions to words, and the similarities subtest which asked individuals to describe how two words are alike (e.g. "chocolate and cookies" or "capitalism and socialism"). Performance IQ was tested using the Block Design subset in which individuals were required to copy patterns made of blocks with different coloured sides and the Matrix Reasoning subtest where individuals were required to select the missing part of a visual pattern from a selection of options. The WASI manual states that the full-scale IQ can be measured in 30 minutes in the standardisation sample, however, this differed across individuals and an hour slot was used to administer the WASI. All WASI tests were conducted by the author who was familiar and had used the WASI before testing in this study. The IQ test was presented to individuals as a series of quizzes and puzzles that are used to assess your strengths and weaknesses. Two individuals from the TD group were unable to be IQ tested. One individual could not be tested due to location and one was too familiar with the Wechsler intelligence measures. An additional individual with ASD was unable to be IQ tested as he completed questionnaires first and did not want to continue but was happy for his results to be included.

*4.2.2.2.1.3.2 The Adult/Adolescent Sensory Profile (ASSP):*

The Adult/Adolescent Sensory Profile (Brown & Dunn, 2002) is a 60-item questionnaire probing sensory behaviours. Each item asks how often the respondent performs a particular behaviour with answers "Almost Never", "Seldom", "Occasionally," "Frequently" and "Almost Always'' scored 1, 2, 3, 4 and 5 points respectively. For example "I dislike having my back rubbed" or "I like to go to places that have bright lights and that are colourful."

*4.2.2.2.1.3.3 The Sensory Preferences Questionnaire:*

The design of the SPQ is described above and the items included presented above. It contains the items from the DISCO (21 items) that have been identified as sensory behaviours. The scoring system was developed from the five point Likert scales in the AASP, this asks individuals how frequently they undertake this behaviour or feel this way from "almost never" or "1" to "almost always" or "5". The SPQ consists of measures of visual, auditory and proximal symptoms. In addition the proximal symptom scales is made up of items on smell/taste, other oral, touch and kinaesthetic behaviours. The reliability of the SPQ will be tested in the study.

Two scores can be gathered by the Sensory Preferences Questionnaire. One method followed the Sensory Profile and used the total score, adding the raw score for each item, which correspond to the scale (1-5, almost never to almost always). The second method used the

approach adopted in the DISCO, the design of the SPQ was also completed so that to two codes "frequently" and "almost always" could be collapsed to create a "marked" code equivalent to the scoring system in the DISCO. Therefore, scores were also recoded in this manner and the number of "marked" scores were counted to create a total severity score for the SPQ. SPQ therefore has a total severity score (SPQseverity) and a total count score (SPQtotal).

*4.2.2.2.1.3.4 Autism Spectrum Quotient:*

The Autism Spectrum Quotient (AQ; Baron-Cohen et al., 2001) is a short, self-report questionnaire, which determines the degree to which someone has traits similar to those of individuals on the autism spectrum. The questionnaire is made up of five scales of behaviour: social skills; attention switching; attention to detail; communication; and imagination. The items from each scale were selected from symptoms in the literature, international classification systems as well as research on cognitive abnormality in autism (Baron-Cohen et al., 2001). The AQ has been found to be reliable and valid (Baron-Cohen et al., 2001; Hurley, Losh, Parlier, Reznick, & Piven, 2007; Woodbury-Smith, Robinson, Wheelwright, & Baron-Cohen, 2005) and can distinguish between individuals with typical development and those with autism (Baron-Cohen et al., 2001) or Asperger Syndrome (Woodbury-Smith et al., 2005) using cut-offs on the AQ.

Group comparisons were only made between individuals whose AQ scores and diagnosis aligned, cut-offs for the AQ have been proposed as 32 for diagnosis (Baron-Cohen et al., 2001) and 26 for screening (Woodbury-Smith et al., 2005). These cut-offs were used to define group comparison with TD individuals scoring above 26 removed from analyses and individuals with ASD scoring below 32 removed from the analysis. Ethics guidelines meant participants did not have to be informed about their AQ scores or cut-offs as this is a measure of autistic traits that are present throughout the population. Although, the AQ is published as a screening tool, it is not a diagnostic tool.

*4.2.2.2.1.4 Analytic strategy:*

The first analyses took Sample A as a whole to assess the internal consistency of the Sensory Processing Questionnaire (SPQ) using Cronbach's alpha level and ensured all items were correlated with the total score on the SPQ.

Correlations between the SPQ and AASP as well as the AQ were conducted across all individuals in Sample A (N=52). Correlations were also conducted between sub-scale scores on the SPQ (visual, auditory, proximal) and AQ sub-domains, in order to investigate if sensory behaviours measured by the SPQ were related to any area of autistic traits in particular. The total SPQ score was also correlated with the AASP scores on the four quadrants to investigate how well these concepts were covered by the SPQ. Finally, both the sensory measures have modality scores and the

scores for each modality of each measure was entered into correlational analyses to address how well the SPQ items cover the modalities in the AASP.

#### *4.2.2.2.1.5 Group Assignment:*

The groups were divided into ASD or TD on both their clinical diagnosis and score on the AQ, anyone scoring above the screening or diagnostic cut-off in the TD group was excluded (N=2) and any ASD participants scoring below the diagnostic or screening cut-off were removed from the group by group analyses (N=8). Group differences were assessed using univariate statistics with Bonferroni corrections applied for multiple comparisons.

The results section was designed to include the following analyses:

- assess the internal consistency of the SPQ
- analyses the correlations between the total scores on the SPQ, AASP and AQ (total and sub-domains)
- correlate the total SPQ score with the AASP quadrants
- compare the modality scores for the SPQ and AASP using correlational analyses
- compared the ASD and TD groups on scores across the total and modality SPQ scores

### *4.2.2.2.2 Results*

Tests of normality were conducted in the sample as a whole and in the ASD and TD groups separately. The total scores for each instrument (SPQ, AQ, AASP) were all normally distributed and therefore pearson's r correlation was used, however, the SPQ severity score and the majority of the sub-domain scores across all three measures were not normally distributed (significant Kolmogorov-Smirnov tests) and therefore correlations using these variables used Spearman's rho correlation.

The SPQ had excellent overall reliability with a Cronbach's alpha value of .89. In addition, the removal of any item did not significantly improve Cronbach's alpha and all items inter-correlated above .3. None correlated higher than .75 and only three pairs above 0.7: dislikes being washed correlated with auditory distress at .706 and mouthing of objects at .706 and negative reaction to gentle touch correlated with negative reaction to firm touch (.746 ). Removal of any of these items reduced the Cronbach's alpha and so all were retained for the following analyses. The modality scores of the SPQ were strongly related to each other and the total SPQ scores as shown in Table 4-10.

**Table 4-10: Table showing the correlational analyses between the total and severity score of the Sensory Preferences Questionnaire (SPQ) with the total scores on each modality of the SPQ in Sample A.**

| | SPQtotal | SPQseverity | Auditory | Visual | Proximal |
|---|---|---|---|---|---|
| SPQtotal | 1 | .950** | . 905** | . 757** | . 959** |
| SPQseverity | | 1 | . 883** | . 727** | . 893** |
| Auditory | | | 1 | . 625** | . 834** |
| Visual | | | | 1 | . 591** |
| Proximal | | | | | 1 |

The correlations between the SPQ total and severity scores in comparison to the total AASP score and total AQ score can in seen in Table 4-11. Both scores from the SPQ were significantly highly correlated with the Adult/Adolescent Sensory Profile and significantly related to the total AQ score. Total score on the SPQ was not correlated with verbal (.064, p=.664), performance (-.197, p=.175) or full scale IQ (-.080, p=.586).

**Table 4-11: Table showing the correlational analyses (r) between the total and severity score of the Sensory Preferences Questionnaire with the total scores on the Sensory Profile and the Autism Quotient**

| | SPQtotal | SPQseverity | Sensory Profile | AQ |
|---|---|---|---|---|
| SPQtotal | 1 | .950** | .819** | .567** |
| SPQseverity | | 1 | .807** | .599** |
| Sensory Profile | | | 1 | .728** |
| AQ | | | | 1 |

To assess whether the SPQ covers the same information as the AASP correlations between the total SPQ score and the AASP quadrant scores were conducted (Table 4-12). These revealed that the SPQ had strong correlations with sensory seeking, sensory avoidance, sensory avoidance but not with the low registration quadrant (-.229, p=.103).

**Table 4-12: Table showing the correlation (r) between the SPQtotal score and the quadrant scores on the Sensory Profile**

| | Sensation seeking | Low registration | Sensory sensitivity | Sensation avoiding |
|---|---|---|---|---|
| SPQtotal | .686** | -.229 (p=.103) | .748** | .743** |

In addition, the modality scores for the AASP and the SPQ showed strong correlations: the AASP smell correlated with APQ smell/taste (.383, p<.01) and SPQ other oral (.348, p<.01); AASP

movement scale (.344, p<.01) and activity scale (.328, p<.01) correlated with SPQ kinaesthetic score; the AASP and SPQ visual (.436, p<.001), auditory (.696, p<.001) and touch (.328, p<.01) scores all correlated.

#### *4.2.2.2.2.1 Group differences*

The ASD (diagnosis and AQ scores above 32, N=22) and TD groups (no diagnosis and AQ scores below 26, N=20) described in the methods sections were compared on total and sub-scale scores. Kolmogorov-Smirnov tests were run for all variables in the ASD and TD groups, significant test statistics were found in one or both groups for age, performance IQ, SPQseverity, all the SPQ severity sub-scales and the majority of SPQ total sub-scales and all but one sub-scale for the AQ. Therefore, all sub-scale analyses were conducted using non-parametric tests, whereas tests for the total scores across the measures were parametric (excluding the SPQseverity score).

The AQ was used to confirm group membership, and therefore, significant differences were found between the groups for total AQ score (t(36) = 15.05, p<.001) and each sub-scale: social (U = 5.5, r = z = -5.15, p<.001); attention switching (U = 18.5, z = -4.76, p<.001); attention to detail (U = 72.5, z = -3.167, p<.001), communication (U = 3.0, z = -5.22, p<.001) and imagination (U = 12.5, z = -4.94, p<.001). But there were no significant differences across age (U = 178, z = -.06, p=.95, n.s.) or IQ (Full Scale: t(33) = -1.23, p=.23, n.s; Verbal IQ: T(33) = -.07, p=.94, n.s; Performance IQ: U = 96.5, z = -1.87, p=.06, n.s.).

The total Sensory Preferences Questionnaire score was significantly higher in the ASD (Mean = 54.83; sd = 14.43) than TD group (Mean = 33.8; sd = 10.07; t(36) = 5.25, p<.001). The SPQ severity score was also significantly higher in the ASD (Mean = 7.22; sd = 4.18) than the TD group (Mean = 1.35; sd = 1.98; U = 35, z = -4.32, p<.001).

The means and standard deviations of the total and modality scores are shown in Figure 4-2. The ASD group had significantly more symptoms in each SPQ sub-domain than the TD group for: proximal (U = 43, z = -4.01, p<.001); and auditory (U = 41, z = -4.09, p<.001) but not visual (U = 107, z = -2.16, p=.033) when controlling for multiple comparisons. In addition, the ASD group scored significantly higher than the TD group for the breakdown of proximal category into Touch (U = 39.5, -4.12, p<.001) but not for the separate scales of smell/taste (U = 122.5, z = -1.74, p=.093), other oral (U = 112, x = -2.22, p=.028), kinaesthetic (136.5, z = -1.44, p=.151) or indifference to pain (115, x = -1.99, .059).

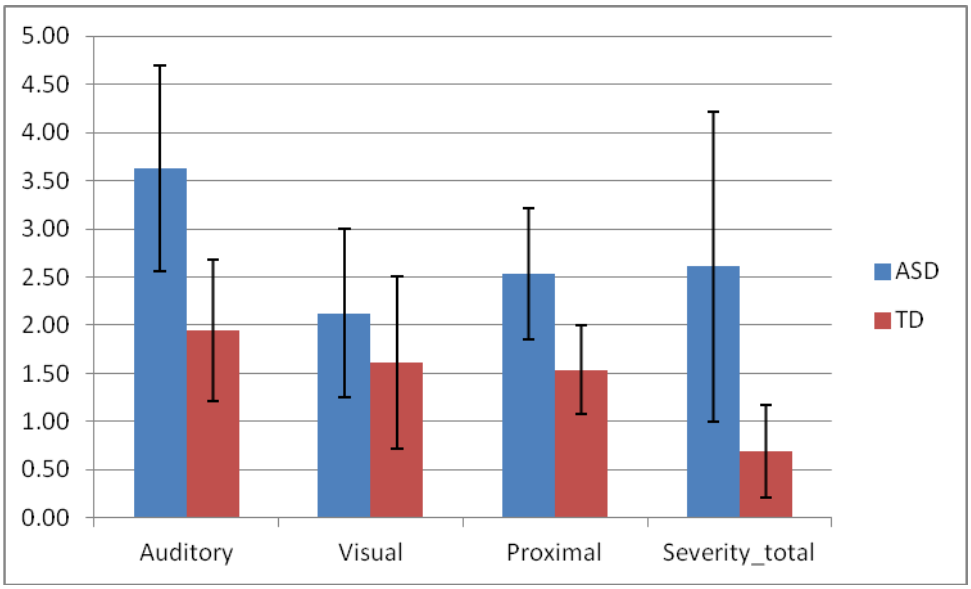



**Figure 4-2: Figure showing mean score for each item the auditory, visual, proximal and total scores on the Sensory Preferences Questionnaire for individuals in the ASD and TD groups of Sample A. Error bars +/- 1 sd.**

Further analyses were conducted to identify if any SPQ items alone significantly differentiated between the ASD and TD groups with Bonferroni correction applied for multiple comparisons. The ASD group scored significantly higher than the TD group on all three of the auditory items (A1, auditory distress, U=22, z=-4,78, p<.001; A2, auditory fascination, U=78.5, z=-3.06, p<.01; and A3, acute hearing, U=90.5, z=-2.67, p<.01) and four of the seven touch symptoms (T3, reaction to gentle touch, U=65.5, z=-3.49, p<.001; T4, reaction to firm touch, U=79.5, z=-3.05, p<.001; T6, dislikes being washed U=62.5, z=-3.91, p<.001) and T7, dislikes sticky hands, U=69.5, z=-3.30, p<.001).

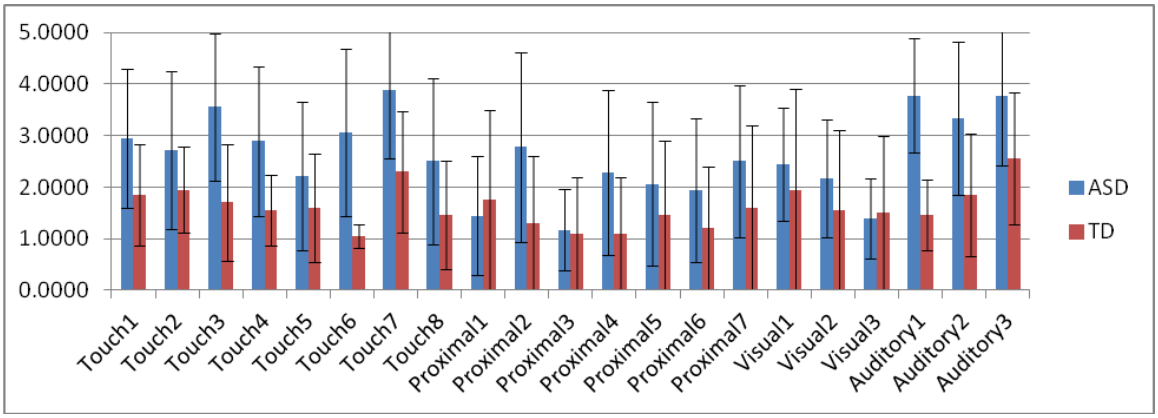



**Figure 4-3: Figure showing the mean scores on each item of the Sensory Preferences Questionnaire for the ASD and TD groups in Sample A. Error bars +/- 1 sd.**

Finally analyses identified the range of scores on the SPQ. Table 4-13 shows the number of marked ("almost always" or "frequently") sensory behaviours. In the ASD group, 94.4% scored to a marked degree on at least one item and 88.8% on more than one, the most marked items is 17 whereas in the TD group, 46% scored one marked behaviour and 36% more than one, the most marked behaviours was six. Furthermore, in the ASD group 16.7% had a marked impairment in one modality, 38.9% in two modalities and 38.9% in three modalities. In the TD group this was 20%, 15% and 10% respectively. In the ASD group 83.3% get at least one marked auditory behaviour and 38.9% score marked on all three auditory items (in TD, 25% get at least one), for visual symptoms 83.9% of the ASD group 38.9% get one marked visual symptom (TD 20%) and 88.9% of the ASD score on one proximal symptom and 83.3% score on more than one marked proximal (TD, 35% get one).

**Table 4-13: Table presenting the number of "marked" sensory behaviours in individuals with ASD and in the typically developing comparison group.**

| Number of "marked" items | ASD | | TD | |
|---|---|---|---|---|
| | Frequency | Percent | Frequency | Percent |
| 0 | 1 | 5.6 | 11 | 55 |
| 1-5 | 4 | 22.3 | 7 | 35 |
| 6-10 | 10 | 55.6 | 2 | 10 |
| 11-15 | 2 | 11.1 | 0 | 0 |
| 15-20 | 1 | 5.6 | 0 | 0 |
| Total | 18 | 100 | 20 | 100 |

## *4.2.3 Sample B*

### *4.2.3.1 Methods*

#### *4.2.3.1.1 Participants:*

Sample B was a secondary dataset. The data were collected by a final year project student (Catherine Walters) at Wales Autism Research Centre, Cardiff University supervised by SL. Fifty (6 male; 44 female) undergraduate Psychology students were recruited through the department's Electronic Management System for research participation. Participants completed a battery of questionnaires including the Sensory Preferences Questionnaire and the Glasgow Sensory Profile as measures of sensory symptoms, the Autism Quotient as well as the Hospital Anxiety and Depression Scale (Zigmond & Snaith, 1983). Individuals had a restricted age range (19-25, mean = 20.76 years old, sd = 1.44).

#### *4.2.3.1.2 Measures:*

The SPQ was used again.

#### *4.2.3.1.2.1 The Glasgow Sensory Questionnaire*

The Glasgow Sensory Questionnaire (Robertson and Simmons, 2012) has 42 items probing abnormal sensory behaviours. Each item asks how often the respondent performs a particular behaviour with answers ‘‘Never’’, ‘‘Rarely’’, ‘Sometimes’’, ‘‘Often’’ and ‘‘Always’’ being scored 0, 1, 2, 3 and 4 points respectively on items such as “Do you find certain noises/pitches of sound annoying?” and “ Do bright lights ever hurt your eyes/cause a headache?”

### *4.2.3.2 Results:*

Again preliminary analyses revealed total scores were normally distributed (persons correlations) but sub-scale scores were not normally distributed and so Spearman’s rank correlations were used. The total SPQ score was correlated with the total GSP (.826, p<.001), the Hyper – responsive scale of the GSP (.775, p<.001) and the hypo-responsive scale of the GSP (.762, p<.001).

Additional analyses found that the modality scores of the two measures also correlated. The GSP and SPQ auditory (.337), visual (.529), touch (.407) scores all correlated as did the GSP vestibular with the SPQ kinaesthetic (.624), the GSP olfactory with the SPQ smell/taste (.525) and the GSP gustatory with the SPQ other oral (.413).

### *4.2.3.3 Discussion Samples A and B*

Study 2 investigated whether the Sensory items from the DISCO have external validity and how they compare with the existing tools used in the literatures. Work for this chapter, also examined whether these items could be successfully converted into a self-report questionnaire and whether these sensory symptoms were more present in individuals with ASD in a sample of high-functioning adults with ASD and age and IQ matched typical control groups.

In Sample A, the SPQ was found to be valid and reliable. It also had good external validity as showed by high correlations with the Adolescent and Adult Sensory Profile, which is part of a family of sensory processing measures that are the most frequently used in the literature. In addition, it was found that 94% of high-functioning adults with ASD reported at least one marked sensory behaviour and 88.8% more than one, in addition individuals with ASD had significantly higher SPQ scores than IQ matched typically developing individuals. The results of Sample B, also found the SPQ to have high correlations with another recently published self-report questionnaire, the Glasgow Sensory Profile (GSP) in a sample of predominately female undergraduate students.

The results from Study 4.2.2 indicated that the sensory items in the DISCO are valid and reliable when used in a self-report format and showed good concordance with existing sensory

questionnaires. This strengthens the results from Study 4.2.1 meaning that they should be comparable to other work presented in the literature.

## 4.3 General Discussion

### *4.3.1 Role of sensory behaviours in ASD*

The findings of Study 1 of Chapter 4 provide some support for the theory proposed by Ornitz and Ritvo (1968) who proposed that brainstem abnormalities resulted in states of arousal, which changed from over-excitation to over-inhibition. It was suggested that in order to compensate for these arousal inconsistencies individuals engaged in repetitive and restricted behaviours, which allowed them to create order or predictability in order to manage arousal states. In turn, these sensory and repetitive behaviours impede communication and social interaction, which were viewed as secondary symptoms of the condition. The results showed that repetitive behaviours were strongly related to sensory behaviours above the effects of individual characteristics of age, ability level and gender and furthermore, the relation between sensory and repetitive features was maintained when the other core features, as well as the associated features, were held constant within a multiple regression model. This extends the results of previous studies which have explored the core features in isolation e.g. comparing questionnaires scores of repetitive and sensory behaviour (e.g. Boyd et al., 2009).

However, this theory does not fully account for the strong relations seen between sensory behaviours and social interaction. Such high correlations would not be predicted by Ornitz and Ritvo (1968) who described social and communicative symptoms as secondary aspects of the condition and therefore that the association between sensory and repetitive behaviours to be the strongest. The findings in Study 1 showed that social interaction was a significant predictor above individual characteristics and remained significant even when the effects of repetitive behaviour were held constant. The fundamental flaw of regression analysis is that it is only correlational and cannot provide information into the causal pathways involved. It is plausible that an individual's social problems may develop in part because of their sensory preferences. For example, communication and interaction may be restricted in individuals who do not like touch (Leekam et al., 2007) or are less responsive to speech sounds (Čeponienė et al., 2003). Alternatively an individual's sensory symptoms may increase because of their autistic symptoms. The strong relationships found in the regression analysis can be argued to stem from many causes such as the influence of currently unmeasured variables. In order to make causal conclusions about these relationships it needs to be shown that manipulation of one of the variables creates an unavoidable change in the other when all other variables are controlled (Tabachnick & Fidell, 1989). The best test of causality, however, is

to examine these variables across time, and longitudinal studies that track both core and sensory behaviours will be needed to fully uncover these causal relationships.

The causal nature or direction of the effect between sensory and maladaptive behaviours also needs to be considered. Again, tracking the appearance of these behaviours over time may highlight causal patterns. There has been some work looking at the overlap between ASD and Oppositional Defiance Disorder (ODD) and Conduct Disorder (see review by Kat and Lecavalier, 2013) to further this work the presence of sensory behaviours across these behaviours could be interesting to examine given the results of the current study. This research should aim to identify if there is a high prevalence of sensory behaviours in these maladaptive behaviour conditions and are these sensory behaviours related to social and communication or repetitive behaviours within these conditions as well within ASD or individuals with a greater number of sensory problems are more at risk of comorbid ASD and conduct problems as would be predicted from the consistent relation between sensory and maladaptive behaviours in Chapter 3 and 4.

This is the first research that took measures of core symptoms direct from a specific diagnostic tool. Hilton et al (2010) explored the relationship with social interaction using an independent social responsiveness scale (SRS) and Boyd (2009) among others, used repetitive behaviour questionnaires. Using measures designed to detect autism such as the DISCO allow measurements specific to the ICD-10 diagnosis. This means the results provide a clearer picture of how sensory behaviour items relate to items that are essential for diagnostic criteria.

It is important to note that even though individual relationships were described above and potential explanations for these relationships were proposed, the sensory measures used in the present study are low level sensory responses to incoming stimuli e.g. distress caused by sounds. These measures, possibly excluding touch, are independent of the measures of social interaction, communication and repetitive behaviours. Such findings are extremely insightful as these low-level behaviours have significant associations with the core features of ASD. Further research needs to explore how such associations exist, which may have implications for the causes and treatments of the primary symptoms in ASD.

### *4.3.2 The Sensory Preferences Questionnaire*

In the second study presented in Chapter 4, the sensory items from the DISCO were converted into a self-report questionnaire and tested with adults with ASD for two reasons; to address the limited work on adults with ASD in the sensory behaviours literature and compare how well this questionnaire, the SPQ, compared to existing tools in the literature (AASP and GSP). The sensory items from the DISCO appeared to work well as a self-report questionnaire – individuals

reported no problems completing the questionnaire and the internal consistency was good – as it was when measuring it when collected with the DISCO, resulting in the SPQ being a valid and reliable tool. The SPQ showed strong correlations with both the Adult/Adolescent Sensory Profile (AASP) in Sample A and the Glasgow Sensory Profile in Sample B as well as showing significant associations with the AQ replicating the results of the AASP and GSP. Furthermore, across the SPQ's modality scores were correlated with the modality scores from both the AASP and GSP, indicating that the SPQ is collecting the same range of behavioural information.

The SPQ differs greatly in the number and type of symptoms being recorded. The sensory items in the DISCO do not conform to the sensory concepts proposed by Dunn as the SPQ does not cover the four quadrants equally (there is only one "low registration" item), however, the total SPQ score highly correlates with both the hyper and hypo responsiveness scores on the GSP and with three of the four quadrants. The SPQ was not significantly associated with the "low registration" quadrant, which is not surprising as only one SPQ items "indifference to pain/heat/cold" measures this.

One advantage of the SPQ is that it is short and can be completed by individuals in much quicker time than the GSP or the AASP. The DISCO is a long interview and if it can be shown that the information collected from questionnaires adapted from the DISCO gathers the same level of reliable information then this may save time in clinical settings where the individuals could complete this questionnaire before assessment. One important stage is required before this is recommended; testing the self-report data against parental interview report to assess how accurate the answers given on the SPQ relate to parents responses on the DISCO. The next study should be to convert the sensory behaviours into parent report in a similar method as the self-report conversion and recruit both individuals and their families to complete both questionnaire. This would provide a further level of validity to the sensory DISCO items but would also greatly improve the literature as little is known about the differences in self and parent report in ASD. It would be hypothesised from previous findings that parents may under-report sensory behaviours in older children than they self-report as Crane et al (2009) found high levels using self-report in adults but Kern et al (2007) found sensory behaviours to decrease with age in parent reports. This is discussed further below. This would allow further work to look at whether parents could also answer these questions before interviews; this has been successful for repetitive behaviours in terms of research. This is the first group of items successfully converted for a self-report tool derived directly from the DISCO. This study provides a clear example of where it may be possible to convert interview items into a questionnaire. It also indicates how such adjustments to DISCO items may benefit the research as

little is known about adult self-report responses to sensory input as well on other associated features.

Study 2 added to the lack of research on adults with ASD and sensory behaviours and limited self-report work. It was found that the high percentage of sensory issues seen in childhood remain high (94.4%) in high functioning adults with ASD. In addition, the auditory and proximal modality scores were significantly higher in the ASD than the TD group. The visual symptoms were the only behaviours to not significantly differ between groups; this replicates the finding that visual symptoms decrease with age (Leekam et al., 2007). This replication was found even though the behaviours were asked about in adults themselves rather than the parental reports of these behaviours. However, it is also possible that it is something about these DISCO items in particular that causes a decrease in report over time.

This high prevalence in adults found in the current study is around the same level shown in children (Tomchek & Dunn; Leekam et al., 2007) and contrasts to the findings of Kern et al (2007) who claimed sensory scores were related to autism severity in children but not adults or adolescents (using the Sensory Profile), however, this study used the parent-report Sensory Profile and findings with the AASP (self-report) also did not find age effects (Crane et al., 2009). The data reported here supports the work of Crane et al (2009) and strengthens the argument for using self-report. The reason for the discrepancies between parent and self-report are that some of the items in the parent-report Sensory Profile may not be relevant to adults for example “has difficultly putting puzzles together or “has trouble staying in between the lines when colouring or when writing” (Crane et al., 2009), another reason may be that adults (especially high functioning) may be better at internalising feelings or have learnt strategies to help cope with the sensory demands placed upon them and therefore although this sensory input is still causing problems to the individual, this may not be apparent to parents of caregivers as their child grows up. The current findings provide research to the autobiographical accounts of adults with ASD stating how sensory behaviours and processing impact on their everyday lives (Grandin, 1995; Grandin & Scarino, 1996).

Sample A (Study 2) did differ in another way to Crane et al (2009), they found that sensory behaviours were not related to AQ score of individuals, and this relation was also absence in Kern et al (2007) in adults. The current study was the second study, however, to find relations between sensory and AQ in adults and the first to show in adults with ASD. The first study was done using the Glasgow Sensory Profile in typically developing adults (Robertson & Simmons, 2013). Differences, between Crane et al (2009) and the current study may reflect the gender bias, as their sample contained females, whereas the ASD sample presented here was purely male, however, this sample

was also larger than Crane et al's (2009) and therefore may have more power to detect patterns in the data.

In addition, a large proportion of individuals with ASD had more than one marked sensory symptom (88.8%), one individual recorded 17 marked problems. The majority (77.8%) also have a marked problem in more than one modality (visual, auditory, proximal). The ASD group scored significantly higher on the SPQ and the three modalities than the TD group. This is further evidence that sensory behaviours are still impacting individuals with ASD across the lifespan and in a sample of very high functioning individuals sensory behaviours are still very prevalent (Sample A). This is informative as even some individuals in the ASD were extremely successful (e.g. one was a lawyer and several had degrees) but they are still reporting sensory problems. However, one caveat of these findings with adults is that recruitment for the study made clear it was about sensory processing in ASD and therefore a respondent bias to individuals with sensory issues may have impacted on the rates of sensory behaviours recorded in this study. Wider use of the SPQ in adults who were not specifically recruited for this purpose are needed as well as the overlap with the Glasgow Sensory Profile in individuals with ASD.

One factor to consider is the that although significantly higher in the ASD group, the TD group also still reported sensory behaviours and 45% of TD still had at least one marked atypicality. Clearly sensory issues affect a large proportion of the typical population as well and consideration of these behaviours in workplaces and public spaces may improve the lives of those beyond the autism spectrum. However, it may be the case that individuals are reporting sensory behaviours but that they may not bother or cause stress like they do in individuals with ASD. One limitation of the SPQ is that is does not measure whether they find the behaviours being asked about as interfering or annoying. Some questions capture this aspect well i.e. "distress caused by sounds" however having acute hearing may be viewed by some as a positive, especially if their work benefits from this. Future work could develop the SPQ or another appropriate questionnaire that measures both how often sensory behaviours occur but also then how much this sensory behaviour impacts on the individual person. For the SPQ, an additional scale for each question could be included that asks the individual to rate the impact of each sensory behaviour.

### *4.3.3 Limitations*

The findings from Study 1 are predominately limited because they cannot make inferences about causal direction. However, as discussed above, proposals of longitudinal studies as well as comparisons across conditions which have overlapping behaviours may benefit this research area. In addition, these results were found using the same data as that reported in Chapter 3 and although

regression models are good at controlling for multiple comparisons within the model replications of these results are needed to strengthen the findings.

There are many limitations to the use of self-report. Although self-report measures have the scope to be more informative and personalised, questionnaires often require the individual to have sufficient language ability to understand and answer the questions. This restricts the information we can gain from personal experience to those at the higher functioning end of the spectrum. This is reflected in the demographics of the current sample (Sample A), that only high functioning individuals were recruited and therefore information gained may be not relevant across the autism spectrum. However, a combination of parent report and self-report from the same validated tool may be useful in overcoming these problems. Not all adults referred for diagnosis have a parent or caregiver to report for them and furthermore not all individuals with ASD can provide accounts of their behaviours. In addition, the SPQ is the first tool specifically designed to measure sensory behaviours and tested in individuals with ASD. Although the GSP was designed for this purpose it has yet to be validated in individuals recruited with a diagnosis of ASD and it also still measures some items that are not directly sensory e.g. repetitive behaviour items – "do you like to listen to the same piece of music/part of a DVD over and over again?" which may impact on correlations with the AQ or core features of ASD. Further work should also focus on identifying research designs that are not dependent on parent report or cognitive ability for self-report such as response to everyday auditory input to capture sensory atypicalities in individuals with high and low IQs (e.g. Blackemore et al., 2006) as these independent empirical measures could then be used to verify the self-reported sensory behaviours of individuals with ASD.

This analysis went some way to providing an external test of the validity of the sensory items from the DISCO. The conversion to a self-report format and data collection with adults was designed for both theoretical and practical reasons. The research on the role that sensory behaviours play across the lifespan of individuals with ASD is inconsistent, firstly little research has focussed on adults specifically and while some reports indicate that sensory behaviours decrease with age (Kern et al., 2007) others do not (Crane et al., 2009). In addition, recruitment for Study 2 was combined with the wider sensory processing and brain imaging study which limited data collection to high functioning individuals. To further test the validity of the DISCO items and make them of stronger relevance in the wider literature two studies should be completed: Firstly, a comparison between the DISCO items in an interview format to parents/caregivers versus a parent-report format. Results from the parent-report questionnaire could also be compared to the Sensory Profile (Dunn, 1999). Secondly, the link between DISCO interview report and the self-report needs to be established, to ensure these two methods of data collection are measuring the same concepts.

### 4.3.4 Future directions

In conclusion, these findings support the idea in the literature that sensory behaviours are strongly related to ASD. As described by Hilton et al (2007) "many variables differ between individuals with ASD and controls, but the acid test for whether a variable is truly related to autism is whether that variable varies as a function of severity within ASD" the findings presented here indicated that the relation between the SPQ and AQ scores and the sensory and social and repetitive behaviours scores in the DISCO suggest sensory is strongly related to autism severity. The score was also shown in Study 1 to differ across IQ, with lower IQ individuals presenting with more sensory behaviours, which reflects the pattern found in core features too (e.g. Mayes et al., 2001).

So why are sensory features not included in the diagnostic criteria? There are discussions about their specificity of these behaviours to autism; sensory features are seen across typical development, blind or deaf individuals and in other clinical disorders, however, more research in ASD is finding significant differences between individuals with clinical conditions and those with ASD and that these sensory behaviours vary as a function of autism severity. The new criteria for DSM-5 as described in Part 1, does now include sensory behaviours. This is a step forward in that their inclusion in the diagnostic criteria may mean that the impairments will be routinely asked about in the diagnostic procedure and therefore management plans and interventions can take these behaviours into account which may ultimately improve individuals with ASD quality of life. However, the addition of these symptoms into the diagnostic criteria as one of four repetitive and restricted sub-domains, of which, individuals only need to score 2/4 on to receive a diagnosis of ASD, does not answer the question of whether sensory behaviours *should* be included as part of the diagnostic description (Wing, Gould, Gillberg, 2011). More work needs to be completed about the specificity of these sensory behaviours across clinical conditions and the impact of the addition of these behaviours to the diagnostic criteria for practice. This will be evaluated in the following section of this thesis (Part 3).

A better assessment will hopefully alleviate some of the distress that individuals with ASD feel when struggling to cope with sensory input. This is especially true of people at the lower functioning end of the spectrum who struggle to explain what they are experiencing and can even act aggressively because of this distress. Therefore, management techniques are essential to deal with these difficulties to enable better experiences on a day to day basis. This will also aid parents to understand their children's experiences with the sensory world and how this is part of the disorder or even heightens other symptoms present in the disorder.

Furthermore it would be worthwhile to investigate using sensory behaviours as a method for early detection of autism. It has been found that unusual responses to sensory input are one of the

first abnormalities that parents notice in their children (Baker et al., 2008) and as these are so prevalent in ASD (e.g. Tomchek & Dunn, 2007) young children presenting with these behaviours should be assessed. This all aids in improving the care of individuals with ASD as the sooner the disorder is recognised then the earlier parents have an understanding of their child and can seek help so more effective interventions can be put in place.

### *4.3.5 Summary*

In Study 2 of this chapter, the sensory items from the DISCO are adapted into a self-report questionnaire (the Sensory Preferences Questionnaire, SPQ) and tested in a sample of adults with high functioning ASD or typical development. This was done for two reasons, firstly to assess the external validity of the DISCO sensory items; the SPQ showed good correlations with total scores on questionnaires already used in the literature. Also, this addressed a gap in the literature on sensory behaviours in adults and using self-report questionnaires. It was found that 94% of high-functioning adults with ASD reported at least one marked sensory behaviour and had significantly higher SPQ scores than IQ matched typically developing individuals. This also provides external validity for the use of the DISCO measure of sensory behaviours in the wider literature, which strengthens the impact of the results found in Study 1.

The first study in this chapter examined the association between the core and associated features of ASD with the sensory behaviours as measured by the DISCO. Replicating the previous chapter, the social and repetitive behaviours had strong associations with the sensory behaviours as did the measures of maladaptive behaviours and pattern of activities. The overall aim for Part 2 was to identify if any of the associated features measured by the DISCO had a significant role than the others. The analyses conducted across both Chapter 3 and 4 highlight the sensory behaviours in ASD to have particularly strong associations with both the core and associated features of ASD and has a key role to play in the behavioural presentation of ASD.

# PART 3: MEASURING AUTISM SPECTRUM DISORDER: DIAGNOSTIC CRITERIA

Part 3 focuses on the diagnostic criteria for autism spectrum disorder. A set of empirical studies explore the measurement characteristics of two different operational definitions of autism spectrum disorder- those of DSM-5 (APA, 2013) and Wing & Gould (Wing, 1996; Leekam et al., 2002). In Chapter 5, I designed several alternative new diagnostic algorithms for DSM-5 criteria (APA, 2013) and tested them using DISCO data in order to establish an acceptable algorithm that optimised sensitivity and specificity. Because the new DSM-5 criteria has been criticised in the literature for potentially missing the diagnosis of individuals with autism when compared with DSM-IV-TR, comparisons were also made between the diagnostic output of DISCO DSM-5 algorithm and diagnostic output of other classification systems and specific attention given to high functioning individuals and those with Asperger profiles who are proposed to be most at risk of being missed. The overall purpose of Chapter 5 was to examine the question of whether individuals who have previously been given a diagnosis of autism or Asperger Syndrome would be captured when the DSM-5 criteria are run using the new DISCO DSM-5 algorithm.

The definition of Autism Spectrum Disorder used by DSM-5 is different from Wing and Gould's original description. The purpose of the second part of Part 3 (Chapter 6 and 7) was to compare the DSM-5 and Wing & Gould sets of criteria. While both definitions describe a lifelong condition that usually appears early in childhood (but may become evident later), and both definitions also allow ASD to be recognised alongside other disorders, the descriptions of the behavioural impairments differ to some extent. DSM-5 describes a disorder with persistent deficits in two domains; (a) social interaction and communication and (b) restricted, repetitive patterns of behaviour, interests or activities. In contrast, Wing & Gould describe a triad of impairments in the domains of social interaction, communication, and imagination. The imagination impairment recently redefined as impairment in "social imagination", is associated with a rigid, repetitive pattern of behaviour. The two definitions of diagnostic criteria also differ in their level of complexity. The DSM-5 description describes multiple criteria (see Box 5.1), while the Wing and Gould ASD algorithm contains only five (chapter 6).

In the final two empirical chapters (6 and 7), I investigate in detail the existing DISCO algorithm for Wing and Gould's Autism Spectrum Disorder (WG-ASD) and particularly the criterion of, "quality of social impairment", which may have diagnostic value used alone. The overall purpose of the work in this second part of Part 3 (Chap 6 and 7) therefore is to evaluate and compare the measurement of these two descriptions for Autism Spectrum Disorder using the DISCO. During Part

3 and in Part 4 (General Discussion) I reflect on the adequacy of the DSM-5 criteria for the diagnosis of Autism Spectrum Disorder, in the light of the earlier debate about its lack of sensitivity and consider how comparisons between DSM-5 and Wing & Gould descriptions and their measurement may help to move forward in our understanding of the autism spectrum.

# 5 Diagnosing Autism Spectrum Disorder: Algorithm Design

The aim of this chapter is to design and test a new diagnostic algorithm for DSM-5 Autism Spectrum Disorder (APA, 2013) using the DISCO. The first part of the chapter presents a review of the literature, related to the new DSM-5 criteria. The second part presents the empirical work. In the study, I present the development and design of a DSM-5 algorithm, comparing the diagnostic output of the final DSM-5 algorithm with the individuals' previous clinical diagnosis. The research reported in this study has been published as a joint first authored paper in the Journal of Child Psychology and Psychiatry (Kent et al., 2013). The work was carried out predominantly in collaboration with the other joint first author with support and input from co-authors. However the content of the article has been changed for my PhD thesis. The second study was carried out independently for the thesis and is not yet published. This provided a further test of the DSM-5 algorithm by comparing the DISCO DSM-5 diagnostic output with the individuals' diagnostic output based on other DISCO algorithms.

The work in this chapter was specifically designed to address a debate in the academic literature and clinical field regarding the planned changes to DSM-5. New research based on the draft criteria for DSM-5 in 2011-2013 indicated that a large number of individuals who had DSM-IV-TR diagnosis of Pervasive Developmental Disorders would miss getting a diagnosis according to DSM-5 because the new criteria lacked sensitivity. The empirical work reported here aimed to operationalise the DSM-5 criteria in order to test who would get a diagnosis of DSM-5 using, for the first time, data from a single diagnostic tool –the DISCO (Studies 1 and 2). The empirical work for the chapter was carried out between 2011 and 2012 before the official release of DSM-5 and was therefore based on the draft DSM-5 ASD criteria that were accessed in 2011. However, further assessment was made when the final criteria were published. As the changes were minimal in the final published criteria (APA, 2013), the DISCO DSM-5 algorithm remains identical (see Method Section)[7].

## 5.1 Literature review

### *5.1.1 Proposed DSM-5 changes to the diagnostic criteria*

As discussed in Chapter 1 of this thesis, a diagnosis of Autistic Disorder (DSM-IV-TR, American Psychiatric Association, 2000) or Childhood Autism (ICD-10, World Health Organisation,

[7] The DISCO DSM-5 algorithm covers all examples of behaviours from the draft DSM-5 ASD criteria (APA, 2011), the final criteria (APA, 2013) included "rituals when greeting others" as an example of behaviour in sub-domain B2, this behaviour was the only example that could not be covered by the DISCO. The DISCO DSM-5 ASD algorithm for the draft criteria remains identical for the final criteria.

1993) is given when an individual has clinical impairments in social interaction, communication, and restricted and repetitive behaviours and interests. The criteria for these two DSM-IV-TR and ICD-10 diagnoses are equivalent (Volkmar & Klin, 2005) with the two diagnostic labels used interchangeably. The diagnostic descriptions of Childhood Autism (ICD-10) and Autistic Disorder (DSM-IV-TR) fall within the umbrella term Pervasive Developmental Disorders (PDD), which also includes Atypical Autism and Asperger Syndrome. Empirical evidence however has not supported the idea of categorical subgroup distinctions within PDD (e.g. Leekam et al., 2000; Lord et al., 2012; Macintosh & Dissanayake, 2004; Prior et al., 1998). Recognising this evidence, the DSM-5 committee introduced a single broad category of Autism Spectrum Disorder (ASD) to replace PDD; the criteria for DSM-5 ASD can be seen in Box 5.1.

Five main changes are proposed within the DSM-5 draft criteria. First, all categorical sub-diagnoses within DSM-IV-TR PDD will be subsumed under this new concept of ASD. This move is strongly supported by the literature presented in Part 1 which found sub-group distinctions under PDDs to be "impossible" to reliably assign individuals to (e.g. Szatmari et al., 1995). It is hoped that this may relieve clinical resources on deciding which categorical diagnosis best fits the individual's needs (Vivanti et al., 2013) especially as the application of diagnostic sub-groups varied across diagnostic centres (Lord et al., 2012).

Second, the social and communication domains from DSM-IV-TR will be combined into one domain. This change reflects a substantial literature which claims the distinction between social and communicative behaviours is arbitrary, for example; non-verbal behaviours such as facial expression or eye contact can be both social and communicative (see Gotham, Risi, Pickles, & Lord, 2007). It also reflects the evidence using factor analytic techniques that the diagnostic triad of behaviours described by DSM-IV-TR lacks validity (Frazier, Youngstrom, Kubu, Sinclair, & Rezai, 2008; Georgiades et al., 2007; Hoekstra, Bartels, Cath, & Boomsma, 2008; Snow, Lecavalier, & Houts, 2009; van Lang et al., 2006). For example, in a sample of 1,170 individuals aged 2-46 years old, exploratory and confirmatory factor analyses were conducted using ADI-R data. It was found that exploratory analyses strongly supported a two factor model of ASD with social interaction and communication on one factor and stereotyped language, restricted, repetitive and stereotyped behaviours on another (Frazier et al., 2008). Snow et al. (2009) also found support for a two factor model in 1,861 4-18 year olds, again using the ADI-R. It is argued that the social-communication and restricted or repetitive behaviour domains have different developmental trajectories (see Lord et al., 2006) and potentially independent causes (Mandy & Skuse, 2008).

**Box 1: Published DSM-5 Autism Spectrum Disorder Criteria - 299.00 (F84.0)**

A. Persistent deficits in social communication and social interaction across multiple contexts, as manifested by the following, currently or by history (examples are illustrative, not exhaustive, see text):

1. Deficits in social-emotional reciprocity, ranging, for example, from abnormal social approach and failure of normal back-and-forth conversation; to reduced sharing of interests, emotions, or affect; to failure to initiate or respond to social interactions.
2. Deficits in nonverbal communicative behaviors used for social interaction, ranging, for example, from poorly integrated verbal and nonverbal communication; to abnormalities in eye contact and body language or deficits in understanding and use of gestures; to a total lack of facial expressions and nonverbal communication.
3.Deficits in developing, maintaining, and understanding relationships, ranging, for example, from difficulties adjusting behavior to suit various social contexts; to difficulties in sharing imaginative paly or in making friends; to absence of interest in peers.

Specify current severity:

Severity is based on social communication impairments and restricted, repetitive patterns of behaviour.

B. Restricted, repetitive patterns of behavior, interests, or activities, as manifested by at least two of the following, currently or by history (examples are illustrative, not exhaustive; see text):

1. Stereotyped or repetitive motor movements, use of objects, or speech (e.g., simple motor stereotypies, lining up toys or flipping objects, echolalia, idiosyncratic phrases).
2. Insistence on sameness, inflexible adherence to routines, or ritualized patterns or verbal nonverbal behavior (e.g., extreme distress at small changes, difficulties with transitions, rigid thinking patterns, greeting rituals, need to take same route or eat food every day).
3. Highly restricted, fixated interests that are abnormal in intensity or focus (e.g, strong attachment to or preoccupation with unusual objects, excessively circumscribed or perseverative interest).
4. Hyper- or hypo-reactivity to sensory input or unusual interests in sensory aspects of the environment (e.g., apparent indifference to pain/temperature, adverse response to specific sounds or textures, excessive smelling or touching of objects, visual fascination with lights or movement).

C. Symptoms must be present in the early developmental period (but may not become fully manifest until social demands exceed limited capacities, or may be masked by learned strategies in later life).

D. Symptoms cause clinically significant impairment in social, occupational, or other important areas of current functioning.

E. These disturbances are not better explained by intellectual disability (intellectual developmental disorder) or global developmental delay. Intellectual disability and autism spectrum disorder frequently co-occur; to make comorbid diagnoses of autism spectrum disorder and intellectual disability, social communication should be below that expected for general developmental level.

The third change is that the DSM-5 repetitive behaviour criteria will include criteria on the presence of hyper or hypo sensitivity to sensory input or the sensory environment. Again, this follows the empirical findings showing that sensory behaviours are both prevalent in individuals with ASD and more likely to be found in individuals with ASD than other clinical groups (e.g. Leekam et al., 2007; Ben-Sasson et al., 2009; Baraneck et al., 2006; Baker et al., 2008). Fourthly, the criterion for early childhood onset includes the caveat that although symptoms must be present in early childhood, they may not become fully manifest until social demands exceed limited capacities. This was done in order to allow all of the previous categorical distinctions to be captured under the umbrella spectrum diagnostic term. The criteria for Asperger Syndrome requires that individuals did not have a language delay and would therefore not be captured if the age criterion for Autistic Disorder/Childhood Autism was adopted. Finally, the requirement for significant severity (i.e. negative impact on daily functioning) is more explicitly stated in the revised guidelines.

The construct validity of the proposed DSM-5 criteria has been supported by confirmatory factor analysis (CFA) using the Developmental, Dimensional and Diagnostic Interview (3di, Skuse et al., 2004). The study demonstrated that ASD was best represented as two dimensions of social-communication and repetitive behaviour, but with an additional sensory subscale (Mandy, Charman, & Skuse, 2012). In addition, the overall change to a spectrum approach (DSM-5, APA, 2013) fits with current terminology and concepts of Autism (e.g. Wing and Gould, 1979; Happé and Frith, 1991).

Any change in diagnostic criteria raises a risk that the symptom profile of some individuals will no longer meet the new criteria, however, if a specific group of individuals are at particular risk this would raise problems for the all-inclusive autism spectrum proposed by DSM-5 which aimed to be more encompassing than the previous subgroup descriptions. Release of the draft DSM-5 criteria caused some concerns about who will get a DSM-5 diagnosis; in particular the difficulty in measuring early onset that manifests itself in later years and the lack of appropriateness of the criteria for all individuals who are currently diagnosed with autism or Asperger Syndrome according to DSM-IV-TR or ICD-10 (e.g. Wing, Gould, & Gillberg, 2011). The diagnostic accuracy and inclusiveness of the specified criteria therefore need to be tested against that of DSM-IV-TR (and ICD-10) before concluding whether these criteria are identifying the correct individuals.

### *5.1.2 Measurement of the effectiveness of the DSM-5 diagnostic criteria*

In order for diagnostic criteria to be accurate they need to be both sensitive and specific. Sensitivity refers to the criteria's ability to capture all individuals with ASD who should receive a clinical diagnosis. Specificity, on the other hand, refers to the criteria's ability to correctly exclude people who should not receive a clinical diagnosis from meeting criteria on a tool or algorithm. In a

statistical sense both of these values can be calculated and rated on a scale from 0 (no ability to be sensitive or specific) to 1 (perfect sensitivity or specificity) or measured as the percentage of individuals correctly classified as having an ASD and the percentage of the control group who do or do not meet criteria. These values are computed using Receiver Operating Characteristic (ROC) curve analyses, which provide a measure that reflects both the sensitivity and specificity of a test or set of criteria. This approach has been used to test the DSM-IV-TR criteria across a range of standardised clinical tools such as the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000), Autism Spectrum Screening Questionnaire (ASSQ; Ehlers, Gillberg, & Wing, 1999) as well as the Autism Diagnostic Interview-Revised (Lord, Rutter, & Le Couteur, 1994) and DISCO (Nygren et al., 2009). Unfortunately, the standard method of comparing algorithm output with an individual's clinical diagnosis is flawed in that a clinical diagnosis itself is based on behavioural features that are also extracted from diagnostic tools. However, as no biological or genetic test exists to confirm a diagnosis, this is the best approach and has been adopted for the majority of psychometric testing for tools in ASD and allows identification of where clinical and research instruments converge and disagree.

Most of the literature reviewed below applies the new DSM-5 criteria in two ways. Firstly, clinicians use the description as an informal checklist and utilise the information gathered about a particular individual to decide if they meet DSM-5 ASD criteria. Secondly, each of the DSM-5 criteria can be directly operationalised or mapped onto a diagnostic tool. This second approach is referred to as creating a specific DSM-5 "algorithm" for that particular tool. An algorithm is the selected set of items that are combined by rules, such as those set out in DSM-5 to provide a binary outcome, whether an individual meets DSM-5 criteria or not.

### *5.1.3 Research evidence on the effectiveness of the DSM-5 criteria*

The findings presented below, raised concerns that individuals might be excluded from a DSM-5 diagnosis due to poor sensitivity of the new DSM-5 criteria. In an epidemiological study of eight-year-old children, clinicians applied DSM-IV-TR and draft DSM-5 criteria to information about children collected who were screened using the ASSQ (Ehlers et al., 1999) and assessed with the ADI-R (Lord et al., 1994) and ADOS (Lord et al., 2000). Clinical diagnoses of DSM-IV-TR were made according to clinical judgement of all available material. In a subset (N=82, IQ>50) of cases, a diagnosis of DSM-5 was then made by applying the draft criteria to the same clinical information. A 54% reduction in diagnostic sensitivity relative to DSM-IV-TR was reported when the draft DSM-5 criteria were strictly applied (Mattila et al., 2011). A 23% reduction in sensitivity was also reported in 2-16-year old children referred for initial diagnostic assessment when the draft DSM-5 criteria

were applied compared to a clinical diagnosis of DSM-IV-TR made according to clinical opinion and guided by ADI-R and ADOS scores (Gibbs, Aldridge, Chandler, Witzlsperger, & Smith, 2012). Comparable reductions in sensitivity were reported for both low functioning adults (37%; Matson, Belva, et al., 2012) and at-risk toddlers (47.9%; Matson, Kozlowski, Hattier, Horovitz, & Sipes, 2012).

Poor sensitivity was also found in studies that included DSM-IV-TR diagnostic sub-groups. In one of the first tests of the draft DSM-5 criteria, a re-analysis of data collected for DSM-IV field trials was used. Information gathered from DSM-IV symptom checklists was used to create a draft DSM-5 algorithm. This algorithm had excellent specificity but the sensitivity varied across diagnostic sub-group, identifying 76% individuals with Autistic Disorder but only 25% of individuals with Asperger Syndrome and 28% of individuals with PDD-NOS. Furthermore, the sensitivity also differed across ability level from 70% sensitivity in low functioning individuals (IQ<70) to 46% in high functioning (IQ>70) individuals (McPartland et al., 2012). Taheri and Perry (2012) also found sensitivity varied by IQ; 89.7% of individuals with an IQ below 70, but only 22.2% of individuals with an IQ above 70 met criteria for DSM-5 using retrospective file reviews of individuals meeting DSM-IV-TR criteria.

Not all studies that used the proposed DSM-5 criteria have reported poor sensitivity, the use of standardised clinical and research tools to map DSM-5 criteria has resulted in better sensitivity. Two recent studies, which both mapped DSM-5 criteria using items from the ADI-R (Lord et al., 1994) and the ADOS (Lord et al., 2000) reported good sensitivity (above .91). However, specificity was poor, 0.53 for the ADI-R and 0.63 for ADI-R and ADOS together (Huerta, Bishop, Duncan, Hus, & Lord, 2012) or not reported (C. A. Mazefsky, McPartland, Gastgeb, & Minshew, 2013).

Only one study to date has reported good levels of both sensitivity and specificity of the draft DSM-5 criteria in children and adolescents (2-18-year olds; Frazier et al., 2012). In this study, items were selected from two parent-report questionnaires, the Social Responsiveness Sales (SRS; Constantino & Gruber, 2002) and the Social Communication Questionnaire (SCQ; Rutter et al., 2003), both of which have strong psychometric properties. Both questionnaires were collected from the Interactive Autism Network, a large registry of siblings with at least one child diagnosed with ASD. The comparison group in this study were the siblings of the affected individuals, including those with and without conditions such as ADHD and anxiety disorder reported by parents. This sample, therefore, was not typical of a clinical comparison group and the reported specificity may be inflated compared with other studies.

### *5.1.4 Limitations of existing research*

One limitation of the majority of existing literature is that the data were typically collected according to DSM-IV-TR criteria and mapped to the DSM-5 descriptions (e.g. Taheri & Perry, 2012).

These data, therefore, may not have included sufficient information to address the full range of behaviours described by DSM-5 (Swedo et al., 2012). Furthermore, the majority of existing literature has focussed on children, and there is a clear need to assess how the criteria perform in adults (Huerta et al., 2012).

It is difficult to make strong conclusions about the diagnostic accuracy of the DSM-5 ASD criteria as the findings are not consistent across studies. It is likely that this is primarily due to the different measures and techniques used to quantify the DSM-5 criteria. The sensitivity of the DSM-5 criteria is reported as being much stronger when standardised research and clinical tools are used to directly map the relevant items onto DSM-5 criteria (Heurta et al., 2012, Mazefsky et al., 2012; Frazier et al., 2012) rather than using clinical judgement. However, in order to reach good levels of sensitivity and specificity items have had to be selected across two instruments, no studies have managed to balance sensitivity and specificity of the DSM-5 ASD criteria using information from one tool only. In addition, even the standardised diagnostic tools such as the ADI-R, ADOS and SCQ were designed to measure autism as described by the previous international classification systems of ICD-10/DSM-IV-TR, these measures may, therefore, not be capturing all the information needed for a DSM-5 diagnosis such as the addition of sensory reactivity.

### *5.1.5 Potential applications of the DISCO*

The newly proposed DSM-5 diagnostic criteria for ASD can be investigated by exploring data not collected specifically for the purpose of diagnosis according to DSM-IV-TR/ICD-10. The DISCO is therefore a good contender for this purpose because the content of the DISCO is influenced by a concept of a spectrum of autistic disorders. This concept predated the earliest ICD and DSM criteria for autism (Wing, 1988; Wing & Gould, 1979), and is therefore not constrained by existing international diagnostic classifications. In addition, the DISCO has a large range of items to map onto the DSM-5 criteria including measures of sensory symptoms and it is designed to be relevant to individuals of all age and levels of ability and could therefore be useful in capturing all individuals in the autism spectrum as defined by DSM-5.

The DISCO is appropriate for measuring the proposed changes in the DSM-5 criteria for several more specific reasons.

First, the prominence of sensory behaviours in the new DSM-5 ASD diagnostic criteria can be easily adopted by the DISCO, which already contains 25 items on sensory processing. Five of these items were used in the ICD-10 DISCO algorithm for Childhood Autism but the range of additional items in the DISCO allows extra behaviours to fully map onto the broader criteria of DSM-5. The criteria are listed as “hyper- or hypo- reactivity to sensory input or unusual interest in sensory

aspects of environment; such as apparent indifference to pain/heat/cold, adverse response to specific sounds or textures, excessive smelling or touching of objects, fascination with lights or spinning objects".

Second, the DISCO can adapt to the change from sub-category definitions to a spectrum definition (APA, 2013): The DISCO was designed according to Lorna Wing's view that "the borderlines of the autistic spectrum merge, at the lower end of the scale of ability, with profound mental retardation. At the upper end of this scale, they merge into mildly eccentric variations of typical development. Within the spectrum, the sub-groups that have been suggested merge into each other" (Wing, 2005, p. 198). The DISCO, therefore, contains items to measure all behaviours present in ASD and not just typical or "core" Autism. Crucially the DISCO was not designed around the previous international classification systems. The resulting wide range of items in the DISCO allows for a range in presentation to be captured and its aim to record all behaviours present across individuals with ASD will be beneficial in meeting the changing criteria specified by DSM-5 toward a spectrum approach (APA, 2013).

Finally, the range of behaviours measured by the DISCO allows it to be flexible to adapt to changes in criteria that are based on a dyad of domains in contrast with the previous triad. The DISCO contains 41 social interaction items, 40 communication items and 55 repetitive behaviours items and the range enables these to be selected from in a flexible manner. Importantly, the DISCO already contains items which capture repetitive speech. This criterion has been incorporated by DSM-5 into the repetitive behaviour sub-domains of "stereotyped or repetitive speech" and "excessive adherence to routines, ritualized patterns of verbal or nonverbal behaviour." The DISCO already covers these criteria with items such as: repetitive questions/repetitive themes; tone of voice; and long winded or pedantic speech.

The empirical work presented below therefore uses the DISCO to design, develop and test a DSM-5 algorithm and allows examination of the question of whether individuals who have previously been given a diagnosis of a Pervasive Developmental Disorder would be excluded when the DSM-5 criteria are run using this algorithm. This work is divided across two studies. The first study (Study 5.2.1) designs and tests the DISCO DSM-5 ASD algorithm across Samples 1 and 3 with particular attention to how the behaviours are mapped from the DSM-5 ASD criteria into a DISCO algorithm. The second study (Study 2) focuses on the concern in the literature that high functioning individuals and those with diagnostic labels outside Autistic Disorder/Childhood Autism are more at risk of being missed by the DSM-5 ASD.

## 5.2 Empirical work:

### *5.2.1 Study 1: Designing and testing the DSM-5 algorithm*

#### *5.2.1.1 Introduction*

The aim of study 5.2.1 was to design and validate a DSM-5 algorithm for the DISCO that was both sensitive and specific across ability level. The design of the algorithm involved several stages. First the mapping of items in the DISCO onto the behaviours specified in the DSM-5 ASD criteria. Second, review and approval of external clinical opinion as previously adopted for research with standardised research tools (Heurta et al., Mazefsky et al., 2012). Third, algorithm design and testing so that particular combinations of DISCO items can be run across samples to correctly classify clinically defined ASD groups with DSM-5 ASD (sensitivity) and distinguish them from individuals with other clinical conditions or typical development according to DSM-5 ASD (specificity).

In order to optimise both the sensitivity and specificity of the proposed DSM-5 algorithm, attention was given not only to the two DSM-5 domains (A) social-communication and B) Restricted, Repetitive patterns of behaviour) but also to the sub-domains embedded within each of these domains (three in domain A and four in domain B; see Box 5.1). In published studies, the number of items or behaviours included in each sub-domain has varied, for example from three to 13 (Frazier et al., 2012). Authors have usually required only one or two items to meet criterion on each sub-domain, as specified by different drafts of the DSM-5 criteria. However, this uniform approach may not always produce the best sensitivity and specificity either at the sub-domain, domain nor algorithm level. For example, the likelihood of an individual having one of 13 behaviours in a sub-domain is greater than having one of just three. The effect of changing this threshold according to each sub-domain has never been tested. A key part this study therefore was to set thresholds for sub-domains in which both sensitivity and specificity levels maximized the clinical utility of the threshold to identify true ASD cases whilst minimising false positives.

##### *5.2.1.1.1 Defining algorithm versions:*

For the development of a new DISCO DSM-5 algorithm several different algorithm versions were compared (“Initial”, “Youden J” and “Modified”); each version of the algorithm included the same items, but the sub-domain thresholds were set using three different criteria.

The first (**Initial DSM-5 DISCO algorithm**) applied the minimum requirements outlined in the proposed DSM-5 criteria as have been applied by previous studies (e.g. Huerta et al., 2012), which also allow direct comparison. Therefore, only one behavioural item (from the DISCO) was required to be present in each sub-domain for an individual to be categorised as scoring on that particular sub-domain.

The second (**Youden J DSM-5 DISCO algorithm**) applied a standardised statistic (Youden J), which is used to identify the best balance between sensitivity and specificity for the threshold of each sub-domain. The Youden J statistic (maximum = (sensitivity + specificity) - 1) (Youden, 1950) has been used in previous research on diagnostic assessment of ASD (e.g. Cohen et al., 2010) and other areas of medicine (e.g. Chiu et al., 2011; Portalez et al., 2012).

The third method (**Modified DSM-5 DISCO algorithm**) involved selection of the highest number of behaviours that maintained the maximum sensitivity of the sub-domain. This method was designed in the current study in order to balance both sensitivity and specificity within sub-domains, while accounting for the control of specificity that is specified at the domain level of the DSM-5 criteria (APA, 2013). Sensitivity was given priority at the sub-domain level. The threshold with the highest level of sensitivity was selected. If a number of threshold values had the same level of sensitivity then the decision on which to select was based on the strictest level of specificity. For example, if a threshold of 1, 2 and 3 all gave a sensitivity value of 1, then a threshold of three would be selected as out of the three thresholds a score of three would ensure the most non-ASD cases were excluded.

The goal was to compare the balance of sensitivity and specificity across these algorithms using two participant samples. Specificity is already controlled at the whole algorithm level according to the rules set out in the DSM-5 ASD criteria, which specify the pattern of behaviour that must be present for an individual to meet criteria (all three social-communication sub-domains and 2/4 repetitive behaviour sub-domains). As the first (Initial) algorithm approach relies purely on these overall algorithm rules (combining of sub-domains) to exclude false positives it is predicted to have excellent sensitivity but lower specificity. Furthermore, it is proposed that this approach may be inappropriate for the design of a DISCO DSM-5 ASD algorithm as mentioned above; the DISCO has many examples of behaviours that individually cover very specific behaviours, compared to other tools such as the ADI-R where each item covers a range of examples of behaviour.

In contrast, the second (Youden J) algorithm approach should more evenly balance sensitivity and specificity for each sub-domain, but may restrict sensitivity of the whole algorithm by controlling for specificity at the sub-domain level and also through the combination of sub-domains. The sub-domain thresholds for the third (Modified) algorithm are raised as high as possible while maintaining maximum sensitivity. We therefore predict that this approach will have improved sensitivity relative to the second approach, and improved specificity relative to the first.

### 5.2.1.2 Method

#### 5.2.1.2.1 Participants

Samples 1 and 3, as described in the Method Chapter (section 2.2.1 and section 2.2.4) were used for the following analyses. Item selection and algorithm design was conducted with Sample 1 only and validated in Sample 3.

To reiterate Chapter 2, for Samples 1 and 3, clinical diagnoses of DSM-IV-TR Autistic Disorder or ICD-10 Childhood Autism were made before recruitment by an independent clinician who did not use the DISCO. The sample used for algorithm design (Sample 1) comprised parents of 82 children from the UK interviewed using DISCO-9 (Wing et al. 2002). Thirty six children (34-140 months; 32 male) had a clinical diagnosis of autism; 18 had higher ability and 18 lower ability. The lower ability comparison group comprised 17 individuals with ID (40-140 months; 10 male). The higher ability comparison groups comprised 14 individuals with LI (49-136 months; nine male) and 15 TD children (51-135 months; nine male). The validation sample (Sample 3) comprised parents of 115 children from the Netherlands interviewed using DISCO-11. Fifty two children (34-137 months; 43 male) had a clinical diagnosis of autism (17 higher ability, 35 lower ability). The higher ability comparison group included 37 TD children (24-49 months; 15 male) and the lower ability comparison group included 26 children with ID (48-134 months; 16 male).

#### 5.2.1.2.2 Measures

The majority of DISCO items are rated for present symptoms (current) and symptoms across lifespan (ever). Consistent with previous research and diagnostic algorithms for the DISCO ever codes were used to develop the DSM-5 algorithm.

#### 5.2.1.2.3 Item selection

The mapping of DISCO items to the DSM-5 description was established using the draft DSM-5 criteria. The official release of DSM-5 (APA, 2013) after this original item mapping meant that this was subsequently rechecked after the analysis and no changes were made; the published DSM-5 ASD diagnostic criteria include one additional example of behaviour: “rituals when greeting other” (Sub-domain B2). No DISCO items were identified that could adequately capture this behavioural description; consequently, item mapping remained identical across the draft and final DSM-5 criteria for the DISCO items. The full set of 320 DISCO items were scrutinised in a three-stage process:

1. All DISCO items mapping onto DSM-5 descriptions (DSM-5, 2011) were assigned to DSM-5 sub-domains (Table 5-1) by the author (R. Kent) and another researcher with experience of

ASD (Dr Sarah Carrington). If any of the items were relevant to two DSM-5 sub-domains then these were highlighted and confirmed in the second stage of item selection (below).

2. One clinician and researcher with extensive knowledge of ASD and the DISCO reviewed item selection and placement. This resulted in the inclusion of four additional items (three in A1: seeks comfort when in pain or distress; gives comfort to others; inappropriate response to others' emotions, and one in A2: understanding of gesture), movement of one item from A1 to A3 (does not interact with age peers), and deletion of one item from B2 (reversal of pronouns). The placement of three verbal items was queried between B1 and B2: long winded or pedantic speech; tone of voice; idiosyncratic use of words or signs.
3. The proposed assignment of all items was reviewed by three experienced DISCO interviewers (two psychiatrists and one psychologist) based in Japan, Canada and the Netherlands. None had been involved in the study's design or implementation. All independently agreed on the placement of all items, giving separate consideration to placement of repetitive verbal items in B1 or B2. All decided these items should be placed in B1.

As with the design of the original ICD-10 algorithms, codes for each item were selected that best met the description in the diagnostic guidelines. To recap, the majority of items in the DISCO are rated on a three-point severity scale: "marked" when a behaviour occurs daily, when no strategy is in action, or whenever the opportunity arises; "minor" when behaviours are less frequent or severe; or "no problem". In line with standard diagnostic coding (e.g. established DISCO and ADI-R algorithms), the majority of items were scored as present (1) only if there was a "marked" (severe) impairment.

Additionally, codes were used according to the type of DISCO item and the behaviour specified by DSM-5. The minor (1) code was used for 12 items (reciprocal communication; sharing of interests and enjoyment; sharing of happiness; seeking comfort when in pain or distress; interaction with age peers; use of imperative gestures; understanding of gesture; imaginative activities; friendship with age peers; quality of friendship; avoidance of age peers; awareness of others' feelings; and response to visitors) in order to capture atypical behaviour that may not have reached criteria for a marked problem but is still present and clinically relevant. The -8 code is available for particular items across the DISCO and usually refers to exclusion criteria such as if the individual is too young, too physically disabled or there has been no opportunity for that individual to present with the behaviour is question. In some cases this -8 code is clinically relevant as it signifies when an individual never displays a behaviour e.g. a "lack of facial expression" or does not

have the opportunity for a behaviour e.g. “has no special friend” but this *may* not be due just to developmental level but could indicate other potential social interaction problems. Seven items included the -8 code: emotional response to age peers (indifferent/no reaction); one sided social approaches (rarely or never makes social approaches); use of body language (uses too little body language to rate); facial expression (little or no facial expression); friendship with age peers (no concept of friendship); interaction with age peers (no opportunity now but never interacted); and limited pattern of activities (no spontaneous activities).

Finally, some DISCO items follow a different coding system; the imaginative activities item is used to assess what level an individual has reached in their development and therefore codes of 0 (no play with model toys), 1 (plays with real household equipment using it for its real purpose) and 2 (holds doll, toy animals as if real, at least some of the time) were used to classify atypical or delayed imaginative development. For domain C, the item “combining 2-3 word utterances” was coded in months and therefore a score of 0 (not yet achieved) or any age in months above 36 was rated as a delay in behaviour. All items scored as present were recoded as “1” and codes not counted were recoded as “0” for use in the algorithm.

The final codes and agreed assignment of items to the sub-domains was then assessed statistically to assess redundancy and reliability of the items used in the algorithm. Internal consistency of the sub-domains ranged from .55-.865. Within each sub-domain, only 2 pairs of items correlated: giving comfort to others and emotional response to age peers in A1 (.79); and descriptive and instrumental gestures in A2 (.71). In both cases, the removal of any of the items reduced the Cronbach’s alpha for the relevant sub-domain and therefore all four items were retained. The overall algorithm consisted of 85 DSM-5 items (Table 5-1), which had excellent overall internal consistency (Cronbach’s alpha = .95), and 80% of the DISCO items used in the algorithm had good/excellent inter-rater reliability (kappa>=.7; Wing et al., 2002) in Sample 1.

**Table 5-1: Table showing the DSM-5 ASD criteria and the DISCO item mapping to each example for the DSM-5 sub-domains. Frequency in the ASD group and chi-square analyses between the ASD group and non-ASD clinical control groups in Sample 1 are presented.**

| **DSM-5 domains and sub-domains** | | **Frequency** | **Chi-square for domains and items** |
|---|---|---|---|
| **Criterion A - Persistent deficits in social communication and social interaction across contexts, not accounted for by general developmental delays, and manifest by ALL THREE of the following:** | | | **52.77*** ** |
| **A1: Deficits in socio-emotional reciprocity, for example:** | | | **52.77*** ** |
| *DSM-5 Example* | DISCO item | | |
| *from abnormal social approach and failure of normal back and forth conversation to response to total lack of initiation of social interaction* | Quality of social communication; | 55.6 | 7.37** |
| | Makes one-sided social approaches | 61.1 | 34.80*** |
| | Seeks comfort when in pain or distress | 72.2 | 25.02*** |
| | Giving comfort to others | 77.8 | 34.87*** |
| | Inappropriate response to others' emotions | 33.3 | 14.70** |
| | Avoidance of age peers | 77.8 | 23.49*** |
| *reduced sharing of interests, emotions, and affect* | Sharing interests and enjoyment | 88.9 | 24.36*** |
| | Reaction to others' happiness | 38.9 | 10.75*** |
| | Emotionally expressive gestures | 50 | 20.04*** |
| | Emotional response to age peers | 77.8 | 40.50*** |
| **A2: Deficits in non-verbal communicative behaviours used for social interaction, for example:** | | | **25.02*** ** |
| *DSM-5 example* | DISCO item | | |
| *poorly integrated verbal and nonverbal communication* | Use of body language | 63.9 | 16.67*** |
| | Using other people as mechanical aids | 47.2 | 9.99** |
| *abnormalities in eye contact and body-language* | Eye contact | 52.8 | 13.16*** |
| | Brief glances | 25 | 7.42** |
| | Blank, unfocussed gaze | 50 | 8.47** |
| | Stares too long and hard | 16.7 | 1.20 |
| *deficits in understanding and use of nonverbal communication to total lack of facial expression or gestures* | Nonverbal communication | 5.6 | 0.66 |
| | Understanding of gesture and miming | 22.2 | 2.96 |
| | Facial expression | 44.4 | 7.14** |
| | Instrumental gestures | 69.4 | 13.78*** |
| | Declarative gestures (joint referencing) | 50 | 20.04*** |
| | Imperative gestures | 13.9 | 6.80** |
| | Descriptive gestures | 75 | 17.66*** |
| | Use of nodding and shaking head | 41.7 | 12.33*** |

| DSM-5 domains and sub-domains | | Frequency | Chi-square |
|---|---|---|---|
| **A3: Deficits in developing and maintaining relationships appropriate to developmental level (beyond those with caregivers), for example:** | | | **3.29** |
| *DSM-5 example* | DISCO item | | |
| *difficulties adjusting behaviour to suit different social contexts* | Psychological barriers | 55.6 | 14.88*** |
| | Interrupting conversations | 33.3 | 4.85* |
| | Personal modesty | 47.2 | 7.13** |
| | Anger toward parents | 0 | / |
| | Behaviour in public places | 52.8 | 9.91** |
| | Embarrassing remarks in public | 11.1 | 5.37* |
| | Approaching strangers | 22.2 | .09 |
| *difficulties in sharing imaginative play and in making friends* | Imaginative activities | 44.4 | 11.95*** |
| | Friendship with age peers | 97.2 | 23.25*** |
| | Quality of friendship | 13.9 | 1.29 |
| | Conventions of peer interaction | 27.8 | 3.86* |
| *apparent absence of interest in people* | Does not interact spontaneously with peers | 83.3 | 19.33*** |
| | Lack of awareness of others' feelings | 88.9 | 29.98*** |
| | Response to visitors | 69.4 | 18.79*** |
| **Criterion B – Restricted, repetitive patterns of behaviour, interests, or activities (manifest by at least TWO of the following):** | | | **25.02*** ** |
| **B1** | | | **33.47*** ** |
| **Stereotyped or repetitive speech, motor movements, or use of objects, for example:** | | | |
| *DSM-5 example* | DISCO example | | |
| *simple motor stereotypies* | Rocking (standing up) | 8.3 | 3.98* |
| | Complex movements | 22.2 | 11.33*** |
| | Unusual movements of hands or arms | 47.2 | 13.6*** |
| | Self-spinning | 30.6 | 2.77 |
| *Echolalia* | Immediate echolalia | 44.4 | 5.90* |
| | Delayed echolalia | 61.1 | 31.43*** |
| *repetitive use of objects* | Interest in parts of objects | 16.7 | 3.48 |
| | Elaborate repetitive activities with objects | 25 | 2.85 |
| | Abstract properties of objects | 27.8 | 8.87** |
| | Quality of pattern activities | 58.3 | 21.01*** |
| *idiosyncratic phrases and stereotyped speech* | Long winded pedantic speech | 8.3 | 3.98* |
| | Tone of voice in speech | 33.3 | 9.71** |
| | Idiosyncratic use of words, signs | 13.9 | 4.09* |

| DSM-5 domains and sub-domains | | Frequency | Chi-square |
|---|---|---|---|
| **B2** | | | |
| **Excessive adherence to routines, ritualised patterns of verbal or on-verbal behavior, or excessive resistance to change, for example:** | | | **16.09*** ** |
| *DSM-5 example* | DISCO item | | |
| *motoric rituals, insistence on same route or food* | Maintenance of sameness in routines | 52.8 | 15.05*** |
| | Clinging to home or familiar places | 11.1 | 2.82 |
| | Eats only a small range of foods | 27.8 | 5.19* |
| | Other repetitive routines | 2.8 | 0.31 |
| | Arranging objects | 61.1 | 25.62*** |
| *repetitive questioning* | Repetitive questions | 36.1 | 3.72 |
| | Repetitive themes | 25 | 5.52* |
| | Repetitive acting out of roles | 13.9 | 6.80** |
| *extreme distress at small changes* | Maintenance of sameness of environment | 44.4 | 14.00*** |
| | Insistence on perfection | 30.6 | 3.77 |
| **B3** | | | |
| **Highly restricted fixated interests that are abnormal in intensity or focus, for example:** | | | **10.05** ** |
| *DSM-5 example* | DISCO item | | |
| *strong attachment to or preoccupation with unusual objects* | Fascinated with specific objects | 69.4 | 13.78*** |
| | Collecting objects | 25 | 2.85 |
| | Clinging to objects | 30.6 | 0.82 |
| *excessively circumscribed or perseverative interests* | Fascination with TV/videos | 66.7 | 15.1*** |
| | Collecting facts on specific subjects | 11.1 | 2.82 |
| | Activities related to a special skill | 30.6 | 13.02*** |
| **B4** | | | **16.52*** ** |
| **Hyper or hypo-sensitivity to sensory input or unusual interest in sensory aspects of the environment, for example:** | | | |
| *DSM-5 example* | DISCO item | | |
| *apparent indifference to pain/heat/cold* | Indifference to pain heat or cold | 27.8 | 8.87** |
| *adverse response to specific sounds or textures* | Distress caused by sounds | 61.1 | 8.91** |
| *excessive smelling or touching of objects* | Smelling objects or people | 22.2 | 11.33*** |
| | Touching objects | 38.9 | 8.91** |
| | Repetitive, aimless manipulations of objects (not near eyes) as if seeking sensory stimulation | 30.6 | 10.40*** |

| DSM-5 domains and sub-domains | | Frequency | Chi-Square |
|---|---|---|---|
| *fascination with lights or spinning objects* | Fascination with sounds | 11.1 | 1.36 |
| | Bright lights and shiny objects | 19.4 | 1.19 |
| | Interest in watching things spin | 19.4 | 4.71* |
| | Twists hands or objects near eyes | 22.2 | 8.31** |
| | Interest in looking at objects from different angles | 33.3 | 14.70*** |
| **Criterion C – Symptoms must be present in early childhood, but may not become fully manifest until social demands exceed limited capacities** | | | 14.37*** |
| | Setback in language | 2.8 | 1.29 |
| | Setback in play | 8.3 | 3.98* |
| | Setback in social interaction | 13.9 | 4.09* |
| | Obeying instructions | 83.3 | 5.78* |
| | Combining words | 86.1 | 7.33** |
| | Selective attachment | 50 | 11.53*** |
| | Development of pretend play | 91.7 | 14.86*** |

**(* *p*<.05, ** *p*<.01, *** *p*<=.001)**

#### *5.2.1.2.4 Setting algorithm thresholds*

Each version of the new DISCO algorithm was based on the proposed DSM-5 criteria (accessed Feb, 2012; table 5-1). These specify that individuals must meet all three sub-domains from domain A (social-communication) and two of the four sub-domains from domain B (repetitive behaviours). The threshold or number of items that must be present for an individual to 'score' on each sub-domain differed between the algorithms. Threshold setting was conducted separately for each algorithm version using data from Sample 1 exclusively:

##### *5.2.1.2.4.1 Initial DSM-5 Algorithm:*

All thresholds were set to one item as proposed by DSM-5, and in line with previous literature (e.g. Matilla et al., 2011).

##### *5.2.1.2.4.2 Youden J Algorithm:*

Receiver Operating Characteristic (ROC) curves (a plot of sensitivity against 1-specificity) were used to identify the optimal threshold for each sub-domain. The point at which each ROC curve maximally deviated from the chance line was calculated using the Youden J statistic (maximum = (sensitivity + specificity) - 1). The thresholds selected for each sub-domain according to this method can be seen in Appendix 5 but a summary of the selected thresholds with the corresponding, sensitivity, specificity and Youden J values can be seen in Table 5-2.

##### *5.2.1.2.4.3 Modified DSM-5 Algorithm:*

This algorithm also used sensitivity and specificity values calculated from ROC curves; the threshold was selected that maximised specificity while maintaining the highest level of sensitivity

(see Appendix 5). The thresholds selected for each sub-domain for this approach are higher than the initial algorithm but have not been increased to the same level as the Youden J algorithm. As in McPartland et al. (2012), the original ICD-10 abnormal early development criteria were adopted for domain C (early childhood onset manifesting when social demands exceed capacities). For the DISCO ICD-10 algorithm, at least one of seven possible items must be present (Leekam et al., 2002).

**Table 5-2: Table showing the thresholds and their sensitivity and specificity values for each version of the algorithm across DSM-5 sub-domains**

| Sub-domain | Threshold for Initial | Threshold for Modified | Threshold for Youden J | Sensitivity | Specificity | Youden J |
|---|---|---|---|---|---|---|
| A1 | 1 | / | / | 1 | .258 | .258 |
| | / | 3 | 3 | 1 | .710 | .710 |
| A2 | 1 | 1 | / | 1 | .290 | .290 |
| | / | / | 4 | .833 | .710 | .543 |
| A3 | 1 | / | / | 1 | .032 | .032 |
| | / | 3 | / | 1 | .452 | .452 |
| | / | / | 5 | .917 | .710 | .627 |
| B1 | 1 | 1 | / | .972 | .452 | .424 |
| | / | / | 3 | .750 | .774 | .524 |
| B2 | 1 | 1 | / | .944 | .419 | .363 |
| | / | / | 2 | .750 | .710 | .460 |
| B3 | 1 | 1 | / | .917 | .387 | .304 |
| | / | / | 2 | .750 | .806 | .556 |
| B4 | 1 | 1 | 1 | .972 | .516 | .488 |

### *5.2.1.3 Testing the algorithm*

The three different sets of thresholds were developed using Sample 1 data. Each version of the algorithm was then tested in Sample 1 and in an independent validation sample (Sample 3). ROC curves were used to compute the sensitivity and specificity of each version of the algorithm, while the area under the curve (AUC) was calculated to quantify overall discriminative power. ROC curves plot sensitivity against 1-specificity and the area under the curve (AUC) quantifies the power of the algorithm to correctly classify individuals as belonging to the ASD or non-ASD clinical control group. AUCs of .7 and above are considered acceptable, whereas AUCs of .8 and above are excellent and AUCs of .9 and above outstanding (Hosmer & Lemeshow, 2000).

McNemar's test (McNemar, 1955) was used to compare the proportion of individuals identified as ASD using each version of the algorithm with each of the other versions (Bonferroni corrections for multiple comparisons; $p<.01$). McNemar's test is a comparative method of analysis and allows measurement of the effect of change and is based on the chi-square distribution ($x^2$); like the sign or sign ranked tests it is used on either nominal or ordinal scale data and can be used on

inter-related samples. In this case each individual has two response states, they either have or do not have a clinical diagnosis of ASD and they either do or do not meet DSM-5 DISCO algorithm criteria (according to the three versions of the algorithm). The procedure for calculation of the McNemar's test is shown in below. Yates correction for continuity was applied to all McNemar's tests as the number of discrepancies of individuals across clinical and algorithm criteria were small (McNemar, 1955). The use of McNemar's test in the evaluation of algorithms sensitivity and specificity has previously been used in a comparison of the DSM-5 and DSM-IV-TR criteria (Heurta et al., 2012).

| | | After: − | After: + |
|---|---|---|---|
| Before | + | A | B |
| | − | C | D |

$$\chi^2 = \frac{\left[|A - D| - 1\right]^2}{A + D}$$

### *5.2.1.4 Results*

Sensitivity and specificity for each co-ordinate of the ROC curve are reported in Table 5-2 (above) used for setting thresholds for each sub-domain. The sensitivity and specificity for the overall algorithm using each set of thresholds are presented in Table 5-3.

#### *5.2.1.4.1 Specificity*

The results reported here are for the comparison between the ASD and clinical comparison groups. Specificity was highest in the Youden J algorithm in both Samples 1 and 3 and lowest for the Initial Algorithm (Table 5-3). The difference between these two algorithms was significant in Sample 1($\chi^2_{(1)}$ = 12.02, p<.01) but did not survive correction for multiple comparisons in Sample 3 ($\chi^2_{(1)}$ = 5.04, p<.05). The Modified Algorithm improved specificity relative to the Initial Algorithm in both samples but this effect was only significant in Sample 1($\chi^2_{(1)}$ = 6.04, p<.01). Moreover, the specificity of the Modified Algorithm was not significantly lower than the Youden J algorithm in both samples (p>.01 in both samples).

**Table 5-3: Table showing the sensitivity and specificity of the three proposed algorithms according to current DSM-5 rules (3/3 social and communication; 2/4 repetitive behaviours).**

| | Sample 1 | | | Sample 3 | | |
|---|---|---|---|---|---|---|
| | Initial Algorithm | Youden J Algorithm | Modified Algorithm | Initial Algorithm | Youden J Algorithm | Modified Algorithm |
| LFA | 18 (100%) | 14 (77.78%) | 18 (100%) | 34 (97.14%) | 24 (68.57%) | 30 (85.71%) |
| HFA | 18/18 (100%) | 13 (72.22%) | 18 (100%) | 15 (88.24%) | 5 (29.41%) | 14 (82.35%) |
| ID | 10 (58.82%) | 1 (5.88%) | 5 (29.41%) | 6 (23.08%) | 0 | 3 (11.54%) |
| LI | 5 (35.71%) | 1 (7.14%) | 3 (21.43%) | | | |
| TD | 0/15 | 0/15 | 0/15 | 0/37 | 0/37 | 0/37 |
| ***All controls*** | | | | | | |
| AUC | .84 | .85 | .91 | .92 | .78 | .90 |
| SE | .05 | .05 | .03 | .03 | .05 | .03 |
| Lower | .75 | .76 | .85 | .87 | .69 | .83 |
| Upper | .93 | .95 | .98 | .98 | .87 | .97 |
| Sensitivity | 1 | .75 | 1 | .94 | .56 | .85 |
| Specificity | .67 | .96 | .83 | .91 | 1 | .95 |
| ***Clinical controls*** | | | | | | |
| AUC | .76 | .84 | .87 | .86 | .78 | .87 |
| SE | .06 | .05 | .05 | .05 | .05 | .05 |
| Lower | .64 | .74 | .77 | .75 | .68 | .77 |
| Upper | .88 | .94 | .97 | .96 | .87 | .96 |
| Sensitivity | 1 | .75 | 1 | .94 | .56 | .85 |
| Specificity | .52 | .94 | .74 | .77 | 1 | .89 |

*Note. LFA= low functioning ASD group; HFA = high functioning ASD group*

#### *5.2.1.4.2 Sensitivity*

In both samples, sensitivity was highest for the Initial Algorithm and lowest for the Youden J Algorithm (Table 5-3). This difference was significant in both samples (Sample 1: $\chi^2(1) = 8.03$, $p<.01$; Sample 3: $\chi^2(1) = 19.01$, $p<.01$) using McNemar's test. The sensitivity of the Modified Algorithm was not significantly different from the Initial Algorithm ($p>.01$ in all three samples) but was significantly greater than the Youden J Algorithm (Sample 1: $\chi^2(1) = 8.03$, $p<.01$; Sample 3: $\chi^2(1) =14.02$, $p<.01$).

### *5.2.1.5 Discussion*

To my knowledge, this study was the first to develop and validate a new DSM-5 algorithm from a single standardised diagnostic tool. Three different versions of a new DISCO DSM-5 algorithm were developed from one dataset of children and tested in a separate validation sample. Results using these samples showed that in comparison to a clinical diagnosis the Initial Algorithm had the highest level of sensitivity but lowest specificity, while the Youden J Algorithm had the highest level

of specificity but lowest sensitivity. The Modified Algorithm, which aimed to maximise specificity whilst maintaining the highest level of sensitivity for each sub-domain had comparable sensitivity to the Initial Algorithm and comparable specificity to the Youden J Algorithm in both samples. Overall, the AUC was best for the Modified Algorithm. These results replicate and extend the findings of Frazier et al (2012) by demonstrating good levels of both sensitivity and specificity of the DSM-5 criteria. However, in their study and the current study, the age was limited in these two samples to children and only contained small numbers of individuals.

Comparison of the three algorithm approaches offered a transparent comparison of different methods of applying DSM-5 criteria and the results clearly demonstrate the importance of algorithm design. The thresholds set for each sub-domain had a significant impact on the performance of the algorithm as a whole. This level of sub-domain analysis has had little previous attention in the DSM-5 literature. The performance of these algorithm versions were tested across age and ability level in the second part of this study.

### *5.2.2 Study 2: Further testing of the sensitivity of the final algorithm*

This linked study described below used another independent validation sample (Sample 2). Its purpose was to directly compare between the diagnostic output resulting from the DISCO DSM-5 algorithm solution and the algorithm outputs provided by other DISCO algorithms (independent of the individual's clinical diagnosis). The first aim was follow up on how the three DSM-5 algorithms would perform across age and ability level in individuals who had DISCO algorithm outputs of ICD-10 Childhood Autism or Atypical Autism. Previous literature on the DSM-5 ASD criteria has been predominately limited to children and adolescents, and has found that the sensitivity ranges were higher in children under 4 years old (.90-.98) than in children with language above 10 years of age (.81-.90). Previous research has also found significant differences in sensitivity for DSM-5, with greater sensitivity in low ability individuals as large as 24% (e.g. McPartland et al., 2012; Taheri and Perry, 2012).

The second aim was to carry out further testing of the sensitivity of the final (Modified) algorithm using algorithm outputs provided by other DISCO algorithms, including one previously published algorithm (Gillberg's Asperger syndrome; Gillberg, Gillberg, Rastam, & Wentz, 2001) and another algorithm for Kanner & Eisenberg's Early Childhood Autism originally designed by Lorna Wing for the DISCO, which has not yet been published. As the description of DSM-5 Autism Spectrum Disorder should incorporate the full range of manifestation of autism symptoms, it was expected that individuals who have a diagnosis of Gillberg's Aspgerger syndrome and Kanner and Eisenberg's Autism, should also quality for a diagnosis of DSM-5 ASD. For all these analyses,

sensitivity was calculated. As no comparison group was included in Sample 2, it was not possible to analyse for specificity. However where possible, analyses were also run on the child-only samples of Sample 1 and Sample 3 and the results for these analyses (including specificity) are shown in Appendix 6. Finally, the prevalence of the DISCO items used in the DSM-5 algorithm will be compared across age groups, high and low ability groups and diagnostic sub-groups of childhood autism and Gillberg's Asperger Criteria to identify whether any DISCO items are common to all individuals or whether specific items are more common in a particular subset of individuals. This may be important in determining why the DISCO performs well in comparison to other tools in the literature and in improving the sensitivity of DSM-5 ASD to high functioning individuals.

### *5.2.2.1 Method*

#### *5.2.2.1.1 Participants:*

Sample 2 included 200 individuals whose clinical and demographic characteristics are reported in the Methods chapter and previous reports (Leekam, et al., 2000; Leekam et al., 2007). IQ measures were primarily based on age-appropriate Wechsler Intelligence Scales and the DISCO was conducted with parents/carers during the diagnostic process.

#### *5.2.2.1.2 Measures:*

For the first analysis, the sensitivity of each version of the DSM-5 algorithm (Intial, Youden-J, and Modified) was measured against the DISCO ICD-10 Childhood Autism algorithm output. The original ICD-10 Childhood Autism (CA) algorithm (Appendix 2) was based on 88 DISCO-9 items and a set of rules specifying how these items convert into diagnostic outcome (Leekam et al., 2002). This algorithm, has good inter-rater reliability and discriminant validity (Leekam et al., 2002; Nygren, Hagberg, Billstedt, Skoglund, Gillberg, & Johansson, 2009; Maljaars, Noens, Scholte, & Van Berckelaer-Onnes, 2011) and shows strong agreement with outputs from the ADI-R (Nygren et al., 2009) and ADOS (Maljaars et al., 2011). The full criteria for ICD-10 Childhood Autism can be seen in Appendix 2. As mentioned in Chapter 1, although, the ICD-10 algorithm was selected here, the ICD-10 and DSM-IV-TR diagnoses are equivalent (Volkmar & Klin, 2005) with the two diagnostic labels used interchangeably. ICD-10 terminology was adopted as this algorithm has already been published and tested (see below).

For all subsequent analyses, the sensitivity of the DSM-5 Modified algorithm only was tested on the same Sample 2 dataset by comparing it with the diagnostic outputs for Gillberg's Asperger Syndrome and for Kanner & Eisenberg's Early Childhood Autism algorithm. The full criteria for Gillberg's Asperger Syndrome and for Kanner & Eisenberg's Early Childhood Autism algorithm can be seen in Appendix 2.

As for Study 5.2.1, McNemar's test was used to compare the proportion of individuals identified as ASD using each version of the algorithm (Bonferroni corrections for multiple comparisons; p<.01). Chi-square analyses were conducted to compare sensitivity of each version of the DISCO algorithm as well as to measure the prevalence of DISCO items across age and IQ with Bonferroni correction for multiple comparisons within each sub-domain.

### *5.2.2.2 Results*

#### *5.2.2.2.1 Effect of age and ability level across all three algorithms*

For comparison with the three versions of the DISCO DSM-5 algorithm, the updated (DISCO-11) algorithms for ICD-10 were run on the Sample 2 participants; only participants who met DISCO ICD-10 diagnostic criteria for Childhood or Atypical Autism were included. The final sample (n=190) comprised 112 children (<144 months), 33 adolescents (144-216 months), and 45 adults (>216 months).

Results for the sensitivity of the DISCO DSM-5 algorithms are presented in Table 5-4. Sensitivity was highest for the Initial Algorithm and lowest for the Youden J algorithm, this difference was significant ($\chi^2(1) = 39.01$, p<.01). The sensitivity of the Modified Algorithm was not significantly different from the Initial Algorithm (p>.01) but was significantly different from the Youden J algorithm ($\chi^2(1) = 37.01$, p<.01). Sensitivity did not vary across age-group or ability level for any of the algorithm versions. For all subsequent analysis below, the final (Modified) algorithm only was used.

**Table 5-4: Table showing the sensitivity of algorithms across age and ability (high and low) in individuals in Sample 2 meeting criteria for any ICD-10 diagnosis.**

| | Children | | | Adolescents | | | Adults | | | TOTAL |
|---|---|---|---|---|---|---|---|---|---|---|
| Ability | High (68) | Low (44) | Total (112) | High (19) | Low (14) | Total (33) | High (33) | Low (12) | Total (45) | |
| Initial Algorithm | 96% | 98% | 96% | 90% | 93% | 91% | 91% | 100% | 93% | 95% |
| Youden J Algorithm | 69% | 84% | 75% | 58% | 71% | 64% | 76% | 83% | 78% | 74% |
| Modified Algorithm | 96% | 96% | 96% | 90% | 93% | 91% | 88% | 100% | 91% | 94% |

#### *5.2.2.2.2 Sensitivity of final DISCO DSM-5 algorithm in comparison to ICD-10*

**Table 5-5: Table showing the number of individuals in Sample 2 receiving an algorithm diagnosis of ICD-10 Childhood Autism (CA)**

| DISCO Algorithm diagnosis | | Age and Ability Level | | | | | | |
|---|---|---|---|---|---|---|---|---|
| **ICD-10 CA** | **DSM-5 ASD** | **Children (*n*=119)** | | **Adolescents (*n*=34)** | | **Adults (*n*=47)** | | **Total (%)** |
| | | **Low** | **High** | **Low** | **High** | **Low** | **High** | |
| **+** | **+** | 42 | 64 | 12 | 17 | 12 | 28 | 175 (97.22%) |
| **+** | **-** | 0 | 3 | 0 | 1 | 0 | 1 | 5 (2.78%) |
| **Total** | | 42 | 67 | 12 | 18 | 12 | 29 | 180 |

In Sample 2, 180 individuals met criteria for ICD-10 Childhood Autism (CA). As can be seen in Table 5-5 the majority of these individuals (97.22%) who meet criteria for DISCO ICD-10 CA also meet criteria for DISCO DSM-5 ASD. In individuals who met ICD-10 CA DISCO criteria, there was no effect of ability level ($x^2(1)=.772$, p=.380) or age group ($x^2(2)=1.582$, p=.453) on the sensitivity of the DISCO DSM-5 criteria but there was a significant effect of gender ($x^2(1)=5.636$, $p<.05$), with more males being identified than females.

#### *5.2.2.2.3 ICD-10 Atypical Autism.*

Only ten individuals met criteria for ICD-10 Atypical Autism and no individuals met criteria for ICD-10 Asperger Syndrome. Only three individuals with ICD-10 Atypical Autism were identified by the DSM-5 ASD algorithm.

#### *5.2.2.2.4 Sensitivity of final DISCO DSM-5 algorithm in comparison to other DISCO algorithms*

##### *5.2.2.2.4.1 Gillberg's Asperger Syndrome criteria*

Eighty nine of the sample met criteria for Gillberg's Asperger Syndrome (Leekam et al., 2000; Gillberg, Gillberg, Rastam, & Wentz, 2001) of these 80 also received a diagnosis of DSM-5 (Table 5-6).

**Table 5-6: Table showing the number of individuals from Sample 2 meeting criteria for Gillberg's Asperger Syndrome (GAS) Criteria in comparison to DSM-5 ASD.**

| DISCO Algorithm diagnosis | | Age and Ability Level | | | | | | |
|---|---|---|---|---|---|---|---|---|
| **GAS** | **DSM-5** | **Children (*n*=119)** | | **Adolescents (*n*=34)** | | **Adults (*n*=47)** | | **Total (%)** |
| | | **Low** | **High** | **Low** | **High** | **Low** | **High** | |
| **+** | **+** | 6 | 27 | 3 | 15 | 5 | 24 | 80 (89.90%) |
| **+** | **-** | 0 | 5 | 0 | 1 | 0 | 3 | 9 (10.11%) |
| **Total** | | 6 | 32 | 3 | 16 | 5 | 27 | 89 |

#### *5.2.2.2.4.2 DISCO algorithms – Kanner and Eisenberg's Early infantile Autism*

Seventy one individuals in Sample 2 had a diagnosis of Kanner and Eisenberg's Early Infantile Autism and all of them also qualified for a diagnosis of ASD using the DSM-5 DISCO algorithm (Table 5-7).

**Table 5-7: Table showing the number of individuals from Sample 2 meeting criteria for Kanner and Eisenberg's Early Infantile Autism Criteria in comparison to DSM-5 ASD.**

| DISCO Algorithm diagnosis | | Age and Ability Level | | | | | | |
|---|---|---|---|---|---|---|---|---|
| **KEAUT** | **DSM-5** | **Children (*n*=119)** | | **Adolescents (*n*=34)** | | **Adults (*n*=47)** | | **Total (%)** |
| | | **Low** | **High** | **Low** | **High** | **Low** | **High** | |
| **+** | **+** | 27 | 26 | 3 | 3 | 7 | 5 | 71 (100%) |
| **+** | **-** | 0 | 0 | 0 | 0 | 0 | 0 | 0 |
| **Total** | | 27 | 26 | 3 | 3 | 7 | 5 | 71 |

#### *5.2.2.2.5 Exploration at the item level*

Analyses were conducted to identify items that were more commonly found in particular subsets of individuals across the spectrum. The majority of items (73%) were comparable across age and ability levels. Indeed three items were highly frequent (>90%) in both ability groups and child/adult age groups: sharing interests and enjoyment, friendships; and awareness of others' feelings. Two additional items (giving comfort to others and does not interact with peers) were highly frequent in both children and adults.

Nineteen items were identified with significantly different frequencies in the high and low functioning groups (Table 5-8); nine were more prevalent in the high functioning ASD group than the low functioning these are marked with a * in Table 5-8. (reciprocal communication, interrupting conversations, anger toward parents, long-winded and pedantic speech, maintenance of sameness in routines, repetitive themes, insistence on perfection, collecting facts on specific subjects, repetitive activities related to special skills) and 10 items were significantly more prevalent in the low functioning group (facial expression; descriptive gestures; nodding/shaking of head; using other people as mechanical aids; psychological barriers; personal modesty; rocking while standing up; twisting hands or objects near eyes; and imaginative activities).

Seven of the nine items more prevalent in the high than low functioning individuals were also found to be significantly more frequent in individuals who met criteria for Gillberg's Asperger Syndrome than individuals who met criteria for ICD-10 Childhood Autism only: reciprocal communication, interrupting conversations, anger toward parents, long-winded and pedantic speech, repetitive themes, collecting facts on specific subjects, repetitive activities related to special skills. In addition, embarrassing remarks in public, quality of friendship, conventions of peer interactions, tone of voice and repetitive questions were also significantly more prevalent in the Gillberg's Asperger than the ICD-10 Childhood Autism group.

Similarly, six had significantly different frequencies for adults compared with all individuals below 18 years old, with a further five when adults and children below 12 years were compared. Of these 11 items, six were more prevalent in adults (anger toward parents, imaginative activities, long-winded and pedantic speech, tone of voice, repetitive themes, collecting facts on specific subjects) and 5 were more prevalent in the younger age groups (fascination with TV/videos, food fads; self-spinning; using other people as mechanical aids; and understanding of gesture).

**Table 5-8: Table identifying the DISCO items that differ significantly between high and low ability individuals or between children and adults in Sample 2.**

| Sub-domain | DISCO item | % individuals scoring on DISCO item with significant chi-square | | | | | | |
|---|---|---|---|---|---|---|---|---|
| | | IQ | | Age | | | Gillberg's Asperger | |
| | | LFA | HFA | Children | Children & adolescents | Adults | CA only | Gillberg |
| A1 | Reciprocal Communication* | 34.3 | 63.3 | | | | **37.6** | **72.8** |
| A2 | Facial expression | 75.7 | 54.2 | | | | | |
| A2 | Descriptive gestures | 82.9 | 62.5 | | | | | |
| A2 | Nodding and shaking of head | 58.6 | 30.8 | | | | 56 | 21 |
| A2 | Understanding of gesture | 35.7 | 7.5 | 25.9 | 22.8 | 2.2 | 31.2 | 0 |
| A2 | Instrumental gestures | | | | | | 73.4 | 51.9 |
| A2 | Using adults as mechanical aids | 54.3 | 27.5 | 49.7 | | 20 | 52.3 | 17.3 |
| A3 | Psychological barriers | 68.6 | 40.8 | | | | 60.6 | 38.3 |
| A3 | Interrupting conversations* | 40 | 61.7 | | | | **36.7** | **76.5** |
| A3 | Personal modesty | 75.7 | 40.8 | | | | | |
| A3 | Anger toward parents Anger*# | 4.3 | 25 | 11.6 | 12.4 | 33.3 | **4.6** | **34.6** |
| A3 | Embarrassing remarks in public | | | | | | ***18.3*** | ***55.6*** |
| A3 | Imaginative activities# | 68.6 | 44.2 | 43.8 | | 71.1 | | |
| A3 | Quality of friendship | | | | | | ***9.2*** | ***25.9*** |
| A3 | Conventions of peer interactions | | | | | | ***19.3*** | ***63*** |
| B1 | Rocking (standing up) | 18.6 | 5 | | | | | |
| B1 | Long winded pedantic speech*# | 7.1 | 33.3 | 12.5 | 16.6 | 46.7 | **5.5** | **48.1** |
| B1 | Tone of voice # | | | 41.1 | | 66.7 | ***32.1*** | ***74.1*** |
| B1 | Self-spinning | | | 35.7 | | 11.1 | | |
| B2 | Maintenance of sameness in routines* | 41.4 | 69.2 | | | | | |
| B2 | Repetitive questions | | | | | | ***19.3*** | ***61.7*** |
| B2 | Repetitive themes *# | 27.1 | 53.3 | 29.5 | 37.9 | 62.2 | **17.4** | **79** |
| B2 | Insistence on perfection* | 24.3 | 49.2 | | | | | |
| B2 | Eats small range of foods | | | 41.1 | | 15.6 | | |
| B3 | Collecting facts on | 2.9 | 31.7 | 8 | 15.2 | 40 | **3.7** | **44.4** |

| | | | | | | | | |
|---|---|---|---|---|---|---|---|---|
| | specific subjects *# | | | | | | | |
| B3 | Fascination with TV/videos | | | 48.2 | 46.9 | 22.2 | | |
| B3 | Repetitive activities related to special skills* | 14.3 | 31.7 | | | | **9.2** | **46.9** |
| B4 | Twists hands or objects near eyes | 24.3 | 8.3 | | | | | |

*Note.* *Items more frequent in high vs low ability # Items more frequent in adults than children

### *5.2.2.3 Discussion:*

The study adds new evidence to the debate that some individuals who currently meet DSM-IV-TR criteria may be missed by DSM-5, and the inclusion of adults addresses a clear limitation of the existing literature (e.g. Huerta et al., 2012). All algorithm versions performed comparably in high and low functioning individuals, which is in stark contrast to previous findings, which found significant differences between individuals with IQs above or below 70, however, these studies (McPartland et al., 2012 and Taheri & Perry, 2012) were conducted with DSM-IV data, using checklists or clinical judgement, the same IQ differences were not found when standardised instruments were utilised (e.g. Frazier et al., 2012). Moreover, the performance of the DISCO algorithms was comparable for children, adolescents and adults. The range of the sensitivity for the modified algorithm across age and IQ groups only varied from .90 - .98 in sensitivity. In Heurta et al., (2012) the sensitivity was high for individuals below four years (.90-.98) but lower in children over 10 years of age (.81-.90) but these results were confounded with language level in the children above 10 years and only limited to children and adolescents rather than the large age range in the current study. The current results suggest that according to the DISCO DSM-5 algorithms, and particularly the Modified Algorithm, individuals across a broad range of age and abilities will receive a DSM-5 diagnosis. The Modified algorithm was therefore selected as the recommended version to use with the DISCO.

The good sensitivity results presented in the current study and the combined sensitivity and specificity results from Study 5.2.2 are in contrast to a large portion of research that has previously been conducted which raised concerns that individuals who currently meet criteria especially high functioning individuals are likely to miss a diagnosis according to DSM-5 (e.g. McPartland et al., 2011). However, while sensitivity was good, it was not perfect for those with milder forms of autism or higher ability. Five with higher ability who had ICD-10 Childhood Autism diagnosis did not receive a DSM-5 diagnosis and only 3/10 of those with ICD-10 Atypical Autism did so. Likewise, while every individual who had a diagnosis of Kanner & Eisenberg Childhood Autism also had DSM-5 diagnoses

only 80/89 of those with Gillberg's Asperger Syndrome were captured. Additional analyses exploring where individuals failed to meet DSM-5 ASD criteria are presented in Appendix 7.

Endorsement for the majority of items was consistent across age and ability. Indeed, a small set of items were observed in above 90% of cases in the high and low ability groups and in both children and adults. All five of these items were also identified as being highly discriminating ($p<.001$) in Table 5-1 at the beginning of the current chapter. These highly frequent items were all social in nature. Their descriptions in the DISCO allow them to capture the range of individuals seen across the spectrum.

In addition, a small minority of algorithm items were more relevant for higher functioning and older individuals. These items were more common in domain B, suggesting that items relating to restricted and repetitive language (e.g. pedantic or long winded speech), repetitive activities related to a special skill, or collecting facts in a specific subject might identify higher functioning individuals and adults with ASD. Some of these items such as the repetitive language behaviours described in the DISCO and DSM-5 criteria can only be present in higher functioning verbal individuals, however, this still highlights these symptoms and the other behaviours described in Domain B of the proposed DSM-5 criteria are important to measure correctly as they may be important for capturing individuals who have had concerns raised about their meeting of the proposed criteria.

One explanation for the improved sensitivity of DSM-5 when measured with the DISCO in comparison to the original tests of the proposed criteria is the broad range of items offered. This gives the DISCO an advantage over other measures for measuring the spectrum because it contains items relevant to the whole spectrum but also items that are more likely to capture specific sub-groups of individuals as well. The combination of these global, as well as more specific, items provides a sensitive measure of DSM-5 ASD.

## 5.3 General Discussion

This chapter presented the design and validation of algorithms for the criteria for DSM-5 ASD. The resultant algorithms performed well when sensitivity and specificity was assessed in comparison to individuals' clinical diagnoses. There were significant differences between the three versions of the algorithms. This result highlights how important algorithm design is as it has impacts on sensitivity and specificity. The algorithm version with the best balance and AUC was the Modified Algorithm.

The DISCO appears to have an important role to play in capturing DSM-5 ASD as the many examples of behaviours selected for use in the study allow the DISCO DSM-5 algorithm to capture the high functioning individuals, originally thought to be at risk of being missed by DSM-5 through

previous work with other methods such as DSM-IV checklists (e.g. McPartland et la., 2011). The main difference of the DISCO in comparison to the ADI-R and the DSM-IV checklists is this broad range of items. From the analyses presented in the current chapter and conducted beyond this thesis, it is concluded that the range of examples of behaviour selected from the DISCO by researchers and clinicians that are an important factor in maintaining sensitivity across changes in diagnostic criteria.

### *5.3.1 Limitations*

Compared with some of the published papers testing the DSM-5 criteria (e.g. Frazier et al., 2012; Huerta et al., 2012), the sample size in the current study is relatively modest. Although a range of age and ASD symptoms were included in Sample 2, this is a clear limitation of this work and much more work with other cases of ASD – including the female profile – across the age-span is needed using the DISCO DSM-5 algorithm specifically and also in this area more generally, the effect on the female profile has not been examined.

Another limitation of the data, particularly relevant to the analyses conducted in this Chapter is the recruitment of individuals in Samples 1 and 3 into groups specifically selected for ASD and non-ASD comparison groups. Large-scale representative population studies will be essential to clarify the capacity of the DSM-5 algorithms to differentiate ASD from other developmental disorders. A clear test of the validity of the DISCO DSM-5 algorithms will be their success when used in larger samples of individuals referred for assessment through standard clinical care pathways rather than measured in individuals selected for inclusion according to their diagnostic label. Meanwhile, this limitation is not true for Sample 2 in which individuals were tested during their referral to a tertiary diagnostic centre, this work demonstrates that the majority of individuals with a previous diagnosis according the DISCO algorithms also meet DSM-5 ASD criteria using a validated DISCO algorithm. However, the Lorna Wing centre for Autism where these individuals were referred to does have a particular focus on the spectrum nature of ASD and ideally the effect of the DISCO items capturing the spectrum versus the potential bias of the clinicians at the centre collecting the information needs to be balanced. Again, this will be overcome by testing the DISCO algorithm in large scale representative studies.

### *5.3.2 Summary*

Overall, the DISCO appears to provide a reliable and valid algorithm to capture the new DSM-5 ASD criteria. The DISCO was originally based around the concept of an autism spectrum and therefore predicted that it should perform well. However, the DISCO also has its own algorithm designed to measure the autism spectrum and it would be valuable to examine how these two

descriptions align and the impact on sensitivity and specificity according to the two spectrum approaches.

# 6 Measuring Autism Spectrum Disorder: Wing and Gould's Autism Spectrum

Chapter 6 and 7 examine Wing & Gould's definition of Autism Spectrum Disorder. The main goal of these chapters is to compare the measurement of Wing & Gould's ASD criteria with the measurement of DSM-5 ASD using the DISCO and to evaluate the effectiveness of the Wing and Gould criteria for describing and discriminating key elements of the autism 'spectrum'. The first chapter (Chapter 6) examines the psychometric properties of the Wing and Gould Autism Spectrum Disorder (WG-ASD) algorithm for the DISCO in comparison to the DISCO algorithm for DSM-5. The second chapter (Chapter 7) is dedicated to analysing a single element within the Wing and Gould algorithm, Wing & Gould's Quality of Social Interaction.

## 6.1 Literature review

### *6.1.1 Background: Wing and Gould ASD algorithm*

The Wing & Gould algorithm is based on the main observations from Wing and Gould's (1979) epidemiological study. The first criterion they described relates to a dimension of social interaction style (Quality of social interaction). This was described as ranging from aloofness or indifference to others, to passive acceptance and lack of spontaneity to active but odd social approaches, in which individuals attempted to engage with everyone (friends, family and strangers) but in a limited form such as asking everyone the same set of repetitive questions (see Wing, 1991a). The second criterion refers to communication impairments varying from no attempts to communicate, communicating needs only to a lack of reciprocal communication such as asking repetitive questions or engaging in monologues. Finally a dimension of imagination impairment was described. This ranged from absent, or limited play copied from others to having spontaneous imaginative play that is repetitive or cannot be modified by others. This impairment in imagination was described as being related to a limited pattern of behaviours, also graded from absence of all activities to a single or limited focus self-chosen interest. When the DISCO was designed, the spectrum graded nature of these impairments was measured against the individual's developmental level, either recording the level achieved or whether there was a delay developing these skills.

The algorithm for Wing and Gould's autism spectrum was published in 2002 (Leekam et al., 2002). It is based on only four criteria (5 items) covering the quality of social interaction, communication, pattern of activities and imagination (2 items), the items are listed below. This algorithm attempts to capture the overall quality; the selected items tend to summarise the

information collected from other items in the DISCO sections e.g. limited pattern of activities. In order to receive a diagnosis all four criteria must be met.

**Quality of social interaction**

0 Does not interact; aloof and indifferent to everyone
1 Interacts to obtain needs, otherwise indifferent
2 Responds to (and may initiate) physical contact only; including rough and tumble games, chasing, cuddling, but is otherwise indifferent
3 Generally does not initiate but responds to social (not just physical) contact, if others, including age peers, make approaches. Joins in passively, e.g. as baby in game of mothers and fathers, or for adults, in adult social situations. Tries to copy, but with little understanding. Shows some pleasure in passive role (unlike Groups 0,1,2, who move away once physical needs are satisfied).
4 Makes social approaches actively, but these are usually inappropriate, 'one-sided', naive and peculiar. The behaviour is not modified according to the needs, interests and responses of the person approached.
5 Over formal, stilted, rigid, over polite and calmly outspoken in social interaction. This can be a subtle problem, but becomes more apparent on prolonged acquaintance
6 Shy, but social contacts appropriate for mental age with well-known people, including age peers. Also use this rating for children who refuse to talk to adults, but interact with other children.
For older children and adults, this rating can be used for those who are not gregarious, but who can interact appropriately with people they like. It can also be used for those who have periods of social withdrawal due to psychiatric illness or moodiness but who otherwise interact appropriately
7 Social contacts with children and adults appropriate for A's level of ability. Looks up with interest and smiles when approached. Responds to the ideas and interests of people of similar mental age and contributes to the interaction. Non-mobile people without speech can show social interest by means of eye contact and 'eye pointing'
8 Selective mutism, not socially impaired as in autism spectrum disorders.
9 Shy, withdrawn but socially aware behaviour typical of Fragile X syndrome.
10 Chatty, friendly behaviour typical of Williams syndrome.

**Reciprocal communication**

If A communicates in any way, is this a two-way communication, or is it one-sided on A's terms only, concerning only A's needs or interests? Does A respond with interest to replies and follow the theme of an interchange?

0 Communicates needs only
1 Communicates only on own terms, one-sided, repetitive
2 Enjoys reciprocal communication at level of ability
-8 Does not communicate

**Imaginative activities – *rate the highest level achieved.***
Does/Did A have any pretend play or other imaginative activities? (Rate on level of imaginative activities, even if they are repetitive. If A has no play or other imaginative activities for any reason, use 0. If A is too old for pretend play, ask about past behaviour and rate on level reached).

0 No play with model toys (e.g. no interest in the function of trains, cars and dolls, although A may handle them in the same way as any other objects)
1 Plays with real household equipment using it for its real purpose. No interest in miniatures,(e.g. sweeps with real broom, digs with real spade)
2 Holds doll, toy animals as if real, at least some of the time (e.g. hugs and kisses toys)
3 Goes through simple sequences of actions with toys as if they are real (e.g. pushes toy trains and cars along floor as if real, and makes appropriate noises, or tucks doll in bed)
4 Will pour out and give a pretend cup of tea to other person spontaneously (If A only drinks from cup rate 3)
5 Goes through longer sequences of actions with toys e.g. has a doll's tea party, sets up a garage, road and road bridges for play with toy cars

**Repetitive pretend play**
If A has pretend play, is this varied and showing development, or is it repetitive, always repeating the same series of actions?

0 Marked repetition
1 Sometimes repetitive
2 Play is varied
-8 No spontaneous pretend play

**Limited pattern of self-chosen activities**
What does A do if left to his / her own devices or is left to choose activities? Can you give me a list of A's usual activities when nothing is provided or suggested?

0 Engages only in repetitive activities
1 Has some varied interests but repetitive activities are a prominent part of S' repertoire
2 Activities varied and flexible
-8 No self-chosen activities

The psychometric properties of the Wing and Gould ASD algorithm were tested using the same sample used in this thesis (Sample 1), comprising 36 children with ASD, 31 clinical comparison and 15 typically developing individuals aged 35-140 months old (Leekam et al., 2002). The WG-ASD algorithm was found to significantly related to diagnostic outcome ($k$ = .79) according to clinical diagnoses made by independent clinician's before recruitment to the study and this was not significantly related to non-verbal IQ, chronological age or language comprehension or expression. However, there were some minor discrepancies, where the WG-ASD algorithm failed to identify three individuals with a clinical diagnosis, these individuals failed to score on the imagination criterion (2 cases) or for reciprocal communication (1 case). In addition, the WG-ASD algorithm had better specificity than the DISCO ICD-10 algorithm which identified 14 of the clinical comparison group whereas the WG-ASD algorithm only identified four. The WG-ASD algorithm identified 92% of

the individuals meeting criteria for ICD-10 Childhood Autism. The algorithm output for two independent raters were also extremely accurate for the WG-ASD algorithm (*k*=.82) as well as the coding of the items in the algorithm. It has also been shown that 66.7% for ever codes and 50% of current codes of the WG-ASD items have inter-rater reliability above r=.75 (Nygren et al., 2009).

To date, no other research has been carried out to study the effectiveness of the diagnostic criteria of the WG-ASD algorithm. Some research has been conducted on the Quality of Social interaction item alone, and this will be reviewed in Chapter 7. In the current Chapter 6, the focus is on the combination of items (the W&G triad of impairments) that comprise the W&G algorithm for Autism Spectrum Disorder and their comparison with the DSM-5 algorithm for Autism Spectrum Disorder.

The algorithms for DSM-5 and WG-ASD are remarkably similar in the behaviours that they are measuring, except the dramatic difference of 85 items to five items. Comparison between the items in WG-ASD that are also found in DSM-5 DISCO algorithm can be seen from Table 6-1. Both item sets cover social and communication impairments focussing on the reciprocal nature and quality of these behaviours. However, the DSM-5 algorithm has a wide range of items measuring restricted and repetitive behaviours that cover much more than just "limited pattern of activities". In contrast, the WG-ASD algorithm also puts emphasis on the imaginative activities of individuals with ASD, a factor that has been given less focus in the new DSM-5, although one item still falls with the social and communication (A) domain: "imaginative activities". If these behaviours are measuring the same behaviours then it would be predicted that the DSM-5 and WG-ASD measures of the spectrum would identify the same individuals.

**Table 6-1: Table showing the Wing and Gould ASD algorithm and overlap with the DISCO DSM-5 algorithm**

| Item | Code | In DSM-5? |
|---|---|---|
| 1. Quality of social interaction | 0,1,2,3,4,5 | No |
| 2. Reciprocal Communication | 0,1,8 | Yes |
| 3. Limited pattern of activities | 0,1,-8 | Yes |
| 4. Either: | | |
| a. Imaginative activities | 0,1 | Yes |
| OR b. Repetitive pretend Play | 0,1 | No |

Recent findings using the DSM-5 ASD criteria have shown the criteria to have good sensitivity when applied to individuals who already receive a diagnosis according to DSM-IV if standardised diagnostic tools are used (Heurta et al., 2012). Furthermore, evidence from Chapter 5 indicates that the DISCO DSM-5 algorithm was accurate at identifying individuals with a previous clinical diagnosis of Autistic Disorder/Childhood Autism and those with ICD-10 Childhood Autism according to the DISCO algorithms for this diagnosis. However, the original DSM-5 algorithm only

captured 30% (3/10) of the ICD-10 Atypical Autism cases and 89% of the individuals who met criteria for Gillberg's Asperger Syndrome. Although, these missed cases are the minority of individuals across the samples, a different spectrum algorithm may be more successful at identifying all diagnostic sub-groups. It is useful to compare DSM-5 ASD to the Wing and Gould Autism Spectrum (WG-ASD) measure and to compare how well the WG-ASD captures the diagnostic sub-groups described in the previous chapter (ICD-10 atypical autism, Kanner's early infantile autism and Gillberg's criteria for Asperger Syndrome) in comparison to DSM-5. If the WG-ASD captured all these individuals meeting criteria for ICD-10 categories and Asperger Syndrome then it would have a clear strength over the DISCO DSM-5 algorithm and could highlight that the spectrums are not identical.

The purpose of this chapter is to compare the two algorithms designed to measure the spectrum (DSM-5 ASD and WG-ASD). A serious challenge confronts this enterprise however, because the items for both these algorithms were designed by Lorna Wing and are used within the same interview. Therefore there is a measurement bias. Not only may both algorithms be driven by the same underlying concept of ASD but also the same measurement items may be creating an effect of convergence. This is a difficult challenge to overcome by comparing the outputs of the two algorithms but in the first two studies of this chapter, I attempt to some way to guard against this bias.

In Study 6.2.1 the sensitivity and specificity of WG-ASD algorithm as a whole was first tested using ROC curve analysis. I explored the WG-ASD items further to examine how well they perform independently in comparison to the full WG-ASD algorithm. The logic was to test the original claim of Wing and Gould (1979) that these items form a triad of behaviours: social interaction, social communication and social imagination (now social imagination) that co-occur together and are associated with a pattern of repetitive and restricted behaviours. Comparisons were then made between the two spectrum algorithms WG-ASD and DSM-5 as well as between WG-ASD and other diagnostic algorithms (e.g. Gillberg's Asperger Syndrome, Kanner & Eisenberg Early Autism). In Study 6.2.2 the focus was on the overlap between the two spectrum algorithms at the item level. The hypothesis outlined above is that the algorithms perform similarly because a small number of items in the WG-ASD algorithm are summary items that cover the behaviours covered by the many items in the DSM-5 ASD algorithm. To attempt to investigate this, I reasoned that if the WG-ASD items are essential to performance of the DSM-5 algorithm then removal of these WG-ASD items would result in loss of sensitivity and/or specificity. In Study 6.2.2 I tested the DSM-5 algorithm with the overlapping WG-ASD items removed. I also examined whether each domain of the DSM-5 algorithm would be able to be substituted for WG-ASD items without a loss of sensitivity.

### 6.1.2 General Methods

#### 6.1.2.1 Participants

The same three samples used to assess the DSM-5 ASD algorithm will be used again in the current chapter. Samples 1 and 3 allow both sensitivity and specificity measures to be computed as both contain clinical control groups as well as a clinically defined ASD group. Sample 2 offers a larger range of age and ability level for measuring the DISCO algorithms but has no control group to assess specificity.

#### 6.1.2.2 Statistical analyses

All ROC curve analyses and Chi-square discriminations were conducted between the ASD group and the clinical control groups. The typically developing individuals are included in tables for descriptive purposes. Chi-square analyses were also conducted in Sample 2 to compare across age groups, ability level and gender.

## 6.2 Empirical work

### 6.2.1 Study 1: Testing the WG-ASD algorithm

The aim of the first study was to run the algorithms for Wing and Gould's ASD across the three samples to assess the sensitivity and specificity of this algorithm in comparison to the results achieved by the DSM-5 ASD algorithm. This algorithm has been previously published (Leekam et al., 2002), however, the published study did not use ROC curve analysis. Therefore the results for sensitivity and specificity were extrapolated from Leekam et al (2002) to allow comparison across studies. The second aim was to assess the overlap in diagnostic outcome of the DSM-5 and WG-ASD algorithms and to examine how individuals who met criteria for WG-ASD also met criteria for ICD-10 Childhood Atypical Autism, Gillberg's criteria for Asperger Syndrome and Kanner's Early Infantile Autism).

There were three aims of Study 6.2.1:

- Assess the sensitivity and specificity of the WG-ASD algorithm and the individual items using ROC curve analysis.
- Examine the associations between the four criteria utilised in the WG-ASD algorithm.
- Compare output of DISCO WG-ASD algorithm with the output for DISCO algorithms for DSM-5, Gillberg's Asperger, Kanner & Eisenberg Infantile Autism and ICD-10 Atypical autism.

### 6.2.1.1 Procedure

The sensitivity and specificity of the WG-ASD algorithm was tested in Sample 3 using ROC curve analyses to assess the sensitivity and specificity of the WG-ASD algorithm in comparison to clinical comparison groups. Typically developing cases are included in the table for descriptive purposes only. Sensitivity was also assessed in Sample 2, chi-square analyses were used to measure the association between the WG-ASD algorithm and individual's age, ability level and gender. To assess how sufficient these items are independently, two sets of analyses will be conducted: ROC curve analyses will assess each of the four criterion separately against individuals' clinical diagnoses (ASD or non-ASD clinical controls) to provide sensitivity and specificity scores; and chi-square analyses will assess each items association with diagnostic group (ASD or non-ASD clinical controls). In addition, the strength of these items across age and IQ will be assessed using chi-square analyses with Bonferroni correction for multiple analyses in Sample 2.

Finally, assessing the relation between items will be conducted using correlation analysis (Pearson's r).

In order to assess how well the algorithm for WG-ASD covers the previous categorical sub-diagnoses of ICD-10 Atypical Autism and Gillberg's Asperger Syndrome, Mcnemar's tests were conducted on the difference in sensitivity and specificity between the algorithms presented in the previous chapter (Gillberg's Asperger criteria, ICD-10 Atypical Autism and Kanner and Eisenberg's Early Infantile Autism) as well as the general overlap with the DSM-5 algorithm and the new WG-ASD algorithm.

### 6.2.1.2 Results

#### 6.2.1.2.1 Aim 1: Sensitivity and specificity of the WG-ASD algorithm

The results from Leekam et al (2002) were extrapolated to create sensitivity and specificity scores for Sample 1 to allow comparison (Table 6-2). This was done by identifying the numbers of individuals meeting criteria for WG-ASD across the clinically defined ASD and comparison groups to plot the ROC curves. The number of individuals meeting criteria for the Wing and Gould algorithm for ASD (WG-ASD) and the ROC curve analysis results for Sample 3 can be seen in Table 6-2.

**Table 6-2: The diagnostic accuracy of the Wing and Gould Autism Spectrum Disorder algorithm in Samples 1 and 3.**

| | Sample 1 (%) | Sample 3 (%) |
|---|---|---|
| ASD – low ability | 88.89 | 88.60 |
| ASD – high ability | 94.44 | 70.60 |
| Intellectual disability | 23.53 | 3.85 |
| Language impairment | 0 | |
| Typically developing | 0 | 0 |
| AUC (standard error) | .894 (.044) | .894 (.039) |
| Sensitivity | .917 | .827 |
| Specificity | .871 | .962 |

The sensitivity of the WG-ASD algorithm was also high across age and ability level in Sample 2. Of the 200 individuals referred with impairments in social and communication 110 children (68 high; 42 low), 30 adolescents (18 high; 12 low) and 44 adults (32 high; 12 low) met criteria for WG-ASD (total=184). Using chi-square tests there were no significant differences for the WG-ASD algorithm across ability level ($x^2(1)=.764$, p=.382, n.s.), age group ($x^2(2)=.853$, p=.653, n.s.) or gender ($x^2(1)=2.746$, p=.097, n.s.). Of the individuals that were missed by the WG-ASD algorithm (n=16), the D rule (limited pattern of activities) missed the most (50%) and both the B (reciprocal communication) and the C rule (imagination) missed 37.5% of these individuals, whereas the A rule (quality of social interaction) only missed three (19%) of the missed cases.

The success of each WG-ASD item alone at predicting diagnostic outcome was assessed using ROC curve analyses in both Sample 1 and 3. As shown in Table 6-3, each of the WG-ASD items and criteria perform relatively well and each of the 5 items and 4 criteria significantly predict whether an individual has a clinical diagnosis of ASD or not using chi-square statistics.

**Table 6-3: Table showing the diagnostic accuracy and discriminative power of the Wing and Gould ASD items alone.**

| WG-ASD rule | AUC | | Standard Error | | Sensitivity | | Specificity | | Chi-square | |
|---|---|---|---|---|---|---|---|---|---|---|
| Sample | 1 | 2 | 1 | 2 | 1 | 2 | 1 | 2 | 1 | 2 |
| A – Social | .839 | .913 | .054 | .044 | 1 | .981 | .677 | .846 | 35.52** | 57.01** |
| B – Communication | .647 | .856 | .069 | .055 | .972 | .981 | .323 | .731 | 10.55** | 46.03** |
| C – Imagination | .636 | .740 | .069 | .066 | .917 | .942 | .355 | .538 | 7.43* | 23.51** |
| *Imaginative activities* | *.646* | *.712* | *.067* | *.059* | *.389* | *.500* | *.903* | *.923* | *7.51** | *13.48*** |
| *Repetitive pretend play* | *.636* | *.740* | *.069* | *.066* | *.917* | *.942* | *.355* | *.538* | *7.43** | *23.31*** |
| D – pattern of activities | .712 | .798 | .066 | .057 | .972 | .827 | .452 | .769 | 17.22** | 26.38** |

** p<.001, * p<.05

In Sample 2, a series of chi-square analyses were conducted for each WG-ASD criteria and item across age group (child, adolescent and adults) and ability level (high or low) with Bonferroni correction for multiple tests. The descriptive statistics are shown in Table 6-4. The only significant chi-square statistics was found for age ($x^2(2) = 15.899$, $p>.001$) and ability ($x^2(1) = 8.378$, $p<.01$) groups for imaginative activities (C1 rule).

**Table 6-4: Table presenting the sensitivity of the WG-ASD algorithm items across age and ability level in Sample 2.**

| | Children | | Adolescents | | Adults | |
|---|---|---|---|---|---|---|
| | Low | High | Low | High | Low | High |
| A – Social | 95.5% | 92% | 85.7% | 95% | 100% | 94.3% |
| B – Communication | 95.5% | 96% | 85.7% | 95% | 100% | 94.3% |
| C – Imagination | 95.5% | 97.3% | 85.7% | 95% | 100% | 91.4% |
| *Imaginative activities* | 50% | 28% | 71.4% | 50% | 91.7% | 60% |
| *Repetitive pretend play* | 95.5% | 96% | 85.7% | 90% | 100% | 88.6% |
| D – pattern of activities | 95.5% | 98.7% | 85.7% | 90% | 100% | 97.1% |
| WG-ASD | 95.5% | 90.7% | 90% | 90% | 100% | 91.4% |

#### *6.2.1.2.2 Aim 2: Examine the associations between the five items (and four criteria) utilised in the WG-ASD algorithm.*

Correlation analyses were conducted across the four criteria specified in the WG-ASD algorithm. As shown in Table 6-5 there were statistically significant relations between all four criteria in the WG-ASD algorithm. In order to confirm that the same level of significant relations between items cannot be found across any selection of items, randomly selected items were also compared and the highest correlation was .206 (see Appendix 8 ).

**Table 6-5: Table showing correlational analyses (r) between the WG-ASD algorithm items in Sample 2**

| | Quality of social interaction rule | Reciprocal communication rule | Imagination rule | Limitied pattern of activities rule |
|---|---|---|---|---|
| Quality of social interaction rule | 1 | .777* | .684* | .671* |
| Reciprocal communication rule | | 1 | .789* | .773* |
| Imagination rule | | | 1 | .656* |
| Limitied pattern of activities rule | | | | 1 |

*p<.001

#### *6.2.1.2.3 Aim 3: Overlap between the DISCO algorithms for WG-ASD and algorithms for DSM-5, ASD, Gillberg's Asperger Syndrome, Kanner & Eisenberg Infantile Autism and ICD-10 Atypical Autism*

The overlap between WG-ASD and DSM-5 was assessed across all three samples; 92% of individuals with ASD meeting DSM-5 criteria in Sample 1 met WG-ASD criteria; 89% in Sample 3, however the WG-ASD algorithm also identified individuals that were missed by DSM-5. Of the individuals with WG-ASD, the DSM-5 algorithm identified 91% (Table 6-6). In both samples there was no difference in the number of individuals identified with each algorithm in terms of either sensitivity, in individuals with ASD (Sample 1: $x^2(1)=2.08$, n.s.; Sample 3: $x^2(1)=0.25$, n.s.) or specificity in the control groups (Sample 1: $x^2(1)=3.06$, n.s.; Sample 3: $x^2(1)=0.25$, n.s.) using McNemar's test. The breakdown of the overlaps can be seen in Table 6-6.

**Table 6-6: Table showing the overlap between WG-ASD and ICD-10 Childhood Autism and Atypical Autism in Sample 1 and 3.**

| **DISCO Algorithm Diagnosis** | | **Clinical Group and Ability Level** | | | | |
|---|---|---|---|---|---|---|
| **DSM-5** | **WG-ASD** | **Autism** | | **Comparison** | | |
| **Sample 1** | | **Low Ability** | **High Ability** | **Low # Ability** | **High* Ability** | **Total** |
| **+** | **+** | 16 | 17 | 4 | 0 | 37 |
| **+** | **-** | 2 | 1 | 1 | 3 | 7 |
| **-** | **+** | 0 | 0 | 0 | 0 | 0 |
| **-** | **-** | 0 | 0 | 12 | 26 | 38 |
| **Total** | | 18 | 18 | 17 | 29 | 82 |
| **Sample 3** | | | | | | |
| **+** | **+** | 28 | 11 | 0 | 0 | 39 |
| **+** | **-** | 2 | 3 | 2 | 0 | 7 |
| **-** | **+** | 3 | 1 | 1 | 0 | 5 |
| **-** | **-** | 2 | 2 | 23 | 37 | 64 |
| **Total** | | 35 | 17 | 26 | 37 | 115 |

**Sample 1: # 17 intellectual disability *14 language impairment & 15 typically developing**
**Sample 1: # 26 intellectual disability **** typically developing**

The results for WG-ASD and DSM-5 for Sample 2 are shown in Table 6-7. This also contains the overlap between WG-ASD and ICD-10 Atypical Autism, Gillberg's criteria for Asperger Syndrome and Kanner's Early Infantile Autism. In total the WG-ASD algorithm identified 6 more individuals than the DSM-5 algorithm in Sample 2 but this trend was not significant using a McNemar's test

($x^2(1) = 1.89$, n.s.). In addition, the WG-ASD algorithm identified 99% of individuals with Kanner's Early Infantile Autism and 99% of those meeting criteria for Gillberg's Asperger criteria. The WG-ASD algorithms identified significantly more of the Gillberg's Asperger individuals than the DSM-5 algorithm (91%), using a McNemar's test ($x^2(1) = 6.04$, $p<.01$). Finally, the results comparing WG-ASD and ICD-10 Childhood Autism are not shown here as they have been previously published (Leekam et al., 2002), however new data were analysed for the overlap between WG-ASD and ICD-10 Atypical Autism. These results showed that nine of the ten ICD-10 Atypical cases were identified by the WG-ASD algorithm, again the WG-ASD identifies significantly more of the ICD-10 Atypical cases than the DSM-5 algorithm (3/10, $x^2(1) = 5.04$, $p<.01$).

**Table 6-7: Table showing the overlap between Wing and Gould's ASD conceptualisation and the DSM-5 ASD DISCO algorithm in Sample 2.**

| **DISCO Algorithm diagnosis** | | **Age and Ability Level** | | | | | | |
|---|---|---|---|---|---|---|---|---|
| **DSM-5 ASD** | **WG-ASD** | **Children (*n*=119)** | | **Adolescents (*n*=34)** | | **Adults (*n*=47)** | | **Total** |
| | | **Low** | **High** | **Low** | **High** | **Low** | **High** | |
| **+** | **+** | 42 | 62 | 12 | 17 | 12 | 28 | 173 |
| **+** | **-** | 0 | 3 | 1 | 0 | 0 | 1 | 5 |
| **-** | **+** | 0 | 6 | 0 | 1 | 0 | 4 | 11 |
| **-** | **-** | 2 | 4 | 1 | 2 | 0 | 2 | 11 |
| **Total** | | 44 | 75 | 14 | 20 | 12 | 35 | 200 |
| **Gillberg's** | **WG-ASD** | | | | | | | |
| **+** | **+** | 6 | 31 | 3 | 16 | 5 | 27 | 88 |
| **+** | **-** | 0 | 1 | 0 | 0 | 0 | 0 | 1 |
| **Total** | | 6 | 32 | 3 | 16 | 5 | 27 | 89 |
| **K&E** | **WG-ASD** | | | | | | | |
| **+** | **+** | 27 | 25 | 3 | 3 | 7 | 5 | 70 |
| **+** | **-** | 0 | 1 | 0 | 0 | 0 | 0 | 1 |
| **Total** | | 27 | 26 | 3 | 3 | 7 | 5 | 71 |
| **ICD-10 Atyp** | **WG-ASD** | | | | | | | |
| **+** | **+** | 1 | 1 | 2 | 1 | 0 | 4 | 9 |
| **+** | **-** | 1 | 0 | 0 | 0 | 0 | 0 | 1 |
| **Total** | | 2 | 1 | 2 | 1 | 0 | 4 | 10 |

### *6.2.1.3 Discussion*

This study had three main aims. The first was to assess the sensitivity and specificity of the WG-ASD algorithm. The WG-ASD algorithm performed well across the three samples. Ninety-two percent of individuals with a clinical diagnosis of autism were identified in Sample 1 and 83% in Sample 3. In addition, the WG-ASD was sensitive across a large sample of all ages and ability levels and there were no significant differences across the age, ability level or gender sub-groups. The individual criteria of the WG-ASD algorithm also had good diagnostic accuracy alone. Across both samples, the sensitivity of all four criteria were in the excellent range but the specificity is variable across these criteria and poor for some items. The AUC was highest (above .80 in both samples) when only the quality of social interaction item was used independently as this item is both sensitive to individuals with ASD but also has good levels of specificity, indicating it did not identify too many individuals from the clinical control group.

Significant correlations were found between all four of the WG-ASD criteria. This supports the original proposal of a triad of impairments of social interaction, social communication and social imagination along with a restricted and repetitive pattern of behaviours (Wing 1988; Wing and Gould, 1979). Importantly, quality of social interaction and reciprocal communication were highly correlated with imagination as well as all these three criteria being related to repetitive behaviours.

The third aim was to examine the overlap between the WG-ASD and DSM-5 ASD algorithms and to assess the overlap of the WG-ASD algorithm and additional DISCO algorithms for, Kanner and Eisenberg's Early Infantile Autism and Gillberg's criteria for Asperger Syndrome and ICD-10 Atypical Autism. The AUC of the WG-ASD was slightly better (.89) than the DSM-5 algorithm (.87) for both Samples 1 and 3 but overall the sensitivity and specificity of the two algorithms was comparable. In the previous chapter the DSM-5 ASD algorithm had missed a minority of the individuals who met criteria for Gillberg's Asperger Syndrome. This was predominately because these individuals failed to score on the early childhood (C) domain. The WG-ASD algorithm does not have this criterion and the results reflect this. The WG-ASD and DSM-5 algorithm identified comparable numbers of individuals meeting criteria for Kanner's early infantile Autism; the DSM-5 algorithm captured 100% whereas the WG-ASD algorithm only missed one individual. However, the WG-ASD algorithm was significantly better at identifying the 89 individuals with Gillberg's Asperger and the 10 individuals with ICD-10 Atypical Autism. This indicates that the WG-ASD algorithm captures "the spectrum" as measured by the DISCO algorithms.

### 6.2.2 Study 2: Comparing the WG-ASD items to DSM-5.

The previous study extended previous research using WG-ASD to show that the WG-ASD algorithm performs extremely well at identifying individuals with ASD as well as excluding individuals with other clinical diagnoses from meeting the criteria. In addition, the results of the two spectrum algorithms were comparable. In order to explore this overlap further it is important to consider the overlap at an item level. The WG-ASD algorithm is made up of four criteria (five items), which have a summary aspect to the behaviours they measure i.e. the quality of an individual's social interaction overall and the reciprocal nature of an individual's overall communication. These items on their own are performing at an equivalent level to the 85 items included in the DSM-5 algorithm. As there is an overlap in some of the items because three WG-ASD algorithm items are also included in DSM-5, one proposal may be that these WG-ASD items are carrying a disproportionate weight in the DSM-5 algorithm's diagnostic power and the removal of these items would therefore be detrimental to the sensitivity of the DSM-5 algorithm. The strength of these items can be tested using the DSM-5 algorithm as its performance is known across the samples. The removal or addition of items in the WG-ASD algorithm can be compared in different versions of the DSM-5 algorithm.

The DSM-5 and WG-ASD algorithms have been shown to identify, in the majority of cases, the same individuals. One explanation for this is that the small number of items in the WG-ASD algorithm may be summarising the larger number of items in the DSM-5 algorithm. Again, the DSM-5 algorithm could be used to test this by measuring its performance when the Domain A DSM-5 items are replaced with the social-communication items from the WG-ASD algorithm, or the Domain B DSM-5 items being replaced with the repetitive behaviour item from the WG-ASD algorithm. If these WG-ASD items are summarising the items for the DSM-5 items then replacing the items for each DSM-5 domains with these summary items would not affect the sensitivity of the DSM-5 algorithm. Furthermore, the WG-ASD algorithm performs extremely well across Asperger and Atypical Autism criteria, which the DSM-5 algorithm was not as able to achieve. Therefore, the addition, of WG-ASD items not previously included in the DSM-5 algorithm (repetitive pretend play and quality of social interaction) could therefore be key in improving sensitivity of the DSM-5 algorithm and their addition to the DSM-5 algorithm would be able to measure this.

The aim of Study 6.2.2 is to assess the strength of the items in the WG-ASD algorithm compared to the DSM-5 algorithm. In order to do this two sets of analyses will be conducted. The first will assess how essential are the inclusion of the items in the WG-ASD algorithm that are already in the DSM-5 algorithm (imagination, reciprocal communication and limited pattern of activities) by removing them from the DSM-5 algorithm and testing for a loss of sensitivity in contrast to the removal of three other randomly selected items.

The second set of analyses will assess the discriminative strength of the WG-ASD items by adapting the DSM-5 algorithm. This will be investigated through a series of edited DSM-5 algorithms. First, the WG-ASD algorithm items not included in the DSM-5 algorithm (repetitive pretend play and quality of social interaction) will be added to the DSM-5 algorithm. They will be placed in domain A (social-communication domain), which is where they best cover the DSM-5 description. Secondly the DSM-5 sub-domains will be replaced with the summary items used in the WG-ASD algorithm. The DSM-5 algorithm will be run with all the items in the B domain (repetitive behaviour) removed and instead individuals will be required to score on "limited pattern of activities." In another modification individuals were required to score on "reciprocal communication", "quality of social interaction" and either "imaginative activities" or "repetitive pretend play" and the usual DSM-5 items for the social-communication (A) domain will be removed.

This procedure will be able to examine how well the WG-ASD items are summarising the DSM-5 domain items. Previous studies have already examined how the DSM-5 algorithm performs across the samples and therefore the effects of editing this algorithm are easy to measure in comparison to the results of the original DSM-5 algorithm. In addition, using the WG-ASD items in a novel way, in some cases independent from the other WG-ASD items, it allows the strength of the items to be identified rather than just the overall algorithm. If the WG-ASD items are summary items then the overlapping items removal from the DSM-5 algorithm should not have a major impact as there are enough of the other items to capture the behaviours being measured by imaginative activities, limited pattern of activities and reciprocal communication, in addition, as the WG-ASD are hypothesised to be summary items from DSM-5 then replacing the whole DSM-5 domain with the relevant WG-ASD should also not result in significant changes to the DSM-5 algorithm's performance.

This study had two main aims:

- To assess the role that three overlapping WG-ASD algorithm items are playing in the DSM-5 algorithm.
- To assess the discriminative strength of the WG-ASD items by replacing DSM-5 domains with WG-ASD items

### *6.2.2.1 Procedure:*

All three samples were used in the following analyses. For the first aim, the importance of the three items from the WG-ASD algorithm already included in DSM-5 (imaginative activities, reciprocal communication and limited pattern of activities) was assessed by running the DSM-5

algorithm, without each of these items both individually and as a group of three items. In order to capture the expected change in results with the removal of any items, three random items were also removed in the same way. These three items were selected from the same sub-domains are the WG-ASD items were removed from (A1, A3 and B1). Each item in these sub-domains was assigned a number and a number (and therefore an item) was selected at random for each sub-domain. This resulted in three random items: makes one sided approaches; has a special friend; and unusual movements of hands or arms.

Secondly (Aim 2), in order to test how well the WG-ASD items are representative of the DSM-5 items, five adjustments were made to the original modified DSM-5 algorithm (see Chapter 5 for full details of DSM-5 algorithm design):

- Addition of "quality of social interaction" to the social-communication (A) domain
- Replaced the social-communication (A) domain with "quality of social interaction", "quality of reciprocal communication", and the two items to measure imagination ("imaginative activities" and "repetitive pretend play"). Individuals were required to score on both quality of social interaction and reciprocal communication and either of the imagination items.
- Replaced the social-communication (A) domain with "quality of social interaction"
- Replaced the repetitive behaviour (B) domain with "limited pattern of activities"

ROC curve analyses were used again to identify the sensitivity and specificity of the DSM-5 ASD algorithm when each modification above was made and re-run. McNemar's tests were used to assess the change in sensitivity and specificity when items were removed or added.

### *6.2.2.2 Results*

#### *6.2.2.2.1 Aim 1: Assess whether the three overlapping WG-ASD algorithm items play an essential role in the DSM-5 algorithm.*

The effects of removing all of the WG-ASD items both separately and in combination from the DSM-5 algorithm are reported in Table 6-8. The removal of all of the WG-ASD items from the DSM-5 algorithm in Sample 1, resulted in 4 less ASD individuals being identified (and one less control individual), this effect was predominately explained by the removal of reciprocal communication, which caused three out of four clinically diagnosed ASD cases and one control case to be missed. This effect, however, was not significant using a McNemar's test ($x(2) = 3.06$, n.s.). The removal of all WG-ASD items in Sample 3, only resulted in the loss of one individual and only three individuals in Sample 2. These results are similar to those of the three random items that were selected to remove as a comparison (Table 6-9). In both cases, removal of any or all three items did not result in more than five ASD cases being missed in Sample 1, and the sensitivity and specificity (Sample 3

only) across the other two samples were relatively consistent whichever item was removed from the DSM-5 algorithm.

**Table 6-8: Table showing the diagnostic accuracy of the DSM-5 algorithm when the overlapping WG-ASD items are removed.**

| | DSM-5 | No "reciprocal communication" | No "imaginative activities" | No "limited pattern of activities" | None of the WG-ASD items |
|---|---|---|---|---|---|
| **Sample 1** | | | | | |
| ASD – low ability | 18/18 | 18 | 17 | 18 | 17 |
| ASD – high ability | 18/18 | 15 | 18 | 18 | 15 |
| ID | 5/17 | 4 | 5 | 5 | 4 |
| LI | 3/14 | 3 | 3 | 3 | 3 |
| TD | 0/15 | 0 | 0 | 0 | 0 |
| AUC (standard error) | | .845 (.053) | .857 (.051) | .871 (.049) | .832 |
| Sensitivity | | .917 | .972 | 1 | .889 |
| Specificity | | 1-.226 | 1-.258 | 1-.258 | 1-.226 |
| **Sample 3** | | | | | |
| ASD | 44/52 | 43 | 44 | 44 | 43 |
| ID | 3/63 | 3 | 3 | 3 | 3 |
| AUC (standard error) | | .856 (.048) | .865 (.047) | .865 (.047) | .856 (.048) |
| Sensitivity | | .827 | .846 | .846 | .827 |
| Specificity | | 1-.115 | 1-.115 | 1-.115 | 1-.115 |
| **Sample 2** | 178/200 | 176 | 177 | 178 | 175 |

**Table 6-9: Table showing the diagnostic accuracy of the DSM-5 algorithm when three randomly selected items are removed, in order to be compared to when the overlapping WG-ASD items are removed from the DSM-5 algorithm.**

| | No "one sided social approaches" | No "friendship with age peers" | No "unusual movements of hands or arms" | None of the comparison items |
|---|---|---|---|---|
| **Sample 1** | | | | |
| ASD – low ability | 16 | 16 | 16 | 16 |
| ASD – high ability | 15 | 18 | 18 | 15 |
| ID | 5 | 5 | 5 | 5 |
| LI | 2 | 2 | 2 | 2 |
| TD | 0 | 0 | 0 | 0 |
| AUC (standard error) | .807 (.056) | .823 (.054) | .823 (.054) | .807 (.056) |
| Sensitivity | .879 | .939 | .939 | .879 |
| Specificity | 1-.265 | 1-.294 | 1-.294 | 1-.265 |
| **Sample 3** | | | | |
| ASD | 42/52 | 43/52 | 44/52 | 41/52 |
| ID | 3/63 | 2 | 3 | 2 |
| AUC (standard error) | .846 (.049) | .875 (.044) | .865 (.047) | .856 (.046) |
| Sensitivity | .808 | .827 | .846 | .788 |
| Specificity | 1-.115 | 1-.077 | 1-.115 | 1-.077 |
| **Sample 2 (178/200)** | 178/200 | 175 | 177 | 174 |

#### *6.2.2.2.2 Aim 2: Assess the discriminative strength of the WG-ASD items by replacing DSM-5 domains with WG-ASD items*

The results for the modifications made to the DSM-5 algorithm can be seen in Table 6-10. The addition of repetitive pretend play on its own had no impact on the DSM-5 algorithm and was therefore not reported here. The sensitivity of the DSM-5 algorithm in Sample 1 was already at ceiling and was therefore not improved by adding quality of social interaction but in Sample 3 the addition of quality of social interaction significantly improved sensitivity ($x^2$=4.05, p<.05) using a McNemar's test. The addition of reciprocal communication and imagination did not significantly affect sensitivity ($x^2$(1)=1.13,n.s.).

The replacement of the A domain in DSM-5 with all the social WG-ASD items (social interaction, reciprocal communication and imagination items) improved sensitivity compared to the DSM-5 algorithm but replacement of Domain A with quality of social interaction by itself improved sensitivity further. The significant increase in sensitivity seen in Sample 3 with the addition of quality of social interaction was maintained when only quality of social interaction was included. In Sample 1, the replacement of Domain A with quality of social interaction also captured all individuals with a

clinical diagnosis of ASD. In Sample 1, the replacement of the B rule by limited pattern of activities resulted in similar results to when the DSM-5 B domain items were used, with loss of only one ASD individual. However, the replacement with limited pattern of activities resulted in a significant drop in sensitivity ($x^2$=4.05, p<.05) in Sample 3.

In Sample 2, the addition of quality of social interaction significantly improved the sensitivity of the DSM-5 algorithm ($x^2$(1)=4.05, p<.05) . Replacing the A rule with quality of social interaction and the B rule with limited pattern of activities improved the sensitivity (but not significantly; $x^2$(1)=1.13, n.s.) whereas the addition of the four items to the A rule, maintained the sensitivity level.

**Table 6-10: Table showing the DSM-5 algorithm re-run with modifications made according to WG-ASD items.**

| | DSM-5 algorithm | Add qualsoc to A rule | A rule replaced by WG-ASD items | A rule replaced with qualsoc | B rule replaced by ltdact |
|---|---|---|---|---|---|
| LFA | 18/18 (100%) | 18 | 17 | 18 | 17 |
| HFA | 18/18 (100%) | 18 | 17 | 18 | 18 |
| ID | 5/17 (29.41%) | 5 | 4 | 5 | 6 |
| LI | 3/14 (21.43%) | 4 | 1 | 5 | 2 |
| TD | 0/15 | 0 | 0 | 0 | 0 |
| AUC | | .855 | .892 | .839 | .857 |
| Standard error (upper-lower) | | .052 (.753-.956) | .045 (.803-.980) | .054 (.733-.944) | .051 (.757-.957) |
| Senstivity | | 1 | .944 | 1 | .972 |
| Specificity | | 1-.290 | 1-.161 | 1-.323 | 1-.258 |
| **Sample 3** | | | | | |
| LFA | 30/35 (85.71%) | 33 | 32 | 33 | 28 |
| HFA | 14/17 (82.35%) | 16 | 16 | 16 | 11 |
| ID | 3/26 (11.54%) | 3 | 1 | 2 | 2 |
| TD | 0/37 | 0 | 0 | 0 | 0 |
| AUC | | .913 | .942 | .933 | .837 |
| Standard error (upper-lower) | | .041 (.833-.994) | .031 (.882-1) | .036 (.863-1) | .048 (0743-.930) |
| Senstivity | | .942 | .923 | .942 | .750 |
| Specificity | | 1-.115 | 1-.038 | 1-.077 | 1-.077 |
| **Sample 2** | | | | | |
| 176 | 176 | 181 | 176 | 178 | 178 |

Qualsoc = quality of social interaction, A rule replaced by quality of social interaction, reciprocal communciation and imagination (imaginative activities or repetitive pretend play). Ltdact = limited pattern of activities.

### *6.2.2.3 Discussion*

The first aim of Study 6.2.2 was to assess the role that the three overlapping WG-ASD items are playing in the DSM-5 algorithm. The overlap between the two algorithms designed to measure the Autism spectrum was further compared at the item level for the items included in the Wing and Gould Autism Spectrum Disorder. Firstly, the power of these items was assessed by removing the WG-ASD items from the DSM-5 algorithm (three items were found in both). However, the removal of any one item or the combined removal of all three items did not produce any significant changes in the sensitivity or specificity of the DSM-5 algorithm and resulted in a similar performance of when three random other DSM-5 items were removed.

This indicates that for reciprocal communication, imaginative activities and limited pattern of activities, these items are inter-changeable with the items included in DSM-5. The DSM-5 algorithm contains enough other items (85 items) that cover the same constructs of behaviour that these WG-ASD items were measuring. This finding goes some way to addressing the serious challenge outlined earlier in the chapter regarding the same author designing both sets of algorithm items. These three WG-ASD items are neither necessary nor essential for an autism spectrum diagnosis described by DSM-5, although they may be interchangeable with other items. For example, impairments in reciprocal communication was likely also captured by “makes one sided social approaches” or “use of body language in social interaction” and for limited pattern of activities, the DSM-5 algorithm contains items which capture the repetitive aspect of this measure e.g. “elaborate repetitive activities with objects” as well as the restricted patterns such as “maintenance of sameness in routines” and this fits with the conclusion in the previous chapter, that it is the range of examples in the DISCO that allow the DISCO DSM-5 ASD algorithm to be sensitive across the whole autism spectrum. The exception to this is for imaginative activities, as this is the only imagination item included in the DSM-5 algorithm. This item has been given less influence in the new conceptualisation according to DSM-5 and it appears that the same results are found whether it is included or not. The answer to whether these items are essential to capture ASD, as conceptualised by DSM-5, is not strong and these findings are limited in furthering knowledge about the necessity of the items in the Wing and Gould ASD algorithm. The second aim was to assess how sufficient these items could be.

The second aim of this study was to assess the discriminative strength of the WG-ASD items by replacing DSM-5 domains with WG-ASD items. In order to assess the strength of the WG-ASD algorithms as summary scores for the range of behaviours measured by DSM-5, the items were substituted for whole sections of DSM-5 items. In this second set of analyses, any WG-ASD items not previously in the DSM-5 algorithm were added in order to examine whether it would improve the

sensitivity or specificity of the results. The addition, of quality of social interaction to the DSM-5 algorithm significantly improved the sensitivity of the DSM-5 algorithm in Samples 2 and 3, sensitivity was already at ceiling in Sample 1. Removal of the DSM-5 social and communication (A) domain (38 items) with the social WG-ASD items (quality of social interaction, reciprocal communication, imaginative activities and repetitive pretend play) was successful in maintaining the same level of sensitivity and specificity as when all the DSM-5 items were used.

More importantly, the replacement of all the DSM-5 items in Domain A (Social-Communication) with the one item of quality of social interaction maintained the improved level of sensitivity that resulted from the addition of quality of social interaction to the A domain. This level of sensitivity was primarily due to quality of social interaction, as when individuals were required to also score on reciprocal communication and either imaginative activities or pretend play, the sensitivity dropped compared to quality of social interaction alone, sensitivity of the original DSM-5 algorithm was maintain (as described above). On its own the one WG-ASD item performed comparably to all the items in Domain A. This implies that these WG-ASD items, especially, the quality of social interaction are excellent summary or proxy measures of the items previously selected to measure DSM-5.

Limited pattern of activities is the only WG-ASD item that falls in Domain B of the DSM-5 algorithm. This item already belongs to both algorithms and the previous study revealed its removal did not have a significant effect, however, when limited pattern activities was used instead of all DSM-5 Domain B items, it performed extremely well in Sample 1, with only one ASD case missed by this replacement and Sample 2, where the sensitivity was improved (by two cases) relative to the unaltered DSM-5 algorithm. This exchange was less successful in Sample 3 however and resulted in a significant drop in sensitivity.

## 6.3 General discussion:

In Study 6.2.1 the sensitivity and specificity of WG-ASD algorithm as a whole was first tested using ROC curve analysis. I explored the WG-ASD items further to examine how well they perform independently in comparison to the full WG-ASD algorithm. The logic was to test the original claim of Wing and Gould (1979) that these items form a triad of behaviours: social interaction, social communication and social imagination (now social imagination) that co-occur together and are associated with a pattern of repetitive and restricted behaviours. The Wing and Gould Autism Spectrum Disorder (WG-ASD) algorithm performs with a high level of accuracy at identifying individuals with a clinical diagnosis of Autistic Disorder or Childhood Autism; ninety-two percent of individuals with a clinical diagnosis of autism were identified in Sample 1 and 83% in Sample 3. The

WG-ASD algorithm identified 99% of the individuals meeting criteria for Gillberg's Asperger Syndrome and Kanner's Early Infantile Autism. In addition, the WG-ASD algorithm captured 9 of the 10 ICD-10 Atypical Autism cases. This was more successful than DSM-5, as shown in the previous chapter, who missed 11% of cases meeting Asperger criteria and 70% of atypical cases. In addition, significant correlations were found between all four criteria of the WG-ASD algorithm.

Comparisons were then made between the two spectrum algorithms WG-ASD and DSM-5, which showed a high level of overlap. In Study 6.2.2 the focus was on the overlap between the two spectrum algorithms at the item level. One hypothesis is that the algorithms perform similarly because a small number of items in the WG-ASD algorithm are summary items that cover the behaviours covered by the many items in the DSM-5 ASD algorithm. To attempt to investigate this, I reasoned that if the WG-ASD items are essential to performance of the DSM-5 algorithm then removal of these WG-ASD items would result in loss of sensitivity and/or specificity. In Study 6.2.2 I tested the DSM-5 algorithm with the overlapping WG-ASD items removed. The removal of the overlapping items did not have a large impact on the diagnostic accuracy of the DSM-5 algorithm both alone or in combination and resulted in comparable results to the removal of three other randomly selected items. I also examined whether each domain of the DSM-5 algorithm would be able to be substituted for WG-ASD items without a loss of sensitivity. Further analyse explored whether the WG-ASD items are summarising the information captured by DSM-5, the replacement of the A domain with each of the four social-communication WG-ASD items was successful in maintaining the same level of sensitivity and specificity when all the DSM-5 Domain A items were used and when quality of social interaction was used by itself, however, the replacement of the Domain B items with limited pattern of activities resulted in a significant drop in sensitivity in Sample 3, but maintained sensitivity in Sample 1 and 2.

### *6.3.1 Limitations*

The specific limitations to this chapter are that the WG-ASD algorithm and concept are fundamental to the DISCO and a major limitation of the results from Sample 2 in this chapter are that the clinicians who collected the data are the authors of the DISCO and could therefore be scoring the WG-ASD items in a specific way or giving these items more weight than others because they know that they are important to their conceptualisation of ASD. This limitation could also be relevant, in a less severe way, to researchers collecting data because all professionals trained on the DISCO are trained in the conceptualisation of Wing and Gould's spectrum approach, however, in data collection for both Samples 1 and 3 researchers were blind to an individual's diagnosis. Future research needs to identify if appropriate items to measure the WG-ASD items can be found in other

diagnostic tools and whether they perform equally well if they are not measured in the DISCO. One potential way to overcome this is for professionals to make clinical judgements on the WG-ASD items after collecting information from the DISCO and after collecting information from another tool or diagnostic interview to see if the ratings on the WG-ASD items are the same.

### *6.3.2 Summary*

The Wing and Gould ASD algorithm performs well in comparison to clinical diagnosis, other DISCO algorithms and across age and ability level. The good overlap with DSM-5 indicated that the change to a spectrum in the diagnostic criteria is similar to spectrum approach already proposed in the literature. Evidence for the WG-ASD items summarising the information collected by the DSM-5 ASD algorithm is mixed, however, one item consistently performs well whichever method it is incorporated in. Addition of quality of social interaction to the DSM-5 ASD algorithm improved sensitivity, and when it was used to replace all the Domain A DSM-5 ASD algorithms it maintained this higher level of sensitivity. In addition, this item performs extremely well diagnostically on its own and is consistently found across age and ability level. This item and its respective coding system are explored in more detail in the following chapter.

# 7 Measuring Autism Spectrum Disorder: Wing and Gould's Quality of Social interaction

This chapter is dedicated to analysing a single element within the Wing and Gould algorithm, Wing and Gould's Quality of Social Interaction. The DISCO item "Quality of social interaction" played an important role in the WG-ASD algorithm in Chapter 6 as well as performing extremely well when added or even used to substitute aspects of the DSM-5 DISCO algorithm. The quality of social interaction binary score was more successful than the other WG-ASD variables and comparable to the DSM-5 and WG-ASD full algorithms at identifying individuals with ASD and distinguishing them from individuals with non-ASD clinical conditions. This item will be further examined in this Chapter.

The summary item at the end of the DISCO schedule asks the clinician to make a judgement based on the information they have collected: "when rating, consider all the available evidence, - particularly from social interaction – with adults, social interaction – with age peers and social play or leisure activities" (DISCO manual, Wing, 2002). However, the relation between the information collected using the DISCO items on social interaction and the quality of social interaction score at the end of the DISCO has never been empirically tested. It is important to consider, whether this previous information aids the clinician in making a judgement or whether the interviewer rates this quality of behaviour on additional features.

This study will explore how individuals are rated by clinicians as "impaired" or "typical" on the summary item of quality of social interaction. Analyses will explore whether this relies on items from the DISCO and if so which sections or items in particular. Further analyses will explore how the binary divide of "impaired" or "typical" social interaction can be used to distinguish individuals on other impairments in the triad and their further behavioural manifestation.

## 7.1 Literature review

### *7.1.1 Quality of Social interaction and the core features of ASD:*

The concept of the quality of social impairment was both formed and used to categorise individuals in the Camberwell epidemiological study conducted by Wing and Gould (1979). Wing and Gould, searched for any child in the London Borough who had one or more of the features described by Kanner or any child whose behaviour appeared to be peculiar, odd or strange for any reason according to teachers or professionals (taking into account the child's level of development). The authors categorised individuals into two groups: those who took pleasure and had interest in social interaction (appropriate for their mental age); and those who had inappropriate social interaction.

Wing described the types of social interaction that constituted "impaired" quality. The codes for the DISCO item "quality of social interaction" (see Chapter 6) reflect the sub-classification of individuals with a social impairment into three groups of "aloof and indifferent" (codes 0,1,2), "passive" (code 3) and "active but odd" (codes, 4 & 5; Wing and Gould, 1979; Wing, 1996, Wing, 1997). Aloof children were characterised by neither responding to social approaches nor seeking any social interactions. Passive children do not initiate social interaction but responds to others. In contrast, active-but-odd children actively seek social interactions with others but in inappropriate or unusual ways (Wing, 1996).

In the Camberwell Study, it was found that individuals with a social impairment (i.e. had impaired quality of social interaction) had more delays or impairments in language and repetitive behaviours than individuals without a social impairment. Specifically, socially impaired individuals were significantly more likely to have no symbolic activities, no speech, more elaborate repetitive routines and have a pattern of activities that was repetitive only. In addition, socially impaired children were more likely to have echolalic speech, use idiosyncratic speech and/or reversal of pronouns and have repetitive symbolic activities but these differences did not reach significance level. Although, the majority of comparisons were made on an individual's current level of functioning, one additional comparison was based on "ever" scores and found that the children with a social impairment were significantly more likely to have a history of childhood autism than the non-socially impaired children.

The main limitation of the work conducted by Wing and Gould (1979) was that their selection criteria meant only individuals with a low ability level were included in their sample. In the past thirty years the recognition of ASD across all levels of ability is widely accepted and research samples now reflect this by collecting information on both low and high functioning individuals with ASD. Considering that some of the behaviours measured by Wing and Gould are extremely dependent on developmental level, for example, the development of speech, it is important to consider whether these differences between the social and non-social groups remain when high functioning individuals are also included. Higher functioning individuals are likely to have more language ability both across individuals with social interaction impairments and other clinical and typical groups and may mean that the symptoms found to be unique previously may be less so when all ability levels are measured.

Overall, little work has explored the binary distinction between typical quality of social interaction and limited or impaired quality of social interaction as measured by the DISCO "Quality of social interaction" item. Following, the conceptualisation made by Wing and Gould (1979) that a triad of impairments tend to co-occur, it is predicted that individuals with and without impaired

social interaction would differ in the "core" features which make up this triad (communication, imagination and restricted and repetitive behaviour). This was already shown in individuals with lower ability levels in the original study. However, what is yet to be considered is whether this social judgement is also useful in determining the presence or absence of associated problems beyond those found in the triad.

The aim of this study is to examine the differences between individuals with and without a social impairment in several ways. First following the approach of earlier studies in the thesis, I look at the Quality of social impairment measure in relation to core symptoms, in this case the focus is on the original Wing & Gould (1979) items, of communication, imaginative and repetitive behaviours (Wing and Gould, 1979). Second I look at the Quality of Social Impairment measure in relation to the 'associated' behaviours of sensory features, maladaptive behaviours, pattern of activities and daily living skills that were the target of Part 2 of the thesis. The current study is novel in including both low functioning individuals and high functioning individuals.

## 7.2 Study 7: Measuring quality of social interaction

From a measurement perspective, no research has been conducted on how interviewers assigned a code for quality of social interaction at the end of the DISCO interview. Therefore, one aim of this study was to assess how the final quality of social interaction judgement is related to the other social items found across the DISCO sections.

This study therefore has three aims:

- Assess the relation between the summary item "quality of social interaction" (qualsoc) to the other social interaction items in the DISCO that are collected in the interview with the informant. Analyses have never directly tested this relation, however, it is predicted that the social items from the DISCO will significantly predict the interviewer's judgement at the end.
- Examine the differences between the social and non-socially impaired groups (according to the quality of social interaction score) on the core features of ASD as measured by the Wing and Gould ASD algorithm and in the original study (Wing and Gould, 1979) and the DSM-5 ASD algorithm items. The proposal by Wing (1996) states that quality of social interaction is the fundamental impairment in ASD and therefore all diagnostic behaviours are predicted to significantly differ between individuals with typical and impaired social interaction.
- Furthermore, the final aim of this study is to assess whether the number of associated features previously investigated in Part 2 of this thesis are found to differ between the

socially impaired group and individuals with typical social interaction styles. This has never been tested before.

### 7.2.1 Methods

#### 7.2.1.1 Participants:

Individuals from Sample 1 will be utilised in the following study. This sample has a relatively even split of typical and impaired social interaction styles as the individuals recruited for the original datasets include individuals with ASD, clinical comparison groups as well as typically developing individuals.

#### 7.2.1.2 Procedure:

The assignment of groups was made independently of an individual's clinical diagnosis. Individuals were rated as having typical or impaired quality of social interaction according to the binary score on the DISCO item. This binary code was computed by collapsing the scores 0-5 to represent impaired social functioning and the codes 6 or 7 to represent typical social functioning, no individuals scored above the code of 7, which represent social behaviour seen in other conditions (see Box above). In Sample 1, 42 individuals were rated as currently having impaired social interaction and 40 were assigned to the typical social interaction group. The current codes for both quality of social interaction and any comparison DISCO items will be used in line with Wing and Gould, 1979 who used current functioning and also allows direct comparison with an individual's current age and level of functioning.

In order to further assess how the quality of social interaction binary code is related to the social items in the DISCO, a total DISCO social score was computed by adding together all the untypical social interaction items that were rated as a "marked" behavioural impairment across Part 3 of the DISCO. In addition, three social sub-scales will be computed for each of the social DISCO sections (Section xii: social interaction with adults (19 items), Section xiii: Social interaction with age peers (16 items) and Section xiv: Social play or leisure activities, (10 items).

Correlations between the quality of social interaction (binary) and total social score as well as the three sub-scores will be conducted. All three social sub-scales will also be entered into a binary logistic regression with impaired or typical social interaction as the outcome variable to further assess their predictive validity of the binary quality of social interaction score. In order to test the assumptions of a binary logistic regression, logarithmic transformations will be computed of the three social sub-scales to ensure linearity of the logit. Multicollinearity will also be tested before the results of the logistic regression are reported.

The measures of core features will comprise the binary (present or absent) score for each item in the Wing and Gould ASD algorithm (analysed using chi-square statistics) and for the counts of binary items across the sub-domains of DSM-5, the full description of DSM-5 is described in Chapter 5; the sub-domain headings are shown below for reference. Non-parametric analyses were conducted with these counts of behaviours as the distributions were found to be significantly different from normal using Shapiro-Wilk tests.

| **Sub-domain** | **Description** |
|---|---|
| A1 | Deficits in socio-emotional reciprocity |
| A2 | Deficits in non-verbal communicative behaviours used for social interaction |
| A3 | Deficits in developing and maintaining relationships appropriate to developmental level |
| B1 | Stereotyped or repetitive speech, motor movements, or use of objects |
| B2 | Excessive adherence to routines, ritualized patterns of verbal or on-verbal behavior, or excessive resistance to change |
| B3 | Highly restricted fixated interests that are abnormal in intensity or focus |
| B4 | Hyper or hypo-sensitivity to sensory input or unusual interest in sensory aspects of the environment |

In addition, the presence of speech, echolalia and language peculiarities will be compared across "typical" and "impaired" social groups using chi-square analyses for the relevant DISCO items. Individuals will be scored as engaging in echolalia if they score to a marked degree on "delayed echolalia;" will be scored as having language difficulties if they score to a marked degree on either "idiosyncratic words or phrases" or "reversal of pronouns"; presence of speech was rated using the "no speech" code on the "delayed echolalia" item and finally language level was rated according to "language estimate." Only individuals rated as "has symbolic language approximately at or above chronological age" were coded as scoring on the language measure. These variables map directly on to those used in the original study (Wing and Gould, 1979).

The measures of associated features used to test aim 4, were the scales of associated behaviours from the DISCO as analysed in Part 2. This chapter revealed four classes of behaviour that formed reliable scales and were therefore used as continuous variables in analyses: sensory behaviours, maladaptive behaviours, daily living skills and pattern of activities. Individual items were recoded as 1 (no problem), 2 (minor problem) and 3 (marked problem) and total counts were used to measure each behavioural domain, with higher scores indicating more behaviours present and to a marked degree. Again, non-parametric analyses (Kruskall-Wallis) tests were conducted due to non-normal distributions. Behaviours not found to form reliable scales were converted into binary variables of "at least one marked behaviour" or "no marked behaviour". Chi-square analyses were used to analyse differences between the binary variables and typical or impaired social interaction.

### 7.2.2 Results

#### 7.2.2.1 Aim 1: Assess the relation between the summary item "quality of social interaction" (qualsoc) to the other social interaction items in the DISCO

To explore the relationship between the quality of social interaction score and individual's scores on the social items from the DISCO correlations were computed between the quality of social interaction binary score and the scores of each social sub-scale of the DISCO (social interaction with adults, social interaction with age peers, social play) as shown in Table 7-1. The current quality of social interaction (impaired or typical) is significantly related to two of the three current social sub-scales of the DISCO: social interaction with adults and social interaction with peers, but not current social play score.

**Table 7-1: Table showing the correlations between the current quality of social interaction binary score ("typical" or "impaired") and individuals scores on the social items from the DISCO.**

| | Qualsco2 | age | IQ | Adults2 | Peers2 | Play2 | Total2 |
|---|---|---|---|---|---|---|---|
| Qualsco2 | 1 | -.099** | .109** | .767** | .680** | .180 | .833** |
| Age | | 1 | .056 | -.113 | .025 | .112 | -.066 |
| IQ | | | 1 | -.285** | .129 | -.088 | -.199 |
| Adults2 | | | | 1 | .488 ** | .149 | .907** |
| Peers2 | | | | | 1 | .312 ** | .743 ** |
| Play2 | | | | | | 1 | .390 ** |
| Total2 | | | | | | | 1 |

A binary logistic regression (Table 7-2) was conducted with current quality of social interaction as the dependent binary variable (impaired or typical quality of social interaction) and the score of social items for each sub-section of the DISCO (social interaction with adults, social interaction with peers, and social play), age and IQ score entered as continuous predictor variables. All independent variables met criteria for inclusion in their regression models: all were linearly related to the log of the outcome variable; no interactions between each variable and log of itself

were significant. In addition the assumption of no multicollinearity was also met: the tolerance values were above 0.7; and the VIF values are less than 10 (1.39); the condition indexes were relatively similar across eigenvalues and no predictors had high proportions on the same small eigenvalue (Field et al., 2009).

**Table 7-2: Table showing the results of binary logistic regression with impaired or typical social interaction style as the dependent variable and total scores on DISCO social sub-domains along with age and IQ as predictor variables.**

| | | | | | 95% CI for Odds Ratio | | |
|---|---|---|---|---|---|---|---|
| Model | Variable | B(SE) | SE | Wald | Lower | Odds Ratio | Upper |
| Step 1 | IQ | -0.01 | 0.01 | 0.52 | 0.98 | 1.00 | 1.01 |
| | Age | -0.01 | 0.01 | 0.91 | 0.98 | 0.99 | 1.01 |
| | Constant | 1.00 | 0.83 | 1.45 | | 2.73 | |
| Step 2 | IQ | 0.03 | 0.03 | 0.86 | 0.97 | 1.03 | 1.09 |
| | Age | -0.07 | 0.04 | 2.96 | 0.87 | 0.94 | 1.01 |
| | Soc_adults | 3.02* | 1.48 | 4.14 | 1.12 | 20.38 | 372.33 |
| | Soc_peers | 3.15* | 1.36 | 5.39 | 1.64 | 23.39 | 334.43 |
| | Soc_play | -1.25 | 1.28 | 0.95 | 0.02 | 0.29 | 3.54 |
| | Constant | -3.72 | 3.01 | 1.53 | | 0.02 | |

The full model explained 69% (Cox and Snell $R^2$)/92% (Nagelkerke $R^2$) of the variance in the binary distinction between typical and impaired social interaction. Importantly, the individual characteristics of age and IQ did not significantly predict quality of social interaction on their own in step 1 ($x^2(2) = 1.453$, p=.484, n.s.), the addition of the DISCO social items, significantly improved the variance explained by the model ($x^2(3) = 94.504$, p<.001) and both social interaction with adults and peers were significant predictors.

In addition, the role that individual items may play in the prediction of quality of social interaction was assessed. As the social interaction with peers and adults were consistently the best predictors of quality of social interaction all the items that made up these total scores were then added to a stepwise binary logistic regression to identify the key DISCO items to predict the presence or absence of impaired quality of social interaction. The stepwise procedure took five steps to create the final model with 5 items. This model accounts for a large proportion of the variance in quality of social interaction according to Cox and Snell R (.746) and Nagelkerke R (1.00). This final

model contained: awareness of others feelings, interaction with age peers, conventions of peer interaction, one sided social approaches and greeting of parents. These items fell across both the social interaction with adults and peers sections.

#### *7.2.2.2 Aim 2: Examine the differences between the social and non-socially impaired groups on the core features of ASD as measured by the Wing and Gould ASD algorithm and the DSM-5 ASD algorithm items.*

Analyses were conducted to test for differences between the individuals who were rated as having typical (40) or impaired (42) quality of social interaction in Sample 1. A Chi-square test revealed no significant effect of IQ (<70/>70; $x^2(1) = 1.397$, p=.237., n.s.) and a Mann-Whitney U test revealed no significant effect of age (U = 743.5, Z =-.895, p=.374, n.s.) between individuals in the impaired or typical social groups in Sample 1, so analyses were conducted on the sample as a whole. The results of the comparisons between the impaired and typical social groups are shown in Table 7-3. Analyses were first conducted on the rest of the items from the WG-ASD algorithm: reciprocal communication; limited pattern of activities; and imagination. Chi-square tests revealed the impaired quality of social interaction group had significantly more impaired behaviours for all WG-ASD sub-domains than the typical quality social interaction group.

In addition, the impaired quality social interaction group were significantly more likely to engage in echolalia than the typical quality of social interaction group as well as more likely not to have symbolic language but no significant differences were found for the use of idiosyncratic phrases or reversal of pronouns and there was no effect of presence of speech.

**Table 7-3: Chi-square analyses and percentages of "impaired" and "typical" individuals scoring on the core features of ASD as measured by the Wing and Gould ASD spectrum in addition to items selected for comparison by Wing and Gould (1979).**

| Item | Impaired 42 | Typical 40 | Chi-Square |
|---|---|---|---|
| Limited pattern of activities | 83.30 | 30.00 | 23.82** (p<.001) |
| Reciprocal communication | 83.30 | 22.50 | 30.49** (p<.001) |
| Imagination | 85.70 | 35.00 | 22.14** (p<.001) |
| Idiosyncratic or pronoun reversal | 21.43 | 12.50 | 1.15 p=.283 |
| Del echolalia | 38.10 | 7.50 | 10.77**, p<.001 |
| No speech | 14.29 | 10.00 | 0.35 p=.553 |
| Language level | 19.00 | 57.50 | 12.88**, p<.001 |

** p<.001

A second set of analyses compared the typical and impaired quality of social interaction groups across the sub-domains of the DSM-5 (A1, deficits in social-emotional reciprocity; A2, deficits in nonverbal communication; A3, Deficits in developing and maintaining relationships; B1, stereotyped or repetitive behaviours; B2, insistence on sameness; B3, highly restricted interests; B4, hyper- or hypo-reactivity to sensory input). As can be seen in Figure 1, the socially impaired group had significantly more behaviours than the typical social group across all of the DSM-5 sub-domains: A1 ($U = 24.5$, $r = .85$, $p<.001$), A2 ($U = 136$, $r = .73$, $p<.001$), A3 ($U = 64$, $r = .80$, $p<.001$), B1 ($U = 165.5$, $r = .71$, $p<.001$), B2 ($U = 241.5$, $r = .63$, $p<.001$), B3 ($U = 385.5$, $r = .48$, $p<.001$), B4 ($U = 286$, $r = .57$, $p<.001$). In addition, the total A ($U = 20.5$, $r = 84$, $p<.001$) and B ($U = 135$, $r = .72$, $p<.001$) domain scores were significantly different between the two social groupings.

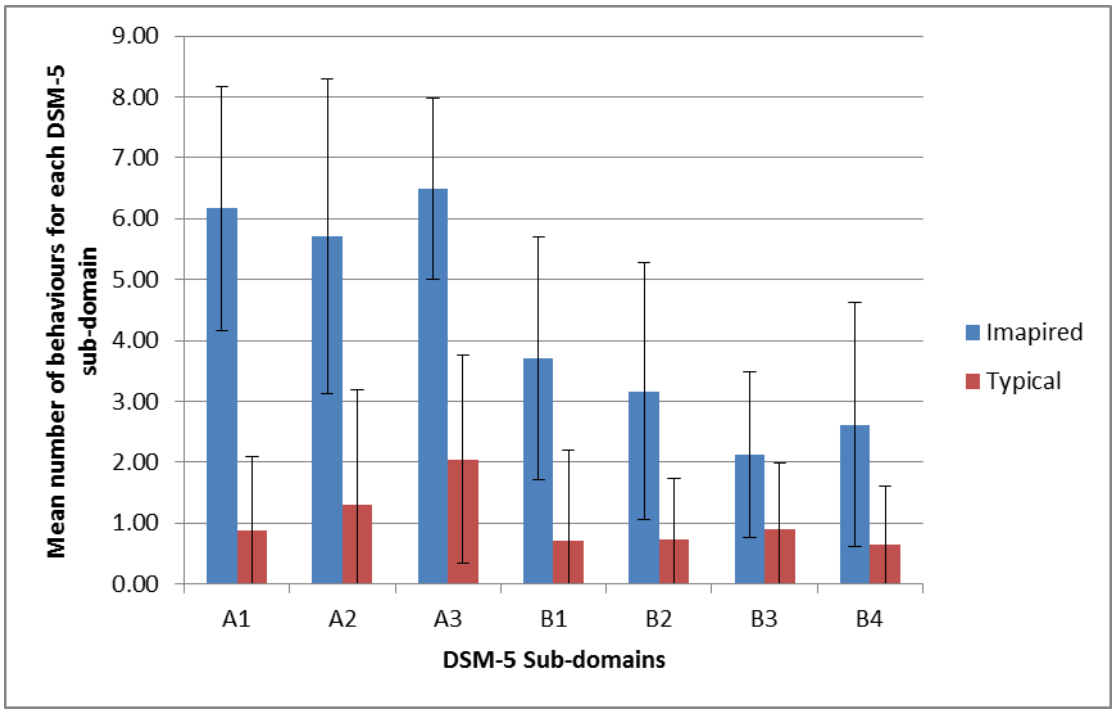



**Figure 7-1: Figure showing the significant differences between the impaired and typical social interaction groups in Sample 1 on the DSM-5 ASD sub-domain criteria. Error bars +/- 1sd.**

### *7.2.2.3 Aim 4: Assess whether differences in the associated features are found between the socially impaired group and individuals with typical social interaction styles*

Finally, the individuals with impaired quality of social interaction were compared to those individuals with typical social interaction across the associated features as measured by the DISCO. As can be seen in Figure 2, the socially impaired group scored significantly higher on maladaptive

behaviours (U = 149.5, r = .71, p<.001), sensory behaviour (U = 198.5, r = .66, p<.001), daily living skills (U = 511.5, r = .34, p<.01) and pattern of activities (U = 271, r = .60, p<.001).

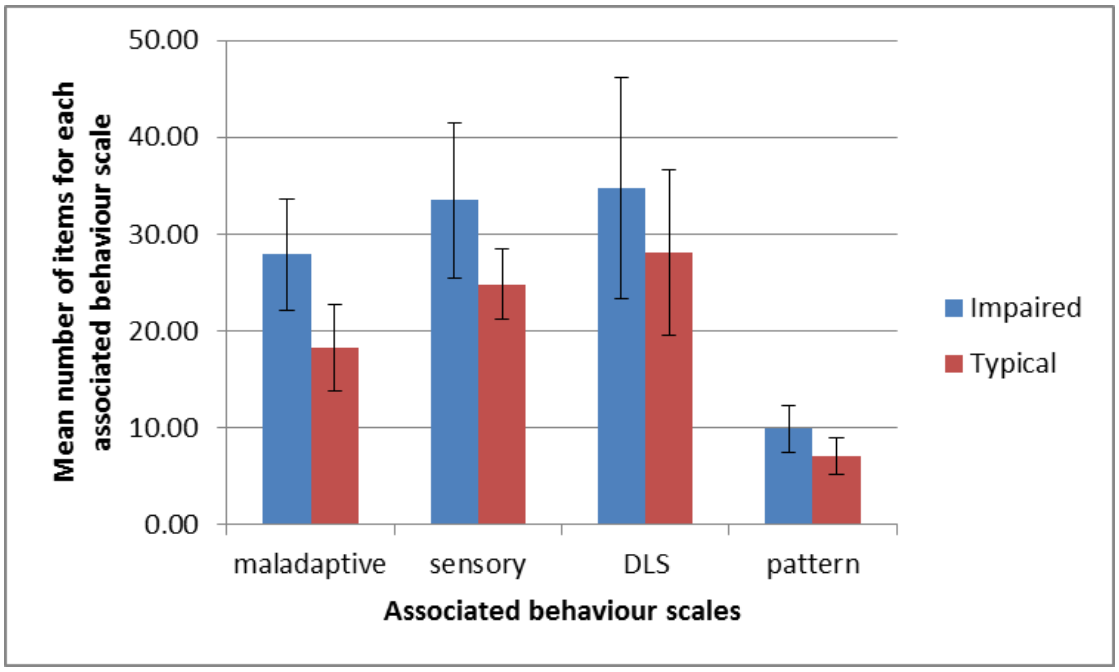



**Figure 7-2: Figure showing the scores on the associated scales (Part 2) for the "impaired" and "typical" social groups in Sample 1. Significant differences were found across groups for all associated scales. Error bars +/- 1sd.**

Associated behaviours that did not form a reliable scale were converted to binary (present or absent) variables and investigated using chi-square tests; the results of motor, emotion and sleep problems across typical and impaired social interaction groups can be seen in Table 7-4, sleep problems was the only associated variable to not significantly differ between the typical and impaired social groups.

**Table 7-4: Chi-square analyses for binary associated features of ASD in the typical and impaired quality of social interaction groups.**

| Behaviour | % in socially impaired group | % in tpyical social group | Chi-square statistic |
|---|---|---|---|
| Emotion | 78.6 | 30 | 19.520, p<.001 |
| Motor | 97.6 | 60 | 17.643, p<.001 |
| Sleep | 40.5 | 22.5 | 3.057, p=.080, n.s. |

## 7.3 General discussion

The binary Quality of social impairment item was significantly correlated with all three social sub-scales of the DISCO and furthermore, in the regression model the amount of variance accounted for by the three total social variables is extremely high. It therefore appears that whether an individual is rated to have impaired or typical quality of social interaction is predominately due to the information gathered from the other social items in the DISCO. This reflects the purpose of this construct as a DISCO item and the instructions that clinicians or interviewers should to use the social information gained from the rest of the DISCO to make this decision. However, a selection of social behaviours were more relevant to the prediction of quality of social interaction score than others: awareness of others feelings, interaction with age peers , conventions of peer interaction, one sided social approaches and greeting of parents were identified as the most predictive. These behaviours have in common key aspects of reciprocal social interactions; the items which measure the reciprocal nature best are most predictive. The items not found to significantly predict the assignment into typical or impaired social interaction were the items which instead measure non-verbal communicative behaviours used in interaction such as eye contact and behaviours related to social play.

Additionally, the aims of Study 7 were to assess whether the quality of the social interaction tells us anything beyond the social functioning of the individual. The second set of analyses tested the quality of social interaction across the core features of ASD including those measured in the original study of impaired social interaction (Wing and Gould, 1979), however, the current sample furthered this analysis by including high functioning as well as low functioning individuals. Wing and Gould (1979) proposed that communication, imaginative and repetitive behaviours would differ across these social groups as they found the co-occurrence of these behaviours across individuals in the epidemiological study. These findings support this proposal that the triad of impairments tend to co-occur together and individuals with impaired quality of social interaction are more likely to display problems in reciprocal communication, a limited or repetitive pattern of activities as well as impairments in imagination. This was further supported by measurement of core features using the DSM-5 algorithm as scores on the behavioural sub-domains were significantly higher in the socially impaired than typical social interaction group. There were, however, minor differences between the results presented here and the original proposals of Wing and Gould (1979). It was previously found that individuals with social impairment were significantly more likely to have no speech, however, in a sample of individuals across a broader range of IQ as sampled in the current study, this difference was no longer significant. However, the level achieved in individual's use of language was

significantly different, with more individuals in the typical social impairment group using a level of language that was appropriate or above their chronological age.

These overlaps in symptom presentation may have an interesting contribution to the research of the new diagnosis of Social (Pragmatic) Communication Disorder in the DSM-5. In order to meet criteria for this disorder, individuals must have "persistent difficulties in the social use of verbal and nonverbal communication" (APA, 2013). Future research should investigate whether these individuals fall under the description of impaired quality of social interaction used in the DISCO. In this case, then the presented evidence makes it likely that these individuals may also show impairments in other areas of the triad if thorough investigation of their past and current behaviour is collected using a broad range of items, such as those covered by the DISCO.

Finally, the binary code of impaired social interaction was found to be associated with significantly more associated behaviours than individuals in the typical social interaction category. This effect was found across all measures of associated features using the DISCO, with sleep problems as the only exception.

The quality of social interaction item, clearly has both diagnostic as well as descriptive power, however, this study only conducted analyses on the binary recode used to define presence or absence of social impairment and this alone did not appear to improve knowledge about the individual beyond the information already gained from the rest of the social items in the DISCO. However, the codes that count towards the code for impaired social interaction are made up of a range of different social impairment descriptions, which characterise individuals with ASD. These codes were explored in Appendix 9 to reveal patterns across individuals with variable social impairment on the autism spectrum.

### *7.3.1 Limitations*

The findings in Chapter 7 indicate that the reason the quality of social interaction item on its own has equivalent diagnostic accuracy to the DISCO DSM-5 algorithm is likely because the quality of social interaction item is rated based on the information gathered from the social interaction items in the DISCO. What is required is an independent measure of quality of social interaction in order to further test the prediction that this item or classification is important diagnostically. Some work has already addressed this by designing the Wing Subgroups Questionnaire (Castelloe and Dawson, 1993), which is designed to look at the social sub-types within social impairment but could also categorise individuals into impaired or typical social interaction groups. It should also be considered whether assignment to social sub-type could be made by clinical opinion (separate to the DISCO) or through observation of the individual. Research would be needed on how well the summary item

from the DISCO can be mapped onto the questionnaires or through observations to ensure that they are equivalent concepts and then further work could assess the relation between the DISCO social items and an independent quality of social interaction measure as well as using this measure to test diagnostic accuracy, and differences across associated and comorbid behaviours in different social sub-types.

To reiterate, Chapter 6 (section 6.3.1) the major limitation to these findings is that the WG-ASD algorithm and concept are fundamental to the DISCO and a major limitation of the results from Sample 2 in this chapter are that the clinicians who collected the data are the authors of the DISCO and could therefore be scoring the WG-ASD items in a specific way or giving these items more weight than others because they know that they are important to their conceptualisation of ASD. Future research needs to identify if appropriate items to measure the WG-ASD items can be found in other diagnostic tools and whether they perform equally well if they are not measured in the DISCO. One potential way to overcome this is for professionals to make clinical judgements on the WG-ASD items after collecting information from the DISCO and after collecting information from another tool or diagnostic interview to see if the ratings on the WG-ASD items are the same.

### *7.3.2 Future directions*

The findings from Chapter 6 and the study showed how successful the binary distinction of quality of social interaction (impaired or typical quality) was at diagnosing individuals with ASD as well as excluding individuals without a diagnosis. In addition, differences were seen across both the core and associated features of ASD. However, this variable comprises a range of scores covering different types of social behaviour. Wing (1996) proposed three socially impaired groups of aloof and indifferent, passive and active but odd. Within these clinical descriptions Wing suggests how additional features such as communication and sensory features may also differ across these sub-types. This area has received some attention in the literature but again samples are limited in age and IQ (e.g. Castelloe and Dawson, 1993; Scheeren et al., 2012; Borden and Ollendick, 1994).

Exploratory analyses presented in Appendix 9 have begun to address this. It was found that although all social sub-types were related to the social diagnostic features of ASD (Domain A of DSM-5) that belonging to the active but odd social sub-group was predictive of significantly more behaviours in sub-domains B2 (Excessive adherence to routines, ritualized patterns of verbal or on-verbal behavior, or excessive resistance to change) and B3 (Highly restricted fixated interests that are abnormal in intensity or focus) but being in the aloof group was most predictive of more B4 behaviours (Hyper or hypo-sensitivity to sensory input or unusual interest in sensory aspects of the environment). Furthermore, the exploratory analyses investigated the presence of associated and

comorbid features across the social sub-types. These analyses revealed that the aloof and active but odd group tend to display more associated behaviours than the passive group in general. The aloof group was a significant predictor of sensory behaviours, a limited pattern of activities, presence of tic disorder and catatonic behaviours. The active but odd group was predictive of more problems with daily living skills, sensory behaviours and more Pathological Demand Avoidance (PDA) behaviours. These results were identified even when the effects of age, IQ and language were held constant within regression models.

### *7.3.3 Summary*

With the conceptualisation of Autism changing in the international classification systems it is important not to forget the findings and proposals made originally about the spectrum approach and classification of Autism. The previous two chapters have shown that the algorithm based on Wing and Gould's concept of ASD that was proposed around 30 years ago performs equivalently and in some respects better than the ASD criteria proposed in 2013 by the diagnostic classification systems (DSM-5). In particular, the quality of social interaction item on its own has equivalent diagnostic accuracy to the DISCO DSM-5 algorithm. Given the limitations set up throughout, Chapter 7, highlights the information that can be gathered from the summary item, quality of social interaction, the analyses show that this is likely based on the information that the interviewer collects throughout the interview. This judgement of quality made by the interviewer is extremely powerful and the same judgement of other areas of impairments could also be important to consider or to include in such diagnostic assessments.

# PART 4: MEASURING AUTISM SPECTRUM DISORDER, A DISCUSSION

*"The most fundamental of the impairments making up the triad is that affecting social interaction. This is the problem that makes autism so special."*

Wing (2008, p. 8)

# 8 General discussion

In Part 1 I introduced the difficulty of how best to measure the autism spectrum. Having now presented two distinct sets of studies focusing on the measurement of autism spectrum in terms of its associated symptoms (Part 2) and diagnostic criteria (Part 3), the aim of this final chapter is to summarise the key results for each part of the thesis, evaluate their interpretation and limitations, and draw conclusions. In addition I consider the design of future studies and the potential direction for future research. At the end of the chapter I make the case that examining behaviours seen in individuals with ASD in isolation will only ever be able to inform us about those behavioural symptoms. However, examining these behaviours in combination will hopefully improve our knowledge about the behavioural manifestation of ASD, help to revise our conceptualisation of autism and in turn identify new avenues for understanding biological mechanisms with aetiological significance. The limitations of the samples and research design for all analyses conducted in the thesis were presented in detail in Chapter 2, and the potential impact that these limitations have on the core findings from the thesis are discussed throughout this chapter.

## 8.1 Overview of findings:

### *8.1.1 Part 2: Measuring the autism spectrum: Associated features*

Part 2 of the thesis focused on the "associated" behaviours present in ASD, that is, behaviours that are not included in the diagnostic algorithms. The reason for including associated behaviours was two-fold. The main aim of Part 2 was to explore the measurement of the boundaries of the 'spectrum' of autism and to explore whether one or more associated feature of ASD had a particularly influential role in the behavioural manifestation of ASD. Firstly, from a purely methodological perspective the associated scales from the DISCO had not been tested for reliability for their use in research or as independent scales of behaviours. In order to address the main aim of Part 2, the first step was to assess how well the DISCO measures associated features of ASD. Preliminary analyses were used to identify which scales within the DISCO were reliable. The main aim of examining the role of the associated features was tested across Chapters 3 and 4 using the

scales identified as being reliable and ASD specific in the DISCO; the maladaptive behaviour and pattern of activities scales were explored together in Chapter 3 and the sensory behaviour scale in Chapter 4.

#### *8.1.1.1 Part 2: Findings*

The aim of Part 2 was to identify if any one type of associated behaviour had a significant role in the behavioural manifestation of ASD. Sensory behaviours were related to both the core and other associated behaviours in ASD. They also appear to influence the relation between other associated features and the core features as demonstrated by the mediating relationship of sensory behaviours on the relation between maladaptive behaviours and both social interaction and repetitive behaviour scores. Furthermore, this thesis addressed the clear lack of research using self-report of sensory behaviours in ASD and found significantly higher rates of sensory behaviours in high functioning adults with ASD compared to age and IQ matched typically developing individuals using the sensory items from the DISCO.

In Part 2, I focussed on the three reliable and ASD specific scales; sensory behaviours, maladaptive behaviours and pattern of activities. The maladaptive behaviour and pattern of activities subsets of items in the DISCO were explored at the same time. The reason for this is that although they form distinct subsets or scales in the DISCO, when these behaviours are measured in other well established child or adult behaviour questionnaires such as the Strengths and Difficulties Questionnaire or the Child Behaviour Checklist, they are often combined together into one measure. Of the remaining associated scales in the DISCO, the daily living skills scale was also reliable but was not of interest in the current thesis as it was not specific to ASD rather it was related to a general developmental delay. The motor, emotion and sleep scales were found to not be reliable; however, binary variables of at least one marked behaviour versus no marked behaviour were used as covariates or predictor variables in the analyses across Part 2.

The analysis reported in the introduction to Part 2 was developed in Chapter 3 by carrying out complex multivariate designs to examine core and associated DISCO items that predicted maladaptive behaviours while controlling for the effects of other variables. One reason for this analysis was that previous studies reported a lack of consistency in findings about gender, IQ and age and these were not being controlled for when maladaptive behaviours were correlated with additional variables. It was argued in Part 2, Chapter 3 that the failure to find relations between IQ or gender in predicting maladaptive behaviours may apply when these behaviours are measured separately, as was the case in these previous studies, but that the interaction between these variables may play an extremely important role. For example, in these analyses (p. 80/81) a

significant IQ by gender interaction was found for predicting maladaptive behaviours; maladaptive behaviours were more prevalent in low functioning males but high functioning females. This effect has not been found in previous research but may contribute to the inconsistent results on the role of gender or ability level at independently predicting maladaptive behaviours. Limitations regarding the sampling bias of males in the sample, restrict the generalizability of this finding but indicates a key reason to control for all variables in regression analyses.

In a model that included both the core features of ASD and all of the associated features, sensory behaviours were found to significantly predict the score on the DISCO scale of maladaptive behaviours. Only limited research has identified a relationship between sensory and maladaptive behaviours (e.g. Tseng et al., 2011). Thus in the model tested in Chapter 3 with all predictors, the core features, in particular, repetitive behaviours had been hypothesised to predict maladaptive behaviours over the associated features (e.g. Bourreau et al., 2009; Oliver et al., 2012). From the analysis presented in the thesis, it was suggested that individuals with a greater number of sensory behaviours are likely to find their everyday surroundings a harsher and more challenging environment than individuals with less sensory problems. Therefore the individuals under additional pressure and stress from the sensory input are more likely to express this in the form of challenging or maladaptive behaviours.

Furthermore, follow up mediation analyses revealed that sensory behaviours were also mediating the relationship between the core features of ASD (social interaction and repetitive behaviours) and maladaptive behaviours. This was shown using Baron and Kenny's (1986) original mediation criteria as well as direct and indirect effects method of modern mediation analysts (e.g. Hayes, 2013). Therefore, this finding represents a robust effect. Again the prediction was that core features were directly associated with maladaptive behaviours (e.g. Dominick et al., 2007; Matson, Wilkins and Macken, 2009; Jang et al., 2009). These findings led to the proposal that the presence of sensory behaviours puts an individual more at risk of maladaptive behaviours than just the core features of ASD themselves. However, the major limitation with both findings is that they do not provide an indication of the causal or directional nature of these relationships and although it was suggested that atypical sensory responsiveness impacts on the presence of maladaptive behaviours in ASD this cannot be assumed.

A major limitation of the analyses in this thesis was that they were exploratory and limited in the type of data analysis that could be employed. In the current literature there is no theory about the link between maladaptive and sensory behaviours in ASD. In order to follow up the findings from Part 2 the best method would be to collect longitudinal data and conduct analyses to distinguish between the following hypotheses. The first is that the presence of sensory behaviours

strengthens the relation between the core features of autism and maladaptive behaviours. This would use analyses based on moderation techniques. The second is whether there is an indirect link between core and maladaptive behaviours via sensory behaviours. Individuals with ASD may be more likely to present with sensory behaviours and that these sensory behaviours are independently associated with higher maladaptive behaviours, this could be tested using path analysis or indirect-effects analysis (Preacher & Hayes, 2008). These models could be tested in both individuals with ASD and those with disruptive, hyperactive or aggressive behaviours or diagnosed with Oppositional Defiance Disorder (ODD) in order to further understand the relation between ASD, sensory and maladaptive behaviours. Furthermore, the addition of wider demographic information and parental coping would be beneficial to measure as these may further influence such models. Parenting stress and household income have previously been shown to have significant predictive effects on child maladaptive behaviour (Bauminger et al., 2010; Kanne & Mazurek, 2012) and in turn findings suggest that hyperactive, disruptive, aggressive, and other co-occurring maladaptive child behaviour increase parental stress beyond the core ASD features (e.g. Hastings et al., 2005; Peters-Scheffer et al., 2012). Future research should include wider variables in such predictive models.

The sensory behaviour scale in the DISCO was also a significant predictor of pattern of activities score. Again, this may have implications for other clinical conditions. The behaviours in the pattern of activities scale closely resemble the diagnostic criteria for ADHD. A number of questions can therefore be considered in future research regarding the role of sensory behaviours in ADHD and comorbid ADHD/ASD, in terms of their prevalence in children with ADHD, their relation to diagnostic symptoms and to social functioning in particular. Finally, although both the maladaptive behaviour and pattern of activities scales form reliable independent scales the next step will be to reliably distinguish these scales in statistical analyses such as factor or cluster analysis. These analyses should also explore whether sub-groups exist within the maladaptive behaviour items both with the DISCO data and other scales of behaviour.

Part 2, also detailed the design of the Sensory Preferences Questionnaire (SPQ), Chapter 4, which consisted of the sensory behaviours from the DISCO that were converted into self-report questions. This was done to overcome a significant lack of research on adults in the literature as well as the use of self-report but also to explore the external validity of the DISCO sensory scale in comparison to existing tools in the literature. The samples recruited in this study had some key limitations, which limit the generalizability of the findings. Firstly, Sample A was restricted to high functioning individuals with ASD (and TD) as self-report is difficult for lower functioning individuals. Methodological considerations need to be made about how best to record individual's reaction to

sensory input beyond questionnaires, which require a certain level of cognitive ability. One avenue is to investigate an individual's skin conductance (galvanic skin response), this is a proxy measure of an individual's sympathetic nervous system and can therefore record physiological arousal, potentially in reaction to sensory input. Although, in an experimental design there are ethical considerations to administering potentially distressing sensory stimuli to the individuals, such instruments could be used to record reaction to everyday sensory input. Secondly the individuals recruited in Sample A may have pre-selected themselves to score high on sensory measures as the recruitment was for a sensory study, therefore, individuals with sensory problems may have been more likely to have volunteered for the study. Further work is needed to replicate these self-report findings in a more representative ASD sample.

The SPQ is the first sensory self-report questionnaire based on the sensory behaviours seen in individuals with ASD through clinical practice and tested on a sample of ASD and typically developing individuals. The SPQ had excellent reliability as found by high levels of internal consistency. Furthermore it showed strong correlations with both the Adult/Adolescent Sensory Profile (AASP, Brown & Dunn, 2002) in a sample of high functioning adults with ASD and typical development (Sample A) and with the Glasgow Sensory Profile (Robertson & Simmons, 2013) in a second sample of typically developing adults (Sample B). This indicates that the SPQ has some degree of convergent validity with the existing tools in the literature and therefore the findings using the DISCO items are relevant and informative to current debates in the sensory literature. These validity findings are relatively independent of the limitations of the sample above but the link between the self-report responses and a parent interview response on the DISCO has not yet been directly compared (this is discussed further in the implications section below).

Sensory behaviours have been shown to consistently occur at extremely high prevalence in individuals with ASD (e.g. Tomchek & Dunn, 2007; Leekam et al., 2007; Kern et al., 2007; Ben-Sasson et al., 2009). The findings in Part 2, Chapter 4 extend these findings to adults as the SPQ revealed that 94% of adults with ASD reported having at least one marked sensory behaviour. In addition, the findings from the SPQ replicate the previous findings using parent report DISCO data, Leekam et al. (2007) found visual symptoms were the only modality to decrease with age and the visual modality was the only modality in the SPQ to not be significantly higher in the ASD than TD group. The high prevalence of sensory behaviours in adults with ASD replicates the one other study that has used self-report in adults (Crane et al., 2009) and therefore adds to the claim that these behaviours persist but that self-report may be the best way of identifying sensory behaviours as parent report has suggested a decrease in sensory symptoms with age (Kern et al., 2006).

### *8.1.1.2 Part 2: Implications and future directions*

This thesis has been extremely beneficial in exploring the non-algorithm items that has implications for their use within the DISCO. Four of the associated scales of behaviour formed reliable scales: maladaptive behaviours, pattern of activities, sensory behaviours and daily living skills. This indicates that the items in these scales provide a good and consistent measure of the scale that is being measured.

The work in Part 2, Chapter 4 converted the sensory items from the DISCO into a self-report format. Although parent-report questionnaires have been used for repetitive behaviours (Leekam et al., 2007), none of the DISCO items have been converted to self-report format before. Overall, this questionnaire converted well and none of the participants reported any problems completing the questionnaire. One reason for this primary data collection was to ensure the work carried out with DISCO sensory items would be relevant to the wider literature, however, in order to use the SPQ in research some more analysis is required. It will be important to compare the overlap between individual's self-report answers and parents responses to the DISCO items and to compare at the same time, interview format and questionnaire format. At the moment little is known about how these two ways of collecting data for the DISCO compared, however, research suggests that there are discrepancies between parent and self-report measures in other behaviour domains such as emotional and behavioural problems (Achenbach, McConaughy & Howell, 1987). This is important to check to ensure the SPQ is measuring the same behaviours as the DISCO. An additional confound is the fact that sensory behaviours are extremely individual and it is suggested that parents may not have a very clear understanding of how an individual is responding to sensory input especially if they have developed coping strategies. For example, parent report suggests sensory symptoms decrease with age (e.g. Kern et al., 2006) but this is not shown in adults using self-report such as the AASP (Crane et al., 2009) and the findings with the SPQ suggest that they are still extremely prevalent. One way to examine this would be in a large scale comparison. It would also be helpful to interview the individual and get the parent to fill in the questionnaire.

Other reliable scales included the maladaptive behaviours and pattern of activities scales, these behavioural items in the DISCO hold great benefit for research about the antecedents and relations of these behaviours with the core and associated features of ASD. However, unlike the sensory items, these behaviours may not be appropriate to convert into self-report measures for use in research as there is likely to be a strong response bias in acknowledging that individuals are performing the behaviours. There is also the confound of individuals social understanding; if an individual does not understand that it is not acceptable to interrupt peoples' conversations then they are unlikely to rate that they engage in this behaviour to a marked degree.

In a clinical sense, the conversion of some aspects into questionnaires may reduce the time taken to conduct the diagnostic assessment, which is beneficial as professionals are usually limited for time, however, the information gathered from an interview and questionnaire will need to be examined to ensure it is the same. The training on the DISCO states that it is the interviewer's responsibility to record accurate codes and this may mean that they have to probe for the answer. This could still be achieved if clinicians were able to review the questionnaire answers before the interview and it is suggested that this would still be better than not collecting this information at all if using a shorter diagnostic interview.

This thesis also identified scales of behaviour that did not have good levels of internal consistency and therefore their use in research in their current format may be limited: emotion, motor and sleep scales. In clinical practice, this may not be a problem. The DISCO still provides a range of items that cover these behaviours which indicate to the clinician that these behaviours should be considered in a care plan or that an individual may need to be referred or followed up on some of these behaviours. However, for research as described above, more psychometric testing of the scales is required before the items can be used and compared to other instruments in the research literature.

The main overall finding from Part 2 is identifying the key role that sensory behaviours have in ASD. They were found to relate to both core and associated features and are prevalent across all ages and levels of ability. These findings suggest that the next step in research should now be to include sensory features as key behaviours in the manifestation of autism. Longitudinal studies with a strong focus from birth onwards will help to identify the developmental trajectory of all key behaviours (social interaction, communication, repetitive behaviours, imagination) and could be significant in identifying the causal nature of these behaviours on each other. Although, limitations are clear from the relatively small sample sizes in the current thesis, I found that the sensory behaviour appear to not only be related to repetitive behaviours but also strongly with the social symptoms present in ASD, opening new lines of consideration in future research.

This research raises the question about how much of a central role sensory behaviours may play in ASD and therefore the need to ensure clinicians and researchers alike are recording individuals' sensory behaviours. The addition of sensory behaviours to the diagnostic criteria for ASD is beneficial in that these behaviours would then have to be recorded at diagnosis and therefore care and management plans would involve sensory functioning from the beginning. However, caution must be taken in adding to the number of criteria that are required for an individual to receive a diagnosis, in case this excludes individuals from receiving interventions or care programmes that come with a diagnosis. The results in Chapter 4 highlighted the sensory

behaviours as impacting on both core and maladaptive behaviours. However, the analyses in this thesis are limited by all being conducted on the same samples using the same measure of behaviour. Replication of the relation between sensory behaviours and the core features of ASD independently are recommended in furthering knowledge about the role of sensory behaviours and how their addition to the diagnostic criteria is likely to impact on sensitivity and specificity.

### *8.1.2 Part 3: Measuring Autism Spectrum Disorder: Diagnostic Criteria*

Part 3 focused on the diagnostic criteria for ASD. A set of empirical studies explored the measurement characteristics of two different operational definitions of autism spectrum disorder- those of DSM-5 (APA, 2012/2013) and Wing and Gould (Wing, 1996; Leekam et al., 2002). The first aim of Part 3 was methodological and the goal was to assess how well the DISCO could measure DSM-5 ASD. In addition, in Part 3, Chapter 5, I investigated the adequacy of the DSM-5 criteria for the diagnosis of Autism Spectrum Disorder, in the light of the earlier debate about its lack of sensitivity and considered how comparisons between DSM-5 and Wing and Gould descriptions and their measurement may help to move forward in our understanding of the autism spectrum.

#### *8.1.2.1 Part 3: Findings*

In terms of measuring the DSM-5, the work described in this thesis has contributed to the literature in two main ways. Firstly, the way in which the DSM-5 criteria are measured and applied to existing tools is extremely important. Comparison of the three algorithm approaches offered a comparison of different methods of applying DSM-5 criteria and the results clearly demonstrate the importance of algorithm design. The thresholds set for each sub-domain had a significant impact on the performance of the algorithm as a whole. This level of sub-domain analysis has had little previous attention in the DSM-5 literature.

Secondly, when the DSM-5 behaviours were mapped accurately onto a tool that is capable of measuring the spectrum, the DSM-5 criteria are found to have both good sensitivity and specificity. This contrasts with much of the research on DSM-5 that has shown either good specificity or good sensitivity but never both using one tool. The DISCO appears to have an important role to play in capturing DSM-5 ASD as the many examples of behaviours selected for use in Part 3, Chapter 5 allow the DISCO DSM-5 algorithm to capture the high functioning individuals, originally thought to be at risk of being missed by DSM-5 through previous work with other methods such as DSM-IV checklists (e.g. McPartland et al., 2011). In the recent DSM-5 literature, the move to mapping the criteria onto existing standardised tools rather than DSM-IV checklists has not substantiated the concern over a lack of sensitivity for the new DSM-5 ASD criteria (e.g. Heurta et al., 2012), however,

the only study to balance both sensitivity and specificity of the criteria before the present DISCO study used two standardised instruments (Frazier et al., 2011). Further work with diagnostic instruments needs to test the DSM-5 algorithms in wider samples, however, the real test of the DSM-5 ASD criteria will be its application in a clinical setting. The results in the current thesis emphasise that collecting a broad range of information that is relevant to high and low functioning individuals should minimise the risk of excluding individuals who would have previously received a diagnosis according to DSM-IV/ICD-10.

The exploration of items used in the DSM-5 ASD algorithm, in Part 3, Chapter 5, found that some behaviours were significantly more likely to be present in high functioning or older individuals with ASD. These items were more common in the repetitive behaviours domain, suggesting that items relating to restricted and repetitive language (e.g. pedantic or long winded speech), repetitive activities related to a special skill, or collecting facts in a specific subject might identify higher functioning individuals and adults with ASD. These findings highlight these symptoms and the other behaviours described in Domain B of the proposed DSM-5 criteria are critical to measure correctly as they may be important for capturing individuals with Asperger-like profiles or Atypical Autism, who were proposed to be at risk of being missed by DSM-5 (e.g. McParland et al., 2011). For example the DSM-IV checklists, which were used in the first tests of DSM-5, may not have covered these impairments in enough detail or did not capture examples that fit the high functioning profile.

One test of the strength of these items more specific to high-functioning individuals would be to remove them from the DISCO DSM-5 algorithm and test for a drop in sensitivity. Further work has investigated the effect of only selecting items that discriminate at the ($p<0.05$) level of significance on the DISCO DSM-5 ASD algorithm, using the same data presented here (Carrington and Kent et al., 2014). This study aimed to further investigate the measurement of the DSM-5 description in order to identify those behaviours that best discriminated between individuals with ASD and non-ASD clinical diagnoses and which could therefore be considered 'essential' to the diagnosis of DSM-5 ASD. This study found that when only discriminating items were used in the algorithm, these items covered all of the DSM-5 sub-domains and sensitivity and specificity was not significantly altered in Samples 1 and 3. However, in Sample 2, which included a wider age range and ability level, reduction of the item set significantly decreased sensitivity of the algorithm in the children group and the proportion of individuals identified in the high functioning group was significantly lower than for the low functioning sub-group in Sample 3. This drop, however, could be remedied by replacing the items identified as relevant to high functioning individual that has been excluded. This provides one level of evidence that these range of behaviours in the DISCO are important in the high levels of sensitivity for DSM-5 present in the thesis. An independent test of

the strength of these DISCO items would be to add these behaviours to the checklists and DSM-5 algorithms that have previously shown a drop in sensitivity of DSM-5 in high functioning individuals (e.g. Frazier et al., 2011; Gibbs et al., 2012; McPartland et al., 2012; Mattila et al., 2011; Matson et al., 2012) in order to assess whether the measurement of these behaviours improves sensitivity outside of the DISCO framework.

Another spectrum approach was also tested in Part 3, Wing and Gould's ASD (WG-ASD), however, a strong confound is present in the test of this algorithm, especially in Sample 2, as the clinicians who collected the DISCO data (J. Gould & L. Wing) designed the WG-ASD algorithm and may therefore have given particular attention to the specific algorithm items. In Chapter 6, the WG-ASD algorithm was shown to perform with a high level of accuracy for identifying individuals with a clinical diagnosis of Autistic Disorder or Childhood Autism. In general, the overlap with the DSM-5 algorithm was high, indicating that the two spectrum algorithms are capturing the same individuals. The WG-ASD algorithm also performed extremely well at capturing the diagnostic sub-groups proposed in DSM-IV-TR/ICD-10 and that have previously been proposed in the literature. The WG-ASD algorithm identified 99% of the individuals meeting criteria for Gillberg's Asperger Syndrome and Kanner's Early Infantile Autism. In addition, the WG-ASD algorithm had an overall agreement of 97% with ICD-10. However, further work needs to be carried out which uses these questions within other clinical interview methods. Only when additional consistent findings are shown can the strength of this algorithm be confirmed. If information from both the DISCO (not collected by L. Wing or J. Gould) and importantly other interviews or clinical assessments found the same behaviours to have strong diagnostic accuracy it would provide a much stronger test of this algorithm.

The strong overlap between the DSM-5 and WG-ASD algorithms was first predicted to be explained by the overlap in items between the two algorithms. However, the set of analyses in Part 3, Chapter 6 found the three overlapping items could not, on their own, explain the overlap as their removal did not significantly affect the sensitivity or specificity of the DSM-5 algorithm. The second proposal was that the WG-ASD items are summaries of the information captured by DSM-5 items. Analyses were conducted which tested the replacement of the large number of DSM-5 items with the "summary" WG-ASD items. The replacement of the DSM-5 social-communication (Domain A) items with each of the four social-communication WG-ASD items was successful in maintaining the same level of sensitivity and specificity as when all the DSM-5 Domain A items were used as was the replacement of the Domain B (repetitive behaviour) items with "limited pattern of activities" in two of the three samples tested. The most significant finding in Part 3, Chapter 6 was the role of the Quality of Social interaction DISCO item. The addition of quality of social interaction to DSM-5 (with

all other items) improved the sensitivity of the DSM-5 algorithm and the strength of this item was shown as even when it was used to replace all of the items previously used in Domain A of the DSM-5 algorithm, it still improved sensitivity relative to the original DSM-5 ASD algorithm.

Consistently, the quality of social interaction items was shown to be important in the diagnosis of DSM-5 ASD and WG-ASD as well as discriminating well between individuals with ASD and clinical controls as a single item. Again, however, this item was rated by clinicians (in Sample 2) and researchers (in Samples 1 and 3) that recognise the importance of this item in the conceptualisation of ASD made by Wing and Gould. What is required is an independent measure of quality of social interaction in order to further test the prediction that this item or classification is important diagnostically. Some work has already addressed this by designing the Wing Subgroups Questionnaire (Castelloe & Dawson, 1993) but it should also be considered whether assignment to impaired or typical social group could be made by clinical opinion (separate to the DISCO) or through observation of the individual. Analyses in Chapter 7 found quality of social interaction to be significantly associated with the social items from throughout the DISCO and these items identified the majority of variance in quality of social interaction. This highlights a unique aspect of the DISCO that gathering the interviewer's judgement at the end of the diagnostic interview may be beneficial, investigation of this summarising in the DISCO and other clinical interviews could be an interesting methodological follow up for this thesis.

The four parts of the WG-ASD algorithm were all found to significantly correlate with each other. This supports the original proposal of a triad of impairments of social interaction, social communication and social imagination along with a restricted and repetitive pattern of behaviours (Wing 1988; Wing & Gould, 1979). Importantly, quality of social interaction and reciprocal communication were highly correlated with imagination as well as all three criteria being related to repetitive behaviours. This finding indicates that it may still be important to consider imagination in diagnostic criteria as an individual presenting with social and communication impairments is also likely to have impairments in social imagination as well. The combination of the findings in Part 3, Chapter 7 on quality of social interaction as well as in Chapters 5 and 6, in which the most prevalent and discriminating items were predominately social, raises a potential challenge in measuring the new diagnostic term "social (pragmatic) communication disorder" in DSM-5 that is diagnosed according to persistent difficulties in the social use of verbal and nonverbal communication (APA, 2013). Concerns have been raised about the validity of this disorder and whether it can truly be distinguished from ASD (Skuse, 2012; Tanguay, 2011). It is not yet clear whether further exploration of the individual's behavioural profile and developmental history may also reveal repetitive behaviours that could be missed using socially oriented diagnostic tools. In addition, the criteria has

no mention of imagination but it could be important to consider when evaluating individuals' social and communicative abilities as imaginative play has been shown to discriminate between individuals with autism and a language impairment (e.g. Barrett, Prior & Manjiviona, 2004).

#### *8.1.2.2 Part 3: Implications and future directions*

In terms of using the DISCO to make a diagnosis, the research presented here adds to existing research (Leekam et al., 2002; Maljaars et al., 2011; Nygren et al., 2009) by designing a DISCO DSM-5 algorithm that performs well. The DISCO items map onto the DSM-5 descriptions without any adjustments required, the only example of behaviour that cannot be directly mapped from the final published version of the DSM-5 ASD criteria is "rituals when greeting others'" but there are already several DISCO items representing examples for "Insistence on sameness, inflexible adherence to routines, or ritualized patterns of verbal or non-verbal behavior," which are the other examples in the same sub-domain.

In general this research indicates that if the DSM-5 ASD criteria are applied to a standardised instrument (or across two diagnostic instruments, Heurta et al., 2012; Frazier et al., 2012) then the sensitivity and specificity of the criteria are acceptable. However, in the clinical setting the training and use of tools is variable and the full impact of the DSM-5 will not be known until the criteria is used in clinical practice by individuals making a diagnosis, in many cases with limited time and resources rather the use of standardised assessments in research studies. Further research into the impact in clinical practice needs to be undertaken (Volkmar & Reichow, 2013).

Furthermore this thesis has highlighted the importance of considering how to use DSM-5 behaviours for research. The mapping of DSM-5 criteria onto standardised clinical tools that has already been conducted in the literature (e.g. ADI-R; Heurta et al., 2012) only required one example of a behaviour (i.e. one marked score on the ADI-R) to meet each sub-domain set out by DSM-5. In Chapter 5, the effect of changing the number of examples resulted in significant changes in sensitivity and specificity of the overall DISCO algorithm. This is important to consider across diagnostic instruments as the measurement characteristics also differ. For example, the ADI-R tends to summarise behaviours such as any atypical response to sensory input whereas the DISCO has around 25 items on separate sensory behaviours. This needs to be considered when measuring DSM-5 across diagnostic instrument to ensure the most reliable test of the DSM-5 ASD criteria.

This research contributes to the work assessing the impact of the DSM-5 changes in adults, which was highlighted as an area that had not been researched (Heurta et al., 2012). However, the oldest individual was 38 years old, so research identifying ASD in older adults will also be essential to thoroughly assess the impact. The research on DSM-5 in Part 3, Chapter 5 did also not include

information about whether the DSM-5 criteria can perform well in infancy or preschool age. An early diagnosis is important for early intervention and outcome but little work has assessed this so far in the literature. The use of the DISCO for this purpose may also be limited as the psychometric properties of the DISCO have not been tested in infancy. The work in Part 3 of the thesis also highlighted the methodological value of using the DISCO's quality of social interaction item, which is a judgement made by interviewers at the end of the DISCO. This judgement item may be useful in predicting an individual's behavioural profile and additional needs and further research on this is warranted.

Part 3 also focussed on the relatively unstudied concept of Wing and Gould's ASD. This work highlights that, like the DSM-5 ASD, the WG-ASD criteria, needs to be able to be applied across tools and clinical settings. Strong findings of diagnostic accuracy were found but currently these are limited to data collected using the DISCO, which emphasises the role of these behaviours and more critically by clinicians who may have been biased in the attention paid to these specific items. Overall, these findings offer exploratory analyses of an exciting avenue into a potentially sensitive and specific measure of the spectrum. As set out in Chapter 1, Wing and Gould's (1979) conceptualisation of a spectrum was limited in the lack of specific criteria required, this in turn limits how this conceptualisation can be tested. Although an algorithm exists to be used within the framework of the DISCO, criteria for measuring WG-ASD independently of the DISCO are still not available in the same way as DSM-5 criteria; these criteria can be compared with clinical records or more reliably mapped onto different tools in order to be tested. This is likely a strong reason as to why Wing and Gould's conceptualisation has been overlooked in research. The next step is to create criteria that can be applied to tools beyond the DISCO.

Finally, Part 3 highlighted the role that an individual's quality of social interaction may play both diagnostically and also in predicating an individual's additional needs. Again these findings are limited in their generalizability as this concept is based on the conceptualisation by Wing and Gould and therefore the DISCO data used here is potentially biased. However, this work uncovered a key future direction. The quality of social interaction item in the DISCO can also be split in social sub-types. Wing (1996) described the atypical social interaction style as ranging in severity across three groups of individuals: from "aloofness or indifference" to others, such that communication was limited to obtaining needs to "passive" acceptance with no spontaneous social behaviours or "active but odd" social approaches, in which individuals attempted to engage with everyone (friends, family and strangers) but in a limited form such as asking everyone the same set of repetitive questions. Exploratory analyses in Appendix 9 found that the "aloof" and "active but odd" group tend to display more 'associated' behaviours than the passive group in general. The aloof group was a significant

predictor of sensory behaviours, a limited pattern of activities, as well as the presence of tic and catatonic behaviours. The active but odd group was predictive of more problems with daily living skills, sensory behaviours and more Pathological Demand Avoidance behaviours. This clinical description of social sub-types proposed by Wing has been largely overlooked in research, however, the move toward a spectrum approach for measuring autism provides an ideal opportunity to explore if clear boundaries can be found between the groups through psychometric testing and whether these are of benefit in providing the best care and management for individuals with ASD.

## 8.2 Summary

The focus of this thesis was on defining the autism spectrum. In order to summarise the findings presented across both the distinct sections in the thesis, I have designed a diagram to represent the concept of the autism spectrum that has developed across the studies in this thesis (Figure 8-1).

In the figure, severity increases toward the centre of the circle, reflected with the darker shades seen across all behaviours toward the centre. The dotted circle reflects the area in which a clinical diagnosis of ASD is given. Within the dotted circle, the primary symptoms of social interaction, communication, repetitive and sensory behaviours can all interact (although the diagram is limited in presenting this). The primary symptoms can also occur independently of ASD as demonstrated by the sections of the circle seen outside of the diagnostic circle and outside of the quality of social interaction circle/continuum. Importantly, it is the pattern of behaviours within the dotted circle that make up ASD and that social interaction is fundamental to this definition of ASD.

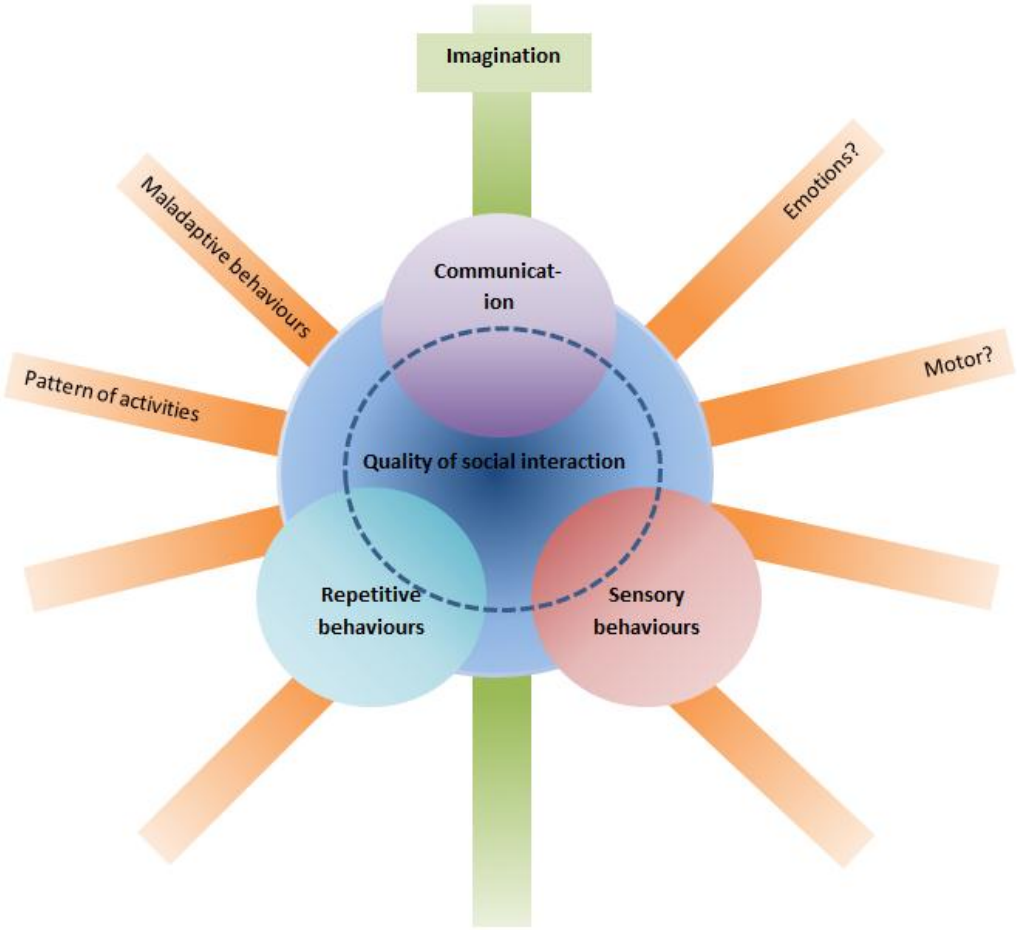



**Figure 8-1: Figure showing the model of the autism spectrum that summarises the results from the thesis.**

The main area of functioning that is atypical in individuals with ASD is the quality of their reciprocal social interaction. Findings from this thesis indicate that the single element of quality of social interaction, which is judged by interviewers at the end of the DISCO, has excellent diagnostic accuracy and may be useful in predicting an individual's behaviours and needs. However, this finding is limited in that, in Sample 2 in particular; the importance placed upon this item is known by the clinicians collecting the data. Nevertheless, the social interaction behaviours measured throughout the DISCO consistently appear in the pattern of behaviours in ASD examined across the thesis; social score was significantly related to both maladaptive and sensory behaviours in Part 2 and the social items in the DSM-5 algorithm were found to be the most prevalent and most relevant to all ages and levels of ability. This is reflected in the diagram by being the focus of the figure; a diagnosis of ASD is impossible without such impairments, the rest of the primary symptoms overlap with social interaction as do the associated or secondary features that cross the spectrum. This

supports the theorising of Wing (1996) who maintained that fundamental impairment in ASD is the lack of social instinct that is present from birth.

### 8.2.1 Comparison with previous spectrum conceptualisations

This model corresponds to both conceptualisations of the spectrum presented in the Chapter 1. DSM-5 describes a disorder with persistent deficits in two domains both of which are captured in Figure 8-1; (a) social interaction and communication and (b) restricted, repetitive patterns of behaviour, interests or activities. The move to capture some communicative behaviour within the repetitive behaviour domain can be modelled in Figure 8-1 as the primary symptoms all interact within the central diagnostic circle (dotted line). The figure also reflects the addition of sensory behaviours to DSM-5 ASD but in this model the sensory behaviours are included as a separate type of symptoms rather than as one of four possible repetitive behaviour sub-domains, this was done because the findings in Chapter 4 found sensory behaviours to be as strongly related to social functioning as they were to repetitive behaviours. One output from this model is that sensory behaviours should be thought about independently from their strong similarity with repetitive behaviours.

This model also fits with conceptualisation of the spectrum according to Wing and Gould's descriptions. Wing and Gould describe a triad of impairments in the domains of social interaction, communication, and imagination; the imagination impairment recently redefined as impairment in "social imagination", is associated with a rigid, repetitive pattern of behaviour. Social interaction and social communication are well placed in the figure and play a large role in the behaviours that would be used to make a diagnosis. The main difference is the role that imagination plays. Wing and Gould described imagination as a primary deficit in individuals with the triad of impairments. However, in the current thesis the imagination items did not have a significant role on their own, as identifying individuals with DSM-5 ASD or Wing and Gould ASD in Chapter 6. Its placement in the current model reflects the idea first proposed by Wing (1996) that impaired imagination is likely to result in a repetition of pretend play or enacting of a character that is strongly related to the primary symptom of repetitive behaviours and an impaired imagination may also impact on individuals understand of others' emotions which is a key aspect in social interaction (see Wing, 1996). It is therefore included as a special type of secondary symptom, however, much more work is needed on the role of imagination and the triad of impairments that Wing and Gould (1979) described. With the move to a spectrum in DSM-5, hopefully this creates a chance to readdress the spectrum approach proposed over 30 years ago (Wing & Gould, 1979).

Finally, the repetitive behaviour domain is classed as a primary symptom in the current model whereas in Wing and Gould's model it is described as a more of a result of the triad of behaviours instead. However, both this model (Figure 8-1) and Wing and Gould include these repetitive or restricted behaviours in their description of ASD. Wing (1996) also detailed the responses to sensory input which may be atypical in individuals with ASD but that these "become less marked with increasing age and may eventually disappear" (p. 51). The results of the SPQ study, however, show that 94% of adults with ASD have at least one marked sensory behaviour, so these behaviours are seen as primary in the model below.

### *8.2.2 Secondary Symptoms*

The orange boxes in Figure 8-1 represent the secondary behaviours that are associated with ASD but not part of the diagnostic criteria. For example, the maladaptive behaviours measured in Chapter 3 were significantly related to the core features of ASD, however, this significant effect was mediated by the sensory behaviours, meaning that maladaptive behaviours were unlikely to play a "core" or causal role in the presentation of ASD but may impact on individuals with ASD. Therefore, it is important to reflect these in the conceptualisation of ASD but separate from the primary features. Any multitude of secondary behaviours can cross the autism spectrum (but fall behind the main features) and all of them can "overlap" in the diagram which represents how they can also affect the presentation of each other for example sleep problems further impacting on the presentation of aggression/maladaptive behaviours (e.g. Mayes & Calhoun, 2009; Sikora et al., 2012).

Further work is needed on these secondary behaviours to discover the mechanisms by which they impact on individuals with ASD and whether these are distinct pathways from those found in typical development. More specifically, research is needed on the role of the motor impairments, emotion and sleep behaviours in ASD in order to assess whether they are best placed as secondary features or should be represented as another primary circle. This could not be explored further in the current thesis as these scales in the DISCO were not found to be reliable.

### *8.2.3 Sub-typing*

Finally, the dashed circle reflects the section that represents individuals who should receive a clinical diagnosis. It is within this section that the possibility of identifying sub-groups with particular needs or causes may be useful. As summarised by Amaral (2011) "given the incredible heterogeneity of this disorder, understanding that one size will never fit all is a reasonable perspective to frame all future findings" (p. 8). The evidence presented in Part 1 concluded that the

evidence for a distinction between the DSM-IV sub-groups was minimal, however, this was due to the application of the DSM-IV criterira for Asperger Syndrome. The move to a spectrum definition may actually allow a less biased review of the clinical descriptions made by Asperger and Kanner rather than the criteria applied to these names by the international classification systems. The rich clinical descriptions across the short history of ASD should be re-examined, for example, this thesis highlights one such description of Wing's social sub-types. This is one of many ways of sub-typing (language, ability, associated features) that should be examined in future research. In conclusion, the move toward a spectrum is certainly helpful in a clinical setting. Resources can be focused on the indivdiuals' needs rather than deciding which sub-group an individual belongs to. However, for furthering knowledge about the causes and aetiology of ASD, sub-typing of the spectrum may provide a useful way to further knoweldge.

To conclude, this model is not a definitive proposal for the behavioural manifestations of ASD but reflects the findings within the current thesis. This model will hopefully be helpful in guiding future research questions with the DISCO as well as other tools. As previously stated, the concepts and ideas of ASD are constantly evolving and our concepts of ASD need to progress with this in order to provide the best individual care to those who fall within the clinical range of the spectrum.

### *8.2.4 Predictions from model*

This model proposes that individuals with ASD would present with impairments in the quality of their social interaction, which would also co-occur with communication impairments and repetitive and sensory behaviours. This would predict that there would be significant direct effects between these primary symptoms. In turn, the secondary symptoms may impact on an individual's core features in an indirect way. This model would predict that secondary symptoms such as maladaptive behaviours may affect an individual's social functioning through another core feature e.g. sensory behaviours or through another associated feature e.g. anxiety. These effects could be tested using path analysis, although a more robust dataset, ideally of a longitudinal nature would be of benefit for these analyses.

This model also predicts that the core features of ASD are usually seen with limited social imagination. This can be tested by examining the imaginative abilities of the individuals that present with the primary features or recording the primary features of ASD in individuals with impaired imagination. An interesting follow up would be whether any of the primary behaviours alone are strongly related to imagination and whether these associations occur outside of individuals with ASD as well as with those with a clinical diagnosis. However, this area has been widely overlooked

because the DSM-IV-TR defined the triad of behaviours as consisting of social interaction, communication and repetitive behaviours rather than Wing and Gould's original hypothesis of social interaction, communication and imagination. However, the difficulty in measuring imagination is finding appropriate measures to test adults who have matured past pretend play.

In addition, this model would predict, within both individuals who fall into a clinical diagnosis and outside this circle, that the severity of impairments could vary and that the primary symptoms can vary independently of each other e.g. very socially impaired individuals would not have to be very sensory impaired as well (but could be). Finally, further exploration is needed on measures of motor and emotions (such as anxiety and depression); reliable measures could be used in statistical models such as the preliminary regression models set out in Part 2 to further identify what roles these behaviours play. The binary variables used in the thesis were of limited predictive value.

We still have a long way to come in our understanding of ASD. It is not a simple condition with one causal or underlying mechanism. The argument presented in this thesis is that ASD consists of a pattern of behavioural symptoms which occur together. It is likely, therefore, that the range of behaviours both central and related to ASD impact on the individual in combination and are likely to have complex interactions in order to create the overt clinical picture. Examining these behaviour in isolation will only ever be able to inform us about that behavioural symptom. Examining these behaviours in combination and the interactions between them will hopefully improve our knowledge about the behavioural manifestation of ASD and in turn improving the potential avenues for identifying biological mechanisms with aetiological significance. This combination of behaviours may be unique across all individuals with ASD and therefore, until further advances are made the best intervention and care programmes are those that assess an individual's strengths and weaknesses.

## 8.3 Conclusion

In conclusion, despite some of the limitations, this thesis provided significant contributions to the literature on the DISCO, sensory behaviours and measurement of the spectrum. The unique contributions of this thesis are as follows: 1) the DISCO was used as a research tool and this thesis validated the use of the maladaptive and sensory symptoms included in the DISCO; 2) the thesis also included the first attempt at converting DISCO items into self-report questions. The Sensory Preferences Questionnaire was designed and (a) found to be reliable and valid and (b) showed good overlap with existing self-report sensory questionnaires and (c) helped substantiate the DISCO sensory findings within the current literature; 3) this was the first study to highlight the role of sensory behaviours in the relationship between the core features of ASD (social interaction and

repetitive behaviours) and maladaptive behaviours; 4) Part 3, was the first study to demonstrate both sensitivity and specificity of the DSM-5 criteria using one diagnostic tool and this thesis raises the awareness of how the selection of items and threshold cut-offs can influence performance; 5) another novel aspect of the thesis was the comparison between the international classification systems (DSM-5) measure of a spectrum with the existing Wing and Gould spectrum (1979).

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# 10 APPENDICES

# APPENDIX 1: COMPARISON OF DIAGNOSTIC INTERVIEWS: THE DISCO, ADI-R & 3DI

This appendix focuses on the measurement of ASD. As discussed in Part 1, the convergence of diagnostic criteria across both DSM-IV and ICD-10 led to a magnitude of new research being conducted and this abundance of research resulted in the development of a range of diagnostic tools that could be used to confirm or aid in the diagnostic process of individuals taking part in research as well as in clinical settings. These measures include questionnaires, observations schedules and diagnostic interviews. Current diagnostic assessments in research and clinical practice rely on assessment of behaviours through observation or interview. Although research has identified potential biological measures that may have diagnostic significance (e.g. brain imaging; Ecker et al., 2010; Lange et al., 2010) further replication is required before such measures were included in routine autism assessments.

The National Institute for Health and Care Excellence (NICE) have published guidelines for diagnosis of Autism in both children and young people (CG128, 2011) and autism in adults (CG124, 2012). The recommendations of what information should be gathered in an assessment can be seen in Table 1. Both sets of guidelines recommend that in clinical practice a diagnosis of ICD-10 or DSM-IV is made from information from all sources and from clinical judgement. They make clear that autism-specific diagnostic tools should not be relied upon alone to make a diagnosis. However, diagnostic tools have an important function in helping the clinician to collect standardised information to be used in making a judgement about differential diagnosis as well as assessment of needs. Instruments that routinely collect a large range of the information specified by the NICE guidelines as standard will be more beneficial, which is why it is important to compare diagnostic interviews to these criteria. Furthermore, standardised clinical tools are important for the defining of research samples in order to look for meaningful differences between ASD and non-ASD groups as well as examining the specificity of behaviours seen in ASD.

**Table 1: Table showing the recommendations made by the National Institute of Health and Clinical Excellence for the diagnosis of children and adults with autism that are referred for a comprehensive assessment**

| CG142 Autism in children and young people (2011, page 2011) | CG142 Autism in adults (2012, page 2012) |
|---|---|
| Assessment (through interaction with and observation of the child or young person) of social and communication skills and behaviours, focusing on features consistent with ICD-10 or DSM-IV criteria | Core autism signs and symptoms (difficulties in social interaction and communication and the presence of stereotypic behaviour, resistance to change or restricted interests) that have been present in childhood and continuing into adulthood |
| Details of the child's or young person's experiences of home life, education and social care | Functioning at home, in education or in employment |
| A developmental history, focusing on developmental and behavioural features consistent with ICD-10 or DSM-IV criteria | Early developmental history, where possible |
| A medical history, including prenatal, perinatal and family history, and past and current health conditions | Past and current physical and mental disorders |

| | |
|---|---|
| A physical examination and consideration of the differential diagnosis and systematic assessment for conditions that may coexist with autism | Other neurodevelopmental conditions |
| Development of a profile of the child's or young person's strengths, skills, impairments and needs that can be used to create a needs-based management plan, taking into account family and educational context | Behavioural problems and individual needs in relation to personal and social functioning and educational, occupational and housing needs. |
| Detailed questions about parent's or carer's concerns and, if appropriate, the child's or young person's concerns | |
| | Hyper- and/or hypo-sensory sensitivities and attention to detail. |
| | The need for an assessment of challenging behaviour, where appropriate, which should be part of the comprehensive assessment outlined above. |

Three inclusion criteria were applied to the tools reviewed in the current review reported here. First, the instruments must have been developed for the assessment of autism (rather than generic range of conditions). Second, in order to measure a developmental history as specified by the NICE guidelines the review was limited to parent report diagnostic interviews as they have been shown to have better accuracy and consistency than other tools and have therefore become the "gold standard" for obtaining clinical histories (Le Couteur & Berney, 2013). Finally, as the research questions in the thesis are based on the autism spectrum across age and ability level, tools must be designed to capture autism as a whole rather than specific sub-groups, for this reason the Asperger Syndrome (and high-functioning autism) Diagnostic Interview (ASDI) that can only be used for individuals with an IQ above 70 was not included (Gillberg, Gillberg, Rastam, & Wentz, 2001).

### *Diagnostic Interviews*

The aim of a diagnostic interview is to gain information about the individual in order to make a decision as to whether the individual meets criteria for the behaviours identified in the international classification systems. Interviews can be undertaken for many reasons from inclusion in a research study to creating an intervention plan. For a diagnostic interview for ASD to be effective Le Couteur and Berney (2013) propose six criteria that the interview should contain:

1) The capacity to record the current concerns or difficulties of the individual that have resulted in the diagnostic interview being conducted at this time, and their development.
2) The systematic collection of information about the behaviour relevant to a diagnosis of ASD as well as behaviours that are known to be associated to ASD such as feeding and bowel problems, motor co-ordination and sensory processing.
3) Ability to capture the individual's environment such as their day to day activities, relationships and abilities.
4) The medical history (including developmental and psychiatric diagnoses) and structure of the family.
5) A developmental history. The interview should be able to record an individual's development of skills from infancy and early childhood as well as through adolescence and after.

6) A report of any current or past relevant behaviours or conditions such as psychiatric or medical disorders, adversity or substance abuse.

Three standardised diagnostic interviews are the most widely used and all broadly cover these criteria, although differ in the amount of additional information that they collect: the Developmental, Dimensional and Diagnostic interview (3di; Skuse et al., 2004), the Autism Diagnostic Interview-Revised (ADI-R; Lord et al., 1994) and the Diagnostic Interview for Social and Communication Disorders (DISCO; Wing et al., 2002). These three diagnostic interviews, their similarities and differences will be reviewed. Primarily, the accuracy of the three interviews was reviewed in terms of making a diagnosis as well as reliability and validity of the tool (see below). In addition, the clinical utility of the tools will be compared using the criteria set out above. The ADI (and ADI-R) is the most frequently used diagnostic interview in research and therefore comparisons will be made between the ADI and the other instruments to highlight the differences.

## *How can we test the accuracy or success of diagnostic criteria?*

### *Sensitivity and specificity*

In order for diagnostic criteria to be accurate it needs to be both sensitive and specific. Sensitivity refers to the ability of the criteria to capture all individuals with ASD who should receive a clinical diagnosis. Specificity, on the other hand, refers to the criteria's ability to correctly exclude people who should not receive a clinical diagnosis from meeting on a tool or algorithm. In a statistical sense both of these values can be calculated and rated on a scale from 0 (no ability to be sensitive or specific) to 1 (perfect sensitivity or specificity) or measured as the percentage of individuals correctly classified as having an ASD and the percentage of the control group who do or do not meet criteria. Following the criteria set out by the NICE guidelines, values of 0.9 and above are "excellent", 0.8-0.9 "good", 0.5-.07 "moderate", 0.3-0.4 "low" and below .30 as "poor" (NICE, 2012).

For the majority of diagnostic instruments the diagnostic criteria are directly mapped onto a diagnostic tool such that the relevant items from that tool are used to decide if a diagnosis should be given. This approach is referred to as creating a specific diagnostic "algorithm" for that particular tool, an algorithm is the selected set of items that are combined by rules, such as those set out in DSM-IV-TR to provide a binary outcome, whether an individual meets diagnostic criteria. In most cases the algorithm scores for the diagnostic interview is compared with an individual's clinical diagnosis. Unfortunately, this method is flawed in that a clinical diagnosis is based on behavioural features that may also be extracted from diagnostic tools and behaviours from diagnostic tools are the items that are being tested for discriminant validity (Leekam et al., 2002). This affects all studies testing the validity of a tool, not just diagnostic interviews. However, as no biological or genetic test exists to confirm a diagnosis of autism, this is the best approach and has been adopted for the majority of psychometric testing for tools in ASD and allows identification of where clinical and instruments converge and disagree.

### *Reliability and validity*

The diagnostic tools measuring a range of behaviours relevant to ASD should be both reliable and valid (Sattler, 2001), the three interviews will be assessed against established levels of reliability and validity. The reliability of a measure refers to how consistently it captures what it has been designed to test. There are three types of reliability: consistency across time or test-retest reliability measures whether the same individual scores the same on a measure at time point 1 as they would at time point 2; consistency across raters (or inter-rater reliability) refers to the ability of the tool to be used by multiple "raters," i.e. that the same individual would score the same regardless of which clinician was conducting the diagnostic interview; finally internal consistency of a tool refers to how well the items in the tool measure the overall construct being measured. The general consensus is that both inter-rater and test-retest values should correlate higher than r=.70 in

order to be rated as reliable and that different methods for measuring internal consistency are considered reliable as follows: r=0.70; α>0.50 (Cronbach's alpha); and *k* = 0.40 (kappa coefficient; NICE, 2012).

In addition, two types of validity can be calculated: criterion and construct. Construct validity can be tested in two ways: discriminant validity measures how well the tool discriminates between different groups e.g. ASD and control cases; convergent validity refers to whether the tool is correlated with other tools that measure similar constructs. Criterion validity is an index of how well the tool correlated with an established construct such as a clinical diagnosis. This can also be measured in two ways through predictive validity (how well tool predicts future measure) or concurrent validity (how well tool related to another current measure). For all validity measures, correlations above r=0.50 are referred to as valid (Burlingame, 1995; NICE, 2012; Nunnally, 1994).

### *ADI-R*

The most widely used diagnostic interview in research is the Autism Diagnostic Interview (Le Couteur et al., 1989) and its revised schedule, the ADI-R (Lord et al., 1994). The ADI-R is a semi-structured diagnostic interview. It contains 93 items in total, 43 of which measure reciprocal social interaction, communication and restricted or repetitive behaviours according to ICD-10, which form the ADI-R algorithm when systematically combined. The items included in the ADI-R were selected in order to identify ASD according to ICD-10 and therefore limited in addressing changing diagnostic criteria as well as behaviours that are not part of the diagnostic criteria. It was designed to be used in children with a mental age above two years, although an algorithm based on current behaviour can be used for children from 12-47 months old (Kim & Lord, 2012) and a toddler version of the ADI-R has also been developed (Kim, Thurm, Shumway, & Lord, 2013).

The ADI-R is used to obtain a history of an individuals' behaviour as well as the current clinical picture. The ADI-R captures information about each behaviour for both "current", whether the behaviour has occurred in the last three months and "past," for some behaviours this captures whether then behaviour was present between 4-5 years or age and for other behaviours it measures whether the behaviour has ever been present. Being a semi-structured interview that is led by the interviewer, it requires more training than the 3di and advises users to maintain inter-rater reliability and consistency by regularly comparing codes.

#### *Diagnostic validity and Reliability of ADI/ADI-R*

The original ADI was tested across 16 individuals with autism and 16 with intellectual disabilities (mean age of 12.28 years), the majority of ADI items significantly discriminated between the two groups, however, the non-ASD group consisted of only individuals with an intellectual disability and no high functioning comparison group was used. In addition, intra-class correlations were excellent (>0.94) for sub-domain and domain scores and inter-rater kappas for individual items ranged from 0.25 to 1 (Le Couteur et al., 1989). The revised schedule was tested in a similar way using 25 children with autism and 25 children with an intellectual disability, again the majority of items could significantly discriminate between these two groups. Furthermore, the intra-class correlations for the domain and sub-domain scores were excellent (>.92) and inter-rater reliability was also good for the majority of items (*k* ranged .63-.89; Lord et al., 1994).

The ADI-R had diagnostic algorithm cut-offs, which have been shown to have excellent sensitivity and specificity (>.90) between individuals with ASD versus other clinical conditions in 25 children over three years old with ASD and 25 children with an intellectual disability or language impairment (Lord et al., 1994). Further testing of these algorithms have not found such high levels of diagnostic accuracy but the levels of sensitivity are usually above .70 across all samples. In a sample of 78 children aged 22 months to 8 years, the diagnostic algorithm in the ADI-R identified 75% of children with a team diagnosis of Autism and 28% of the non-autism group, giving sensitivity of .75 and specificity of .73 (Mazefsky & Oswald, 2006). However, the levels of specificity have also been found to be below acceptable levels. In 184 individuals (5-20 years old) with an intellectual

disability, cut-offs for Autistic disorder provided sensitivity of .771 and specificity of .632 and cut-off for PDD resulting in a sensitivity of .716 and specificity of .787, compared to DSM-IV-TR classifications (De Bildt et al., 2004). Sensitivity was .88 but specificity .69 in a sample of 77 individuals from 33months – 22 years (Papanikolaou et al., 2009). However, the sample used by Papanikolaou et al. (2009) used a referral sample rather than individuals specifically recruited into ASD or non-ASD groups. This is a more accurate test and therefore these results are comparable to higher values in pre-selected groups.

The ADI-R diagnostic algorithms were not as successful at diagnosing preschool children and discrimination was poor between non-verbal children with or without ASD under two years old (Lord et al., 1994). The problem of low specificity in younger children has been replicated (Risi et al., 2006) and others have also found low sensitivity in 16-37 month olds (Gray, Tonge, & Sweeney, 2008; Ventola et al., 2006; Wiggins & Robins, 2008). The ADI-R now has algorithms specifically designed for toddlers from 12-47 months, Kim and Lord (2012) analysed 491 toddlers with ASD, 136 toddlers with other disorders and 67 typically developing children to give good sensitivity (>.80) and moderate specificity (>.70). The also designed research algorithm cut-offs with higher specificity and lower sensitivity which also performed well in toddlers (sensitivity, .80-.84; specificity, .85-.90).

### *The 3di*

The 3di (Skuse et al., 2004) is unique in that the completion of the interview is completely computerised. It was developed to assist clinicians produce reliable diagnoses with less training than the ADI-R or DISCO. The computer programme can output symptom analysis, an automated written report as well as run diagnostic algorithms for DSM-IV-TR categorical diagnoses and common childhood co-morbidities according to ICD-10 (Skuse, 2013). A positive of this approach is the benefit for research as data from the programme can be directly transferred to statistical software for analysis. The 3di has a large range of items: around 200 collecting information on family background and developmental history; 300 on questions relevant to a diagnosis of PDD disorders; as well as 300 additional items relevant for other diagnoses. The items are broken into modules to allow selection of how much information is required to collect about each individual and just the algorithm items can be asked when a diagnosis is strongly suspected according to an abbreviated 3di (Santosh et al., 2009).

The 3di is described as neither a structured nor semi-structured interview but a combination of both techniques. Questions must be asked in the way they are written which is appropriate for individuals over the age of three. It can be administered to assess adults with only minor modification to the questions asked to the informant (Skuse, 2013). A three scale severity scale is used to code “no such behaviour,” “minimal evidence of such behaviour” or “definite or persistent evidence of such behaviour.” Unlike the other two interviews, the 3di does not collect separate codes for ever and current scores and instead adopts the approach of if a behaviour is currently present then it scores for the ever measurement (Skuse et al., 2004). This is similar to the DISCO (Wing et al., 2002, see below) in that if an individual is rated as having a marked behaviour currently then the marked code is also assigned to ever, however, both codes are recorded and the current code can be less marked than the ever.

#### *Diagnostic validity and reliability of the 3di*

Four samples of children were recruited for a reliability study of the 3di. The majority of participants were randomly selected from referrals to either the psychiatric services who were suspected of having a PDD or general paediatric services. Although, individuals were excluded from the control group (general paediatric) if they had behavioural problems, this selection process is less biased than specifically recruiting individuals with ASD for the ASD group and non-ASD individuals for the control groups, like the ADI above, however, ASD and control individuals were still recruited separately. The best test would be between those referred to psychiatric services who went on to receive a diagnosis and those who did not, this only constituted a minority of the participants in this

study. It was found that that the correlations were excellent (>0.86) for both inter-rater and test-retest reliability for all algorithm scores used to measure DSM-IV-TR in individuals with ASD and clinical controls, although scores were lower for typically developing participants, which were reported as "most in excess of 0.70" (Skuse et al., 2004, p. 522). The 3di identified all individuals with ASD (sensitivity = 1.0) and also reported excellent specificity (>.93; Skuse et al., 2004).
In addition, the 3di was compared with the ADI-R in another sample of 29 children. The interviews were counter-balanced and conducted an average of 2.4 years apart. Complete agreement on diagnosis was achieved for 19 individuals (according to the ADI-R algorithm applied to the ADI-R and 3di data), for 9 cases the instruments disagreed on one domain score and in one case on two DSM-IV-TR domains. This resulted in a correlation between the instruments of 0.64 for reciprocal communication and communication and 0.53 for repetitive and stereotyped behaviours. The latest version (3di-5) computes diagnostic probabilities for comorbid conditions such as ADHD, Conduct Disorder and Tic disorders according to DSM-5 using data collected from the full 3di. Additional 3di modules can be used to gather the same information for anxiety disorders, Depression, eating disorders and schizotypal disorders, bipolar affective disorder, post-traumatic stress disorder and Dysthymia (Skuse, 2013).

### *The DISCO*

The Diagnostic Interview for Social and Communication Disorders (Leekam et al., 2002; Wing, 2003; Wing et al., 2002) is a semi-structured interview conducted with a parent of caregiver of an individual of any age or level of ability. It is the interviewers responsibility to gain enough information to ensure accurate coding and interviewers are also required to make judgements about individuals overall behaviours. The purpose of the interview schedule is to collect information about a broad range of behaviours both relevant to the diagnosis of ASD as well as the individual's development, skills and difficulties. The wide range of information collected is interpreted within the individuals' broad developmental and behavioural background. The DISCO was designed around Wing and Gould's concept of a spectrum of autistic disorders as well as the behaviours seen frequently in clinical practice and is intended to measure the autism spectrum. A full description of the information collected with the DISCO can be found in the general methods chapter (chapter 2). The key strengths of the DISCO are described by (Leekam, 2013) as collecting information about a wide range of behaviours that go beyond the core features of autism, these include sensory processing, motor impairments, maladaptive behaviours, emotional disturbances as well as behaviours that related to other clinical condition such as ADHD or DCD. This is a key strength for research with the DISCO as all of the information is collected from the same informant and in the same style which improves reliability and could help to overcome the multitude of measures used across research studies on the associated features of ASD. In addition, the DISCO can be used to collect information about an individual of any age and it also focuses on the development of the individual as it captures information about both delays in developmental milestones as well as the individual's current level of ability. This allows the collection of further information to be placed within a broader framework on that individual's developmental level i.e. if the individual is capable of dressing themselves but refuses to do so, which will improve the reliability of recommendations about individual's needs and management.

The DISCO contains over 300 items, which cover 8 sections of developmental and untypical behaviours. A full description can be found in the "DISCO Methods" below. Untypical behaviours are recorded for both "ever," which captures the worst manifestations of the behaviours and "current" scores, which refer to whether the individual currently presents with that behaviour. The DISCO has 147 items that are used to form DISCO diagnostic algorithms for the categorical diagnoses in both DSM-IV-TR and ICD-10 as well as algorithms for Kanner and Eisenberg's (1956) Early Infantile Autism, Gillberg's criteria for Asperger Syndrome (Ehlers & Gillberg, 1993; Gillberg & Gillberg, 1989; Wing, 1981), Autistic Spectrum Disorder (Wing & Gould, 1979) and Wing and Gould's social impairment. In addition, the DISCO collects information about comorbid conditions. Checklists of

behaviours are provided, which help to identify whether an individual should be referred for further assessment for ADHD, tic disorders, developmental co-ordination disorder, catatonia and Pathological Demand Avoidance (Newson, Le Maréchal, & David, 2003). Furthermore, there is a section of the DISCO mainly for use in adolescents and adults that collects information on psychiatric disorders such as schizophrenia, personality disorders, sleep problems and eating disorders.

### *Comparison of the DISCO to the ADI*

The DISCO and the ADI/ADI-R were designed for two distinct purposes, the ADI was primarily designed to be used to enable clinicians to make a diagnosis according to international classification systems in contrast to the DISCO, in which the aim was to systematically record information on developmental history from birth as well as a measure of current level of functioning (Wing et al., 2002). As Nygren et al. (2009) describes, the DISCO differs from the ADI-R at both the "section" and "item" level. The DISCO contains several "sections" that are not covered by the ADI-R such as emotional disturbance, psychiatric disorders, catatonia, problems with sexual behaviour or crime, sleeping problems, maladaptive behaviours as well as a range of daily living skills. In addition, at the item level the DISCO tends to have more items which capture a range of examples of behaviour whereas the ADI-R summarises these in one question. For example, "unusual sensory interests" is covered by a range of items in the DISCO covering responses to auditory, visual and proximal stimuli. Both these tools collect information about current untypical behaviours as well as past or ever codings of the specified behaviour, however, for the DISCO this refers to the behaviour at its worst rather than a specific time point and interviewers rate behaviours that the parent or caregiver remember well. On the other hand, the ADI-R asks for information about the individual between the ages of 4 and 5. However, recall of when behaviours occurred has been shown to be unreliable, Angold, Erkanli, Costello, and Rutter (1996) found that the onset of behaviours was uncertain if it had occurred for a year or more. The DISCO "ever" approach may therefore by more reliable, for the DISCO interviewers are trained to record behaviours that the parent or caregiver remembers clearly (Wing et al., 2002).

### *Diagnostic validity and Reliability of the DISCO*

Inter-rater reliability scores were conducted for over 300 items from the DISCO and the majority of items had high inter-rater reliability; kappa coefficients or intra-class correlations were above .75 in 80% of items. The DISCO also enables algorithm diagnoses according to ICD and DSM. The original ICD-10 Childhood Autism algorithm was based on 88 DISCO-9 items and a set of rules specifying how these items convert into diagnostic outcome (Leekam et al., 2002). This algorithm was originally tested along with the Wing and Gould ASD algorithm in a sample of 36 children with autism, 31 individuals with a non-ASD clinical condition and 15 typically developing children. The items in both algorithms had good inter-rater reliability and the two algorithms were significantly related to individual's clinical diagnoses (Leekam et al., 2002).

The ICD-10 algorithm has also been shown to have good criterion and convergent validity in a study using the Dutch translation of the DISCO-11 (van Berckelaer-Onnes et al., 2008). Again individuals with autism (n=52), non-ASD intellectual disability (n=26) and typically developing children (n=37) were compared. The ICD-10 DISCO algorithm had good sensitivity (.96) and specificity (.79), although the comparison to clinical diagnosis was better in individuals with average intelligence or a mild intellectual disability than those with moderate to severe intellectual disabilities. The DISCO also showed strong agreement with scores on the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000, k=.69), a structured observation task designed to be used in conjunction with a development history interview (the ADI-R) to also make a diagnosis. However, lower agreement was found with the Social Communication Questionnaire (SCQ; Rutter et al., 2003, pp., k=.49), which is parent report questionnaire on an individual's social behaviours (Maljaars et al., 2012). The DISCO ICD-10 algorithm was also tested against the ADI-R and showed excellent agreement with it and clinical diagnosis and again the inter-rater reliability of these items were excellent for 70% of the items (Nygren et al., 2009). The DISCO, however, has not been tested

in very young children and therefore it is unknown whether additional cut-offs or algorithms may be required like for the ADI-R.

### *Summary of diagnostic interviews*

In terms of reliability, the results for the 3di, ADI-R and DISCO are relatively consistent and show each is capable of being used to provide a diagnostic outcome. For the 3di, criterion validity was excellent and correlations were above 0.50 for construct validity in comparison to the ADI; inter-rater and test-retest reliability was good but internal consistency was not reported and only one validity study has been conducted (Skuse et al., 2004). The advantage of this study on the 3di is that participants were recruited from individuals referred to clinics suspected of having a PDD or to a general paediatric clinic, with no behavioural problems, this contrasts with the original ADI and DISCO samples who were recruited specifically as already having an ASD, however, the sample selected all had average or high ability levels. More reliability and validity studies are needed on the 3di across age and ability level.

The DISCO has been shown to have good inter-rater reliability for the majority of responses to items (Nygren et al., 2009; Wing et al., 2002). The construct validity of the ICD-10 algorithm was good and significantly related to clinical diagnosis (Leekam et al., 2002) and scores on the ADI-R (Nygren et al., 2009) and the ADOS but just below acceptable levels with the SCQ (Maljaars et al., 2012). In addition, the sensitivity and specificity of the ICD-10 algorithm for the DISCO were good (Maljaars et al., 2012). The DISCO and 3di have not been directly compared, although both correlate well with the ADI-R. Validity of the DISCO is clear for children but still to be tested in adults and very young children, which is also true of the 3di. Finally, the ADI-R is by far the most well validated in the literature. Several studies have found excellent sensitivity and specificity (Kim & Lord, 2012; Le Couteur et al., 1989; Lord et al., 1994) across individuals of all ages. However, in less well defined samples (i.e. not recruited as ASD or non-ASD) specificity has been shown to drop below acceptable levels, limited work with the 3di and DISCO has assessed this possibility. The inter-rater reliability and intra-class correlations have also been shown to be excellent in the ADI-R (Lord et al., 1994).

The content of the interviews was also reviewed in comparison to the criteria set out by both the NICE guidelines (2011; 2012) and Le Couteur and Berney (2013). The three interviews are all successful in recording the current concerns of the individual and systematically collecting information about behaviours relevant to a diagnosis of ASD according to ICD-10. In comparison to the ADI-R, the DISCO and 3di collect a large amount of information on the associated features of ASD as well as behaviours that are relevant to signposting individuals for related clinical conditions. This is incredibly important for differential diagnosis in order to systematically assess for other disorders that may coexist with ASD. The 3di and ADI-R have less of a focus on the developmental level of the individual; the DISCO provides the best developmental perspective and in comparison to the other two instruments it collects much more detailed information about individual's delays, current functioning and untypical behaviours for a range of developmental skills as well as capturing behaviours in infancy. Development of a profile of the child's or young person's strengths, skills, impairments and needs, taking into account family and educational context is ideal for identifying the need for intervention and care (NICE, 2011) and many of these areas are covered in the DISCO. The focus of the current thesis is on the DISCO. The DISCO has been widely overlooked in research, however, its developmental nature, coverage of associated and comorbid behaviours and relevance across age and ability level make it a useful standardised tool for the research questions posed in this thesis. In addition, it shows comparable reliability, validity and diagnostic accuracy with the ADI-R as well as strong overlap with the ADI-R, which is the most frequently, used measure in research. The following section sets out why the DISCO is capable of measuring Autism Spectrum Disorder, according to the new criteria set out by DSM-5. This is followed by a review of the research that has previously been conducted using the DISCO, to highlights its broad application across research questions and its relevance to the research questions in this thesis.

# APPENDIX 2: UPDATING THE ALGORITHMS FOR DISCO 11; SUMMARY OF CHANGES

The items for each algorithm and changes between the DISCO 9 and 11 versions of the algorithms are presented in the tables below along with the frequency of each item in the ASD group and the ability of each item to discriminate between ASD and clinical comparison according to the chi-square statistic (ICD-10, Kanner and Eisenberg's Early Infantile Autism and Gillberg's Asperger Syndrome). The algorithm for Gillberg's Asperger Syndrome criteria changed substantially between DISCO 9 and DISCO 11 versions and therefore only DISCO 11 was included.

## *ICD-10 (& DSM-IV)*

| Variable | Label | DISCO 9 | DISCO 11 | Changes required? | Frequency in ASD group | Chi-square statistic |
|---|---|---|---|---|---|---|
| | | Codes | | | | |
| **Onset before three years** | | | | | | |
| Set-back language | LOSSLANG | 001-035 | 001-035 | | 2.78 | 0.874 |
| Set-back play | LOSSPLAY | 001-035 | 001-035 | | 8.33 | 2.704 |
| Set-back social | LOSSOC | 001-035 | 001-035 | | 13.89 | 2.323 |
| Obeying instructions | BINSTRUC | 0 | 0 | | 83.33 | 0.082 |
| Combining words | B3WORDS | 000 Or 36 thru 500 Or 777 (late) | 000 Or 36 thru 500 Or 777 (late) | | 86.11 | 0.066 |
| Selective attachment | BAFECTIN | 0 | 0 | | 50.00 | 5.354 |
| Development pretend play | BPLAYAL | 0 | 0 | | 91.67 | 2.663 |
| **Section B1. Social impairment** | | | | | | |
| **Item B1a. Failure to use non-verbal behaviours to regulate social interaction** | | | | | | |
| Imperative gestures | AIMPERAG | 0,1 | 0,1 | | 13.89 | 4.653* |
| Declarative | ADECLARG | 0 | 0 | | 50.00 | 15.085*** |
| Nodding head | ANODSHK | 0 | 0 | | 41.67 | 6.783** |
| Instrumental | AINSTRUG | 0 | 0 | | 69.44 | 6.363** |
| Descriptive | ADESCRG | 0 | 0 | | 75.00 | 7.569** |
| NVC with social interaction | CUSENVC1/2 | 0, -8 | 0, -8 | | 63.89 | 8.111** |
| Eye contact | CEYECON1/2 | 0 | 0 | | 52.78 | 6.396** |
| Brief glance | CGLANCE1/2 | 0 | 0 | | 25.00 | 4.176* |
| Blank gaze | CBLANK1/2 | 0 | 0 | | 50.00 | 3.044 |
| Stares | CSTARES1/2 | 0 | 0 | | 16.67 | 0.186 |
| **B1b: Failure to develop peer relationships** | | | | | | |
| | DPEERIN1/2 | 0,-8 | 0,1 | Remove "-8" and add code "1" | 77.78 | 12.248*** |
| | DINPEER1/2 | 0,-8 | 0,1,-8 | Add code "1" | 83.33 | 14.18*** |
| | DQUALIN1/2 | 0 | 0,-8 | Add code "-8" | 88.89 | 30.106*** |
| | DEMPEER1/2 | 0 | 0,-8 | Add code "-8" | 77.78 | 28.098*** |

| | | | | | | |
|---|---|---|---|---|---|---|
| | DPEERAD1/2 | 0 | 0 | | 11.11 | 0.361 |
| | DFRIEND1/2 | 0 | 0,1,-8 | Add code "-8" and "1" | 97.22 | 13.752*** |
| | DQUALFR1/2 | 0 | 0,1 | Add code "1" | 13.89 | 0.066 |
| | DTURNS1/2 | 0,-8 | 0,-8 | | 72.22 | 6.281** |
| | DTEAM1/2 | 0,-8 | 0 | Remove "-8" code | 36.11 | 0.822 |
| | DOLDCH1/2 | 0 | 0 | | 11.11 | 0.444 |
| **B1c: lack of socio-emotional reciprocity** | | | | | | |
| Emotionally expressive gestures | AEMOTG | 0 | 0 | | 50.00 | 12.585*** |
| Greeting parents | CGREETP1/2 | 0 | 0 | | 36.11 | 8.433** |
| Greeting others | CGREETO1/2 | 0 | REMOVED | Remove item | | |
| Response to visitors | CREPVIS1/2 | 0,1 | 0,1 | | 69.44 | 10.884*** |
| Mechanical aids (adults) | CTOOL1/2 | 0 | 0 | | 47.22 | 4.399* |
| Comfort when hurt | CPAINR1/2 | 0,1,-8 | 0,1 | Remove "-8" code | 72.22 | 14.357*** |
| Social comfort | CCOMFOT1/2 | 0,1,-8 | REMOVED | Remove item | | |
| Giving comfort | CGIVEC1/2 | 0 | 0 | | 77.78 | 25.328*** |
| One sided approaches | CONESIS1/2 | 0,-8 | 0,-8 | | 61.11 | 24.757*** |
| Aware of feelings | CEMPTAH1/2 | 0,1 | 0,1 | | 88.89 | 20.664*** |
| Laughs at distress | CLAUGHX1/2 | 0 | 0 | | 33.33 | 9.655** |
| Response to others injury | CRESINJ1/2 | 0 | REMOVED | Remove item | | |
| "intellectual" reaction to injury | CREANJ1/2 | 0 | REMOVED | Remove item | | |
| Behaviour in public places | BEHAPUB1/2 | 0 | 0 | | 52.78 | 3.861* |
| Personal modesty | MODEST1/2 | 0 | 0 | | 47.22 | 2.321 |
| Psychological barriers | PSYCHOB1/2 | 0 | 0 | | 55.56 | 7.528** |
| Approaching strangers | TALKSR1/2 | 0 | 0 | | 22.22 | 0.118 |
| Embarrassing remarks | REMARK1/2 | 0 | 0 | | 11.11 | 3.663 |
| Interrupts conversations | INTERP1/2 | 0 | 0 | | 33.33 | 1.656 |
| Paradoxical responses | PARADX1/2 | 0 | 0 | | 33.33 | 9.655** |
| B1d: lack of shared enjoyment | | | | | | |
| React to happiness | CSHAPY1/2 | 0,1 | 0,1 | | 38.89 | 5.725* |
| Sharing interests | CSHAREP1/2 | 0,1 | 0,1 | | 88.89 | 13.048*** |
| **B2: Communication impairment** | | | | | | |
| **B2a: lack of or delay in spoken language and failure to compensate through gesture** | | | | | | |
| Development of expressive language | ADEVEXPR | 00,01 | 0,1 | | 11.11 | 1.5 |
| No NVC | CNVCOM1/2 | 0,1 | 0,1 | | 22.22 | 0.01 |
| **B2b: Relative failure to initiate or sustain two way conversation** | | | | | | |
| Reciprocal communication | CRECIPR1/2 | 0,1 | QUALCOM 2,3 | Item changed to summary | 55.56 | 1.895 |

| | | | | | | |
|---|---|---|---|---|---|---|
| | | | | (Part 7 item). Codes for crecipr changed to "1" to reflect Qualcom "2" and "3" codes. | | |
| **B2c: Stereotyped, repetitive or idiosyncratic language** | | | | | | |
| Immediate echolalia | CECHOIM1/2 | 0 | | | 44.44 | 1.692 |
| Delayed echolalia | CECHODE1/2 | 0 | | | 61.11 | 21.647*** |
| Pronoun reversal | CPRONOU1/2 | 0 | | | 38.89 | 0.083 |
| Idiosyncratic | CIDIOSY1/2 | 0 | | | 13.89 | 2.323 |
| Long winded | CPEDAN1/2 | 0 | | | 8.33 | 2.704 |
| Content of speech | CSPCONT1/2 | 0 | | | 22.22 | 0.001 |
| Repetitive questions | QUESREP1/2 | 0 | | | 36.11 | 1.456 |
| Repetitive themes | THEMREP1/2 | 0 | | | 25.00 | 4.176* |
| **B2d: Lack of varied spontaneous make believe or social imaginative play** | | | | | | |
| Imitation of social/domestic activities | ADOMIMI | 0,1,2,-8 | 0,1,2,-8 | | 30.56 | 1.104 |
| Imaginative activities | AIMAGACT | 00,01 | 0,1,2, | Include "2" code | 44.44 | 6.206** |
| Absent or repetitive | CPRETEN1/2 | 0 | 0,-8 | Include "-8" code | 88.89 | 3.663 |
| Copying imaginative activities | COPYIM1/2 | New | 0,-8 | CREATED – an individual has to score above 3 on imaginative activities and a marked score (0) on acting out roles. | 11.11 | 11.392*** |
| **B3: repetitive activities** | | | | | | |
| **B3a: Encompassing preoccupation or circumscribed pattern of interest** | | | | | | |
| Clings to objects | OBJCLG1/2 | 0 | 0 | | 30.56 | 0.539 |
| Collects objects | OBJCOL1/2 | 0 | 0 | | 25.00 | 6.219* |
| Fascination with objects | OBJFAS1/2 | 0 | 0 | | 69.44 | 7.727** |
| Sameness of environment | SAMENV1/2 | 0 | 0 | | 44.44 | 9.91** |
| Wants perfection | PERF1/2 | 0 | 0 | | 30.56 | 2.987 |
| Limited pattern of activities | LTDACT1/2 | 0 | REMOVED | Remove item | | |
| Fascination with violence | CRESINJ1/2 | New item | 0 | New addition to DISCO 11. No relevant item in DISCO 9. | | |
| **B3b: apparently compulsive adherence to non-functional routines or rituals** | | | | | | |
| Arranging objects | OBJARR1/2 | 0 | 0 | | 61.11 | 18.838* |
| Repetitive actions with objects | OBJREP1/2 | 0 | 0 | | 25.00 | 0.793*** |

| | | | | | | |
|---|---|---|---|---|---|---|
| Eats only few foods | FADDY1/2 | 0 | 0 | | 27.78 | 5.153* |
| Routes, bedtime, etc. | SAMERT1/2 | 0 | 0 | | 52.78 | 7.955** |
| Acting out roles | ACTROLE1/2 | 0 | 0 | | 13.89 | 4.653* |
| Repetitive special skills | ACTSPSK1/2 | 0 | 0 | | 30.56 | 8.462** |
| Amasses facts | FACTCOL1/2 | 0 | 0 | | 11.11 | 3.663 |
| Watches same videos etc | TVFAS1/2 | 0 | 0 | | 66.67 | 7.89** |
| Other repetitive routines | ROUTOTH1/2 | 0 | 0 | | 2.78 | 0.012 |
| **B3c: Stereotyped and repetitive motor mannerisms** | | | | | | |
| Hand and arm flapping | HANDUN1/2 | 0 | 0 | | 47.22 | 7.302** |
| Self spinning | SPIN1/2 | 0 | 0 | | 30.56 | 0.539 |
| Standing rocking | ROCKUP1/2 | 0 | 0 | | 8.33 | 2.704** |
| Complex moves (other) | MOVEM1/2 | 0 | 0 | | 22.22 | 7.823 |
| **B3d: Preoccupations with part objects or non-functional elements of materials** | | | | | | |
| Smelling | SMELL1/2 | 0 | 0 | | 22.22 | 7.823** |
| Touching | TOUCH1/2 | 0 | 0 | | 38.89 | 4.247 |
| Aimless manipulation | MANIP1/2 | 0 | 0 | | 30.56 | 6.188** |
| Fascination with sound | AUDFAS1/2 | 0 | 0 | | 11.11 | 0.444 |
| Bright lights, shiny | LIGHTS1/2 | 0 | 0 | | 19.44 | 2.418 |
| Watching spinning | SPINVIS1/2 | 0 | 0 | | 19.44 | 2.418 |
| Twist objects near eyes | TWISTH1/2 | 0 | 0 | | 22.22 | 5.169* |
| Inspecting angles | ANGLES1/2 | 0 | 0 | | 33.33 | 9.655** |
| Parts of objects | OBJPTS1/2 | 0 | 0 | | 16.67 | 1.653 |
| Abstract properties | OBJABS1/2 | 0 | 0 | | 27.78 | 5.153* |

## *Kanner and Eisenberg's Early Infantile Autism*

| **Variable** | **Label** | **DISCO 9** | **DISCO 11** | **Changes required?** | **Frequency in ASD group (%)** | **Chi-square statistic** |
|---|---|---|---|---|---|---|
| **A. Social aloofness** | | | | | | |
| Quality of social interaction | QUALSOC1/2 | 0,1,2 | 0,1,2 | | 69.44 | |
| **B. Elaborate repetitive routines** | | | | | | |
| Nature of chosen activity | LTDACT1/2 | 0,1 | 0,1 | | 58.33 | 12.494*** |
| Clinging to objects | OBJCLG1/2 | 0 | 0 | | 30.56 | 0.539 |
| Collecting objects | OBJCOL1/2 | 0 | 0 | | 25.00 | 6.219** |
| Fascination with objects | OBJFAS1/2 | 0 | 0 | | 69.44 | 7.727** |
| Arranging objects | OBJARR1/2 | 0 | 0 | | 61.11 | 18.838*** |
| Interest in parts of objects | OBJPTS1/2 | 0 | 0 | | 16.67 | 1.653 |
| Repetitive action with objects | OBJREP1/2 | 0 | 0 | | 25.00 | 0.793 |
| Abstract properties of objects | OBJABS1/2 | 0 | 0 | | 27.78 | 5.153* |
| Sameness of environment | SAMENV1/2 | 0 | 0 | | 44.44 | 9.91** |
| Insistence on perfection | PERF1/2 | 0 | 0 | | 30.56 | 2.987 |
| Sameness of routine | SAMERT1/2 | 0 | 0 | | 52.78 | 7.955** |
| Other repetitive routines | ROUTOTH1/2 | New addition | 0 | Add "routoth" | 2.78 | 0.012 |

## *Gillberg's Asperger Syndrome Criteria for DISCO 11.*

| Variable | Label | Codes | Changes | Frequency in ASD group (%) | Chi-square statistic |
|---|---|---|---|---|---|
| **1: Social Impairment** | | | | | |
| **1a: Considerable difficulties interacting with peers** | | | | | |
| Quality of interaction with peers | DQUALIN1/2 | 0,1 | | 47.22 | 0.188 |
| Convention of interaction | DHOST1/2 | 0 | | 27.78 | 1.301 |
| Mechanical use of peers | DPEERAD1/2 | 0 | | 11.11 | 0.361 |
| Friendship with age peers | DFRIEND1/2 | 1 | | 5.56 | 1.103 |
| Quality of friendship | DQUALFR1/2 | 0,1 | | 13.89 | 0.066 |
| Bullied by peers | DTEASE1/2 | 0 | | 22.22 | 0.408 |
| **1b: lack of interest in making friends of interacting with peers** | | | | | |
| Avoids age peers | DPEERIN1/2 | 0,1 | | 77.78 | 12.248*** |
| Interaction with peers | DINPEER1/2 | 0,-8 | | 66.67 | 15.078*** |
| Friendship with age peers | DFRIEND1/2 | 0 | | 8.33 | 2.704 |
| **1C: Problems appreciating social cues** | | | | | |
| Quality of communication | QUALCOM1/2 | 2,3 | DISCO 9 "crecipr" code "1" to cover "qualcom" "2" and "3" | 55.56 | 1.895 |
| Unaware of own identity | CIDENT1/2 | 0 | New item, no DISCO 9 equivalent | | |
| Giving comfort | CGIVEC1/2 | 0,1 | | 86.11 | 27.386*** |
| Unaware of others' feelings | CEMPATH1/2 | 0,1 | | 88.89 | 20.664*** |
| Reaction to others' happiness | CSHAPY1/2 | 0 | | 36.11 | 6.403** |
| **1d: socially or emotionally inappropriate behaviour** | | | | | |
| Fascination with violence | CRESINJ1/2 | 0 | New item, no DISCO 9 equivalent | | |
| Anger towards parents | ANGERP1/2 | 0 | | 0.00 | / |
| Harassment of others | HARAS1/2 | 0 | New item, no DISCO 9 equivalent | | |
| Emotional response to age peers | CEMPEER1/2 | 0 | | 22.22 | 3.263 |
| Approaching strangers | TALKSR1/2 | 0 | | 22.22 | 0.118 |
| Inappropriate response | PARADX1/2 | 0 | | 33.33 | 9.655** |
| Quality of interaction | QUALSOC1/2 | 3,4,5 | | 30.56 | 6.188* |
| **2. Narrow interests** | | | | | |
| Special skills | ACTSPSK1/2 | 0 | | 30.56 | 8.462** |
| Collecting facts | FACTCOL1/2 | 0 | | 11.11 | 3.663 |
| Repetitive themes | THEMREP1/2 | 0 | | 25.00 | 4.176* |
| Intense interest in a person | PERS1/2 | 0 | New item, no DISCO 9 equivalent | | |

| | | | | | |
|---|---|---|---|---|---|
| **3. Repetitive routines** | | | | | |
| Acting out roles | CTROL1/2 | 0 | | 13.89 | 4.653* |
| Sameness of environment | SAMENV1/2 | 0 | | 44.44 | 9.91** |
| Insistence of perfection | PERF1/2 | 0 | | 30.56 | 2.987 |
| Sameness of routine | SAMERT1/2 | 0 | | 52.78 | 7.955** |
| Food fads | FADDY1/2 | 0 | | 27.78 | 5.153* |
| Clinging to home | CLINGH1/2 | 0 | | 11.11 | 1.5 |
| Repetitive questions | QUESREP1/2 | 0 | | 36.11 | 1.456 |
| Other repetitive routines | ROUTOTH1/2 | 0 | | 2.78 | 0.012 |
| **4. speech and language peculiarities** | | | | | |
| Development of expressive language | ADEVEXPR | 8,9 | | 36.11 | 0.11 |
| Appreciation of humour | AHUMOUR | 1,2,3 | | 86.11 | 0.014 |
| Literal understanding | CLITUND1/2 | 0 | | 25.00 | 2.66 |
| Long winded, pedantic | CPEDAN1/2 | 0 | | 8.33 | 2.704 |
| Tone of voice | CTONE1/2 | 0 | | 33.33 | 5.364* |
| **5. Non-verbal communication problems** | | | | | |
| Facial expression | CFACEXP1/2 | 0,-8 | | 44.44 | 2.517 |
| Body language | CUSENVC1/2 | 0,-8 | | 63.89 | 8.111** |
| Stares | CSTARES1/2 | 0,1 | | 36.11 | 0.378 |
| **6. Motor clumsiness** | | | | | |
| Clumsiness | CCLUMSY1/2 | 0 | | 36.11 | 2.363 |
| Immature gait | CGAIT1/2 | 0 | | 36.11 | 0.238 |
| Poor at games, PE | CCOORD1/2 | 0,1 | | 47.22 | 1.55 |
| Hand eye co-ordination | AHEYE | 0,1,2,3,4,5 | | 66.67 | 0.143 |
| Clumsy fine motor | CFINEMT1/2 | 0 | | 25.00 | 6.245*# |
| Abnormal walking | WALKPRO1/2 | 0 | | 19.44 | 4.167* |
| #more frequent in control group | | | | | |

## *Wing and Gould's ASD and social impairment*

| Algorithm item | Changes in variable name or coding | | Change to text? | Changes needed for study? |
|---|---|---|---|---|
| | DISCO 9 | DISCO 11 | | |
| Quality of social interaction | QUALSOC 0-5 | QUALSOC 0-5 | / | / |
| Reciprocal communication | CERICPR1/2 0,1,8 | QUALCOM1/2 0,1,2,3 | | QUALCOM "0" = CRECIPR "8"<br>QUALCOM "1" = CRECIPR "0"<br>Need to find equivalent items for QUALCOM "2" and "3" |
| Imaginative activities | AIMAGACT – 00, 01 | QUALIM 0,1,2,3,4 | | The items from DISCO 9 do not fully cover the information provided by DISCO 11. |
| Repetitive play | CPRENTEN1/2 – 0,1 | | | |
| Limited pattern | LTDACT1/2 – 0,1,8 | QUALACT | / | / |

An attempt was made to replicate the DISCO 11 item quality of imagination in order to update the algorithm (see below), however, as shown in the results section, this was not successful

and therefore the decision was made to retain the DISCO 9 algorithm with the current data when measuring Wing and Gould's ASD.

| DISCO 9 WG-ASD Imagination | DISCO 9 WG-ASD Imagination |
|---|---|
| Imagination limited to imitation e.g.:<br>"imitates sounds and simple movements made by other"<br>"plays with real household equipment using it for its real purpose"<br>"Holds doll, toy animals as if real, some of the time"<br>OR<br>Limited imagination e.g.<br>"Engages in role play alone or just copies others or videos"<br>"repetition in pretend play" | Quality of imagination |

### Updating the algorithms for DISCO 11: The results

Data from Sample 1 was used to run the DISCO 9 and DISCO 11 algorithms. The number of individuals from each diagnostic sub-group who meet criteria for each diagnostic algorithm can be seen in the table below. Statistics were conducted in comparison with the ASD and clinical control groups only (17 ID and 14 LI), the TD group was removed from the analyses to provide a stricter test of each algorithms ability to discriminate between ASD and clinical conditions. In all cases, except Wing and Gould's ASD, the updated algorithms performed equivalently or better than the previous DISCO 9 version (see AUCs). Updating the ICD-10/DSM-IV algorithm for DISCO 11 improved the specificity of the algorithm, which was weak beforehand.

| | | Ever | | Current | |
|---|---|---|---|---|---|
| | | DISCO 9 | DISCO 11 | DISCO 9 | DISCO 11 |
| ICD-10 Childhood Autism | Autism -70 | 18 | 18 | 16 | 16 |
| | autism +70 | 18 | 18 | 16 | 17 |
| | learning | 10 | 8 | 7 | 7 |
| | language | 4 | 4 | 2 | 2 |
| | AUC | .774 | .806 | .799 | .813 |
| Wing and Gould ASD | Autism -70 | 16 | 17 | 13 | 12 |
| | autism +70 | 17 | 16 | 13 | 12 |
| | learning | 4 | 4 | 2 | 2 |
| | language | 0 | 0 | 0 | 0 |
| | AUC | .894 | .894 | .829 | .801 |
| Gillberg's Asperger criteria | Autism -70 | 1 | 1 | 2 | 0 |
| | autism +70 | 8 | 9 | 4 | 8 |
| | learning | 1 | 0 | 0 | 0 |
| | language | 0 | 2 | 0 | 0 |
| | AUC | .609 | .607 | .583 | .611 |
| Kanner and Eisenberg's Early Infantile Autism | Autism -70 | 11 | 11 | 4 | 5 |
| | autism +70 | 5 | 5 | 1 | 1 |
| | learning | 1 | 1 | 1 | 1 |
| | language | 0 | 0 | 0 | 0 |
| | AUC | .706 | .706 | .567 | .567 |

# APPENDIX 3: MEASURING THE ASSOCIATED FEATURES OF ASD USING THE DISCO

This appendix presents the preliminary analyses of the items measured by the DISCO that do not belong to the diagnostic algorithms and instead capture the many associated features of ASD and are used in clinical use of the DISCO to create a clinical picture of the individual's strengths, weaknesses and pattern of behaviours. The DISCO covers a large range of such items, however, varying numbers of items are used to capture each section and with the exception of the sensory processing items, none of these associated features as measured by the DISCO have been directly tested for reliability and validity in research. The first step in using these DISCO items to address conceptual or research questions is to assess the reliability of these scales of behaviours to accurately and comprehensively measure the behaviour in question, as detailed in the following chapter.

**Aim of analyses – to describe the measurement of DISCO associated features, are there reliable scales and how do individuals with ASD score on these measures of behaviour compared to individuals with other clinical conditions or typical development as well as to investigate the role of age and IQ.**

This aim was measured in two separate analyses. The first assessed the frequency of all associated items across individuals with ASD, individuals with clinical diagnoses other than ASD and individuals with typical development. The second set of analyses assessed the reliability of the scales of associated items as they are arranged in the DISCO: maladaptive behaviours, daily living skills, motor behaviours, sensory behaviours, pattern of activities, sleep disturbances and emotion behaviours.

A four stage process was endorsed in which items had to: meet acceptable levels of inter-rater reliability (above .6; taken from Wing et al., 2002); be endorsed by at least a minority of individuals with ASD (at least 10%); contribute to the internal consistency of the scale; and correlate with the total scale. Items with item-total correlation below .30 were removed from the scale as were items whose removal would improve the overall Cronbach's alpha by greater than 0.5.

**Method**

*Participants:* This study will utilise data from both Sample 1 (82 cases) as previously described in the methods section.

*Measures*: The items for each "associated" scale were taken directly from the DISCO sub-sections. As described in the methods section, there are three types of DISCO items, for each associated behaviour domain, all the "untypical behaviour" and "developmental stages" items were selected for analysis.

*Scoring of items:* The majority of items selected to measure associated behaviours are "untypical" behaviour items. These untypical behaviours are coded on a three-scale severity score: marked problem, minor problem, or no problem, coding was reversed here from the codes described in the methods section: marked = 3; minor problem = 2 and no problem = 1, so that a higher score indicates more problems for ease of interpretation. Previous work with DISCO items, with diagnostic algorithms and sensory behaviours have, instead, assigned a code of one to any item rated as marked and 0 to "minor" or "no problem" codes (e.g. Leekam et al., 2007), however, the level of coding selected here provides a wider level of variability to both each individual item and to the total scale scores, which a larger range of analyses (including multivariate designs) to be applied to the data. A minority of the items selected to measure associated behaviours, mainly those selected to measure Daily Living Skills and Motor behaviours, are "developmental stages" items. These items were recoded as follows, however, a selection of these items were coded as "age in months when skill achieved" and therefore has to be recoded as delayed or typical depending on the developmental milestones set out in the DISCO, the recodes for this specific items can be seen in Table 1.

**Table 1: Table showing the recode of developmental items for the motor and daily living skills associated scales in the DISCO**

| | Independent sitting | Walking | Riding Tricycle | Clean and dry in day | Clean and dry at night |
|---|---|---|---|---|---|
| Generic codes | | | | | |
| 555 "early" | 1 | 1 | 1 | 1 | 1 |
| 666 "average" | 1 | 1 | 1 | 1 | 1 |
| 777 "late" | 3 | 3 | 3 | 3 | 3 |
| Scored as 3 (late) when recorded in months | <12 | <24 | <48 | <36 | <48 |

Finally, two items from the overall pattern of activities sub-section, "time attends to own interests" and "time attends to tasks others give" are coded according to a five point scale: "no self-chosen activities" and "attention fleeting" were recoded to 3 (marked), "engages in such activities for less than 15 minutes and then needs attention" to 2 (minor problem) and "engages in such activities for 15 minutes or more" plus "Engages for an hour or more" to 1 (no problem).

*Ever or current:* Differences between diagnostic groups on separate items were run for both ever and current scores; the ever scores are reported, however, if there were any differences when the comparisons were run with current this will be made clear in the text. In contrast, it was essential to use the current ratings for the analyses in Sample 2 in order to allow accurate and direct comparisons with the individuals' current age and level of ability.

*Analytic Strategy:* Data analysis in this chapter will be conducted in two parts. Firstly, the frequency of all of the items measured by the DISCO for each associated scale (sensory, maladaptive, emotion, motor, daily living skills and pattern of activities) will be compared across diagnostic group. Secondly, the reliability of the items in each scale will be assessed using the criteria in Part 2. Further analyses will be conducted on the differences across total associated features for each scale will be examined between the five groups of individuals in Sample 1. In contrast to analyses conducted on the individual items (step 2) analysis of the total scale score will test for differences across individuals with ASD (both the high and low functioning groups), language impairments, intellectual disability and typical development.

a. Low functioning ASD compared to the low functioning comparison group of individuals with intellectual disability.
b. High functioning ASD individuals compared to the high functioning comparison group of individuals with a language impairment
c. High functioning ASD individuals compared to another high functioning comparison group of typically developing individuals

*Data Screening:* Multivariate non-parametric analyses were used for all analyses. These consisted of Kruskall-Wallis tests to first identify if individuals from the different diagnostic groups differed on each item overall (when more than two grouping options) or primary Mann-Whitney U tests for two group comparisons.

There are two methods to following up a significant Kruskall-Wallis test (Field, 2009), the first is to run a series of Mann-Whitney U tests on each pairwise grouping. When a specific direction of effect is expected an alternative is to run a Jonckheere-Terpstra trend test (J-T test). Both methods are equal and can both be used to compare groups aslong as, when using multiple Mann Whitney U tests, corrections are made to the p value to avoid making a type I error through multiple comparisons. Therefore, for the key analyses using the total scale scores Mann-Whitney U tests will be used to confirm the significant differences between each diagnostic sub-group, however, for the individual items in each scale Kruskall-Wallis tests will be supplemented with Jonckheere test in order to be both efficient and to give an overview of the pattern of the data, which will also be

clearly displayed in the presented figures. For all sets of tests, exact significant values were used where processing speed allowed or Monte Carlo exact tests for complex tests to be as accurate as possible. Exact tests are recommended when samples are small (Sample 1) or when data is especially non-normal (Field, 2009), the exact tests are the most reliable but Monte Carlo using the distribution of the sample applied to additional samples (10,000) to estimate the exact values and is therefore still reliable.

**Maladaptive Scale:**

All tests are reported at p<.01 to control for the multiple comparisons with these data. Kruskall-Wallis tests revealed a significant difference across the three groups and follow up Jonckheere trend tests revealed a significant trend with higher scores (more marked behaviour) being reported for individuals with ASD, then clinical comparison individuals followed by the least being reported for typically developing individuals for 13 items as shown in Table 2. The same pattern was found across the "current" measures except current destructive activities was no longer significant (H(2) = 8.364, p=.012, n.s.) but current temper tantrums was (H(2) = 11.308, p<.01).

**Table 2: Table presenting the results of Kruskall-Wallis tests and follow up Jonckheere tests for items labelled as maladaptive behaviours in the DISCO**

| Item | Code | Kruskall-Wallis test (H), df=2 | Jonckheere trend test (J) | Z score | Power (r) |
|---|---|---|---|---|---|
| Wandering | Wander | 27.45** | 514.50 | -5.23 | 0.58 |
| Destructiveness | Destruc | 11.93* | 746.50 | -3.44 | 0.38 |
| Noisiness | Noise | 12.44* | 706.50 | -3.54 | 0.39 |
| Temper Tantrums | Temper | 4.805 | | | |
| Physical aggression | Aggress | 16.22** | 643.50 | -4.02 | 0.44 |
| Anger toward parents | Angerp | | | | |
| Behaviour in public places | Behapub | 14.43** | 674.50 | -3.67 | 0.40 |
| Personal modesty | Modest | 17.67** | 494 | -4.14 | 0.46 |
| Psychological barriers | Psychob | 22.53** | 468.50 | -4.72 | 0.52 |
| Approaching strangers | Talksr | 3.256 | | | |
| Embarrassing remarks in public | Remark | 8.444 | | | |
| Interrupting conversations | Interp | 9.78* | 158.50 | -2.85 | 0.31 |
| Inappropriate response to others' emotions | Paradox | 18.88** | 628.50 | -4.41 | 0.49 |
| Difficult or objectionable personal habits | Habits | 15.77** | 687.50 | -3.98 | 0.44 |
| Scatters or throws objects around | Scatter | 15.53** | 685.50 | -3.92 | 0.43 |
| Lack of co-operation | Lackcop | 14.61** | 665.50 | -3.79 | 0.42 |
| Needs constant supervision | Superv | 19.44** | 667 | -3.70 | 0.41 |
| Demands carer's attention | Attseek | 6.463 | | | |
| Other | Behsoth | | | | |

**p<.001,* p<.01

*Reliability:* All items met acceptable levels of inter-rater reliability. "Other behavioural problems" and "anger at parents" were not endorsed by greater than 10% of the individuals with

ASD and were therefore removed from the reliability analysis. The remaining items were entered into the reliability analysis, the first run (Cronbach's alpha = .913) revealed one item ("temper") which had an item-total correlation of .269 and was therefore removed from the analysis as this was below the criterion of .30. This resulted in a scale of 16 items (as shown in Table 3 with an overall Cronbach's alpha of .918 (excellent).

**Table 3: Table showing the selected items for the Maladaptive behaviour scale of the DISCO**

| | Corrected Item-Total Correlation | Cronbach's Alpha if Item Deleted |
|---|---|---|
| Wandering | .695 | .911 |
| Destructiveness | .645 | .913 |
| Noisiness | .545 | .915 |
| Physical aggression | .610 | .914 |
| Behaviour in public places | .623 | .913 |
| Personal modesty | .643 | .913 |
| Psychological barriers | .752 | .909 |
| Approaching strangers | .685 | .912 |
| Embarrassing remarks in public | .753 | .909 |
| Interrupting conversations | .704 | .911 |
| Inappropriate response to others' emotions | .484 | .917 |
| Difficult or objectionable personal habits | .380 | .919 |
| Scatters or throws objects around | .568 | .915 |
| Lack of co-operation | .451 | .919 |
| Needs constant supervision | .649 | .912 |
| Demands carer's attention | .724 | .910 |

Finally, analyses were run for the total score on the maladaptive scale (sum of the 16 final maladaptive items) and differences tested across the five diagnostic groups (high and low functioning ASD, intellectual disability, language impairment and typical development). All results are reported at $p<.0125$ as corrected for the follow up comparisons of interest using bonferonni correction, all tests run using the Monte Carlo exact method. A Kruskall-Wallis test revealed significant differences across groups ($H(4)=37.92$, $p<.000$). Follow up Mann-Whitney tests revealed a significant differences between the HFA and both the LI ($U = 43.5$, $r = -0.56$, $p<.01$) and TD groups ($U = 0$, $r = -.85$, $p<.001$) as shown in Figure 1.

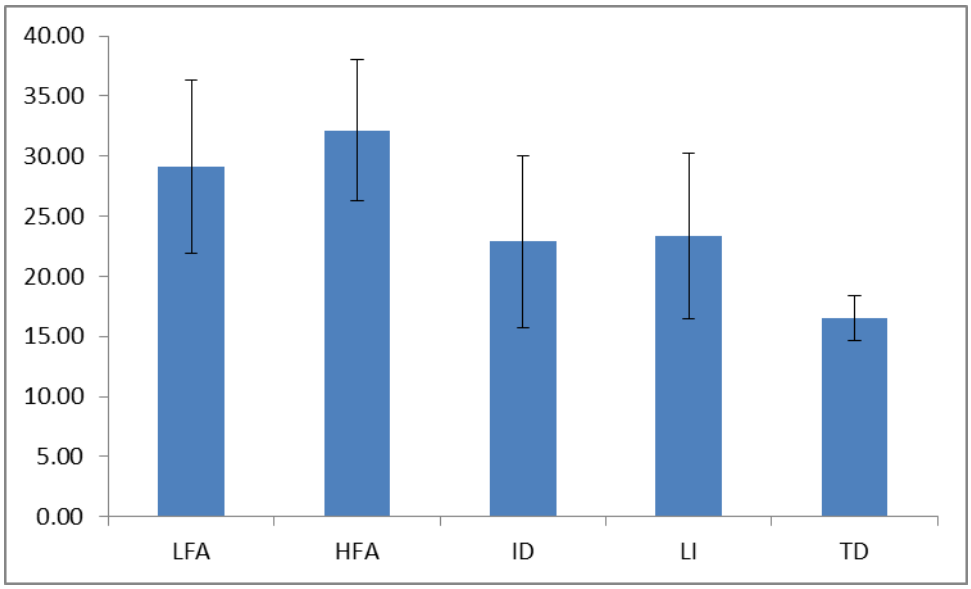



**Figure1: Figure showing the mean total maladaptive score across diagnostic sub-groups in Sample 1. Significant differences were found between the HFA group and the LI and TD groups.**

**Emotion Scale:** All tests are reported at p<.01. Kruskall-Wallis tests revealed a significant difference across the three groups with a significant trend for the most marked behaviours in ASD, followed by clinical and then typically developing individuals for items marked with a * in Table 4. The same pattern was found for current scores.

**Table 4: Table showing the results of Kruskall-Wallis tests and follow up Jonckheere tests for each item classified as an emotion item in the DISCO**

| Item | Code | Kruskall-Wallis test (H), df=2 | Jonckheere trend test (J) | Z score | Power (r) |
|---|---|---|---|---|---|
| Lack of emotional expression | Lackexp | 9.80* | 767 | -3.03 | -0.33 |
| Unhappiness, misery | Misery | 3.175 | | | |
| Changeable mood | Moodch | 0.872 | | | |
| Crying and moaning | Moan | 5.497 | | | |
| Laughing for no reason | Laugh | 25.55** | 570.5 | -4.99 | -0.55 |
| Puzzlement | Puzzle | 15.52** | 653 | -4.18 | -0.46 |
| Anxiety | Anxiety | 13.639** | 665 | -3.71 | -0.41 |
| Special fears | Fears | 8.202 | | | |
| other | Emototh | | | | |

**p<.001,* p<.01

*Reliability*: All items met acceptable levels of inter-rater reliability. "Other emotional problem" was not endorsed by any individuals with ASD and was therefore removed from the reliability analysis. All other items were entered into the analysis resulting in an overall Cronbach's alpha of .683, however, three items (misery, moodch and laugh) had item-total correlations below .3 and were therefore removed and the analysis was re-run. The final items included in the emotion scale are shown in Table 5. These five items had a total Cronbach's alpha of .697 (acceptable) and could not be improved further.

**Table 5: Table showing the selected items for the Emotion scale of the DISCO**

| | Corrected Item-Total Correlation | Cronbach's Alpha if Item Deleted |
|---|---|---|
| Lack of emotional expression | .443 | .652 |
| Crying and moaning | .413 | .664 |
| puzzlement | .592 | .588 |
| Anxiety | .490 | .631 |
| Special fears | .344 | .698 |

Finally, analyses were run for the total score on the emotion scale (sum of the five emotion items) and differences tested across diagnostic groups. All results are reported at .01 using the Monte Carlo level of significance. Analyses across the five diagnostic groupings revealed a significant Kruskall-wallis test (H(4)=26.136, p<.000). Follow up Mann-Whitney tests revealed the HFA group had significantly more emotion behaviours than the LI group (U = 52.5, r = -0.50, p<.01) and the TD group (U = 10, r = -0.79, p<.001) as shown in Figure 2.

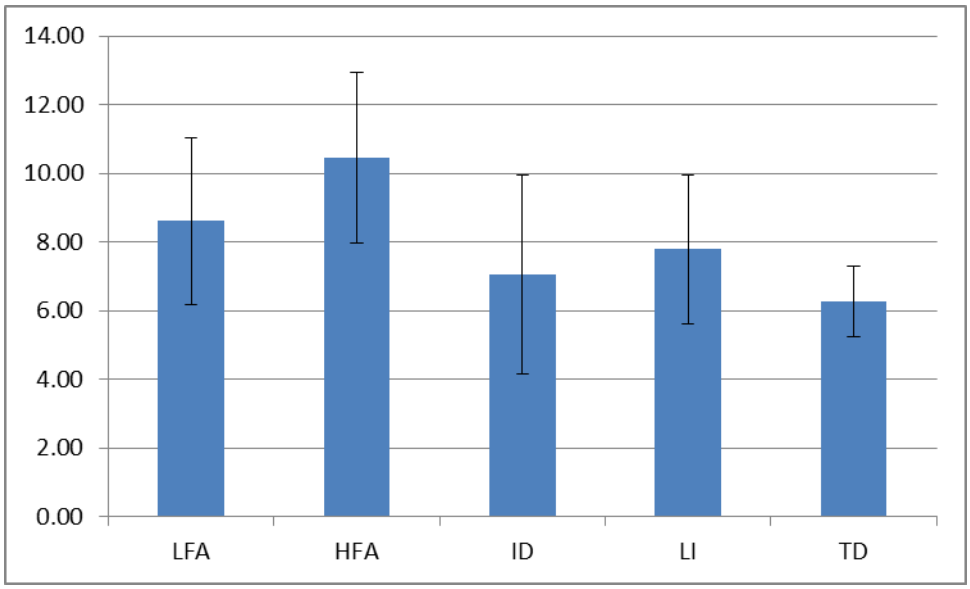



**Figure 2: Figure showing the mean total emotion score across diagnostic sub-groups in Sample 1. Significant differences were found between the HFA group and the LI and TD groups.**

**Sensory Scale:** Kruskall-Wallis tests were conducted to identify diagnostic group differences for each sensory item, significant differences across groups, with follow-up Jonckheere tests revealing significant trends for the most marked behaviours reported for the ASD group, then the clinical comparison group and then typically developing individuals were found for seven items in Table 6. Smelling objects or people was also significantly different across groups (H(2) = 16.1, p<.001) but the TD group had more behaviours than the clinical comparison group. The same pattern was found for the current scores except for current reaction to gentle touch (H(2) = 8.08, p=.015, n.s.) which was no longer significant once multiple comparisons were controlled for.

**Table 6: Table showing the Kruskall-Wallis test and follow up Jonckheere tests for items classified as sensory according to the DISCO**

| Item | Code | Kruskall-Wallis test (H), df=2 | Jonckheere trend test (J) | Z score | Power (r) |
|---|---|---|---|---|---|
| Smearing | Smears | 20.21** | 672.5 | -4.37 | -0.48 |
| Mouthing or swallowing of objects | Mouth | 6.28 | | | |
| Self injury | Sib | 6.571 | | | |
| Self stimulation without injury | Selfst | 7.836 | | | |
| Smelling objects or people | Smell | 16.109** | 814 | -3.00 | -0.33 |
| Touching objects | Touch | 13.26** | 715.5 | -3.42 | -0.38 |
| Scratching and tapping surfaces | Scratch | 8.064 | | | |
| Repetitive destructive activities | Repdest | 8.053 | | | |
| Repetitive, aimless manipulation of objects | Manip | 8.858 | | | |
| Being spun round | Spun | 6.729 | | | |
| Indifference to pain, heat, cold | Painind | 14.95** | 698.5 | -3.86 | -0.43 |
| Reaction to gentle touch | Gentle | 10.84* | 803.5 | -3.27 | -0.36 |
| Reaction to firm touch | Firm | 8.29 | | | |
| Overbreathing | Breath | 7.359 | | | |
| Other | Sensoth | 2.375 | | | |
| Distress caused by sounds | Audist | 4.469 | | | |
| Fascination with sounds | Audfas | 2.53 | | | |
| Acuteness of hearing | Hearac | 6.371 | | | |
| Bright lights and shiny objects | Lights | 3.437 | | | |
| Interest in watching things spin | Spinvis | 9.328 | | | |
| Twisting hands or objects near eyes | Twisht | 13.96** | 792.5 | -3.59 | -0.40 |
| Interest in studying angles | Angles | 11.79* | 732 | -3.76 | -0.42 |
| Self-spinning | Spin | 9.008 | | | |
| Food fads | Faddy | 6.754 | | | |
| Dislikes being washed etc | Cwshre | | | | |
| Dislikes having dirty, sticking hands | Cwshsti | | | | |
| Refuses food that is lumpy or hard to chew | Clump | | | | |

**p<.001,* p<.01

All items met acceptable levels of inter-rater reliability. Three of the Sensory items "other Sensory problems," "other auditory behaviours," and "other visual behaviours" were not reported in

any individuals with ASD were therefore removed from the reliability analysis. The first set of analyses revealed four items with inter-total correlation less than 0.3: mouth (.161), faddy (.210), cwshsti (.170) and clump (.221) and so these items were removed from the sensory scale. This resulted in a sensory scale of 22 items from the DISCO that can be seen in Table 7. This scale had an overall cronbach's alpha value of .859

**Table 7: Table showing the selected items for the Sensory scale of the DISCO**

| | Corrected Item-Total Correlation | Cronbach's Alpha if Item Deleted |
|---|---|---|
| Smearing | .414 | .854 |
| Self injury | .417 | .854 |
| Self stimulation without injury | .455 | .853 |
| Smelling objects or people | .561 | .850 |
| Touching objects | .443 | .853 |
| Scratching and tapping surfaces | .418 | .855 |
| Repetitive destructive activities | .460 | .852 |
| Repetitive, aimless manipulation of objects | .593 | .848 |
| Being spun round | .377 | .855 |
| Indifference to pain, heat, cold | .570 | .848 |
| Reaction to gentle touch | .387 | .855 |
| Reaction to firm touch | .420 | .854 |
| Overbreathing | .386 | .855 |
| Distress caused by sounds | .416 | .855 |
| Fascination with sounds | .407 | .855 |
| Acuteness of hearing | .400 | .855 |
| Bright lights and shiny objects | .343 | .856 |
| Interest in watching things spin | .445 | .853 |
| Twisting hands or objects near eyes | .466 | .853 |
| Interest in studying angles | .393 | .855 |
| Self-spinning | .491 | .851 |
| Dislikes being washed etc | .329 | .858 |

Again, analyses assessing the frequency of the total sensory scale score across the five diagnostic groups was performed (Figure 3). A Kruskall-Wallis test was significant ($H(4) = 43.01$, $p<.001$). The LFA group had significantly more sensory behaviours than the ID group ($U = 51$, $r = -0.57$, $p<.001$) and the HFA group had significantly more behaviours than the LI ($U = 18.5$, $r = -0.72$, $p<.001$) and TD group ($U = 3$, $r = -0.83$, $p<.001$).

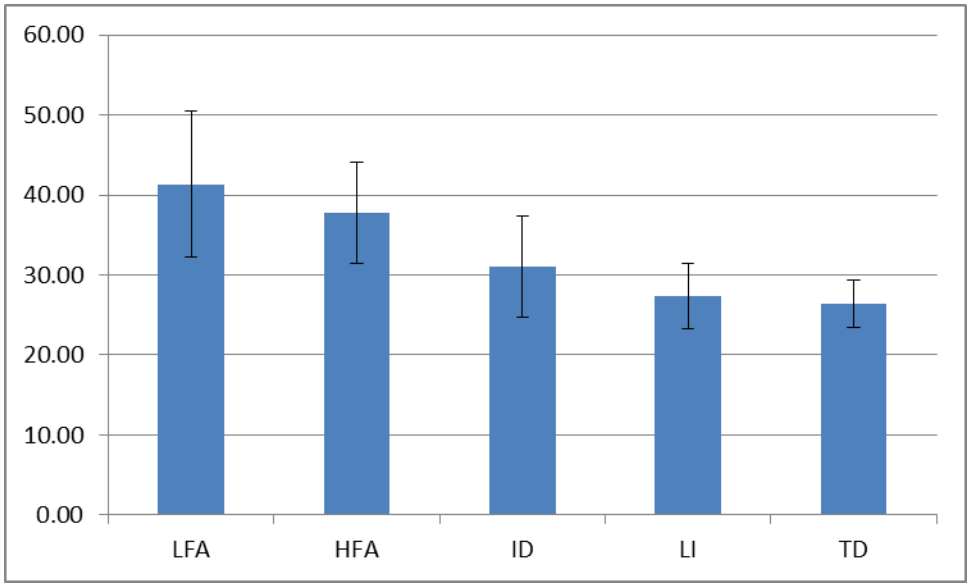



**Figure 3: Figure showing the mean total sensory score across diagnostic sub-groups in Sample 1. Significant differences were found between the LFA and ID group as well as between the HFA group and the LI and TD groups.**

### Motor Scale

The motor items with both significant group differences as demonstrated by a Kruskall Wallis test and a significant trend for more behaviours reported in the ASD then the clinical comparison group and more in the clinical comparison than typically developing group are reported in Table 8. Significant group differences were also found in other items, which did not follow the same trend: the clinical comparison group had the most delay learning to walk ($H(2) = 12.72$, $p<.001$) and learning to put together jigsaws ($H(2) = 16.27$, $p<.001$); they were more clumsy ($H(2) = 10.52$, $p<.01$); and had more fine motor difficulties ($H(2) = 16.94$, $p<.001$).

All items met acceptable levels of inter-rater reliability. "Other gross motor problems" and "other visuo-manual and spatial skills" were not endorsed by any of the ASD group, in addition, a delay in independent sitting was only present in 5.56% of the ASD group and reluctance to use hands in only 8.33%. These four items were therefore removed from the scale for reliability analyses. Removal of items with item-total correlations less than .3 resulted in six items being removed from the motor scale leaving five items to form a scale with a cronbach's alpha of .719 (items included are shown in Table 9).

**Table 8: Table showing the results of Kruskall-Wallis and follow up Jonckheere tests for all items classified as motor behaviours according to the DISCO**

| Item | Code | Kruskall-Wallis test (H), df=2 | Jonckheere trend test (J) | Z score | Power (r) |
|---|---|---|---|---|---|
| Independent sitting | Bsit | 8.161 | | | |
| Walking | Bwalk | 12.722 | | | |
| Riding tricycle | Btrike | 5.905 | | | |
| Climbing | Cclimb | 10.30* | 772 | -3.01 | -0.33 |
| Clumsiness | Cclumsy | 10.522 | | | |
| Immature gait when walking | Cgait | 9.804 | | | |
| Poor co-ordination in PE and games | Ccoord | 12.19** | 414.5 | -9.80 | -1.08 |
| Other | cmototh | 1.412 | | | |
| Jigsaws of 10 or more pieces | Bjigsaw | 16.721 | | | |
| Drawing recognisable objects/people | bdrawsy | 21.39** | 582 | -4.88 | -0.54 |
| Unusual dexterity | Cdesxter | 9.821 | | | |
| Clumsiness with fine motor tasks | Cfinemt | 16.935 | | | |
| Reluctance to use hands | Cnohand | 1.487 | | | |
| Reluctance to draw/use pencils | Cnodraw | 9.07* | 827 | -2.90 | -0.32 |
| Other | cvmsoth | 1.412 | | | |

**p<.001,* p<.01

**Table 9: Table showing the selected items for the Motor scale of the DISCO**

| | Corrected Item-Total Correlation | Cronbach's Alpha if Item Deleted |
|---|---|---|
| Immature gait when walking | .459 | .679 |
| Poor co-ordination in PE and games | .533 | .649 |
| Jigsaws of 10 or more pieces | .438 | .688 |
| Drawing recognisable objects/people | .438 | .687 |
| Clumsiness with fine motor tasks | .520 | .655 |

The best-attempt at creating a valid scale resulted in a grouping which was did not meet an acceptable level of reliability, however, for consistency analyses assessing the frequency of the total motor scale score across the three diagnostic groups was again performed. A Kruskall-Wallis test revealed a significant difference across the five groups ($H(4) = 26.136$, $p<.001$) the only significant difference was between the HFA and TD group ($U = 21$, $r = -0.73$, $p<.001$) using Mann-Whitney U follow up tests (Figure 4).

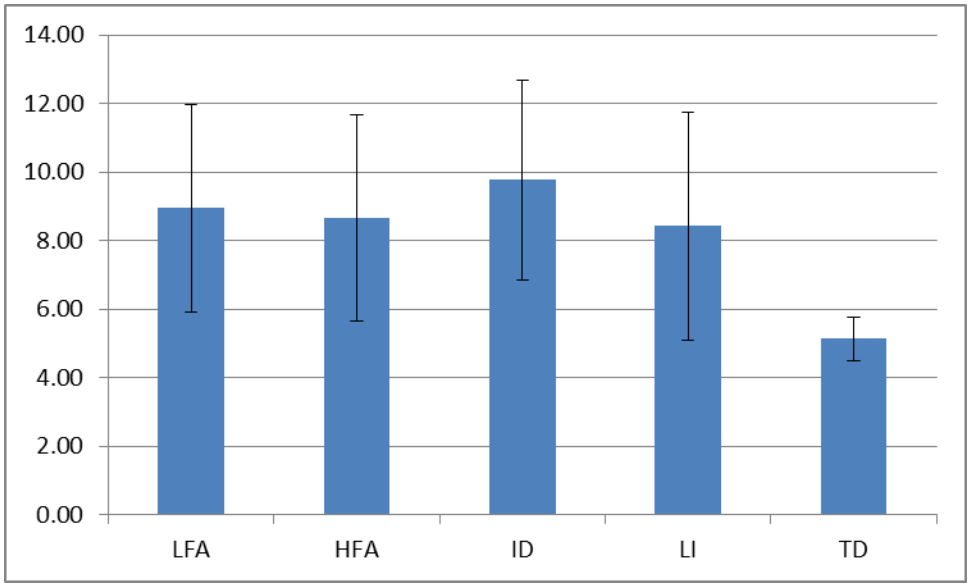



**Figure B: Figure showing the mean total motor score across diagnostic sub-groups in Sample 1. The only significant difference was between the HFA and TD group.**

**Daily Living Skills**

Eight daily living skill items had both significant group differences according to Kruskall-Wallis tests and significant Jochkerene trends indicating individuals with ASD had the most behaviours and TD the least (Table 10).

**Table 10: Table showing the Kruskall-Wallis and follow up Jonckheere tests for items classified as Daily living skills according to the DISCO**

| Item | Code | Kruskall-Wallis test (H), df=2 | Jonckheere trend test (J) | Z score | Power (r) |
|---|---|---|---|---|---|
| Clean and dry in day | Bdryday | 7.611 | | | |
| Clean and dry at night | Bdryngt | 15.60** | 744 | -3.10 | -0.34 |
| Resistance to using pot or lavatory | Btiolre | 12.50** | 575.5 | -3.30 | -0.36 |
| Smearing | Csmears | 14.73** | 744 | -3.76 | -0.42 |
| Constipation | Cretent | 3.99 | | | |
| Eating solid food | Bsolids | 4.02 | | | |
| Giving up bottle or breast | Bbottle | 1.622 | | | |
| Feeding self with spoon and fork | Beatself | 9.95* | 738.5 | -2.79 | -0.31 |
| Using knife and fork | Bknfork | 9.66* | 308 | -3.08 | -0.34 |
| Refuses food that is lumpy or hard to chew | Clump | 1.769 | | | |
| No interest in food | Coffood | 0.155 | | | |
| Excessive drinking of fluid | Cpolydi | 4.853 | | | |
| Refuses to feed self | Crefreed | 0.616 | | | |
| Other | Ceatoth | 4.629 | | | |
| Pulling pants down and up | Bpants | 12.715# | | | |
| Independent dressing, not buttons | Bdresind | 19.05** | 516 | -4.11 | -0.45 |
| Tying laces | Blaces | 10.8* | 284.5 | -2.86 | -0.32 |
| Willingness to dress self | Cdreself | 6.435 | | | |
| Slowness dressing | Cdrslow | 8.026 | | | |
| Awareness of suitability of clothing | Cdroth | 1.531 | | | |
| Dries hands without help | Bhands | | | | |
| Bathing and drying without physical | Bbathe | | | | |

| | |
|---|---|
| help | |
| Dislikes being washed etc | Cwshre |
| Willingness to wash etc | Cwshwil |
| Awareness of need for cleanliness | Cwshreq |
| Dislikes having dirty hands | Cwshsti |
| Fetching, carrying, taking simple messages | Bfetch |
| Completing a simple task without supervision | Btaskal |
| Going in garden alone | Bgarden |
| Going to local shop on own | Bshops |
| Could be left alone at home for half day | Bhomeal |
| Lack of common sense | Csense |

**p<.001,* p<.01

All items met acceptable levels of inter-rater reliability. Four items were not rated as marked for any individuals with ASD (other toileting problems, awareness of suitability of clothing, other washing problems, other independence) as well as only 5.56% of individuals with ASD scoring marked on giving up bottle or breast, 2.78% on other dressing problems and 2.78% on home alone. These items were therefore removed from the analysis. The reliability analysis for daily living skills went through three stages with four items being removed at the first stage (cronbach's alpha = .954) due to item-total correlations being below .3 and an additional one item in the second stage (cronbach's alpha = .952). The final set of items can be seen in Table 11 and this set resulted in an excellent reliable scale with a cronbach's alpha of .956.

**Table 11: Table showing the selected items for the Daily Living Skills scale of the DISCO**

| | Corrected Item-Total Correlation | Cronbach's Alpha if Item Deleted |
|---|---|---|
| Clean and dry at night | .702 | .953 |
| Resistance to using pot or lavatory | .473 | .956 |
| Constipation | .804 | .952 |
| Feeding self with spoon and fork | .311 | .958 |
| Using knife and fork | .408 | .957 |
| Refuses food that is lumpy or hard to chew | .526 | .955 |
| Excessive drinking of fluid | .706 | .954 |
| Other feeding | .770 | .953 |
| Pulling pants down and up | .777 | .954 |
| Independent dressing, not buttons | .742 | .953 |
| Tying laces | .801 | .952 |
| Willingness to dress self | .724 | .953 |
| Slowness dressing | .473 | .957 |
| Dries hands without help | .807 | .952 |
| Bathing and drying without physical help | .889 | .951 |
| Willingness to wash etc | .764 | .953 |
| Awareness of need for cleanliness | .717 | .953 |
| Dislikes having dirty hands | .505 | .956 |
| Fetching, carrying, taking simple messages | .787 | .952 |
| Completing a simple task without supervision | .859 | .952 |
| Going in garden alone | .777 | .954 |
| Going to local shop on own | .787 | .952 |
| Lack of common sense | .942 | .951 |

The total score for daily living skills was again compared across diagnostic sub-groups. The Kruskall-Wallis test revealed a significant difference across groups (H(4) = 20.29,p<.001) with follow up Mann-Whitney tests revealing that the only significant difference of interest was between the HFA and TD group (U = 36.5, r = -0.62, p<.001) as shown in Figure 5.

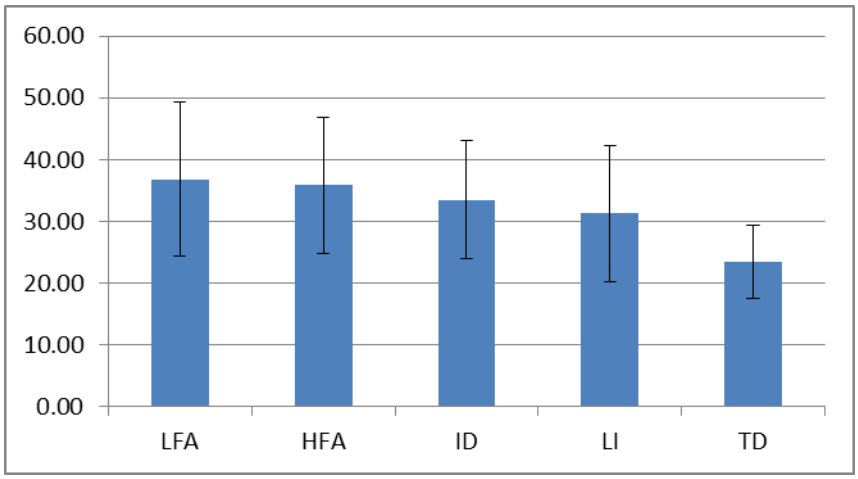



**Figure5: Figure showing the mean total daily living skills score across diagnostic sub-groups in Sample 1. The only significant difference was between the HFA and TD group.**

**Pattern of activities**

Significant group differences were seen across four of the pattern of activities items using Kruskall-Wallis tests and these items (Table 12) had significant trends (according to Jockhenere statistic) of the most problems for ASD and least for TD.

**Table 12: Table showing the Kruskall-Wallis and follow up Jockheere tests for items classified as pattern of activity items according to the DISCO**

| Item | Code | Kruskall-Wallis test (H), df=2 | Jonckheere trend test (J) | Z score | Power (r) |
|---|---|---|---|---|---|
| Limited pattern of activities | Ltdact | 36.35** | 380 | -6.07 | -0.67 |
| Attention span (self-chosen activities) | Attenst | 9.77 | | | |
| Attention span (other activities) | Attento | 14.84** | 746.5 | -3.16 | -0.35 |
| Inability to remain sitting | Inabsit | 20.123 | | | |
| Continual motor restlessness | Restl | 8.294 | | | |
| Hyperactivity | Hypact | 9.70* | 754 | -2.88 | -0.32 |
| Fixed, repeated motor stereotypes | Steroc | 13.45* | 734.5 | -3.67 | -0.40 |
| Excessive repetition of activities | Repact | 8.428 | | | |

**p<.001,* p<.01

All items met acceptable levels of inter-rater reliability. Attention span for self-chosen activity (2.78%) and for other activities (8.33%) as well as other pattern of activities problems (0%) were not endorsed by at least 10% of the ASD individuals and were therefore removed from the reliability analysis. This left six items for the reliability analysis, which created a reliable scale (cronbach's alpha = .807) as shown in Table 13.

**Table 13: Table showing the selected items for the Pattern of Activities scale of the DISCO**

| | Corrected Item-Total Correlation | Cronbach's Alpha if Item Deleted |
|---|---|---|
| Limited pattern of activities | .483 | .799 |
| Inability to remain sitting | .711 | .741 |
| Continual motor restlessness | .701 | .745 |
| Hyperactivity | .706 | .742 |
| Fixed, repeated motor stereotypes | .410 | .809 |
| Excessive repetition of activities | .463 | .803 |

The difference of the total score on all pattern of activity items was assessed using a Kruskall-Wallis test. This revealed a significant difference for total pattern of activities score across the five diagnostic groups (H(4) = 33.02, p<.001) and follow up tests showed significant differences between the LFA and ID groups (U = 75, r = -0.44, p<.01), the HFA and LI groups (U = 53, r = -0.49, p<.01) and the HFA and TD groups (U = 6.5, r = -0.83, p<.001) as shown in Figure 6.

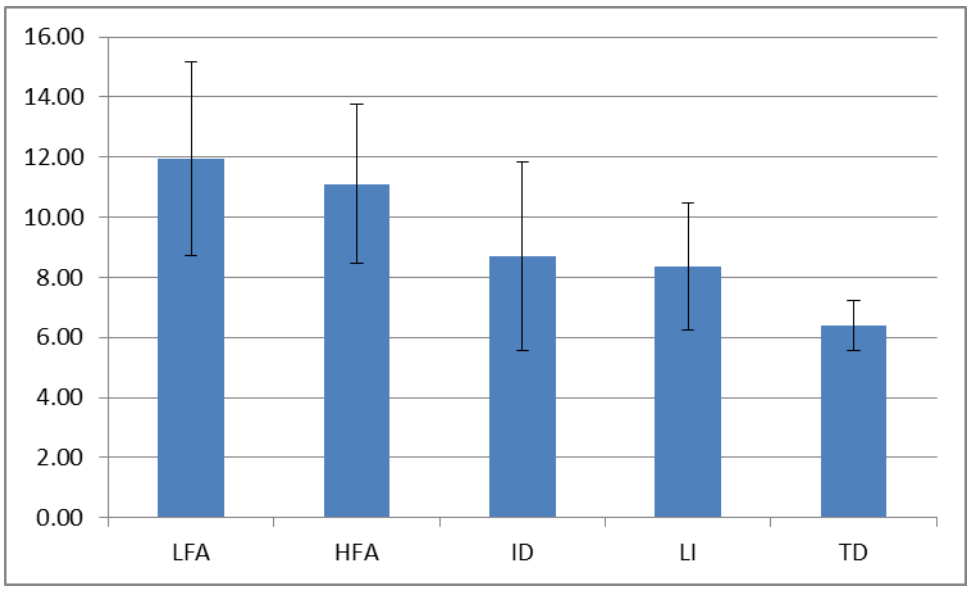



**Figure 6: Figure showing the mean total pattern of activities score across diagnostic sub-groups in Sample 1. The only significant difference was between the HFA and TD group.**

**Sleep disturbances**

Finally, there was only one sleep disturbance item, which was significantly different across the groups using a Kruskall-Wallis test, which also showed a significant trend of being most prevalent in the ASD group and least in the TD group as shown in Table 14.

**Table 14: Table showing the results of Kruskall-Wallis and follow up Jockheere tests for items classified as sleep behaviours in the DISCO**

| Item | Code | Kruskall-Wallis test (H), df=2 | Jonckheere trend test (J) | Z score | Power (r) |
|---|---|---|---|---|---|
| Night sedation | Nighsed | 1.959 | | | |
| Difficulty falling asleep | Gosleep | 12.06* | 726.5 | -3.37 | -0.37 |
| Difficulty in remaining asleep | Stayslp | 8.441 | | | |
| Night terrors or nightmares | Terror | 0.43 | | | |
| Other | Slpoth | 1.412 | | | |

**p<.001,* p<.01

All items met acceptable levels of inter-rater reliability. "Night sedation" was only marked in 8.33% of individuals with ASD and "other sleep problems" was not endorsed by any individuals with ASD and were therefore removed from the reliability analysis. However, this only leave three items (in Table 15) which create an overall reliability of .630. Moreover, The overall Kruskall-Wallis test across the five diagnostic groups was not significant at the p<.01 level if significance (H(4) = 12.749, p=.011, n.s.) as shown in Figure 7.

**Table 15: Table showing the selected items for the Sleep Disturbances scale of the DISCO**

| | Corrected Item-Total Correlation | Cronbach's Alpha if Item Deleted |
|---|---|---|
| Difficulty falling asleep | .443 | .527 |
| Difficulty in remaining asleep | .551 | .362 |
| Night terrors or nightmares | .336 | .661 |

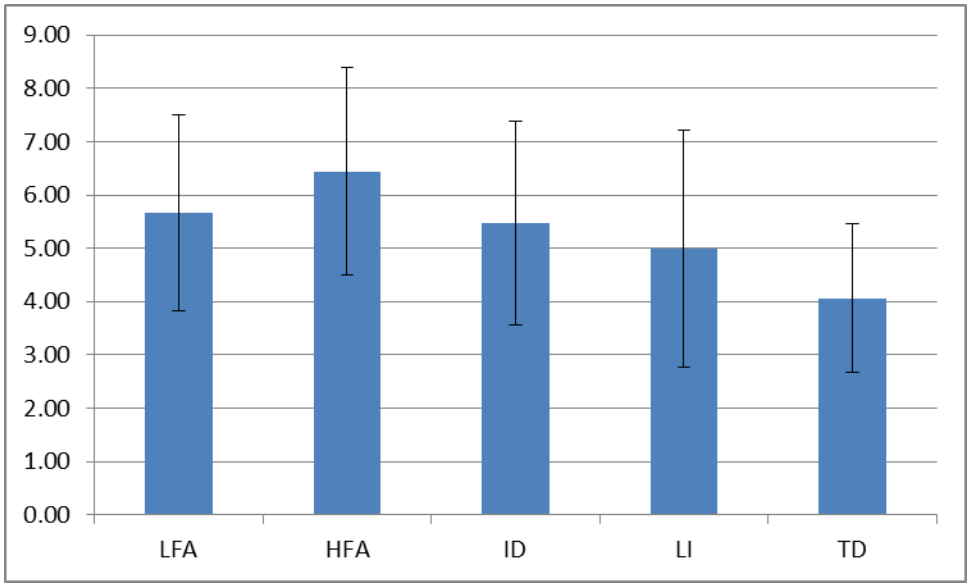



Figure 7: Figure showing the mean total sleep score across diagnostic sub-groups in Sample 1. The only significant difference was between the HFA and TD group.

## APPENDIX 4: THE SENSORY PREFERENCES QUESTIONNAIRE

# Sensory Preferences Questionnaire

## INSTRUCTIONS

Please check the box that best describes how often you feel or engage in the behaviours that are described. Please answer all of the questions using the following guidelines:

| | |
|---|---|
| **Almost Never** | Tick this box if you **almost never** respond in this way or **almost never** feel this way. |
| **Seldom** | Tick this box if you **seldom** respond in this way or **seldom** feel this way. |
| **Occasionally** | Tick this box if you **occasionally** respond in this way or **occasionally** feel this way. |
| **Frequently** | Tick this box if you **frequently** respond in this way or **frequently** feel this way. |
| **Almost Always** | Tick this box if you **almost always** respond in this way or **almost always** feel this way. |

| Qu. | Responses to Auditory Stimuli | Almost Never | Seldom | Occasionally | Frequently | Almost Always |
|---|---|---|---|---|---|---|
| 1 | I am upset by some sounds that do not affect other people (e.g. vacuum cleaners, aeroplanes, fire engines or road drills). | | | | | |
| 2 | I have an unusual interest in some sounds (e.g. bells, water hissing in pipes, records played at the wrong speed) and I spend time listening to these sounds. | | | | | |
| 3 | I have unusually acute hearing (e.g. I can hear the jangle of car keys, the rustle of a sweet paper or a quiet sound from a long distance away). | | | | | |

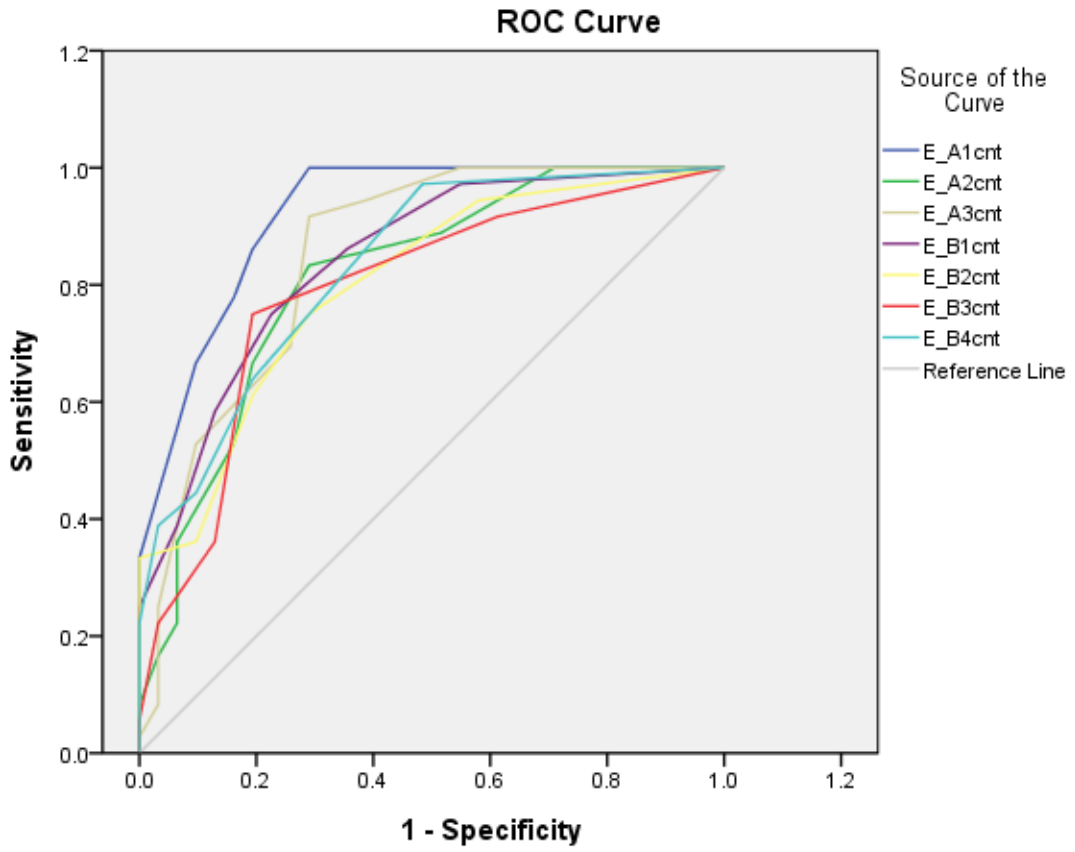
| Qu. | Responses to Visual Stimuli | Almost Never | Seldom | Occasionally | Frequently | Almost Always |
|---|---|---|---|---|---|---|
| 1 | I am unusually interested in shiny things (e.g. silver paper, tinsel, patches of sunlight, street lights or lights on a motorway). | | | | | |
| 2 | I get unusually excited at seeing things spin. | | | | | |
| 3 | I twist or flick my hands or objects near to my eyes. | | | | | |
| 4 | I like to look at objects from many different angles for no obvious reason or I examine objects, such as lines of toy trains, by eyeing them closely on their level, perhaps kneeling down or lying on the floor to do this. | | | | | |

| Qu. | Responses to Proximal Sensory Stimuli | Almost Never | Seldom | Occasionally | Frequently | Almost Always |
|---|---|---|---|---|---|---|
| | **Touch** | | | | | |
| 1 | I have an unusual interest in the feel of surfaces (e.g. fur, velvet, hair or smooth or rough surfaces). | | | | | |
| 2 | I scratch or tap on different surfaces in order to feel the sensation. | | | | | |
| 3 | I react negatively if gently touched (e.g. if someone softly touches my arm or shoulder). | | | | | |
| 4 | I react negatively to being held firmly or tightly (e.g. if someone hugs me). | | | | | |
| 5 | I flick things like pieces of string or sticks, tap two objects together or roll pieces of cotton in my fingers because I like the sensation. | | | | | |
| 6 | I dislike or resist being washed, having my hair washed, my nails cut or my hair cut. | | | | | |
| 7 | I am bothered by having dirty or sticky hands or dislike handling Play-Doh, sand, glue or other messy materials. | | | | | |

| Qu. | Responses to Proximal Sensory Stimuli | Almost Never | Seldom | Occasionally | Frequently | Almost Always |
|---|---|---|---|---|---|---|
| | **Smell/Taste** | | | | | |
| 1 | I tend to explore people or objects by smelling them. | | | | | |
| 2 | I have very unusual food fads (e.g. I will only eat marmite sandwiches or I will insist on one brand of drink only). | | | | | |
| | | | | | | |
| | **Other Oral** | | | | | |
| 1 | I tend to put inappropriate objects in my mouth or I swallow inedible objects (e.g. cigarette ends, small pieces of metal or paper). | | | | | |
| 2 | I refuse to eat food that is lumpy or hard to chew. | | | | | |

| Qu. | Responses to Proximal Sensory Stimuli | Almost Never | Seldom | Occasionally | Frequently | Almost Always |
|---|---|---|---|---|---|---|
| | **Kinaesthetic** | | | | | |
| 1 | I enjoy being spun round or going on roundabouts, more than other people my age. | | | | | |
| 2 | I like spinning round or running round in circles, more than other people my age. | | | | | |
| | | | | | | |
| | **Pain** | | | | | |
| 1 | I DO NOT react to unpleasant or painful sensations (e.g. cuts and bruises, toothache, sore throat or broken bones). | | | | | |

# APPENDIX 5: SETTING THRESHOLDS FOR SUB-DOMAIN ANALYSES USING ROC CURVE ANALYSIS




Diagonal segments are produced by ties.

**A1**

| Threshold | Sensitivity | Specificity | Youden J |
|---|---|---|---|
| 1 | 1 | .258 | .258 |
| 2 | 1 | .484 | .484 |
| 3* # | 1 | .710 | .710 |
| 4 | .861 | .806 | .667 |
| 5 | .778 | .839 | .617 |
| 6 | .667 | .903 | .570 |

**A2**

| Threshold | Sensitivity | Specificity | Youden J |
|---|---|---|---|
| 1# | 1 | .290 | .290 |
| 2 | .944 | .387 | .331 |
| 3 | .889 | .484 | .373 |
| 4* | .833 | .710 | .543 |
| 5 | .667 | .806 | .473 |

**A3**

| Threshold | Sensitivity | Specificity | Youden J |
|---|---|---|---|
| 1 | 1 | .032 | .032 |
| 2 | 1 | .226 | .226 |
| 3# | 1 | .452 | .452 |
| 4 | .944 | .613 | .557 |
| 5* | .917 | .710 | .627 |
| 6 | .694 | .742 | .436 |

**B1**

| Threshold | Sensitivity | Specificity | Youden J |
|---|---|---|---|
| 1# | .972 | .452 | .424 |
| 2 | .861 | .645 | .506 |
| 3* | .750 | .774 | .524 |
| 4 | .583 | .871 | .454 |

**B2**

| Threshold | Sensitivity | Specificity | Youden J |
|---|---|---|---|
| 1# | .944 | .419 | .363 |
| 2* | .750 | .710 | .460 |
| 3 | .611 | .806 | .417 |
| 4 | .361 | .903 | .264 |

**B3**

| Threshold | Sensitivity | Specificity | Youden J |
|---|---|---|---|
| 1# | .917 | .387 | .304 |
| 2* | .750 | .806 | .556 |
| 3 | .361 | .871 | .232 |
| 4 | .222 | 968 | .190 |

**B4**

| Threshold | Sensitivity | Specificity | Youden J |
|---|---|---|---|
| 1*# | .972 | .516 | .488 |
| 2 | .639 | .806 | .445 |
| 3 | .444 | .903 | .347 |

* Threshold selected using the Youden J statistic; # Threshold selected for the modified DISCO algorithm

# APPENDIX 6: OVERLAP BETWEEN DSM-5 AND ICD-10 CHILDHOOD AUTISM AND ATYPICAL AUTISM IN SAMPLES 1 AND 3

In Sample 1, all individuals with a clinical diagnosis of ASD (18 high; 18 low) met criteria for ICD-10 CA as did 12 individuals in the clinical comparison groups. Table 1 reveals that in Sample 1 100% of the clinical ASD group received a diagnosis of both DISCO ICD-10 CA and DISCO DSM-5 ASD, however, in the comparison group more individuals met criteria for ICD-10 CA (n=12: 8 low; 4 high) than DSM-5 ASD (n=8: 5 low; 3 high) but this effect was not significant ($x^2(1)=0.61$, n.s.).

*Table 1: The number of individuals receiving a DISCO ICD-10 diagnosis of Childhood Autism (CA) in comparison to DISCO DSM-5 criteria in Sample 1.*

| **DISCO Algorithm Diagnosis** | | **Clinical Group and Ability Level** | | | | |
|---|---|---|---|---|---|---|
| **ICD-10 CA** | **DSM-5 ASD** | **Autism** | | **Comparison** | | |
| | | **Low Ability** | **High Ability** | **Low # Ability** | **High* Ability** | **Total** |
| **+** | **+** | 18 | 18 | 4 | 3 | 43 |
| **+** | **-** | 0 | 0 | 4 | 1 | 5 |
| **-** | **+** | 0 | 0 | 1 | 0 | 1 |
| **-** | **-** | 0 | 0 | 8 | 25 | 33 |
| **Total** | | 18 | 18 | 17 | 29 | 82 |

In Sample 3, 47 individuals in the clinical ASD group and four comparison individuals met criteria for ICD-10 Childhood Autism. Table 2 reveals that Sample 3 showed a lower sensitivity of DSM-5 in comparison to ICD-10 CA (82.69% individuals with ICD-10 CA get DSM-5), however, the difference in individuals meeting just ICD-10 CA (4) or DSM-5 (1) criteria was not significant ($x^2(1)=1.25$, n.s.). As in Sample 1, the specificity was better for DSM-5 (n=3) than ICD-10 CA (n=4, $x^2(1)=0.03$, .n.s.).

Table 2 *The number of individuals receiving a DISCO ICD-10 diagnosis of Childhood Autism (CA) or Atypical Autism (Atyp) in comparison to DISCO DSM-5 criteria in Sample 3.*

| **DISCO Algorithm diagnosis** | | **Clinical Group** | | | | |
|---|---|---|---|---|---|---|
| **ICD-10 CA** | **DSM-5 ASD** | **Autism Group** | | **Comparison** | | |
| | | **Low Ability** | **High Ability** | **Low# Ability** | **High* Ability** | **Total** |
| **+** | **+** | 29 | 14 | 1 | 0 | 44 |
| **+** | **-** | 3 | 1 | 3 | 0 | 7 |
| **-** | **+** | 1 | 0 | 2 | 0 | 3 |
| **-** | **-** | 2 | 2 | 20 | 37 | 61 |
| **Total** | | 35 | 17 | 26 | 37 | 115 |

As can be seen in Table 3, in Sample 1 none of the clinical ASD group met criteria for ICD-10 Atypical Autism. When comparing ICD-10 Atypical cases only, the DSM-5 ASD algorithm identifies one more comparison case (n=8) than the ICD-10 atypical algorithm (n=7). However, as can be seen when the ICD-10 CA and atypical cases are combined, a large proportion of comparison individuals

are already identified by the ICD-10 CA algorithm. Nineteen comparison cases met ICD-10 criteria (CA or atypical) and only eight control cases met DSM-5 criteria. A McNemar test revealed significantly more of the comparison individuals received an ICD-10 diagnosis (CA or atypical) and not a DSM-5 ASD than DSM-5 ASD and not an ICD-10 diagnosis ($x^2(1)=10.02$, $p<.01$). This revealed a greater level of specificity for the new diagnostic criteria than previous categorical approaches.

*Table 3: The number of individuals receiving a DISCO ICD-10 diagnosis of Childhood Autism (CA) or Atypical Autism (Atyp) in comparison to DISCO DSM-5 criteria in Sample 1.*

| **DISCO Algorithm Diagnosis** | | **Clinical Group and Ability Level** | | | | |
|---|---|---|---|---|---|---|
| **ICD-10 atyp** | **DSM-5 ASD** | **Autism** | | **Comparison** | | |
| | | **Low Ability** | **High Ability** | **Low # Ability** | **High* Ability** | **Total** |
| **+** | **+** | 0 | 0 | 1 | 0 | 1 |
| **+** | **-** | 0 | 0 | 2 | 4 | 6 |
| **-** | **+** | 0 | 0 | 4[#] | 3[#] | 7 |
| **-** | **-** | 18 | 18 | 10 | 22 | 68 |
| **Total** | | 18 | 18 | 17 | 29 | 82 |
| **ICD-10 CA or Atyp** | **DSM-5 ASD** | | | | | |
| **+** | **+** | 18 | 18 | 5 | 3 | 44 |
| **+** | **-** | 0 | 0 | 6 | 5 | 11 |
| **-** | **+** | 0 | 0 | 0 | 0 | 0 |
| **-** | **-** | 0 | 0 | 6 | 21 | 27 |
| **Total** | | 18 | 18 | 17 | 29 | 82 |

Table 4, shows the results of the ICD-10 Atypical Autism algorithm in Sample 3; three individuals with clinically defined ASD met ICD-10 Atypical criteria as did eight of the comparison individuals. In the ASD group, the combined results of ICD-10 Childhood and Atypical Autism show the same pattern as the previous Childhood Autism results; the ICD-10 criteria (CA and atypical) is more sensitive than DSM-5 ASD. Only 84.65% of the combined ICD-10 group meet DSM-5 criteria; a McNemar's test revealed that significantly more individuals met ICD-10 CA or Atypical (n=50) and not DSM-5 ASD criteria, than met DSM-5 ASD (n=44) and not ICD-10 CA or atypical criteria ($x^2(1)=5.042$, $p<.05$). However, the specificity was again better for DSM-5 (n=3) than both ICD-10 diagnoses (n=12), which was significant using a McNemar's test ($x^2(1)=8.03$, $p<.01$).

Table 4: *The number of individuals receiving a DISCO ICD-10 diagnosis of Childhood Autism (CA) or Atypical Autism (Atyp) in comparison to DISCO DSM-5 criteria in Sample 3.*

| **DISCO Algorithm diagnosis** | | **Clinical Group** | | | | |
|---|---|---|---|---|---|---|
| | | **Autism Group** | | **Comparison** | | |
| | | **Low Ability** | **High Ability** | **Low# Ability** | **High* Ability** | **Total** |
| **ICD-10 Atyp** | **DSM-5** | | | | | |
| **+** | **+** | 1 | 0 | 2 | 0 | 3 |
| **+** | **-** | 2 | 0 | 6 | 0 | 8 |
| **-** | **+** | 29 | 14 | 1 | 0 | 44 |
| **-** | **-** | 3 | 3 | 17 | 37 | 60 |
| **Total** | | 35 | 17 | 26 | 37 | 115 |
| **ICD-10 CA or Atyp** | **DSM-5** | | | | | |
| **+** | **+** | 30 | 14 | 3 | 0 | 42 |
| **+** | **-** | 5 | 1 | 9 | 0 | 9 |
| **-** | **+** | 0 | 0 | 0 | 0 | 2 |
| **-** | **-** | 0 | 2 | 14 | 37 | 62 |
| **Total** | | 35 | 17 | 26 | 37 | 115 |

# APPENDIX 7: FURTHER TESTING OF THE SENSITIVITY OF THE FINAL ALGORITHM AT THE DOMAIN AND SUB-DOMAIN LEVEL.

This study consists of supplementary analyses to identify why the individuals did not meet the criteria for DSM-5. Exploring the sub-domains on which individuals failed to score could highlight potential adjustments to improve sensitivity.

## *Method*

### *Participants:*

Individuals included in the following analyses were the individuals who did not meet DSM-5 criteria from Sample 2, these consisted of five individuals who met ICD-10 Childhood Autism Criteria, seven individuals who met ICD-10 Atypical Autism criteria and nine individuals who met criteria for Gillberg's Asperger Syndrome.

## *Results*

Of the individuals who met ICD-10 Childhood Autism criteria but not DSM-5 ASD criteria all were high functioning, the three children (2 male; 1 female), one female adolescent and one male adult consistently failed to score on Domain A ("Persistent deficits in social communication and social interaction"), specifically all five individuals do not meet criteria for the sub-domain A2 ("Deficits in non-verbal communicative behaviours used for social interaction") and the adult also failed to meet criteria for A3 ("Deficits in developing and maintaining relationships appropriate to developmental level").

Of the seven atypical cases that were missed by DSM-5, three (2 female; 1 male) were low functioning and four high functioning (3 male; 1 female). Neither domain was specifically missed by these individuals; six individuals failed to meet criteria for domain A and five failed to meet criteria for Domain B ("Restricted, repetitive patterns of behaviour, interests, or activities").

Finally, the individuals that met Gillberg's Asperger criteria but not DSM-5 ASD consistently failed to score on Domain C "Symptoms must be present in early childhood, but may not become fully manifest until social demands exceed limited capacities"; only one of the nine individuals met criteria for this domain. In addition two individuals failed to score on A2 ("Deficits in non-verbal communicative behaviours used for social interaction"). All nine individuals were high functioning: five children were all male; one female adolescent and the three adults were all male.

## *Discussion*

Analyses revealed that individuals meeting criteria for ICD-10 Childhood Autism are consistently failing to score on DSM-5 ASD because they do not meet criteria for deficits in non-verbal communicative behaviours used for social interaction (sub-domain A2), however, individuals who met criteria for ICD-10 Atypical Autism are not as a group consistently failing to score in one particular area. Individuals meeting Gillberg's Asperger individuals because they fail to score on Domain C (age on onset). In consideration it is not surprising that Gillberg's cases were missed by the C rule as this was just taken from the ICD-10 criteria for Childhood Autism (criterion A). It is, therefore, restricted to individuals who present with a delay or impairments before three years of age and does not account for the new addition to DSM-5 that states impairments should be present in early childhood "or when social demands exceed functioning".

As described in the literature review in section 5.2, the DSM-5 ASD criteria has been shown to under-diagnose those meeting criteria for DSM-IV, especially high functioning or Asperger individuals. In the literature, one potential adaptation has been proposed to improve sensitivity of DSM-5, "relaxing" the number of sub-domains required to meet each domain. According to these adjustments, individuals are required to meet two rather than three of the social-communication sub-domains (persistent deficits in social communication and social interaction; e.g. Matson, Hattier

& Williams, 2012). Previous research has found that adjustment to this criterion increased sensitivity by approximately 10-12%, however specificity was also compromised (Frazer et al., 2012; Matson, Hattier, & Williams, 2012; McPartland et al., 2012). Wing, Gould and Gillberg (2011) also highlighted Domain C (age of onset criterion) as being insufficient and that could potentially limit the sensitivity of DSM-5 ASD to the wider spectrum, however, previous research has not yet established the need for a change or removal of this criterion, for example, the same criteria as previously used for DSM-IV-TR Autistic Disorder has been adopted in DSM-5 measurement (e.g. McPartland, et al., 2012).

If the adjustment proposed in the introduction of "relaxing" of the A rule was applied to the current Sample then a "relaxed" DSM-5 ASD algorithm would identify all of the ICD-10 Childhood Autism cases (meaning that 100% of these individuals would be captured) but only one of the ICD-10 Atypical Autism cases and none of the Gillberg's Asperger individuals. Therefore, this adjustment would not be successful in improving the sensitivity of the criteria to the cases identified in Study 5.2.2. The previous literature identified that this adjustment may also have detrimental effects on specificity (Frazer et al., 2012; Matson, Hattier, & Williams, 2012; McPartland et al., 2012).

In contrast, seven of the nine missed Gillberg's Asperger individuals would be captured of the age of onset criterion (Domain C) was removed. This would improve the sensitivity of DSM-5 in individuals with an Asperger profile from 89.89% to 97.75%. In addition, if this modification was made to an algorithm run in Samples 1 and 3, the same level of specificity as the original algorithm was found. This raises the possibility that criterion C is too restrictive for individuals outside of those meeting criteria for Childhood Autism. The items already included in the age of onset criterion are not by themselves restrictive and do not explicitly require delays before the age of three but attempt to capture whether there were delays in behaviours that commonly appear before or around that age such as obeying instructions, use of phrases, selective attachment and the development of pretend play. However, at least one of the specified behaviours must be present in order to meet criteria for DSM-5 and potentially sensitivity could be improved if the strict criteria were relaxed. Another option, is that this criterion could instead be part of the clinical judgement made by the clinician rather than a count of DISCO items. The DISCO already contains an item that utilises the interviewer's judgement of "onset of untypical behaviour" in age in months. This one item could be modified to incorporate the specifier that impairments should be present in early childhood "but may not become full manifest until social demands exceed functioning" so the interviewer codes both the age and whether this age is relevant to the individual's functioning level and therefore diagnosis.

# APPENDIX 8: CORRELATION BETWEEN RANDOMLY SELECTED ITEMS

Correlations between three randomly selected items in Sample 3

| | One sided approaches | Having a special friend | Unusual movements of hands or arms |
|---|---|---|---|
| One sided approaches | 1 | .206* | .116 |
| Having a special friend | | 1 | .110 |
| Unusual movements of hands or arms | | | 1 |

*p<.001

# APPENDIX 9: EXPLORATORY ANALYSES OF THE SOCIAL SUB-TYPES (ALOOF, PASSIVE AND ACTIVE BUT ODD)

Study 2 has four parts:

- Assess the frequency of social sub-groups across age, ability and gender.
- Assess the prevalence of core diagnostic features across social sub-type.
- Assess the prevalence of associated features across social sub-type.
- Assess the scores on DISCO co-morbid disorders checklists across social sub-type (see methods chapter for items for co-morbid diagnoses)

## *Results*

### *Aim 1: Assess the frequency of social sub-groups across age, ability and gender*

Table 1 presents the percentage of individuals in Sample 2 scoring across the three social sub-types of interest as well as those in the typical social group. Chi-square analyses found that the social sub-types significantly differed across the four age groups ($x^2(3) = 31.18$, $p<.001$) and across high and low functioning individuals ($x^2(1) = 23.873$, $p<.001$). There was no significant association between the quality of social interaction sub-types and gender ($x^2(1) = 1.03$, $p=.599$, n.s.).

**Table 1: Table showing the percentage of individuals in Sample 2 scoring across the social sub-types in high and low ability pre-school children, older children, adolescents and adults.**

| | Prechool (45) | | Children (74) | | Adolescents (34) | | Adults (47) | |
|---|---|---|---|---|---|---|---|---|
| | High (22) | Low (23) | High (22) | Low (52) | High (14) | Low (20) | High (12) | Low (35) |
| Aloof and indifferent | 68.2 | 56.5 | 40.9 | 13.5 | 21.4 | 0 | 41.7 | 2.9 |
| Passive | 9.1 | 0 | 0 | 5.8 | 35.7 | 20.0 | 33.3 | 25.7 |
| Active but odd | 22.7 | 39.1 | 50.0 | 71.2 | 28.6 | 75.0 | 25.0 | 65.7 |
| Typical | 0 | 4.3 | 9.1 | 9.6 | 14.3 | 5.0 | 0 | 5.7 |

### *Aim 2: Assess the prevalence of core diagnostic features across social sub-type.*

The number of items that individuals in the Aloof, Passive and Active but odd group are scoring on each associated scale is shown in Figure 1.

A series of multiple regression analyses were run with age, IQ and language entered in the first step and quality of social interaction sub-group added in the second step when predicting individual scores on seven sub-domains for DSM-5. For all cases the addition of the social sub-types in the second step significantly improved the variance explained by the model (from 7.36% in B2 to a 23.5% increase in A2). In addition, for all three social-communication sub-domains each social sub-type was a significant independent predictor in a model with all three social sub-types, age, gender and IQ.

The social sub-types, however, differed in their predictive power across the B or repetitive behaviour domain in DSM-5. For sub-domain B1 (Stereotyped or repetitive speech, motor movements, or use of objects) the aloof ($\beta = .657$, $p<.001$), passive ($\beta = .284$, $p<.01$) and active but odd ($\beta = .601$, $p<.001$) variables were significant predictors. However for both B2 (Excessive adherence to routines, ritualized patterns of verbal or on-verbal behavior, or excessive resistance to change) and B3 (Highly restricted fixated interests that are abnormal in intensity or focus) the only independently significant social sub-type was the active but odd group (B2, $\beta = .371$, $p<.01$; B3, $\beta = .455$, $p<.001$). Finally, it was the aloof group that was the only significant predictor of the B4 (Hyper

or hypo-sensitivity to sensory input or unusual interest in sensory aspects of the environment) sub-domain (β = .467, p<.001).

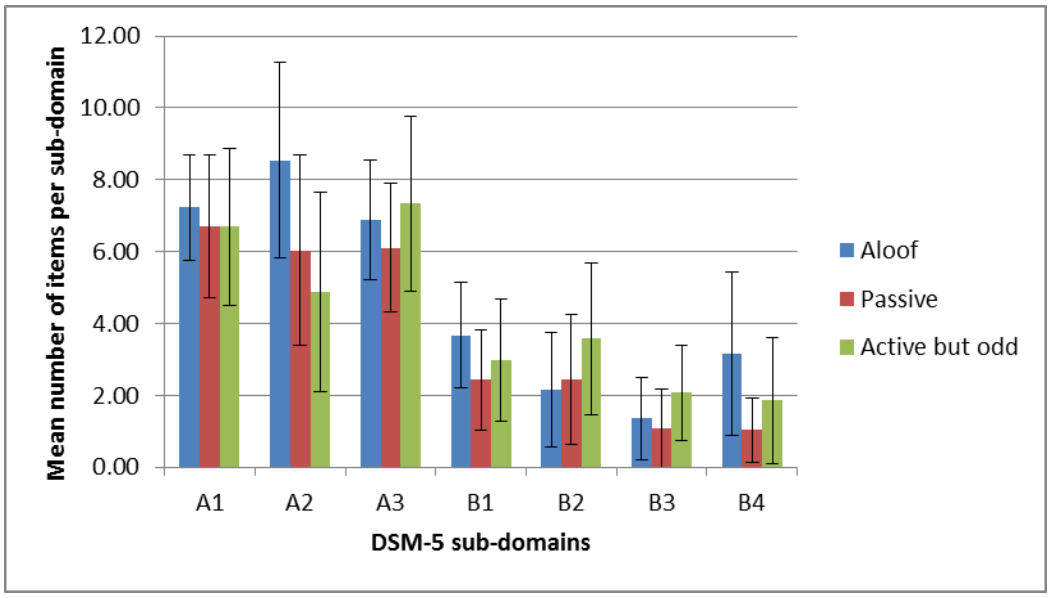



**Figure1: Figure showing the mean number of items that individuals in the Aloof, Passive and Active but odd social sub-types score for each sub-domain of DSM-5**

*Aim 3: Assess the prevalence of associated features across social sub-type.*

The number of items that individuals in the Aloof, Passive and Active but odd group are scoring on each associated scale is shown in Figure 2.

A series of multiple regression analyses were run with age, IQ and language entered in the first step and quality of social interaction sub-group added in the second step when predicting individual scores on the sensory, maladaptive, pattern of activities and daily living skills scales. ANOVA analyses were used to test whether the addition of social sub-group significantly contributed to the prediction of the dependent variables (associated features). For all associated variables the addition of social sub-group significantly improved the model above age, IQ and language: in the sensory model the addition of social sub-type improved the variance explained by 9.1% ($F(3,193) = 7.072$, $p<.001$); in the maladaptive model by 7.1% ($F(3,193) = 5.142$, $p<.01$); in the pattern of activities model by 5.9% ($F(3,193) = 4.886$, $p<.01$); and in the Daily living skills model by 5.2% ($F(3,193) = 3.913$, $p<.01$).

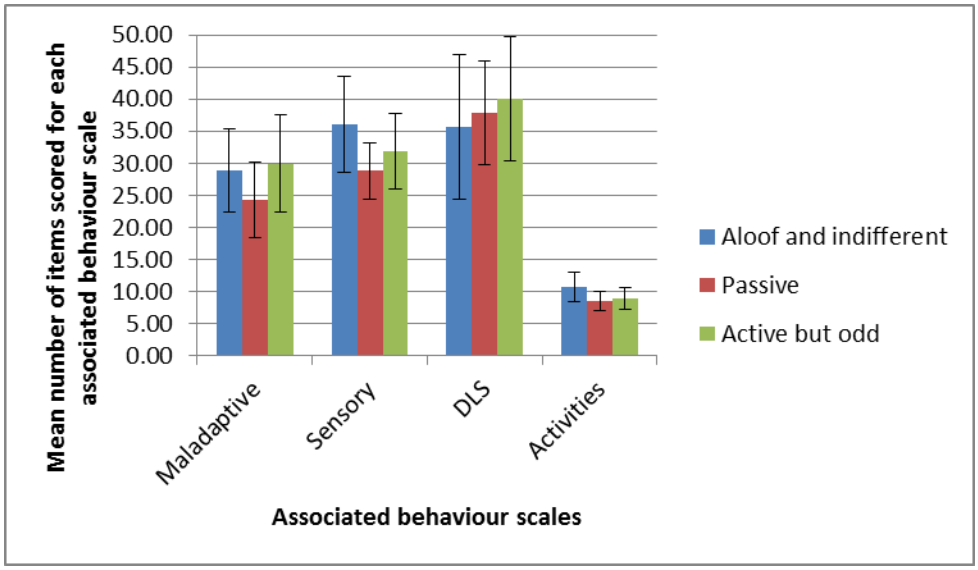



**Figure 2: Figure showing the number of items that individuals in the Aloof, Passive and Active but odd social sub-types score for each associated behaviour scale.**

Being in the aloof group significantly predicted the presence of more sensory behaviours (β=0.447, p<.001) as did being in the active but odd group (β = 0.277, p<.05). Being in the aloof group also predicted more behaviours on the pattern of activities scale (β=0.296, p<.05) and being in the active but odd group predicted more problems with daily living skill behaviours (β = 0.391, p<.001). Although the addition of social sub-type significantly improved the model for maladaptive behaviours no social sub-type on its own was a significant predictor in the model.

### *Aim 4: Assess the scores on DISCO co-morbid disorders checklists across social sub-type*

Comorbid features were measured according to the symptom counts used in the DISCO that are described in Table 2.

**Table 2: Table presenting the co-morbid scales in the DISCO and the corresponding items for each co-morbid scale.**

| Item | Code |
|---|---|
| **Attention Deficit/Hyperactivity Disorder** | |
| Time attends to own interests | 0,1,2 |
| Time attends to tasks given to A by others | 0,1,2 |
| Understanding future events | 0 |
| Climbs everywhere regardless of danger | 0 |
| Does not sit down | 0,1 |
| Continual restlessness | 0 |
| Always running round, jumping etc. | 0 |
| Noisy | 0 |
| Creates aimless chaos | 0 |
| Cannot take turns | 0 |
| Constantly interrupts others | 0 |
| **Developmental co-ordination disorder** | |
| Clumsiness | 0,1 |
| Odd gait | 0 |
| Poor motor co-ordination | 0 |
| Poor fine motor co-ordination | 0,1 |
| **Tic disorder including Tourette's syndrome** | |
| Jerky involuntary movements | 0 |

| | |
|---|---|
| Involuntary facial grimaces | 0 |
| Shrieks, grunts, other involuntary noises | 0 |
| **Catatonia** | |
| ***Features described in catatonia and autistic disorders*** | |
| Immediate echolalia | 0 |
| Delayed echolalia | 0 |
| Stares too long and hard | 0 |
| Complex body movements | 0 |
| Tiptoe walking | 0 |
| Facial grimaces | 0 |
| Self-tapping, twiddling, finger flicking etc. | 0 |
| Repetitive aimless manipulation of objects | 0 |
| Repetitive talking on same theme | 0 |
| Fixed, repeated motor stereotypies | 0 |
| Impulsive behaviour – may be bizarre | 0 |
| Other catatonic features | 0 |
| ***Features described as typical of catatonia and/or parkinsonism*** | |
| Odd hand postures | 0 |
| Freezing interrupting actions | 0 |
| Repeated approach and withdrawal | 0 |
| Turning up of eyes | 0 |
| Marked lack of any spontaneous activity | 0 |
| Odd gait or mannerisms when walking | 0 |
| Echopraxia | 0 |
| **Pathological demand avoidance** | |
| Unusually quiet and passive in infancy | 0,1 |
| Clumsy in gross movements | 0,1 |
| Uses peers as mechanical aids; bossy and domineering | 0,1 |
| Repetitive role play – lives the part not usual pretence | 0,1 |
| Hands seem limp and weak for unwelcome tasks | 0,1 |
| Repetitive questioning | 0,1 |
| Lack of co-operation; strongly resists | 0,1 |
| Lies, cheats, steals, fantasises, causing distress to others | 0,1 |
| Difficulties with others tease, bully, refuse to take turns, make trouble | 0,1 |

The number of items that individuals in the Aloof, Passive and Active but odd group are scoring on each comorbid checklist scale are shown in Figure 3.

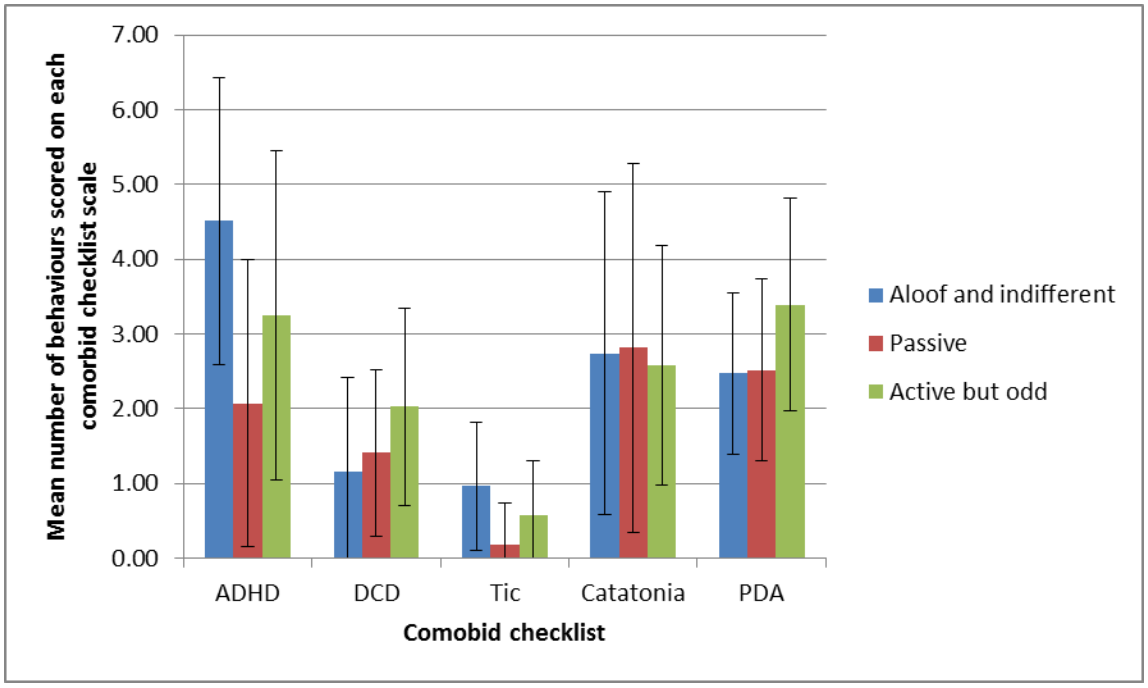



**Figure 3: Figure showing the number of items that individuals in the Aloof, Passive and Active but odd social sub-types score for each comorbid checklist in the DISCO.**

A series of multiple regression analyses were run with age, IQ and language entered in the first step and quality of social interaction sub-group added in the second step when predicting individual scores on the ADHD, DCD, tic disorder, catatonia and PDA DISCO behaviour checklists. ANOVA analyses were used to test whether the addition of social sub-group significantly contributed to the prediction of the dependent variables (comorbid checklists). For all comorbid checklists variables the addition of social sub-group significantly improved the model above age, IQ and language: in the model predicting ADHD the addition of social sub-type improved the variance explained by 4.5% ($F(3,193) = 3.266$, $p<.05$); in the DCD model by 7% ($F(3,193) = 5.036$, $p<.01$ ); in the tic disorders model by 7.2% ($F(3,193) = 5.474$, $p<.001$); in the PDA model by 11.4% ($F(3,193) = 8.916$, $p<.001$). There was one exception, the addition of the social sub-types to the catatonia model was not significant (3.1%; $F(3,193) = 1.885$, $p=.134$, n.s.).

Being in the passive group significantly predicted an individual having fewer ADHD behaviours present ($\beta = -.246$, $p<.05$). Being in the aloof was found to significantly predict more likely to have more tic behaviours ($\beta = 0.315$, $p<.05$) and catatonic behaviours ($\beta = 0.438$, $p<.05$) whereas being in the active but odd group was a significant predictor for having more PDA behaviours ($\beta = .405$, $p<.01$). Although the addition of social sub-type significantly improved the model for the DCD checklist no social sub-type on its own was a significant predictor in the model.