INTRODUCTION
I first heard the term ‘Asperger syndrome’ 20 years ago when I accompanied my sister-in-law Elizabeth and her son Ivan on a visit to a child psychiatrist. Ivan was 19 years old and had an odd pattern of behaviours since childhood. As none of the experts he had seen had been able to clearly diagnose his problem, we hoped that this new consultation would give a better insight into his difficulties. That day, Ivan received a diagnosis of Asperger syndrome, a condition that was virtually unknown at the time. The psychiatrist was Lorna Wing and the date was 1984, only a few years after she had written her paper on Asperger syndrome (Wing 1981).

It is a long time since that day when I first met Lorna Wing and she diagnosed Ivan, but the impact of that day still remains. Although Ivan was too old by then to benefit from specialized education, he was still able to gain access to adult services and residential accommodation that was more tailored to his needs. All this, and the fact that his diagnosis brought increased understanding of his condition by the people who work with him, has helped him and his family enormously. There is no doubt that Ivan’s life was changed immeasurably by being able to gain an understandable diagnosis.

Since Lorna Wing’s landmark paper on Asperger syndrome, there has been an extraordinary increase in the clinical, research and media attention given to this condition. The term Asperger syndrome has come far from the unknown status it had when she first wrote her landmark paper. It is now virtually a household term. In a recent article, Wing (2004) describes how public and professional conceptions about Asperger syndrome have changed since her original writings. There is now greater recognition of
Wing’s original idea that autism is a broad a spectrum of disorders, forming a continuum with normal functioning (Wing 1988). In reflecting on the changes in thinking about Asperger syndrome and its impacts, both Frith (2004) and Wing (2004) argue for the benefit of this increased understanding by the public and professionals.

Alongside the benefits of an expanded understanding of Asperger syndrome there has also been great controversy about its status as a separate subgroup. In 1993 and 1994, the international classification systems were revised leading to the description of a separate subgroup category for Asperger syndrome. This gave it a separate status in both behavioural and developmental terms. The new appearance of separate diagnostic criteria for Asperger syndrome caused considerable debate.

Asperger himself had not delineated any specific criteria for Asperger syndrome. Wing (1991) also described Asperger syndrome as part of the autistic spectrum. While the new official subgroupings were seen as an advantage in terms of giving recognition to a milder form of autism, there were many unanswered questions about what actually distinguished Asperger syndrome and autism as two separate subgroups.

In the 1990s I had the opportunity to explore some of these questions with Margot Prior and Richard Eisenmajer in Australia and Lorna Wing and Judith Gould in England. Our work together allowed us to investigate in detail the behavioural patterns of children with autistic spectrum disorders. We were interested in carrying out research that would address the debate about the utility versus the futility of defining distinct subgroupings. However we also wanted to understand better the types of behavioural patterns that made up the spectrum of autism and how these connected to various cognitive and developmental variables.

In this chapter I discuss our collaborative work on subgroupings and the conclusions that we drew from this work. One thing that has emerged from this research and from the work of others in the field has been an increased interest in questions about development. Recent findings about the relationship between Asperger syndrome and autism have made us wonder about the way that the behavioural profiles we see in autism emerge from early childhood to later life and how the behaviours themselves constrain the potential for development. So in this chapter I also explore some new ideas about the way that early behavioural and developmental profiles might help to identify developmental pathways for children with autistic spectrum disorders.

**ARE THERE SUBGROUPS IN THE AUTISTIC SPECTRUM?**

Shortly after Ivan’s diagnosis I trained as a researcher in autism. In my initial research I was investigating potential cognitive markers for autism. A compelling new idea at that time was the view that it was possible to identify a specific cognitive impairment that could explain the social and communicative difficulties in autism. The idea was that this cognitive impairment could also be linked to a brain system or genetic locus providing a causal account that connected underlying impairment to the overt behavioural pattern.
or phenotype. This idea was enormously influential in providing a key turning point in research in autism (Leslie 1987; Frith 1989; Baron-Cohen 1995).

The central hypothesis was that children with autism have a mentalising deficit—that they are unable to detect and take account of other people’s inner mental states. This lack of awareness of what other people were thinking or feeling by even high-functioning children, constrasts with the awareness of typically developing children who are able to make inferences about others’ mental states from the first few years of life. The marked failure to mentalise or show a ‘theory of mind’, was first demonstrated in a research study carried out by Baron-Cohen, Leslie and Frith (1985). These authors used a simple story-based task in which the child makes an inference about another person’s mistaken belief. Evidence for this deficit was striking (see Baron-Cohen 2000 for review) and was widely replicated using this and similar tasks.

Initially the mentalising hypothesis was tested out with high-functioning children who had ‘core’ or classic autism. As broader definitions of autism become the norm however, children with milder features and/or high mental age came to be included in the research studies. Increasingly, when a broader view of autism was used, the results looked different. The specific cognitive impairment for children with autism compared with those who did not have autism was no longer found. These results raised questions for researchers and clinicians needed to think about the association between particular sets of symptoms within the autistic spectrum and particular cognitive or neurocognitive impairments. Was it the case that specific subgroups of children with particular behavioural or developmental characteristics would be more likely to have this particular mentalising deficit? If children with a cognitive disability in mentalising were found to belong to a different subtype of autistic disorder then perhaps this could be used as a diagnostic marker to distinguish subgroups within the autistic spectrum of disorders. Our study therefore aimed to investigate whether this cognitive ability might provide a measure of external validity for the existence of subgroups.

As part of this enterprise, we first needed to understand more about the behavioural patterns themselves. What are the different behavioural patterns found in children with diagnoses of autistic spectrum disorder? Do particular kinds of symptoms naturally cluster into subgroupings? Could these subgroupings be looked at in a ‘bottom-up’ way, by seeing what types of features are statistically most likely to connect together? In our first collaborative study we aimed to elicit subgroupings in exactly this way by using a statistical technique called cluster analysis.

In this project we collected detailed information about the range of behavioural features of a large group of high-functioning children, all with autistic spectrum disorders, in both South-East Australia and in England (Prior et al. 1998). This project had several components to it. The first was to identify what kinds of behavioural items cluster together in children with autism. The second was to investigate whether a specific cognitive ability might account for these groupings. There was a third component however. Since we also took measures of the child’s developmental history and
other measures of their current language, it was possible to examine the extent to which both language delay and current verbal ability might be accounting for subtypes of autistic features.

We started the study by recruiting a large sample of 135 individuals aged from 3 to 21 years that had been assessed and diagnosed by different clinicians at assessment centres in both England and Australia. Children were recruited who had a range of sub-group diagnoses of pervasive developmental disorders such as high-functioning autism (HFA), Asperger syndrome (AS), pervasive developmental disorder not otherwise specified (PDD-NOS) or simply ‘autistic features’. All these children were high-functioning with a minimum verbal mental age of approximately three to four years.

Parents of all the children in both countries were then interviewed using the Autism Spectrum Disorders Checklist (Rapin 1996). The interview collected two kinds of information - information about diagnostic behaviours specifically seen in autism and information about background history. Behavioural information included information about Wing and Gould’s (1979) three domains of the ‘triad of impairments’. These are impairments in social interaction, communication/imagination and restrictions/repetitions in self-chosen behaviour. There were 110 items in the checklist covering social interacton (body language, greeting, comfort seeking/giving, awareness of others’ feelings, awareness of social rules, imitation and play, joint referencing), communication/imagination (comprehension and use of language, speech characteristics, non-verbal communication, imagination and pretence) and restrictions and repetition in self-chosen behaviour (stereotyped movements, preoccupations with objects, sensory responses, patterns of interest, maintenance of sameness). We also collected background history variables, including information on pregnancy and birth, developmental milestones, health problems, family history, age of onset. In addition we took a measure of current vocabulary comprehension. We then carried out a cluster analysis that would empirically separate the data into the groupings that best classified the behavioural, verbal ability and background history variables.

As mentioned earlier, we aimed to discover what the subgroupings emerging from these cluster analyses would look like. For example, would the clusters that emerged from the analysis resemble the children’s original clinical subgroupings? Three clusters emerged that best fitted the data. We called these Cluster A, Cluster B and Cluster C. Interestingly these empirically directed clusters did not map perfectly onto the clinical groupings that the children had when they came into the study. Fifty per cent of children who had clinical diagnoses of high functioning autism (HFA) were in Cluster A but the rest were evenly distributed across the two clusters. More than more than half of the children who had clinical diagnoses of Asperger syndrome (AS) fitted a second cluster but 30 per cent of them were in Cluster A which fitted the more autistic-like picture. This result itself supported the intuition that clinicians are diagnosing children in ways that resemble a continuous spectrum of disorders rather than in separate groupings.
Leaving aside the original clinical diagnoses, what kinds of current behaviours were distinguished by each of the empirically derived clusters and how did they differ from each other? To answer this, we looked at the diagnostic symptoms within the categories of social interaction, communication and restricted/repetitive behaviours. Three groupings of children emerged from the analysis. Their behaviours differed from each other in terms of the severity. Cluster C children had fewer problems across all domains, Cluster B children were most like the children we would recognize as having Asperger syndrome while Cluster A children were most autism-like. Cluster B and Cluster C children were more aware of friendships and social relations than Cluster A children and were keen to communicate their interests. Half of Cluster B children and most of Cluster C children had joint attention skills, compared with only very few of Cluster A children. In contrast Cluster A children were more socially isolated and socially impaired. While Cluster A and B children were similar in some respects, they were differentiated by their social and communicative development. Yet in other areas of behaviour, for example, unusual sensory responses (smelling or scratching surfaces, fascination with sounds or lights), Cluster A and B were alike and together these two clusters differed from Cluster C.

Given that the Cluster A (autism-like) and Cluster B (Asperger-like) groupings could be differentiated on the basis of their social and communication development, does this mean that their social and communication behaviours were quite different from each other? When we looked in more detail at the social and communication features that were different between the cluster groupings, the critical distinction was not a qualitative difference in the actual presence or absence of particular features. Instead there seemed to be a difference in the degree or severity of symptoms. Severity of impairment in this respect refers to the nature of differentiating symptoms. For example in the social domain, children in Cluster A tended to ignore others and isolate themselves whereas those in Cluster B made approaches and sought friendship, although this was unsuccessful. Yet there were behaviours that did not differ at all between Cluster B and Cluster A (e.g. problems with greeting, discrimination of persons to whom to show affection, peer friendships) and there were also behaviours that did not differ between any of the three sub-groups (e.g. impaired eye contact, recognizing another’s personal space, relationships with peers, response to others’ emotions, imaginative ability, tone of voice, non-verbal communication, idiosyncratic speech).

Next we looked at the extent to which the background variables such as language delay and current language level discriminated the cluster groups. We found that language delay did not separate the groups. This was a surprising result as language delay had previously been considered to be as a candidate feature for differentiating Asperger syndrome from autism. What about current language ability? Here the result was quite different. There was a strong difference between the clusters in terms of current verbal ability (vocabulary comprehension) factors. Children in the A group had a lower verbal ability/mental age than Children in B or C. Again this was an interesting
finding given that the cluster groups were not selected on the basis of their mental age but groups were empirically driven on basis of a range of behaviours. Given that Cluster A children had a lower level of current language, this might account for some of the social-communication differences between this group and Cluster B.

To summarise, the cluster analysis produced three cluster groups that differed in their patterns of behaviour. These clusters did not map perfectly onto the original clinical diagnosis. The critical difference between them relied on the severity of diagnostic behaviours rather than absolute differences and the groupings were differentiated by current verbal mental age but not by language delay.

As mentioned earlier, our study aimed to investigate whether particular behavioural symptom profiles in autism might link to the mentalising deficit identified in earlier studies. If so, and this cognitive ability could meaningfully differentiate certain groups of children, then this might be an important marker for clinical subtypes and account for why some children in the autistic spectrum pass while others fail tests of ‘mentalising’. All the children in the sample were given a standard experimental task of ‘mentalising’ (also known as ‘theory of mind’ tests). These tests assessed children’s ability to infer another person’s mental state.

The results showed that children in Cluster A were significantly less likely to pass tests of theory of mind than those in Clusters B or C. Does this then mean that theory of mind provides a reliable marker for subtypes of pervasive developmental disorder (PDD)? Before drawing this conclusion it is important to remember that the groups could be differentiated not only on their theory of mind skills but also on the basis of their current verbal mental age. So one possibility is that there are not one but two markers for subgrouping – theory of mind ability and verbal ability. Each would provide independent markers for the subgrouping. However, our own research and the research of others in the field, shows that theory of mind (mentalising) and verbal ability are themselves strongly related to each other. So if there should be just one marker of the subgroupings, which one should it be? This question has been widely debated in the literature (see Garfield, Peterson and Perry 2001 for discussion). Many now make the case that language experience is critical for theory of mind ability although the direction of the causal relationship is not fully understood. On the basis of the data from this study, we argue that theory of mind is not a specific marker for subtypes of pervasive developmental disorder (PDD). Instead, given the overall picture of functioning found in these children we suggest that language competence has a general influence on all domains of functioning of which mentalising ability is one.

What then should we conclude from this study of subgroupings of the autistic spectrum and the connection between mentalising and behavioural profiles? Our results showed that it is possible to empirically subdivide the autistic spectrum into separate subgroups. However it seems that these subgroupings do not represent distinct, all or none categories but can be distinguished on the basis of severity of social and cognitive impairment and verbal ability and theory of mind and it is suggested that these abilities
are linked. So we would argue that the best way to see the subgroupings is as within a
spectrum of autistic disorders with severity of social and cognitive impairment being the
primary basis for group differences. This is not to say that meaningful groupings cannot
be identified where it is helpful to do so. As Prior (2003), points out ‘the label so often
depends on the mental lens through which the labeler looks’ p. 296. We simply
conclude that given the graded nature of severity and the number of exceptions appear-
ing when subcategorizations are applied, that it is usually more sensible to see the sub-
groups as part of a wider autistic spectrum.

THE BEHAVIOURAL AND DEVELOPMENTAL PROFILES OF INDIVIDUALS
WITH ASPERGER SYNDROME

In the study of high-functioning children described above, we found three empirically
derived subgroupings. These subgroupings did not map directly onto the clinical
groupings identified by clinicians. Furthermore, when we applied the behavioural and
developmental criteria for Asperger syndrome defined by the official international clas-
sification systems of DSM-IV (American Psychiatric Association 1994) and ICD-10
(World Health Organization 1993), we found that some official behavioural and devel-
opmental criteria did not fit our subgroupings at all. For example, ICD-10 research
criteria for Asperger syndrome specifies that children with Asperger syndrome have ‘no
clinically significant delay in spoken or receptive language or cognitive development’. Yet
language delay did not differentiate any of the cluster groupings in our study.

Could the DSM-IV and ICD-10 criteria for Asperger syndrome be wrong? The
clinical experience of members of our team together with the findings from the research
study gave us good reason to question the validity of these criteria. There were two
aspects of the criteria in particular that did not seem to fit the clinical picture. The first
was the requirement for normal cognitive development and lack of developmental delay.
This means that the child is required to have normal cognitive functioning currently and
should not have had any delays in the acquisition of single words or phrases nor any
delays in their self-help and adaptive skills. The second was the absence of any criteria
requiring impairment in current speech and language suggesting that the child might
have no abnormalities in their use of communication or language. Neither of these
requirements seems to fit Asperger’s original descriptions.

Asperger (1994/1991) himself did not lay down diagnostic criteria but amongst
his descriptions he did describe a range of abnormalities of language including odd
intonation, inappropriate use of speech and pronoun reversal. Also in two case histories
he described children who were delayed in starting to speak. Although he described
their high intelligence, he noted that clinical picture could be found in those with poor
ability, even in children with severe mental retardation. The official DSM/ICD criteria
therefore seemed to be describing a different sub-group than the group that Asperger
originally described.
We wondered, as did other researchers at the time, what would happen if the official criteria for DSM-IV and ICD-10 were to be literally applied? How many cases of Asperger syndrome would we expect to see amongst children referred to clinicians with suspected autistic spectrum disorders if these official criteria were used? We attempted to answer this question in our next study (Leekam et al. 2000a). We started out on this study in the mid-1990s when the specific ICD-10 and DSM-IV criteria for Asperger syndrome were still new and just beginning to be adopted by clinicians. We wanted to know how useful these precise criteria would actually be if applied in the way that was intended by these international classification systems. As ICD-10 and DSM-IV criteria are similar, the criteria we worked with for this study were the ICD-10 diagnostic criteria for research (World Health Organization 1993).

The study had another aim. This was to compare the diagnostic criteria of ICD-10 with a different set of criteria proposed by Gillberg (1991) that more closely resembled Asperger’s own descriptions. Although other criteria for Asperger syndrome also existed at this time (Tantam 1988; Szatmari, Brenner and Nagy 1989), we chose the Gillberg criteria because these criteria included all the behavioural features covered by two other systems and because the only prevalence study at the time had used Gillberg’s criteria (Ehlers and Gillberg 1993). A few adaptations were made to the Gillberg criteria in order to fit them more closely with Aperger’s own original descriptions.

The adapted Gillberg criteria differed from the ICD-10 criteria in two important ways. The first concerned the ICD-10 requirement for lack of developmental delay. The adapted Gillberg’s criteria did not include this requirement. This meant that unlike ICD-10, Gillberg’s Asperger syndrome could be diagnosed even in cases where an individual had early language delay or had language levels below their chronological age or had a low IQ.

The second difference between the ICD-10 criteria and Gillberg’s criteria relates to the current speech and language behaviour. For ICD-10, the diagnostic criteria for Asperger syndrome include no criterion for language and communication deficits. In contrast, Gillberg’s criteria for Asperger syndrome includes a category specifically devoted to abnormalities of speech and language. This category includes the following items – lack of appreciation of humour, interprets language literally, non-reciprocal communication, long-winded speech, odd tone of voice and uses different voices for no reason. To obtain a diagnosis of Asperger syndrome according to the Gillberg criteria you must have at least three of these impairments in speech and language.

Gillberg’s criteria then rely on current behaviours rather than on developmental or ability variables. The criteria consist of six categories of impairment. These include speech and language peculiarities (described above), social interaction (odd quality of social interaction, lack of empathy, poor peer interaction, inappropriate behaviour in public), narrow interests (limited pattern of activities, repetitive routines, collects facts), repetitive routines (rituals, talks repetitively), non-verbal communication problems (body language, facial expression, staring) and motor clumsiness (gross, gait, games,
hand–eye, head bowed, fine motor). The Gillberg criteria therefore include more numerous criteria for current behaviour overall as well as more specific criteria that can identify problems such as motor clumsiness.

For this study, the clinicians L. Wing and J. Gould obtained detailed information from parents or other carers over a four-year period as part of their assessment process at the National Autistic Society’s Centre for Social and Communication Disorders. Almost all of the referrals to this Centre have disorders in the autistic spectrum. Information was collected using the Diagnostic Interview for Social and Communication Disorders (the DISCO), a semi-structured interview schedule that elicits information on more than 300 aspects of development and behaviour. The DISCO has been designed to include a set of algorithms or rules that correspond with specific diagnostic categories. These include the diagnostic criteria for pervasive developmental disorders and their subgroupings according to DSM-III-R, DSM-IV (American Psychiatric Association 1994) and ICD-10 (World Health Organization 1993) as well as Gillberg’s criteria for Asperger’s syndrome (Ehlers and Gillberg 1993), in addition to the criteria for other diagnostic systems such as Kanner’s (Kanner and Eisenberg 1956) and Wing and Gould’s (1979) criteria. The reliability of the DISCO has been established (Wing et al 2002) and its algorithms have been reported (Leekam et al. 2002).

The sample for this study consisted of 200 children and adults aged 32 months to 38 years. The complete DISCO interview was carried out, enabling information to be extracted that fitted the criteria for ICD-10 subgroups of both childhood autism and Asperger syndrome as well as Gillberg’s Asperger syndrome. Information about lack of developmental delay, a requirement for the ICD-10 diagnosis, was obtained from the DISCO items relating to toileting, self-feeding, dressing, washing, independence and response to instructions. In addition, formal psychological assessments were carried out to collect information about current IQ level and language ability.

The first question we were interested in was how many individuals would qualify for a diagnosis of Asperger’s syndrome when the ICD-10 criteria for Asperger syndrome were applied to each case. We found only three individuals who fitted the ICD-10 criteria exactly. However, not only did these individuals meet ICD-10 criteria for Asperger syndrome, they also met the criteria for either childhood autism or atypical autism. This finding appeared to justify our concerns about the official ICD-10 criteria for Asperger syndrome. This finding also supported our earlier finding that language delay does not predict different subgroups of current clinical behaviours. These findings support Szatmari’s conclusion that obtaining an Asperger syndrome diagnosis as defined by the official international classifications systems is ‘virtually impossible’ (Szatmari et al. 1995 p. 1669).

Our second aim was to compare ICD-10 and Gillberg’s criteria for Asperger syndrome. We found that in contrast to the strict criteria for ICD, Gillberg’s criteria identified 45.5 per cent of our population of 200 participants with Asperger syndrome. This is a comparable percentage to that found by other research studies (Ehlers et al. 1995).
1997). Although ability level was not itself included with the criteria, Gillberg’s criteria selected a group that tended to have better language and more intellectual interests. Nearly two-thirds of individuals with the Gillberg diagnosis had current cognitive and language levels in the age-appropriate range. However, high ability and normal development was by no means the only profile to be found. Half of the individuals had language delays in childhood, one-third had language levels below their chronological age and a quarter had low IQ.

Examination of the current behavioural items used in the Gillberg diagnosis revealed that four of the categories – repetitive behaviour, speech and language, non-verbal communication and motor clumsiness discriminated the Asperger group from the rest of the sample, while two, social impairment and narrow interests did not. The reason for this was that the incidence of these criteria was already very high in the non-Asperger group (92 per cent and 94 per cent respectively). It should be noted that all the other Gillberg criteria were also commonly found in many of the individuals who were not diagnosed with Gillberg’s Asperger syndrome. As many as 80 per cent of the group who did not qualify for a Gillberg Asperger diagnosis also had motor clumsiness and 83 per cent had odd or absent non-verbal communication.

To summarise, we found only three individuals in our large group of 200 individuals with autistic spectrum disorders who could be diagnosed with Asperger syndrome using ICD-10 criteria. So ICD-10 may create a subgroup but it is not very helpful one. Indeed Eisenmajer (1996) suggests that clinicians are not even using ICD criteria but diagnosing Asperger syndrome on basis of research and case studies. Given the inadequacy of the current diagnostic criteria to meaningfully identify Asperger syndrome and the growing view that high functioning autism and Asperger syndrome are the same thing, Mayes and Calhoun (2004) recommend that the current version of DSM-IV criteria for Asperger syndrome should be deleted. Our results suggest that the same should apply for ICD-10.

How then should we describe the behavioural and developmental profiles of individuals with Asperger syndrome? If we apply Gillberg’s criteria in contrast with the ICD-10 criteria a clearer pattern of behaviours is identified that more closely fit Asperger’s original description. The most common picture is of an intellectually able individual with repetitive routines, peculiarities in speech and language, odd, one-sided non-verbal communication and motor clumsiness. Again, this pattern indicates a mildness of degree of impairment rather than any distinct behavioural profile. Furthermore, this pattern may belong to the majority but it by no means fits all.

What does seem clear from the results of both of our studies is that early developmental history in terms of language delay and early adaptive skills cannot differentiate children with Asperger syndrome from other children with autistic spectrum disorders. While this discredits the ICD-10 criteria it is also an interesting finding in its own right. In our large sample, we found that between 80–90 per cent of all individuals with ASD had delays in adaptive functioning skills such as self-help, curiosity and fetching and
carrying and 60 per cent had delays in language. What is remarkable is that delays in self-help and adaptive skills and in language acquisition are so prevalent even in children with fluent language and high ability. This lack of motivation to carry out self-care skills that other people will do for you and the lack of curiosity in a wider sense may be an important part of the developmental picture for children with autistic spectrum disorders. These kinds of problem may also be some of the most difficult to change as Howlin (2000) notes in her follow-up study on the outcomes of older individuals with autism.

**SUBGROUPINGS BASED ON CURRENT LANGUAGE, COGNITIVE AND BEHAVIOURAL PROFILES**

Both of the studies described above suggest that when you find differences between subgroups within the autistic spectrum that these are best characterized by differences in degree of current behavior. Our two studies as well as other research studies (see Mayes and Calhoun, 2003 for review) also suggest that it is not a successful strategy to subdivide groupings on the basis of early developmental variables such as language delay or early adaptive function. Groupings are not clearly distinguished on that basis. Instead, both of the studies described above and in other research have shown that behavioural groupings are best distinguished on the basis of current verbal and cognitive profiles.

Although the picture emerging from recent research is that degree of severity in terms of current behavioural features and ability level (current language and cognitive function) provides the best discriminators of subgroupings, nevertheless, even this criterion does not always apply. In our study using Gillberg’s criteria for Asperger syndrome, for example, we found that 20 per cent of individuals fitting the description for Asperger syndrome had IQs below 70 and 36 per cent had language below the level of their chronological age. Frith (2004) in tackling the question of what criterion should be used to separate Asperger syndrome from autism confirms the view that mildness of symptoms and ability level are not sufficient factors on which to separate Asperger syndrome from autism. This point is confirmed by some of the cases that Asperger himself described and by the findings of our studies reported above. In these studies we see that some individuals have learning disabilities combined with milder symptoms, while there are others who have high ability and more extreme symptoms. Therefore, although the subtyping of children with autism according to mildness of their symptoms and ability level might provide a guide for the clinician, the application of hard and fast categorization rules based on these factors will always be imprecise.

Given the difficulty that has plagued previous research attempts to identify distinct subgroupings and the conclusion that a spectrum concept best characterizes these disorders, is there any point in continuing to look for more precise criteria on which to subcategorize children within autistic spectrum? Surprisingly, despite the concerns sur-
rounding previous research, many still argue strongly for the need to identify subtypes. What distinguishes these more recent research studies however, is the attempt to break out of the circularity of attempting to describe existing subgroupings on the basis of the symptoms or ability factors that are already part of the diagnosis. These new approaches to subtyping involve research with children who have diagnoses across the autistic spectrum. The research investigations then look at patterns of behaviour and/or neuropsychological tests that are not part of the standard diagnostic picture and attempt to link them to specific patterns of neuropathology and/or to clinical characteristics of other disorders.

The case for subtyping is summarized by Szatmari (1999) and Rutter (2000). These authors argue that identifying subtypes is important in order for progress to be made in our understanding of the genetics of autism and underlying patterns of neuropathology. Tager-Flusberg and Joseph’s (2003) work for example aims to focus on aspects such as language difficulties that are not identifying features for the disorders. They then connect these aspects to patterns of brain structure or genetic markers that are found not only in autism but also in other language disorders.

In one study, Kjelgaard and Tager-Flusberg (2001) took children who had a range of autistic spectrum disorders (ASD). They looked at their language profiles using a battery of language tests that measure a range of different skills and then used MRI scanning to detect brain activation differences that might be related to these profiles. To conduct this study they first tested the children across a range of language functions. This resulted in three subgroups that had different levels of overall language ability – normal, borderline and impaired. They then tested these groups on a marker test for specific language impairment (SLI), a phonological skills task that involved assessing their ability to process language sounds. This task has not previously been linked to problems in autism. However, the authors found that this test distinguished between children with ASD who had normal language from those with impaired or borderline language. They also found that children with impaired language were poor on past tense markers (grammatical errors). This detailed examination of the language profiles therefore provided evidence to support the case that autism and SLI are overlapping disorders.

To complete their study of these subgroups, Kjelgaard and Tager-Flusberg (2001) then asked whether children with ASD and language impairment showed the same atypical brain structure as seen in children with SLI. In fact as predicted, they did find reversed asymmetry in the frontal language area, a similar finding to that found in SLI. Joseph and Tager-Flusberg (2003) suggest that dividing groups on the basis of language may also be useful for identifying genes associated with autism in cases where autism is either associated or not associated with SLI.

The usefulness of dividing groups on the basis of current language ability has also been proposed by Reitzel and Szatmari (2003). Szatmari makes the interesting claim that the traditional distinction between autism and Asperger syndrome could be turned
on its head if we consider language impairment to be separately associated with each
group. He argues that instead of Asperger syndrome being thought of as a variant of
autism proposes that autism should be thought of as a variant of Asperger syndrome in
which children have autistic spectrum disorder together with an additional handicap of
SLI. These children frequently also have mental retardation in addition to the pragmatic
language problems found in Asperger syndrome. This simple distinction may not
provide a precise conceptualization but it offers an important new way of thinking
about the problems experienced by children with autism. It is also in line with recent
changes of thinking about the similarities in language impairment across different
clinical groups (Bates 2004).

So far, I have outlined a proposal for subtyping on the basis of language impairment.
Is there also a case for subtyping on the basis of nonverbal ability also? Recently, Joseph,
Tager-Flusberg and Lord (2002) have subgrouped children in terms of their relative
rather than absolute differences in both verbal and non-verbal ability. Traditionally it
has long been claimed that people with autism have a standard psychological profile in
autism in which non-verbal ability is higher than verbal ability. Subsequent researchers
including Joseph and Tager-Flusberg have refuted this standard picture. Instead they
have shown that when children are tested on standard psychological tests, some have a
profile in which non-verbal ability is better than verbal ability, others have a profile of
verbal ability greater than non-verbal ability and some show equal ability on verbal and
non-verbal tests. Joseph and Tager-Flusberg have then explored the link between these
three profiles in the light of both behavioural symptoms and neural pathology. In terms
of the link between cognitive profile and symptoms, they found that individuals with
discrepantly high non-verbal IQ (NVIQ) relative to verbal IQ scores, have more impair-
ments in reciprocal social interaction that was greater than would be expected on the
basis of their absolute level of ability and their level of language skills.

Following this finding, the authors considered the possible neuropathology that
might be related to this unbalanced pattern of high NVIQ, low language and poor
reciprocal interaction. One proposal they make follows the developmental explanation
that this pattern of functioning could be the outcome of a different pattern of
neurocognitive development early in life. The suggestion is that isolated visuo-percep-
tual strengths may be related to increased neuronal growth or reduced connectivity in
the brain. One way of testing this would be to look at head and brain size. Joseph and
Tager-Flusberg examined head circumference and brain volume and found that the
group with discrepantly high NVIQ relative to verbal IQ had enlarged brain volume in
addition to enlarged head circumference.

Nonverbal IQ is a very general basis on which to subtype children. Should the basis
for subtyping go beyond general cognitive factors such as non-verbal IQ and include
more specific cognitive variables? Frith (2004) agued for two specific cognitive candi-
dates, mentalising difficulties and weak central coherence indicated by lack of
top-down cognitive control. The case of mentalising ability, discussed earlier, has been
used to trace impairments in brain functioning. For example, a recent study by Castelli (2002) used tests of theory of mind to demonstrate reduced activation in parts of brain considered to be the ‘social brain’ in ten individuals with autism.

While links between neural pathology and specific cognitive function are certainly suggested by these studies, whether they will provide good candidates for subtyping remains to be tested. What is not clear is whether any subtyping relating to mentalising ability would be independent or tied to subtyping based on language ability. One of the most exciting challenges that lies ahead is the task of disentangling the nature of the causal relation between theory of mind, language and social communicative symptoms of autism. We know that language is an important mediator for symptoms of autism including social interaction. We also know that mentalising ability is related to verbal ability (de Villier 2000). Further developmental research following the transactional approach of Mundy and Neal (2001) is needed to trace the links between symptoms, theory of mind and language and their neuropathological origins.

Finally, could subtyping of the autistic spectrum rely on specific behavioural rather than cognitive or neural markers? Recently our own research has focused on specific behavioural symptoms that are not part of required criteria for autism but like the impairments in language are showing themselves to be strongly associated with a proportion of high-functioning individuals on the autistic spectrum. It is expected that these aspects of impairment will also show neuropathological markers, although research on this is yet to be conducted.

One pervasive problem is the difficulty with sensory responsiveness. Several studies show that this might form a potential marker for the social and communication problems in autism warranting further investigation along the lines taken by Joseph and Tager-Flusberg by looking at the patterns of dysfunction associated with this problem. In the first study of high-functioning children, described earlier in this paper (Prior et al. 1998), we found three different empirically derived subgroupings when we put children’s diagnostic features into a cluster analysis. Clusters A and B differed from Cluster C by being more likely to have unusual sensory responses. Clusters A and B, however, differed from each other to some extent by their degree and awareness of social interactional problems. This finding suggests that meaningful subgroups might be based on the combination of sensory and social interactional features.

In a new study with colleague Carmen Nieto from Spain (Leekam et al. under review), we have been able to make a more detailed description of both the behavioural and the developmental characteristics that are associated with these sensory impairments. First, we collected details from parents using the Diagnostic Interview of Social and Communication Disorders (Wing et al. 2002; Leekam et al. 2002). Details were collected of abnormalities in three domains of sensory abnormalities. Proximal abnormalities (including a range of different tactile and olfactory behaviours), auditory abnormalities (including unusual responses to or fascination with particular sounds) and visual abnormalities (including unusual attention to specific aspects of visual stimuli such as
their brightness or movement). To test the degree to which sensory abnormalities might be related to language problems and ability levels we included comparison groups that had typical development, a group that has mental retardation and a group that had language impairments but no mental retardation.

The results showed that children with autism had significantly more sensory abnormalities than all these comparison groups. A particularly marked contrast was found between the high-functioning children with ASD on the one hand and children with only language impairment (SLI) or the typical development group. Approximately 85 per cent of children with autism had sensory abnormalities regardless of whether their IQ was above or below 70. Another finding was that these ASD children were more likely to have multi-modal abnormalities. That is, they were likely to have abnormalities in more than one sensory domain while comparison groups of learning disability or language impairment did not. Amongst the autism group, by far the most common abnormality was the proximal class.

Having established that a large proportion of autistic individuals have sensory abnormalities and that these abnormalities differ from comparison children and are found across several different modalities, we carried out another study with a larger sample of 200 individuals with a range of impairments across the autistic spectrum. We found that sensory abnormalities persist across age and IQ. Of the three main categories outlined above (proximal, auditory and visual) only the visual category showed significant reduction in symptoms with age and IQ. Unusual visual responses were more frequent in younger children in those with lower IQ than in older and more able individuals. In future research we plan to study the behavioural, cognitive and physiological profiles of children with and without sensory abnormalities in different modalities. These profiles may, like Joseph and Tager-Flusberg’s (2003) research, provide an interesting comparison with the profiles of children from other atypical groups such as children with visual or hearing impairment.

Another problem that provides a potential behavioural subgroup within the spectrum of autistic disorders is the occurrence of disorders of posture and movement in the autistic spectrum (Wing and Shah 2000). As Wing (2004) reports, these catatonic features affect approximately 10 per cent of adolescents and adults with autistic spectrum disorders. Like the sensory difficulties, this problem is not only found in people with low ability. ‘Autistic catatonia’ (Hare and Malone 2004) may therefore provide a meaningful subgroup within the autistic spectrum.

To summarise, research to date has shown that there is no consistent profile in which individuals with Asperger syndrome have either higher verbal ability, poorer non-verbal ability or more motor problems than individuals with autism. As Frith (2004) points out in her review of the literature genetic studies point to the same aetiology for both groups and neuroanatomical studies also show certain features of brain pathology that are present regardless of IQ. Given the common basis at both the biological and behavioural level for an autistic spectrum, the notion of differentiating sub-groups of Asperger
syndrome and autism according to the international classification systems is unjustified. However, many researchers consider that other distinctions are helpful. For example, recent attempts to subdivide the autistic spectrum according to neurocognitive profiles help to narrow down the definition of the phenotype, providing implications for future genetic and neurobiological research. These studies are providing insights into overlaps between clinical groups outside of autism and challenging previous assumptions about the separateness of different disorders. Finally, 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 association and risk factors for autism.

DEVELOPMENTAL QUESTIONS ABOUT AUTISTIC SPECTRUM DISORDERS

As discussed above, although research findings over the last ten years have undermined the idea of distinct diagnostic subgroups based on ICD-10 and DSM-IV criteria, new attempts are being made to identify subgroupings using language, cognitive and behavioural factors that are not part of the essential diagnostic picture. These studies may help us to understand better the conditions under which certain behavioural profiles might overlap with other disorders and/or be associated with biological markers. However, many of the distinctions used for sub-grouping are based on the child’s current profile of functioning. What about early development factors? Surely if we are interested in understanding how autism emerges and what the potential is for improvement, we need to understand better the how these behavioural profiles change over time.

Given that developmental considerations have been strongly highlighted in the clinical literature (Wing 1996), it is surprising that little research attention has been paid to this dimension, especially as there seem to be so many important developmental questions to answer. For example, why is it that two individuals may start off with very impaired early development, for example with delays in language and adaptive skills but then one child will improve and show few related difficulties later in childhood while the other will not? Or why is it that for some children, problems do not appear to be apparent early in development but then appear later leading to a late diagnosis? Could there have been some subtle problems in early development that were missed by parents?

The focus on these kinds of developmental questions is now gaining momentum (e.g. Charman et al. 2001; Mundy and Neale 2001) but theoretical and methodological approaches still tend to be rooted within non-developmental traditions. These traditions tend to focus on trying to uncover common underlying factors that may explain the cause of autism. Attention to developmental issues is still underrepresented compared to the study of other developmental disorders such as SLI or Williams syndrome (Bishop 1997; Karmiloff-Smith 1998). There may be many reasons for this. One reason may be that some of the research investigating developmental outcomes in
autism tends to focus on stability in outcome rather than change, although there are exceptions to this (Piven et al. 1996). In addition, a good proportion of previous research on outcomes has focused on ‘core’ autism rather than the broader autistic spectrum. Once the focus on the discrimination between autism and Asperger syndrome was no longer important, the concern has been with the broader autistic spectrum, and perhaps there should be renewed interest in the conditions under which change does or does not occur.

One of the puzzling findings that emerges from this research is that about early developmental delay. Our research findings and those of others shows that when you compare the groups who do have language delay and those who do not, you do not see any differences in these two group in terms of their autistic behaviours when they are older. These findings are supported by longitudinal research by Szatmari (2000). Szatmari found that delayed language onset did not predict later language outcomes but a language score a few years later did predict subsequent language outcome. Reitzel and Szatmari (2003) propose an important developmental explanation for these findings. They argue that until the age of three years, children with autistic spectrum disorders are not easily differentiated from each other. Once some children start to develop speech in two to three word phrases after the age of three however, they are able to use this as an opportunity to acquire better social and communication skills and academic skills. Even children who develop this ability after three years can still take advantage of what these newly acquired language skills have to offer later on.

Could this also help to account for an unusual finding by Eisenmajer et al. 1998? Richard Eisenmajer followed up the cluster analysis study described earlier and divided the children into two groups, according to whether they had language delay or did not. He then compared these two groups in terms of whether the language delay group would have a different pattern of autistic symptoms than the normal language onset group. He also looked at whether other developmental history variables such as delay in reaching motor milestones. There were two main findings. The first was that children who had language delays also had other general developmental delay in achieving their motor milestones and lower receptive language abilities. The second was that language delayed children did not differ in their autistic behaviours from the non-language delayed children except if you took into account the age at which they were diagnosed. The children who had language delay and who were diagnosed earlier (before six years) had more autistic symptoms than the non-language delay group. However, those who had language delay and were also diagnosed at an older age (after six years) showed no difference in behavioural profile to the non-language delay group.

What does this result mean? First, it is important to remember that many children in this large sample (about half of them) had language delay. Second, if we assume that children in the early diagnosis group had more autistic features early in life as well as later, it is likely that problems related to these features combined with their language delay and led them getting their early diagnosis. What kinds of early associated difficul-
ties might put this particular language delay group at greater risk for a poor outcome than the language delay group who were picked up for diagnosis later? From the data we have analysed so far, it is not clear. One would expect that delays in language would be related to other basic delays in developmental milestones such as delayed toileting or self-help skills. However this is not the case. Our Asperger syndrome study revealed that as many as 80–90 per cent have these kinds of problems. This means that even children without language delay have these kinds of developmental delays.

Another contender for the difference in developmental trajectory for the two language delay groups may be the difference in early non-verbal communication skills, especially in joint attention. Joint attention refers to the ability to coordinate attention between people and objects and is strongly related to language ability in typically developing children. Joint attention ability is also believed to be related to the development of theory of mind. If children do not show these early signs of non-verbal interaction and also have language delay they might be more likely to be referred for an early diagnosis.

Sigman and Ruskin (1999) carried out one of the few longitudinal studies of autism to follow up children’s joint attention and other problems. Sigman and Ruskin followed up 51 of 70 children who had been diagnosed at age three to five years. The children were seen eight to nine years after their original diagnosis. Sigman and Ruskin predicted that children who showed more joint attention and symbolic play would make greater gains in language. This is what they found. Long term gains in language were predicted by responsiveness of others’ bid for joint attention and by play. Joint attention ability was also related to changes in IQ amongst a subset who moved across the borderline from mental retardation to non-mental retardation. The authors found that individual differences in joint attention and play and emotional responsiveness has consequences for later development. If more non-verbal communication is shown early in development then more language is also shown later in development. Even more specifically, if the child is able to follow gaze early then they will make more long-term gains in language. Interestingly the impact of joint attention on later development is independent of early intelligence though this clearly helps. Early joint attention also predicts greater social engagement with peers.

We have also been carrying out a series of studies on joint attention in children with autism. Our early work took children of different ages and verbal abilities and we tested gaze-following experimentally and observationally. We found very strong developmental effects that were related to verbal mental age (Leekam, Hunnisett and Moore 1998). Subsequently we worked with preschool children and again found that the problem was largely confined to the children with greatest cognitive and language delay - when compared with non-autistic developmentally delayed children their problems were extremely marked (Leekam, Lopez and Moore 2000b). Our most recent research shows that even when you take an extremely basic element of social interaction - social orienting in response to name call – this is highly associated with language ability (Leekam
and Ramsden, in press). Furthermore, this association is more strongly associated in the autism group than in the non-autism group.

It is difficult to explain in a simple way the different developmental trajectories that might appear as a consequence of early delays and difficulties. One idea we are working on relates to the bi-directionality of language and joint attention development (Leekam 2005). This proposal is that as language improves, children can use it to build their non-verbal skills. Another idea is that there is a dynamic relationship between sensory experience and social interaction. We suggest that early sensory difficulties may restrict opportunities for the infant to develop both social and sensory responsiveness. For example, a restricted response when hearing a voice does not facilitate the infant to come into visual contact with that person which further restricts the opportunity for further engagement and thereby does not provide further sensation that is socially modulated (Leekam and Wyver 2004). This explanation might help to account for common patterns of associated impairments in sensory responsiveness, social impairments and adaptive self-directed behaviour.

To summarise, the answers to many developmental questions only become possible when we start to move away from the focus on prescribed subgroups and think about development. One question that still requires an answer is why one child with developmental delay goes on to develop more severe autistic impairments while another does not. Future longitudinal studies that trace developmental changes in infants with language and other developmental delays may help to identify the risk factors and critical points of timing that lead children towards different outcomes.

CONCLUSION
My research story started with a visit to a child psychiatrist 20 years ago. Progress in understanding Asperger syndrome has come a long way since then. Public recognition of Asperger Syndrome has promoted awareness of the broader concept of a spectrum of disorders, with milder edges forming a continuum with normal functioning. This concept of an autistic spectrum (Wing 1996) has helped to change our thinking because it challenges the idea about rigid categories that separate distinct psychological conditions, influencing the way that other conditions are thought about, for example, forms of specific language impairment and attention deficit disorder. In terms of education and management, this awareness of milder forms of autism has also led to greater understanding and support for people like Ivan that would not have been possible years ago.

Looking back over the years, Ivan had so many unmistakeable behavioural features of autism, that despite his good language, it is inconceivable that his diagnosis would have been missed if he were a young child today. Yet even now, many children who come to receive a diagnosis of Asperger syndrome still do not have their condition recognized until later in childhood or even adulthood. This is ironic because these are the children who may have the greatest prospects for learning and improvement. Hopefully, accep-
tance of the broader concept of the autistic spectrum, together with an increased focus on developmental issues may enable us not only to identify these children better but also to assist more effectively in their potential for development.

REFERENCES


