Diagnosing Autism Spectrum Disorders in Children:

Medical and Social Perspectives

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Abstract

In this submission, five articles are presented examining one theme: diagnosis of autism spectrum disorders (ASD) in children. Three articles provides perspectives on various social and medical factors that influence the diagnosis of ASD, and the others examine social and behavioural outcomes for children diagnosed with ASD. One article provides an in depth examination of the dilemmas of diagnosis from a parental perspective.

The research utilized both qualitative and quantitative methods. A secondary analysis of a longitudinal birth cohort study revealed that there were a number of children who had autistic traits equally severe as those with clinical diagnosis. Further analysis exposed a possible gender bias in diagnosis. Outcomes for children with ASD diagnoses were worse than for those without diagnoses but with comparable behaviours as preschoolers. ASD diagnosis apparently had no positive effect on the developmental trajectory of prosocial behaviour. The implications of these results are discussed.

Analysis of qualitative data collected in semi-structured interviews with parents of both diagnosed and undiagnosed children exposed dilemmas faced by parents as they contemplated an ASD diagnosis and highlighted parental action to de-stigmatise the condition after diagnosis had been applied.

The body of work as a whole falls at the junction of clinical and educational psychology, developmental psychology, social psychology, social psychiatry, sociology and epidemiology. It draws attention to a number of social processes that contribute to ASD diagnosis. Overall, it is argued, the work supports the conceptualisation of ASD as both a biologically and socially determined condition.

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Abbreviations

	Abbreviation
Autism Spectrum Disorder	ASD
International Classification of Diseases	ICD
Diagnostic and Statistical Manual of Mental Disorders	DSM
World Health Organisation	WHO
Asperger's Syndrome	AS
The Avon Longitudinal Study of Parents and Children	ALSPAC
Intelligence Quotient	IQ
Pervasive Developmental Disorder	PDD
Not Otherwise Specified	NOS
Attention Deficit Hyperactivity Disorder	ADHD
Fragile X Mental Retardation – 1	FMR1
Deoxyribose Nucleic Acid	DNA
National Autistic Society	NAS
National Institute of Health	NIH

United Kingdom	UK
United States (of America)	US(A)
American Academy of Pediatrics	AAP
Child and Adolescent Mental Health Service	CAMHS
Autistic Rights Movement	ARM
Applied Behaviour Analysis	ABA
Special Educational Needs	SEN
Chronic Fatigue Syndrome	CFS
Autism Diagnostic Interview	ADI
Picture Exchange Communication System	PECS
Special Educational Needs Coordinator	SENCO
Acquired Immune Deficiency Syndrome	AIDS
Self Fulfilling Prophecy	SFP
Socio-economic Status	SES
Strengths and Difficulties Questionnaire	SDQ
Analysis of Variance	ANOVA
Autism Diagnostic Observation Schedule	ADOS
Randomised Controlled Trial	RCT

Author's declaration

The candidate's contribution to co-authored papers:

All articles: Conception, research design, analysis, reporting, draft writing, revision, literature review. Article 5: Data collection, in addition to above.

Contribution of co-authors

Article 1:

JG**:** Research design, revision for important intellectual content.

CS: Research design, analysis.

TF: Revision for important intellectual content.

Article 2:

JG**:** Research design, revision for important intellectual content.

CS: Research design, checking analysis.

Article 3:

JG**:** Research design, revision for important intellectual content.

CS: Research design, analysis.

BN: Revision for important intellectual content.

AE: Revision for important intellectual content.

TF: Revision for important intellectual content.

Article 4:

JG**:** Research design, revision for important intellectual content.

CS: Research design, analysis.

TF: Revision for important intellectual content.

Article 5:

BN: Suggested revisions for important intellectual content, checking analysis.

Introduction

Section 1: Key themes covered in the collection of articles

The autism spectrum encompasses a wide range of behaviours varying from subtle problems of understanding and social function to severe disabilities (Baird, Cass, & Slonims, 2003). Diagnosis can be made in adult life, but normally the condition is identified in childhood. This submission encompasses an examination of social outcomes for children with autistic traits both with and without an ASD diagnosis. It also reports on the discourses deployed by parents of autistic children to justify or avoid ASD diagnosis. This submission as a whole is focused therefore on diagnosis of autism spectrum disorders in children, and specifically the effect of social and biomedical factors on diagnosis. Although the work resulted in several discrete publications, each dealing with a different aspect of this theme, it was originally conceived as a coherent thesis, addressing debates between biomedical and sociological perspectives, which are described in more detail in the following sections.

To be diagnosed with an autism spectrum disorder (ASD) children must display a number of impaired behaviours from each of three behavioural domains (Bailey, Phillips, & Rutter, 1996). The core symptoms of autism are impairments in social interaction, impairments in communication and impaired imagination, expressed as restricted or repetitive behaviour. This is often referred to as the so called 'triad' of behaviours (Wing & Gould, 1979). The diagnostic criteria most often used in clinical practice are the most recent specified by the International Classification of Diseases (ICD-10), and Diagnostic and Statistical Manual of Mental Disorders (DSM-IV) (World Health Organisation [WHO], 1992; American Psychiatric Association, 1994). The prevalence of ASD has increased dramatically (Fombonne, 2003; Fombonne, Misès, Jousselme, Fujiura, & Guralnick, 2005). Forty-five years ago, it was estimated that approximately 0.04% of children in a UK population displayed autistic behaviours (Lotter, 1966); current estimates are as high as 1% (Baird et al., 2006; Brugha et al., 2008). In the UK, an ASD diagnosis must be assigned by a clinician, usually a child psychiatrist or paediatrician, ideally informed by a multidisciplinary team (The National Autism Plan, 2003; Scottish Intercollegiate Guidelines Network, 2007). Although the diagnostic criteria are standardised, clinical practice varies between practitioners (Skellern, McDowell, & Schluter, 2005) and at regional levels (Bebbington & Beecham, 2007).

ASD is classified by both sources of the diagnostic criteria as a subtype of pervasive developmental disorder. Here, 'pervasive' refers to the fact that multiple domains of development are affected; normally in psychiatry, pervasive means not limited to one situation e.g. both at home and at school (Goodman & Scott, 1997). Of the two sources, ICD is the most commonly used classification system in the UK and has been used throughout this research as a reference point.

The findings of several studies have highlighted a disparity between the prevalence of childhood psychiatric disorders recorded by researchers and specifically autistic spectrum disorders and their recognition as defined by receiving a clinical diagnosis (e.g. Baron-Cohen et al., 2009; Honda, Shimizu, Misumi, Niimi, & Ohashi, 1996; Kadesjö, Gillberg, & Hagberg, 1999; Yeargin-Allsopp et al., 2003). This tradition of work forms a key theme in the thesis. Recent studies with very high estimates of the prevalence of ASD in the UK have included children whose symptoms are documented as reaching clinical thresholds, but have no formal diagnosis (e.g. Baron-Cohen et al., 2009). There are very few empirical studies looking at the effectiveness of the package of services for childhood psychiatric disorders used in the community through use of matched controls e.g. by identifying undiagnosed groups with comparable behaviours, such as those included in prevalence estimates (Angold, Costello, Burns, Erkanli, & Farmer, 2000; Lambert & Bickman, 2004). In psychiatric research generally, prevalence literature indicates that there is a substantial minority of undiagnosed children whose behaviour meets diagnostic thresholds for psychiatric disorders (Ford, Goodman & Meltzer, 2003; Goodman & Scott, 1997), but who have never received a diagnosis because their condition fails to have sufficient impact on their families (Rutter, Tizard, Yule, Graham, & Whitmore, 1976).

Judging whether a child's behaviour adds up to a disorder rather than a normal variant involves a consideration of the symptoms' impact (WHO, 1992). Assessing impact should take both distress for the child and disruption for others into account (Goodman & Scott, 1997). Previous versions of the diagnostic criteria resulted in overestimates of the prevalence of child psychiatric disorders in ethnic cultures, as many children reached diagnostic thresholds but their behaviour did not appear to pose a major problem within

their own cultures (e.g. Bird et al., 1988). Epidemiological studies have indicated that most children do not get referred to health services for psychiatric disorders because families themselves are not concerned about symptoms (Burns et al., 1995; Fergusson, Horwood, & Lynskey, 1993; Leaf et al., 1996).

Aside from the issue of 'impact', discrepancies in wide prevalence estimates for ASD have been found as a function of regional variation. This occurs on a country by country basis, as well as at local levels. Liu and colleagues (2010), for example, found ASD diagnoses were geographically clustered in California, and put this down to social influences. Such social influences form a second key theme in this thesis. The submitted research sought to establish whether parents took an active role in the decision to diagnose children with ASD. The process of weighing up the benefits versus the disadvantages of receiving a diagnosis has been characterized as a dilemma in the research literature (Norwich, 2008, 2009). ASD has been conceptualized in popular representations as a 'male' disorder (Murray, 2008). Recent work has suggested that high levels of foetal testosterone may be linked to the development of autistic traits (Ingudomnukul, Baron-Cohen, Wheelwright, & Knickmeyer, 2007). This has been interpreted as concurring with Baron-Cohen's well known 'extreme male brain' theory of autism (2002).

In addition to issues around undiagnosed children and social influences on ASD diagnosis, another major theme of this thesis concerns tensions between sociological and biomedical perspectives on ASD diagnosis. The biomedical perspective which is generally accepted within the medical literature, within organisations dedicated to the interests of those with ASD, in policy guidelines, and by most parents of autistic children is that ASD is a neurological disorder which is partially genetically determined, and diagnosis should be made as early as possible to treat and manage the condition effectively (Chakrabarti & Fombonne, 2005; Charman & Baird, 2002; Charman, 2003; Landa, Holman, O'Neill, & Stuart, 2011). ASD diagnosis is generally promoted by autism charities, health bodies and the educational institutions. Education professionals often advocate diagnosis because it may be used to secure additional educational resources and support for children. Health institutions have also promoted diagnosis, and particularly early diagnosis. The stated rationale is often that diagnosis will lead to appropriate 'treatment'. The aim of treatment, according to the American Academy of Pediatrics (AAP), is to promote the development of

communication, social, adaptive, behavioural and academic skills as well as lessening maladaptive and repetitive behaviours. In other words, treatment should target core autism symptoms:

> Currently accepted strategies are to improve the overall functional status of the child by enrolling the child in an appropriate and intensive early intervention program that promotes development of communication, social, adaptive, behavioral, and academic skills; decrease maladaptive and repetitive behaviors through use of behavioral and sometimes pharmacologic strategies.
> (Committee on Children with Disabilities, 2001, p.1223).

On the other hand, sociologists, autistic adults and some parents argue that ASD is in part socially constructed and that a shift is required in the norms and values of society that deem autistic behaviours abnormal (Nadesan, 2005; Baggs, 2007). Labelling theory as developed by Becker (1963; 1967) and Scheff (1974) suggests that a medical diagnosis might lead to stigmatisation and a self fulfilling prophecy causing an exacerbation of the condition. As Nadesan (2005, p.8) puts it, children will 'inhabit the identities they have been ascribed' and this will serve to reinforce the 'abnormal' behaviour in question. Once told about a specific childhood diagnosis, it has been shown that perceivers tend to interpret behaviour in the light of the diagnostic frame (Fogel & Nelson, 1983; Rosenthal & Jacobson, 1966).

Perhaps, as argued by some sociologists, such as Rabinow (2005), the forging of a social identity is the most fundamental concept for galvanising responses to biomedical models. Various identities have coalesced around ASD, both predominantly led by parent groups as well as autistic self advocates. These groups assert their political claims on the basis of their 'autistic' identities. One of the defining characteristics of such social movements is how participants become aware of their own role in defining the affected individuals as a group, in the ASD case, as 'autistic' people. This is an important precursor for political mobilisation according to Epstein (1995).

It is the tensions between biomedical conceptualizations of ASD and its diagnosis, and various 'sociological' perspectives (for example, those of labeling theorists, social constructionists and autistic activists) that informed this thesis as it was originally

conceived. A review of the literature pertaining to these perspectives as they relate to the overarching subject of ASD diagnosis is presented in the following section.

Section 2: Literature review related to key themes; current and historical perspectives on ASD diagnosis

Current criteria for diagnosing autism spectrum disorders

Children on the autism spectrum exhibit impairments in three behavioural domains. These are qualitative impairments in social interaction, for instance, poor use of nonverbal behaviours such as eye contact, facial expression, body postures, and gestures. They may also demonstrate a failure to develop appropriate peer relationships, and a lack of social sharing or reciprocity. Secondly, autistic children display impairments in communication, such as a delay in development of, or total lack of, spoken language. In those who do develop adequate speech, there may be no ability to initiate or sustain a conversation, and they may use stereotyped or idiosyncratic language. Finally autistic people also exhibit restricted, repetitive and stereotyped patterns of behaviours, interests, and activities, which include abnormal preoccupations with certain activities and inflexible adherence to routines or rituals. In addition to these core difficulties, many autistic people have difficulties integrating sensory information which leads to hypersensitivity or hyposensitivity. They may experience over-excitement or an aversive reaction in the presence of certain sensory stimuli (Bogdashina, 2003). Most people with typical development are able to filter out irrelevant stimuli in order to focus on particular matters, but many people with ASD are not able to do this. They might find each sound equally significant, whether human voices or car engines running.

Judging whether these behaviours add up to a disorder rather than a normal variant involves a consideration of the symptoms' impact. Assessing impact should take both distress for the child and disruption for others into account. Previous versions of the diagnostic criteria resulted in overestimates of the prevalence of child psychiatric disorders in alien cultures, as many children reached diagnostic thresholds but their behaviour didn't appear to pose a major problem within their native cultures (Bird et al., 1988). Thus a behaviour that may appear to be a problem in one culture or situation may not be problematic in another. Such considerations lead to the adoption of impact as a diagnostic criterion for ASD and other conditions in the latest versions of ICD and DSM; ICD-10 and DSM-IV.

Within the range of behaviours that are classified as ASD there is a huge variation in the severity and persistence of symptoms. Furthermore, the type of symptoms presented can also differ enormously (Table 1).

Table 1: Range of behaviour in ASD (adapted from Baird, Cass & Slonims, 2003).

| | Presentation can range ... | |
	From…	to…
Diagnostic criteria:		
Social interaction	Aloof and indifferent	Makes one sided approaches
Social communication	No communication	Spontaneous but repetitive, one sided
Repetitive behaviour	Simple, bodily directed e.g. face tapping, self injury	Verbal abstract e.g. repetitive questioning
Other characteristics:		
Formal language	No language	Long winded repetitive literal interpretations
Responses to sensory information	Very marked	Minimal or no unusual responses
Unusual movements e.g. hand flapping	Very marked	Minimal or absent
Special skills e.g. rote memory	One skill at high level, very different from other abilities.	Absent

The most common forms of ASD are listed in ICD-10 as autism, Asperger's syndrome (AS), and atypical autism, known as pervasive developmental disorder not otherwise specified (PDD-NOS) in DSM-IV. Both sources of diagnostic criteria include Rett's syndrome and childhood disintegrative disorder, which have similar symptoms to autism but have unrelated causes (Lord, Cook, Leventhal, & Amaral, 2000). The film *Rainman* and the subsequent plethora of autistic autobiographies, novels and popular documentaries have

raised public awareness of autistic individuals and particularly those with savant skills. However savant skills are only evident in an estimated 0.5% to 10% of those with ASD (Treffert, 2007). In 2002, Lainhart et al. stated that approximately 20% of children with ASD have regressive autism; that is they appear to have relatively normal development during the first 12 to 24 months of life. This period of relative normality gradually or suddenly ends and is followed by a period of regression, characterized most prominently by loss of language skills, after which autism becomes evident. The syndrome has also been divided into low-, medium- and high-functioning autism based on intelligence quotient (IQ) thresholds, or on how much support the individual requires in daily life, but these subdivisions are not standardized (Baron-Cohen, 2006).

The classification systems of mental disorders that are used today, the ICD and DSM, were originally developed from the work of the German psychiatrist Emil Kraepelin. Kraepelin adopted a biomedical model which assumed there were a discrete and discoverable number of psychiatric disorders. Although he recognised that symptoms can occur in more than one disorder he argued each disorder has a typical symptom picture with distinct psychopathology and aetiology. A line could be drawn between mental illness and normal behaviour. It was under this influence that the early versions of the classification schemes were drawn up.

The clinical psychologist Richard Bentall delivers a stinging critique of this system in his book *Madness Explained* (Bentall, 2004). He points out that as definitions of disorders became more established, the American Psychiatric Association (APA) came under increasing pressure to develop standardised psychiatric classifications. This pressure came from insurers (medical insurance only treated bona-fide conditions) and pharmaceutical regulators who wanted psychiatrists to specify exactly who their drugs would treat, as well as from those within the medical establishment. Bentall demonstrates that the two classification systems produce varying numbers of diagnoses, and that cross cultural interpretations of the classification systems also lead to wide discrepancies. Many children who have psychiatric disorders fail to meet the full criteria for a diagnosis and have to be given the 'Not Otherwise Specified' (NOS) or 'atypical' diagnosis. Bentall contends that to make DSM exhaustive, the number of definitions in successive editions of the manual was increased, and catch-all 'NOS' definitions were used to sweep up anyone who did not fit

the criteria for a specific disorder. Children who have pronounced autistic features but fall short of the full criteria can be diagnosed in this group, thus the NOS and atypical diagnoses provide a way for diagnosis to be made even when clinicians are unsure of specific categories. The two sets of criteria also be used flexibly: within behavioural domains, if one criteria is not met, others can be substituted, introducing another layer of uncertainty into the process. Molloy and Vasil (2002) claim that inclusion of any diagnostic category in DSM gives it 'official' status whilst masking the fact that a clear consensus on what it actually is may not exist. For this reason, exclusion rules were introduced to make sure people do not fall into more than one category. These mean that the diagnostic categories include the caveat the patient is 'not better described by condition X Y or Z', thus preventing comorbidity from becoming apparent. Bentall argues that strong associations between schizophrenia, mania and depression occur because these diagnoses do not in reality describe separate disorders.

Thus the criteria and their classification into ASD can be seen as pragmatic. The diagnostic criteria themselves and their classification into the larger picture are always in a state of flux. As Goodman and Scott point out in their seminal book *Child Psychiatry* (1997):

> Fashion continues to be important in classification, and there are likely to be minor and major revisions in the schemes for many years yet ….Our current ideas are like maps of largely unexplored territory- better than nothing provided you don't take the details too seriously (p. 26).

Hence the definition of ASD is itself considered by theoretically inclined psychiatrists as 'work in progress'. However, when used in practice, the diagnostic criteria are presented as scientific, and cautions are often held back (Goodman & Scott, 1997).

A brief history of autism
Before the twentieth century most individuals with autistic traits, it is thought, were known as 'feeble-minded' and other frequently used terms such as 'idiots' and 'imbeciles'. Many of these individuals were locked up in institutions.

Henry Maudsley's nineteenth century edition of *The Physiology and Pathology of the Mind*, may have recorded a thirteen year old boy who had Asperger's syndrome (Maudsley, 1868). There is also the well documented case of Victor, 'the wild boy of Aveyron', found as a feral child in 1798 (Wolff, 2004). Victor displayed several traits consistent with autism, and his carer attempted to teach him to speak. Frith's (2003) analysis of the court records of Hugh Blair, from 1747, claims that he displayed many symptoms of autism. The implication of researchers citing references to autistic individuals from historical texts is that the condition has always been present in society, even though it was named relatively recently. This position does not appear conclusive. There seem to be very few accounts of autism-like symptoms although Frith points out that identification via historical texts is very difficult.

At the start of the twentieth century health workers started to look at abnormal child development. They attempted to name sub-categories within the main diagnosis that was used at the time, 'childhood psychosis'. Later in the century, children who developed language and social skills and then lost them were described; children whose symptoms would today be described as regressive autism (Hulse & Heller, 1954).

In 1938, Hans Asperger first used the term 'autism' in a lecture, and subsequently published his description of patients with Aspergers syndrome in 1944. The psychiatrist Leo Kanner used the term 'autism' in a report in 1943. His description of autistic children is still relevant; his description of 'insistence on sameness' and 'autistic aloneness' are still used to diagnose the disorders. He also defined it as an innate inability to form the usual 'biologically provided' affective contact with people. It is not known whether Kanner and Asperger used the term entirely independently of each other (Lyons & Fitzgerald, 2007).

By the mid to late twentieth century, Bruno Bettelheim (1972) and many researchers in psychology interpreted autistic behaviour in Freudian terms. Child psychiatry subsequently focussed on conceptions of autism as an infant's response to a cold and distant 'refrigerator mother'. This blaming lead to intense guilt amongst mothers (Nadesan, 2005). New frameworks for understanding the condition emerged in the 1960s; the neurologist Bernard Rimland (1964) explained infantile autism as a neurological disorder with a genetic component. In the late 1970s, the first monozygotic twin study was published showing a

high degree of heritability of autistic traits (Folstein & Rutter, 1977). Results were consistently replicated by other studies. Today ASD is thought to be one of the most heritable of all psychiatric conditions (Szatmari & Jones, 2007). The rise of cognitive psychology which has become dominant over psychoanalytic models has shifted the conceptualisation of autism to a deficit in cognitive processing, located in neural mechanisms. By the mid 1990s, and since then, most psychiatrists and other professional practitioners in the developed world have considered ASD to be a neurological disorder with a strong genetic component.

This conceptualisation of ASD has its roots in the biomedical understandings developed by Kraepelin. Wing and Gould (1979) identified a triad of impairments in three key areas. These were in *social communication*; e.g. lack of eye contact, in *social understanding;* e.g. understanding of social norms of the group and in *imagination;* e.g. in flexibility of thought. This lack of imagination leads to narrow rigid repetitive activities and interests. Their description of the basic triad of impairments still dominates the diagnostic criteria. It is clear that although the biomedical understanding of ASD still persists, the concept and definition of the disorder has changed greatly over time and ideas that were once held with great conviction are now thought to be unfounded. This illustrates that throughout its history and today, understanding of the condition is a product of its social, historical and political context.

The evolution of the spectrum

The last 20 years has seen the broadening of the category of autism to become ASD, with the inclusion of AS (1994) and PDD-NOS (1987) in the current concept. As well as extending the spectrum to include AS, sub-clinical levels of autistic traits are now conceptualised as being part of the broad autistic phenotype. Twin and family studies show that relatives of autistic people often possess qualitatively similar but minor impairments in these behaviours (Lainhart et al., 2002). Thus individuals who have autistic-like traits may not have ASD but may display the broad autistic phenotype. Such subtle manifestations do not warrant clinical diagnosis. ASD is now considered to be a subset of the broad autistic phenotype (Le Couteur et al., 1996).

Today ASD is conceived as a true spectrum, where autistic traits have a normal distribution in the general population, and an arbitrary cut off point determines who is considered to be on the spectrum and who is not. Constantino and Todd (2003) measured autistic traits in a large community sample, and found no discontinuity between normality and psychopathology, as would be evidenced by bimodal distribution. These findings were repeated in a Scandinavian study (Posserud, Lundervold, & Gillberg, 2006). Individual symptoms of autism occur in the general population and do not appear to associate highly, so if an individual has one symptom of ASD there is a moderately low chance of having another. Furthermore, there is no a sharp line separating pathological severity from common traits (London, 2007).

Bentall (2004) describes this concept of a spectrum that extends into the subclinical and normal range as the 'principle of continuity'. He asserts that:

> Abnormal behaviours and experiences are related to normal behaviours by continua of frequency (the same behaviours and experiences occur less frequently in non-psychiatric populations), severity (less severe forms of the behaviour and experiences can be identified in non psychiatric populations) and phenomenology (non-clinical analogues of behaviours can be identified as part of normal life). (p.115)

The imposition of a cut off between normality and abnormality is therefore 'an arbitrary but convenient way of converting a dimension into a category' as Goodman and Scott (1997 p. 23) point out.

In a review of evidence for single genetic or cognitive causes for autism, Happé, Ronald, and Plomin (2006) note that twin studies suggest combinations of largely non-overlapping genes act on each area of impairment. Their own study found only modest correlations between the three domains of behavioural traits in the general population, in the range of 0.1- 0.4 for the relationship of each domain to the other. This evidence shows that the three types of autistic traits are clustered or linked or co-inherited, but with a weak association. These low correlations could be attributed to social and cultural factors as well as genetic links. If a child is unable to communicate for example, then his social skills will not be practised, so he is unlikely to score highly in these measures. The weak correlation between

repetitive behaviours is harder to explain. Speculation is possible: repetitive behaviours have been shown to have both self-stimulatory as well as calming functions (Turner, 1999). Repetitive behaviours can therefore be interpreted as responses to unwanted stimuli, e.g. social stimuli with which autistic people have difficulty. Williams (1992) has given a first person account of use of repetitive behaviours to ameliorate the stress of social situations. Conversely, the need for stimulatory repetitive behaviours, concentrating on drawing lines or circles for example, may interfere with social abilities. Weak associations do not confirm or deny genetic co-inheritance.

The trajectory of symptoms is also different across each behavioural domain, suggesting these behaviours may be separable (Charman et al., 2005). One contention is that these results occur because autism is not a single disorder, but rather a triad of core aspects that have distinct causes, but the diagnostic label is applied when they co-occur. For these reasons, the neurobiologist Eric London (2007), and others have recently questioned the validity of studying ASD in its currently defined triad of symptoms. The reason that ASD and particularly autism is so widely studied, as opposed to its component traits, is largely historical. The phenotype is well established, with a medical diagnosis allowing a clear breakdown. Researchers can easily examine whether children have been diagnosed or not been diagnosed, whereas measurements of individual autistic traits are less standardised. Much aetiological and epidemiological work on ASD has concentrated on the diagnosis rather than symptoms simply because ASD is a longer established, better described phenotype with a higher profile than any composite of individual traits. If the traits across the triad are not inherited together as a cluster, or appear together as a result of the same organic biological deficit, an argument would be that the diagnostic category is fairly arbitrary. If it happens that a person has a combination of these particular traits, they match the diagnostic category for ASD, rather than ASD having a unique aetiology as Kraepelin believed distinct mental disorders did. It is plausible that the diagnosis could have developed differently historically, and focussed on a different set of traits. In a large population-based sample, many children showed difficulties in only one area of the triad, at a level of severity comparable with those who were diagnosed with ASD (Ronald, Happé, Thomas, Baron-Cohen & Plomin, 2006).

Bentall (2004) advocates abandoning psychiatric diagnoses altogether and instead tries to explain the experiences of psychotic people. According to him, categorical diagnoses fail to capture adequately the nature of psychological complaints for either research or clinical purposes. The argument about classification: whether ASD is one discrete condition or a mixture of completely unrelated conditions, all with different causes, continues to rage.

The major change to the last version of the DSM and ICD was the inclusion of impact as a criterion. Understanding of what is 'normal behaviour' and what behaviour has an 'impact' thus clearly becomes dependent on how the behaviour is viewed by parents, teachers and the clinicians who give the diagnosis. This assessment may be different in different social contexts. A study of teachers looking at attention deficit hyperactivity disorder (ADHD) showed that teachers were likely to identify children as having ADHD at rates higher than the expected prevalence rates specified in DSM-IV. This was influenced by cultural factors such as class size (Havey, Olson, McCormick, & Cates, 2005). Differences between how teachers and parents perceive the impact of children's behaviour have also been documented. In a study of attention and activity difficulties, Norwich et al. (2002) found that teachers perceived disruptive behaviour to last longer and have a greater impact than parents did. The researchers put this down to greater difficulties in coping with children in large classes or behavioural expectations differing at home and school. It may also be that children are more disruptive as they are less able to cope at school than at home. In a study of parental and teacher ratings of ASD, parents were again more likely to give a positive prognosis than teachers (Stone & Rosenbaum, 1988). This optimism in parents is well documented and may serve both to help them cope but also to help their children. Research has shown that positive parental belief that a child has good cognitive abilities, and that these are under the child's control, leads to better outcomes (Miller, 1995).

The key point is that ASD is now defined by its impact. Once the significance of 'impact' is recognised, the definition of 'atypical behaviour' becomes dependent on its social context, or how it is viewed by parents, educators and those making the diagnosis, within their cultural framework. The definition of what the disorder is, what is atypical and what is abnormal has an inherently normative function. Thus there is a need for a biological, psychological but also a social causal framework for ASD.

Aetiology of ASD and the bio-psycho-social model

Most 'causes' are best thought of as risk factors that increase the likelihood of a disorder without guaranteeing it will occur. One model involves three types of risk factors: predisposing, precipitating and perpetuating factors (Goodman & Scott, 1997, p.9). There is no one single cause of ASD but many different causes, and many different claims of what these causes may be. The condition can be conceptualised using a bio-psycho-social developmental model. The biological component is considered to be a genetic or biological cause, and the psychological component the underlying psychological mechanisms that result in the behaviours that are presented. The bio-psycho-social model might be better termed the bio-psycho-social-enviro model as many studies have shown that environmental factors add to risk of developing ASD. A common hypothesis is that autism is caused by a genetic predisposition which is triggered by an early environmental influence (Trottier, Srivastava, & Walker, 1999). Environmental risks are presented here under the 'biological' heading.

Biological causes

Although ASD is behaviourally defined, approximately 6% of cases are the result of several known biological aetiologies. These include prenatal rubella infection, untreated metabolic disorders, anticonvulsants taken in pregnancy, localised lesions and postnatal infections (Newschaffer et al., 2007). In addition, about a tenth of childhood autism cases have an identifiable single-gene condition, chromosomal mutation or other genetic syndrome (Folstein & Rosen-Sheidley, 2001).

Children with ASD have a higher risk of suffering from several other conditions. Comorbidity can reflect an inability to supply a single diagnosis that accounts for all symptoms. Children with ASD have been shown to have higher rates of epilepsy, with 30% of cases have epilepsy comorbid (Danielsson, Gillberg, Billstedt, Gillberg, & Olsson, 2005). Other co-morbidities include hearing impairment (Kielinen, Rantala, Timonen, Linna, & Moilanen, 2004), mental health and behavioural problems (Bradley, Summers, Wood, & Bryson, 2004), anxiety, depression (Evans, Canavera, Kleinpeter, Maccubbin, & Taga, 2005), and attention deficit hyperactivity disorder (Sturm, Fernell, & Gillberg, 2004).

It has also been shown that parents of autistic children are twice as likely to have suffered from psychiatric illness (Daniels et al., 2008).

Although ASD is thought of as a neurological disorder, results from structural brain scans are mixed. Neuroimaging and autopsy have suggested that ASD is caused in part by abnormal brain development as neurodegenerative signs are mostly absent. Brain autopsies have shown abnormalities in the frontal lobes, amygdala, limbic system, brain stem and cerebellum (Penn, 2006). It is not clear whether some or all functional brain systems are affected (Müller, 2007). Brains of ASD individuals are often larger than controls (DiCicco-Bloom et al., 2006). One theory is that this is due to too many neurons causing local over-connectivity in key brain regions (Courchesne et al., 2007). A second hypothesis is that the larger volume is due to disturbed neural development during gestation (Müller, 2007), and Finally, insufficient neural pruning has also been implicated (Benayed et al., 2005). The brain structure most consistently affected in individuals with autism is the cerebellum, with a decrease in the number of Purkinje cells being present in the majority (Benayed et al., 2005). Functional imaging studies have shown that the cerebellum is active during cognitive tasks that may be difficult for people with ASD, including language and attention.

Some 30 to 50 percent of autistic children have abnormally high levels of serotonin, a neurotransmitter that conducts signals in nerve cells. Penn reviewed the neurobiology of autism and found converging evidence suggesting that autism involves both abnormalities in brain volume and neurotransmitter systems (2006). Overall, however, it seems that the neurobiology of ASD is a field where generalizations are difficult to make due to large differences between different cases, with various theories vying with each other and offering competing explanations.

Genetic risk factors
After monozygotic twin studies and family history studies highlighted the heritability of characteristics of ASD, aetiological research began to focus on uncovering the genetic mechanisms involved. For siblings of children with ASD, the risk of having one or more features of the broader autism phenotype is high (Yang & Gill, 2007). Critics point out that environmental factors could play a part in these results. It is hard to separate similar social and environmental influences experienced by twins and siblings from genetic influences.

Widely separated or adopted 'monozygotic twins' form the 'gold standard' of twin research. No references to such studies have been found relating to ASD.

10% of childhood autism cases have an identifiable single-gene condition, chromosome mutation or other genetic syndrome (Folstein & Rosen-Sheidley, 2001). These include Fragile X syndrome, which results in autistic-like symptoms, and is caused by a known gene called Fragile X Mental Retardation – 1 (FMR1). A defect in FMR1 renders the gene ineffective and it cannot manufacture the protein that it normally makes. A large proportion of girls who have Rett's syndrome too have an identified mutation on a known gene on the X chromosome.

As well as cases with known genetic causes, in some cases, underlying social factors may predispose autistic symptoms. Rutter and colleagues (1999) noted a very high instance of autism (6%) in Romanian baby cohorts due to poor early care. These children exhibited typical symptoms of autism at four years old, but unlike cases of autism without maltreatment, symptoms by age 6 were much milder. This case is an illustration of how children who share severe autistic symptoms at young ages may have differing developmental trajectories.

In the majority of cases, however, there appears to be a genetic predisposition that is complex and multi factorial. Over 100 candidate genes have been associated with ASD, most of which encode proteins involved in neural development, but exact mutations within the candidate genes have yet to be identified (Freitag, 2007). Furthermore, different individuals may have mutations in different sets of genes, and there may be interactions among mutations in several genes, e.g. between regulatory genes and coding regions, or between the environment and mutated genes, altering their expression. The effect of a mutation or deletion can depend on processes relating to a gene's expression and regulation as well as the subsequent effects on the expression of other genes.

Although heritability is high, there are also many people with ASD who have no family members affected. This could be due to spontaneous alterations in their deoxyribose nucleic acid (DNA), duplicating or deleting DNA during meiosis, thus the mutations that predispose ASD are not present in the parental genome. There are also genes that are

involved in DNA repair systems, and if these cease to work properly the likelihood of such spontaneous mutations remaining unrepaired is higher.

The advent of genomics and the emphasis placed on this has led to much research to identify genetic predispositions to ASD. The field of psychiatry as a whole has been 'geneticised' according to some social theorists. This refers to the potential reclassification of psychiatric conditions in the light of findings from molecular biology. For example, a particular sub-category of DSM-IV schizophrenia has been linked to a substitution in a single base in the sequence of DNA of a particular gene localised to a precise place on a particular chromosome, leading to a substitution of one amino-acid for another in an enzyme involved in neurotransmission. The increase in research on genetic predisposing factors has lead to a partial 'geneticisation' of many psychiatric conditions. Hedgecoe (2001) provides a discussion of the geneticisation of schizophrenia. The debate as to whether the old psychiatric systems of classification should be overhauled in the light of new genomic knowledge which illuminates genetic aetiologies is ongoing (Ericson & Doyle, 2003).

A simplified model underlying much behaviour genetics research envisages a direct linear relationship between individual genes and behaviours. The reality is likely to be far more complex with gene networks and multiple environmental factors impacting brain development and function, which in turn will influence behaviour (Hamer, 2002).

Environmental risk factors
Many environmental factors have been implicated in ASD but the effect of each is poorly established. After the well publicized paper that linked autism to the MMR vaccination, research has repeatedly refuted a link between the MMR jab and ASD (Rutter, 2005). Deykin and MacMahon (1979) found increased risk due to exposure to, and clinical illness from, common viral illnesses in the first 18 months of life. In this study, mumps, chickenpox, fever of unknown origin, and ear infections were all significantly associated with ASD risk.

Epidemiological studies have shown there is a higher rate of adverse prenatal and postnatal events in children with ASD than in the general population (Zwaigenbaum et al., 2002).

Newschaffer and colleague's 2007 review named associated obstetric conditions that included low birth weight, gestation duration, and caesarean section. It is possible that an underlying cause could explain both autism and these associated conditions (Kolevzon, Gross & Reichenberg, 2007). Some studies have suggested that the risk of autism may be increased with advancing maternal age (Bolton et al., 1997). Using anticonvulsants during pregnancy also appears to increase the risk of ASD (Moore et al., 2000). These drugs are used to combat epilepsy which is commonly often comorbid with ASD. Parental occupational exposure to chemicals during the preconception period has also been higher in ASD families than controls in some studies (Felicetti, 1981).

It is not just aetiological environmental factors that seem to lead to increased risks of displaying autistic behaviours. Aetiological causes can be distinguished from proximate determinates which occur at the same time as symptoms, for example, unpatterned environmental noise or fluorescent lights may exacerbate the expression of ASD symptoms. There are also those influences in the environment that are sometimes referred to in psychiatry as maintenance factors, including stigmatisation and incarceration.

Environmental risk factors have received widespread media coverage within the last few years, perhaps because of the strong degree of public concern (Russell & Kelly, 2011-see thesis Appendix 2). In most health and disease categories, a secondary function of diagnosis is to group together people who have a common aetiology. However, both the specific effects of genetic factors and environmental risk factors that might play a part in abnormal neural development are largely unknown. Goodman and Scott (1997) stress that the current understanding of aetiology for childhood psychiatric conditions more generally will probably look ridiculously simplistic or misguided in years to come.

Psychological theories of ASD

The most accepted psychological theories of autism were reviewed by Happé in 1994. The extreme male brain theory as developed by Baron-Cohen (2002) suggests that autistic individuals can systematize—that is, they can develop internal rules of operation—but are less effective at empathizing and handling events and stimuli generated by other people or externally. Baron-Cohen's Cambridge group has carried out work that has suggested high levels of foetal testosterone may be linked to the development of autistic traits

(Ingudomnukul et al., 2007). Knickmeyer and colleagues (2006) found that talented fathers and grandfathers in the areas of maths, physics, and engineering are highly over-represented in autistic samples. These findings have also been interpreted in the light of the extreme male brain theory. The theory was developed from the earlier 'theory of mind' (Baron-Cohen et al., 1985), which can be described as the ability to understand other peoples' mental states, put oneself in another person's place or imagine what they might be thinking or experiencing. It is suggested that autistic people lack this 'mentalising' capacity. The theory fits in well with the mirror neuron theory of autism (Iacoboni & Dapretto, 2006) which was based on the discovery that the macaque monkey brain contained 'mirror neurons' that fired not only when the animal is in action, but also when it observes others carrying out the same actions. Mirror neurons provided a plausible neurophysiological mechanism for imitation and empathy.

An alternative explanation is provided by Frith whose 'weak central coherence' theory (see Frith, 2003; Happé & Frith, 2006) describes the ability to place information in a context in order to give it meaning. Most people pull together numerous stimuli to form a coherent picture of the world. This allows most people to see the 'bigger picture'. In central coherence theory, the failure to appreciate the whole accounts for the piecemeal way in which people with ASD acquire knowledge. This may manifest itself as interest in minute detail or limited ability to transfer meaning from one situation to another. Autistic people may also show relative strengths in some areas, known as 'islets of ability'; and this accounts for savant skills. Related to this is the theory that autistic behaviours are due to interference in executive function (Russell & Jarrold, 1998). Executive functions coordinate the flow of information processing in the brain and are the mechanisms of transferring attention from one thing to another flexibly and easily. They allow people to plan strategically, solve problems and set objectives. Their absence means autistic people show an inability to plan and attain overarching goals. This manifests as easily distractible behaviour and reliance on routines (Hill, 2004).

Bailey and Parr (2003) criticise such theories of psychological mechanisms as 'narrow cognitive conceptualisations' (p. 27), because they cannot accommodate the broad autism phenotype. Furthermore, models that emphasise social deficits are badly stretched to account for language delay in relatives of autistic people (p.30), and psychological theories

take no account of phenotypic individuality. None of the cognitive theories that explain social deficits adequately explain repetitive behaviours, which may be a result of distraction behaviour to avoid anxiety, for example. In their review, Happé and colleagues (2006) argue that that there has been a failure to find a psychological mechanism that accounts for all impairments in the three core behavioural domains of autism, and further attempts to find a unifying explanation should be abandoned in favour of examining individual aspects of the triad.

The social component in the ASD model

As the characteristic behaviours of ASD occur to a lesser degree in the general population the question arises as to how to distinguish someone who is disordered from someone who is not. Consideration of the 'impact' of the condition is key: how are relatives and carers and the children themselves affected by it? But evaluations by affected persons, relatives and psychiatrists are all subjective. As there is no definitive biological or medical test that will determine that someone has ASD, different psychiatrists will invariably have different opinions as to where this boundary lies.

This social dimension to mental disorders is highlighted by social construct theory (Walker, 2006; Zurifff, 1996). The theory has grown from a criticism of the biomedical model of medicine which in turn has its roots in the pathological anatomy of the eighteenth century. The biomedical approach, which is still the dominant paradigm in medicine today, rests on the assumption that the body can be repaired like a machine; so it adopts a mechanical metaphor. It is reductionist in that its explanations of disease concentrate on biological causes and neglect social and psychological factors. Social constructionists have argued that disease categories and particularly mental disorders are not accurate descriptions of anatomical malfunctioning, but are contingent upon the society which produces them. In addition, sociologists of science like Harry Collins describe how the apparatus of science is presented as a 'certainty rendering machine' (Collins, 1987, p.700) in that the credibility and legitimacy of medical knowledge means that society's values may be transformed into apparent 'facts' (White, 2002). The science and technology studies scholar Sheila Jasanoff put forward a more sophisticated idea of how knowledge is derived in her model of co-production of scientific knowledge, which she crystallised in her book *States of Knowledge: The Co-production of Science and the Social Order* (2004). In her vision,

knowledge is created by different sections of society acting together. However, the voices of some have more weight than others in the co-production of knowledge. For example, when developing policy guidelines for diagnosing ASD, epidemiologists are deferred to over mothers as they are assumed to have the benefit of overview (National Institute of Clinical Excellence, personal communication, January 4th, 2009).

The points made by social constructionists are most easily understood with reference to historical medical 'disorders' which have been shown with hindsight to be constructed by cultural, social and political circumstances:

- In the nineteenth century, women who wanted an education were deemed to be suffering from hysteria.
- In the Soviet Union in the mid twentieth century, dissatisfaction with communism in general was regarded as a symptom of schizophrenia.
- Homosexuality was listed as a disorder in the second edition of the DSM.

By the same token, some conditions that are accepted as normal by our society seem to match the current official criteria for psychiatric disorders. The biologist Richard Dawkins contends in his book *The God Delusion* (2007)that spiritual experiences could be viewed as mental illness, for example. Although these examples are extreme, they illuminate how 'the line between sanity and madness must be drawn relative to the place where we stand' (Bentall, 2004, p.117).

The radical psychiatrist Thomas Szasz made this point more forcefully (1961). He proposed there is no such thing as mental illness, but simply individual traits or behaviours that society deems unacceptable, immoral and deviant. Szasz believed labelling such people as ill harms them - they come to accept the label, and they are treated accordingly. This perspective reiterates that the boundaries of acceptability of normal behaviour are determined by culture and society, not by any distinct or innate biological reality. Szasz elucidated that that 'mental illness' should be dealt with through changing social and environmental factors.

In an ingenious study, Rosenhan (1973) sought to illustrate the constructed nature of schizophrenia. The author worked within a team of researchers who all applied for admission to various psychiatric institutions complaining of auditory hallucinations. They were all admitted, mostly diagnosed as schizophrenic, and subsequently their actions were interpreted as pathological. However, this study did not show that the researchers became schizophrenic, merely that they were treated as such.

Alongside social constructivism, the anti-psychiatry movement also materialised in the 1960s. This argued that much mental illness was created by psychiatrists whose professional medical dominance maintained social control (Ingleby, 2004). The practice of hospitalising people who were deemed to suffer from mental illness was captured as labelling someone as unfit to participate in social and political life. Armstrong (1983), for example, described the development of child psychology and paediatrics as 'policing the normal'. Marxists shared this perspective on diagnosis as a form of censure, arguing that from a position of authority, doctors can make ideological statements that are legitimised by science (Waitzkin, 2000). These messages, according to Waitzkin, tend to direct behaviour into safe, acceptable and non-disruptive channels that maintain the status-quo of capitalist societies. In social construct theory, ASD exists only in relation to the society that constructs it. Some advocates of this approach such as Timini argue that the change leading to increasing prevalence of ASD is primarily ideological, the result of changes in society's emphasis: to intellectual, social and emotional competence (Timimi, Gardner, & McCabe, 2010). Thus social construct theorists also explain the increase in prevalence of ASD in terms of cultural boundaries changing with time. They point out that not fitting into social norms is explicitly considered as one of the diagnostic criteria of ASD for example, social impairments may include 'odd' relationships with adults where children appear either over-friendly or aloof (Baird et al., 2003). Sociologists have argued that what constitutes a 'social norm' is not an objective measure. Bentall (2004) and Bolton (2008) interpret the DSM as the effort of our culture to make sense of breakdowns in behavioural norms.

The French philosopher Michel Foucault analysed how social norms are created, describing the 'normalising judgement' of the individuals and groups within society as they adhere to their roles as teachers, doctors, social workers and so on (Foucault, 1975; 1989). The actions or attributes of each individual are judged by these individuals and groups leading

to the establishment of a norm. This type of community surveillance also leads to self-surveillance, that is, self-judgement ensures that one's own behaviour lies within accepted cultural boundaries. Foucault put forward the concept of 'discourse' which shapes and reports on knowledge and includes events, settings, technologies, activities, and language that shape understandings. Foucault also described the 'gaze' through which knowledge, or more specifically, disease, is conceptualised. Through the 'medical gaze', biomedical phenomena become obvious to the clinician, thanks to training and experience producing expertise (Collins & Evans, 2007). The 'medical gaze' has altered throughout time. These days, the 'medical gaze' increasingly takes patients' experiences into account. This is in part due to a response to such powerful sociological views (Nettleton, 1992).

Deviance or difference, then, in a biomedical sense is taken to be attributes and traits that sit outside a statistical 'normal' taken from population averages, and in a sociological sense that which sits outside what can be considered 'normative' within a given cultural context (Armstrong, 1984).

Tied to the concept of normalisation is *medicalisation*. Conrad (2003) defines medicalisation as the process by which non-medical problems become understood as disorders. Medicine names and defines the disorder in medical language, claiming expertise in areas that were not previously in its realm, such as aging, childbirth, alcohol consumption and childhood behaviour (Conrad, 2005; Conrad & Schneider, 1992). Conrad argues that new pharmacological treatments and medical technologies like genetic testing strengthen the grasp of this extended medicalised domain. It has been argued that clinicians depoliticise the causes of illness by providing pharmaceutical solutions to problems such as depression which are really social in origin (Brown & Harris, 1978). Such dependence on medical expertise strips ordinary people of ability to cope with own problems according to Illich (1999). However, studies have shown this is a simplistic view, as patients do not always accept everything doctors have told them; there is lay resistance to medicalisation as people draw on mixed sources of information including their own knowledge and experience (Gabe & Calnan, 1989).

It is true, however, that in the field of child psychiatry and specifically autism, the range of behaviours perceived as pathological has grown (Porter, 1987). Castel, Castel and Lovell

(1982) refer to this process as the 'psychiatrization' of difference, and suggest abnormal and disordered behaviours of today are a part of the natural range of human variation. Molloy and Vasil (2002) argue that the category of AS effectively reduces the range of behaviour patterns that would count as 'normal' (though eccentric) and 'sentences the child to a lifetime of special needs and interventions' (p. 667). With the broadening of the autistic spectrum, psychiatry has certainly widened the scope of what can be considered as a mental disorder.

Radical constructionist theory denies the existence of a biological base for disease, whereas biological psychiatrists have taken the position that neuropathology will be found. The social constructionist position has been attacked as an outdated style of cultural relativism which is just a reflexive opposition to biomedicine and displays a lack of common sense (Aronowitz, 2008). Interpretations that only take social aspects into account, which perceive the experience of autism as solely a product of social and cultural structures, disregard the evidence pointing to biological predisposing factors and the reality that autism can have a hugely negative impact on a child's experiences. As Molloy and Vasil (2002) comment: 'Various approaches to theorizing about disability can be plotted on an axis from a totally...materialist position (medical) to a totally socially constructed position (post structuralist)' (p. 664). The compromise position is that there is a biological reality, but in the processes of explaining, treating and classifying, ASD must also be viewed in the social context. A dynamic relationship of biology with cultural values and the social order is therefore considered (Lock & Gordon, 1988). In the bio-psycho-social model, difference is thought of as *both* biological *and* as a product of the social world. The bio-psycho-social model assumes that ASD is the label given to an atypical clustering of functional characteristics (with negative impacts) that arise from the interaction of genetic and environmental factors though a developmental process and whose expression and interpretation also depends on the social context. From this point of view, disability is neither the product of biological impairment or of society, rather it has an emergent property involving the interplay of physiological impairment, structural establishments and socio-cultural elaboration over time (Williams, 1999).

The prevalence of ASD

Far more boys display autistic type symptoms than girls according to Rutter (2005).
Estimates for ASD average at approximately 4:1 male-to-female ratio, (Rett's syndrome
being the exception that only occurs in girls), although the most recent estimate from a
population based survey on ASD in the UK is 9:1 (Brugha et al., 2008). This survey also
reported the prevalence of ASD in adults to be around 1%. Measurements of prevalence
vary, with urban areas having higher rates than rural locations. The total prevalence of ASD
in the Thames region of London among 9-10 yr olds was measured as 1.1% in 2006 (Baird
et al., 2006). In the previous year, Fombonne (2005) carried out an overall comparison of
epidemiological studies and estimated that ASD occurs in approximately 0.6% of the
population.

There have been far more diagnoses of ASD in recent years with the United States (US)
Department of Education showing an astounding 556% increase in the number of children
being treated between 1991 and 1997 (Stokstad, 2001). In a 2005 review, Rutter writes:
'the true incidence of autism spectrum disorders is likely to be within the range of 30–60
cases per 10,000, a huge increase over the original estimate 40 years ago of 4 per 10,000'
(2005, p.2). Today, prevalence estimates are even higher. According to Muhle, Trentacoste
and Rapin (2004), ASD now has a greater prevalence in children than that of cancer or
Down's syndrome. There is no doubt that the reported prevalence has increased
spectacularly, and particularly over the last twenty years. The question is, why?

Influential work by Fombonne (2001) suggested that the change is not a true reflection of
more children with ASD in the population now than in the past, but is the result of the
diagnosis being given to more children. Three main reasons have been cited: the extension
of the spectrum in recent times to include milder conditions like AS, the extension of
diagnosis to younger children, and increasing awareness of ASD by parents and clinicians.

'Diagnostic substitution' may provide a partial explanation. Barbaresi, Katusic, Colligan,
Weaver & Jacobsen, (2005) found that increased rates of diagnosis followed increases in
funding for special educational programmes. Where resources are being directed towards
ASD, greater incidence is associated with declines in other diagnostic categories indicating
that clinicians prefer to label children with ASD in order to allow them access to greater

resources (Shattuck, 2006). If this is occurring, children who in the past would probably have been diagnosed as having a specific learning disability or a psychiatric disorder, or not diagnosed at all, are recorded as cases of ASD (Pettus, 2008).

The view that the increased volume of ASD cases is due to changes in diagnostic practice has become the consensus position amongst psychiatrists and epidemiologists. This position conflicts with the opinions of many people in contact with autistic children. Individual parents, for example, have published their view that the true incidence of ASD has increased; i.e. there are actually more children now who display ASD. A sample of the public, most of whom had a direct relationship to autism, either as parents, professionals or autistic people, showed that many lay people believe the increasing incidence of ASD is due to increased exposure to new environmental, medical and technological hazards (Russell, Kelly & Golding, 2010- see thesis Appendix 2). In 2007, an annual review of public health suggested the question of whether incidence of ASD really has increased remains unanswered (Newschaffer et al., 2007).

The reliability of prevalence estimates themselves are also questionable. In his wide ranging reviews, Fombonne noted that the assessment process, sample size, publication year, and geographic location of studies all have an effect on prevalence estimates (Fombonne, 2001; 2003; Fombonne et al., 2005).

ASD is also likely to be diagnosed in different ways by different institutions at a national and local level. Skellern et al. (2005) studied local diagnostic practices in Queensland and found local differences between the practise of paediatricians and child psychiatrists. Whether an ASD diagnosis is given may also be dependent on local cultural factors such as whether a local medical facility specialises in autism, for example. A UK study showed wide variation in the numbers of children diagnosed with ASD throughout the country. This was thought to reflect the wide variations in service provision policies of local agencies (Bebbington & Beecham, 2007). A similar study in the USA reported few consistencies among states in policies and practices regarding the identification and care of infants and toddlers with autism (Stahmer & Mandell, 2007). The phenomena of 'clustering' of autism diagnoses in neighbourhoods in California has been attributed to social influences such as communication between parents (Liu et al., 2010).

Undiagnosed cases of ASD

Throughout research in child psychiatry, there appears to be a large group of children who remain undiagnosed despite appearing to exhibit behaviour that would justify a diagnosis. Most studies have reported that psychiatric disorders are present in roughly 10-25% of children. These high rates are probably an overestimate and in part reflect the lack of impact in the former versions of the diagnostic criteria already discussed (Goodman & Scott, 1997). However, epidemiological studies have indicated that most children do not get referred for psychiatric disorders because families themselves are not concerned about symptoms (Burns et al., 1995; Fergusson et al., 1993; Leaf et al., 1996). If a condition does not impact on carers or children, they are not in need of treatment and are unlikely to be referred to health services. In the seminal Isle of Wight study of 1970, Rutter et al. noted that even when all the research team thought that children had a psychiatric disorder, only half the parents thought their children had definite problems (as cited in Rutter, 1989).

This disparity suggests a large percentage of children may present autistic traits that meet diagnostic thresholds but may never be referred for diagnosis. In the British Child and Adolescent Mental Health Survey of 1999, roughly 1 in 10 children had at least one DSM-IV disorder, involving a level of distress or social impairment that warranted treatment. Of those with emotional, behavioural and concentration difficulties, only 58% had been in contact with health or educational special services (Ford, Goodman & Meltzer, 2003). As is apparent from the Isle of Wight study, a mild condition may not pose enough of a problem to parents, educators or to the children themselves to get a diagnosis. Whether parents seek professional help for their children is likely to be partially dependent on social factors (Zola, 1973). Studies reporting high prevalence rates for ASD often calculate results by including children who have been documented by researchers as having symptoms of ASD but have never received a formal diagnosis in their communities (Baird et al., 2006; Baron-Cohen et al., 2009).

Early diagnosis

There is plenty of medical research literature arguing that ASD is partly genetically determined and diagnosis should be made as young as possible to treat and manage the condition (Chakrabarti & Fombonne, 2005; Charman & Baird, 2002; Charman et al., 2005;

Landa & Garrett-Mayer, 2006). Clinical guidelines have also promoted early identification (Filipek et al., 1999). Against this backdrop, diagnosis has been taking place at successively younger and younger ages. Some clinical guidelines currently recommend screening below 2 years in age (Crais, Watson, Baranek, & Reznick, 2006).

Early diagnosis is supported by the National Autistic Society (NAS) in the UK. Their *National Autism Plan for Children* in collaboration with the Royal College of Paediatrics and Child Health and the Royal College of Psychiatrists, states 'the benefits of the early identification of ASD are recognised by parents and professionals alike'. The NAS recommends the use of screening instruments such as the Checklist for Autism in Toddlers (Allison et al., 2008) which identifies children aged 18 months who are at risk for social-communication disorders. The NAS comment that it is generally accepted that 'the earlier a true diagnosis is made the better for the child, the family and those involved around them' (www.autism.org.uk; 'The Need for Early Diagnosis', accessed 2011). The American Academy of Paediatrics has published a policy statement recommending screening programmes for ASD at 24 months (Gupta et al., 2007).

Diagnosis of ASD is often framed in literature as an essential step to accessing appropriate, effective interventions. The National Autistic Society, for example, state on their website:

> Getting an accurate diagnosis can give families and people with autism access to specialised support and services. The earlier a diagnosis of autism is made, the better the chances of the person receiving the most appropriate help and support. (NAS website, 'The Need for Early Diagnosis', accessed 2011)

Myers and Johnson (2007) found no evidence for claims that preschool intervention is essential. Nevertheless, reviews have concluded that there are enough positive outcomes to indicate that some form of intervention is warranted (Guralnick 1996; Tonge 2006). In the USA, the US National Institute of Health (NIH) has emphasized that for autism, 'the urgency of early identification and treatment puts the quest for infant screening and diagnostic instruments in the forefront of our priorities' (Bristol-Power & Spinella, 1999, p. 435). Much research effort has been directed at developing screening instruments for young children to monitor early signs of autism (see for examples Allen, Silove, Williams, &

Hutchins, 2006; Allison et al., 2008; Baird et al., 2000; Bryson, Zwaigenbaum, McDermott, Rombough, & Brian, 2007). In the biomedical realm, then, ASD is conceptualised as a neurodevelopmental condition, for which appropriate treatment can be obtained. Advocates of early diagnosis argue that it also enables provision for family support and reduction of family stress (Cox et al., 1999). Some have concluded that early diagnosis is vital to ensure that genetic counselling is made available to parents who may be considering other children. Osterling et al. (2002) claim that 12-month-old infants with autism can be distinguished from their same-age peers with idiopathic learning disability. They point out that such behaviours can be identified at even younger ages. The NAS acknowledge there is more work to do on the predictive utility of these findings but are welcoming of the suggestion that autism will eventually be diagnosed as early as 12 months or even younger.

A counter argument to early diagnosis is that throughout childhood, all behavioural traits increase in variability over time as children develop. Thus it is difficult to distinguish a two year old who cannot speak because s/he may continue to display symptoms of ASD from a two year old who cannot speak because he is slower and will catch up later (Landa, 2008). Overall the evidence of the age at which ASD can be reliably diagnosed is unclear (Stone et al., 1999). The psychiatry and medical literature centres on how reliably an early diagnosis can be made, and the accuracy of screening checklists as tools. There are very few if any references to literature questioning the merits of an early diagnosis of ASD per se.

Protocols of medical diagnosis

In the UK, a child with developmental complaints can initially be seen by a wide range of health professionals in primary care, including health visitors and general practitioners. Referral can be made by those in education, social services and of course, parents or carers. Information is gathered at this stage and may suggest the need for specialist assessment. The purpose of assessment is to confirm a medical diagnosis. For preschool children in the UK, referral is usually to the health visitors within a child development team. For school age children the child may be assessed initially by an educational psychologist, then, the referral may be to a paediatrician, child psychiatry or learning disability service. Consequently, for school age children there are many pathways leading to diagnosis; referral could lead to the child and adolescent mental health service (CAMHS) or to the local paediatric team depending on the local authority. The primary assessment may mean

that the referral is labelled as a social/communication difficulty or the referral can be vague e.g. 'in trouble' 'difficult to manage' (T. Ford, ex-CAMHS child psychiatrist, personal. communication, September, 2008). Levels of support and particular interventions provided by services are variable throughout the UK (Jordan, Jones & Murray, 1998).

In the UK, the paediatrician or child psychiatrist commonly performs a preliminary investigation by taking a developmental history and physically examining the child. If warranted, diagnosis and evaluations are conducted with help from ASD specialists who observe and assess cognitive, communication, family, and other factors using standardized tools, and taking into account any associated medical conditions. The national clinical guidelines in Scotland recommend that observations should take place in different contexts both at home and school as well as in the clinic, as contextual information increases understanding of how a child functions in groups, in unstructured settings or in real life tasks (Scottish Intercollegiate Guidelines Network, 2007). Since the publication of the National Autism Plan, most local authorities have adopted a multidisciplinary health assessment procedure. However, this is not standardised throughout the country and the basic diagnosis is in effect made by a paediatrician or child psychiatrist, although in many cases the clinician 'won't write the diagnosis until it goes through the assessment…as a courtesy to colleagues' (A. Emond, paediatrician, Centre for Child and Adolescent Health, personal communication, September, 2008). Psychiatrists and paediatricians may view themselves as 'gatekeepers' for resources, giving a stamp required by local authorities by making a medical diagnosis (A. Emond, paediatrician, Centre for Child and Adolescent Health, personal communication, September, 2008). Although a multi-agency assessment is recommended and in many cases adopted, it may be the responsibility of the clinician on the team to collate reports from different sources. Thus the team may never sit down together in the same place to discuss a case.

The National Autism Plan guidelines (2003) also recommend that 'The outcomes should include immediate feedback to the family, even where the diagnosis remains unclear. The family should have adequate opportunity to discuss the outcome' (Executive summary, p.2). Brogan and Knussen (2003) showed that the way a diagnosis was presented to parents had a major impact on their acceptance of the situation, their long-term attitudes, stress and general coping strategies. In their study, 55% of parents indicated that they were satisfied or

very satisfied with the disclosure, but 45% were not. Those parents who were most satisfied were more likely to have been given the diagnosis of AS and to have children who were not currently in a specialist school for children with special educational needs (SEN). The social scientist Chrissie Rogers conducted a study of 42 parents of children with learning impairments, and published her findings as a book in 2007. Rogers criticised the parent-professional relationship in the light of her findings, pointing out complaints that as diagnosis is given, a professional simply asking 'any questions?' does not reflect the enormity of the situation to a parent. Parenting a child with ASD or other childhood disorder drastically changes the expectations of what becoming a parent involves, she argues. Both Brogan and Knussen (2003), and Mansell and Morris (2004) comment on the importance of encouraging families to participate in any decisions related to their child.

Howlin and Asgharian (1999) carried out a UK-wide survey of parents of children with autism spectrum disorders in the UK, comparing the diagnostic experiences of 614 parents of children with autism and 156 with AS. Although the ages of the children in the two groups were very similar at the time of the survey, parents of children given a diagnosis of AS had experienced significantly longer delays and greater frustration in obtaining a diagnosis than those with a child with autism. In the autism group, the average age when diagnosis was confirmed was around 5.5 years; in the AS group 11 years. According to this study, parents of children with a diagnosis of autism were generally aware of problems in their child's development by 18 months of age; in the AS group concerns emerged later, at around 30 months of age. Initial worries in both groups centred on abnormal social development. Stereotyped or repetitive behaviours were not prominent in the early years in either group.

Guidelines encourage clinicians to routinely check for comorbid problems as well as medical problems or emotional difficulties and disorders. Other studies have also suggested that children's problems are more likely to be addressed if they present externalising behaviours like aggressive or disruptive behaviour (Todman, Justice, & Swanson, 1991). Vardill and Calvert (2000, p.222) describe 'the quiet or amenable child….who never quite gets to the top of the list'. Parents are more likely to perceive a diagnosis as satisfactory according to Brogan and Knussen's study when they receive a single, simple diagnostic label (2003).

Neuro-identity politics

Silverman (2008), in a review of social science work on autism, describes how self-advocates such as Michelle Dawson (2004) propose that autism is a valid identity that should be protected from aggressive behavioural therapies. Such self-advocates have grouped together to form the autistic rights movement (ARM). By emphasising both the validity and distinctiveness of autistic experience, self-advocates have led anthropologists to explore the idea of autism as culture, a perspective that is preceded by studies of 'Deaf' culture (e.g. Skelton & Valentine, 2003). Nancy Bagatell, for example, presents an ethnographic case study of one man's attempt to define his identity in terms of AS via internet support (Bagatell, 2007).

The ARM grew in tandem with the development of the internet as a tool for communication in the mid-1990s. The internet has helped many with ASD to communicate without the need for face-to-face engagement, which those with ASD typically find difficult. Use of symbols allows expression of emotions clarifying intentions that users find difficult to interpret face-to-face. The internet has also allowed people with the condition to work remotely (Biever, 2007). The internet supports activism in that it facilitates communication between widely dispersed people (Nambiar, 2004). Knowledge is shared and interpreted according to experience and the activist group identity emerges. The internet provides a feedback mechanism where communication serves to create and maintain bonds within the activist community, as well as strengthening the hold of the idea around which the movement is organised.

Perhaps, as argued by some sociologists, e.g. Rabinow (2005), this forging of a social identity is the most fundamental concept for galvanising responses to biomedical models. The identity of the ARM can be considered as an incomplete and open-ended concept but one that maintains coherence across multiple social roles and reference groups (Giddens, 1991), and can be regarded as part of the wider disability rights movement, spurred by the social model of disability (Finkelstein, 1979; Oliver, 1997). This model emerged in the UK after the manifesto *Fundamental Principles of Disability* written by the Union of Physically Impaired Against Segregation stated that 'it is society which disables physically impaired people' (1976, p.3). Crucially the social model advocates a distinction between the physical

deficit or 'impairment' and the 'disability' caused by attitudes and infrastructures of society. Dowse (2001) describes the social model as the collective actions frame of the disability movement, and indeed it provoked the formation of numerous disability groups, where the group identity is turned around to become a mark of pride (Bickenbach, Chatterji, Badley, & Üstün, 1999). Epstein (1995) also notes the new wave of health-related activism. This is a politics of identity organized around particular diseases and conditions, for example, cancer or chronic fatigue syndrome. These groups assert their political claims on the basis of these identities. One of the defining characteristics of such social movements is how participants become aware of their own role in defining the affected individuals as a group, in the ARM case, as 'autistic' people. This is an important precursor for political mobilsation according to Epstein.

The ARM's core point is its argument against curing ASD. Their view that ASD is an alternative form of being with its own benefits and drawbacks draws from the social model. The ARM contends that society should learn to tolerate harmless behaviours like hand-flapping or humming (Trivedi, 2005). However, these advocates also deploy biomedical understandings of autism as a physical impairment or neurological difference, in order to form a cohesive identity. The ARM argues that increased awareness should lead to the creation of attitudes and environments within society that are more tolerant to typical autistic behaviours. The movement has also developed its own unique terminology; 'neurodiversity' represents the neurological variation within the brains of the human population and is to be embraced; 'neurotypicals' are those who do not have ASD and 'curbies' is a slightly derogatory term used to describe those parents, professionals and individuals who are pro-cure. Some activists prefer to be referred to as 'autistic people' instead of 'people with ASD' which implies to them that autism can be removed (Sinclair, 1999). Although anti-treatment, the ARM does advocate giving children more tools to cope with the non-autistic world instead of trying to change them into 'neurotypicals'. Thus systems like Picture Exchange Communications System (PECS) are generally supported (Bondy & Frost, 2001).

This fundamental position of ARM to work for a change in society to accept ASD symptoms as they are was described succinctly by Amy Harmon in her 2004 article in the New York Times:

Autism has been seen by the medical establishment as a condition from which a normal child might one day emerge. But some advocates contend that autism is an integral part of their identities, much more like a skin than a shell, and not one they care to shed. Some discussed plans to be more openly autistic in public, rather than take the usual elaborate measures to fit in. Others vowed to create more autistic-friendly events and spaces. *Autreat* participants, for instance, can wear color-coded badges that indicate whether they are willing to be approached for conversation. Common autistic mannerisms, like exceedingly literal conversation and hand-flapping, are to be expected. Common sources of autistic irritation, like casual hugs and fluorescent lighting, are not (Harmon, 2004).

One of the best known activists is Michelle Dawson, a Canadian academic diagnosed with autism. She has questioned the ethics of treatments such as Applied Behaviour Analysis (ABA) and appeared as an expert witness in the 2004 case Auton versus British Columbia where parents of autistic children filed to get medical insurance to cover the cost of treatment, claiming it was a medical necessity. Her position is that ABA's techniques of aversion, which persistently expose children with ASD to stimuli that cause distress and subsequently discourage unwanted behaviours such as hand-flapping are tantamount to cruelty. Dawson considers these unwanted behaviours to be coping mechanisms in stressful situations. She writes in an online article:

> Where ABA needs scrutiny is when its power is used to remove odd behaviours which may be useful and necessary to the autistic (such as rocking, flapping, and analytical, rather than social or 'imaginative' play); and when typical, expected behaviours which may be stressful, painful, or useless to the autistic (such as pointing, joint attention, appropriate gaze, and eye contact) are imposed (Dawson, 2004).

Others within the movement have also criticised therapies which attempt to remove autistic behaviours, claiming the behaviours are attempts to communicate (Baggs, 2007).

Attempts have also been made by the movement to highlight other issues. A petition to Downing Street was launched by *Aspies for Freedom* in 2007, lobbying for the autistic community to be given minority group status. This followed their proposal to the UN in 2004 which stated:

> This is a declaration from the worldwide autism community that from here on we wish to be recognised as a minority group. We mean for this statement to begin a process of official recognition by the United Nations that we are indeed a minority group, and worthy of protection from discrimination, inhumane treatment, and that our differences are valid in their own right and not something that needs to be cured (Nelson, 2004).

A further issue for the ARM is their assertion that prenatal genetic tests could lead to the elimination of ASD. Since those in the ARM see ASD as natural human variation and not a disorder, they are also opposed to what they see as attempts to eliminate ASD. Despite the complexity of genetics in the field, in 2011, RainDance Technologies launched two genetic screening tools for identifying mutations associated with ASD (2011). The ARM vehemently opposes the development of prenatal genetic test for ASD, and have launched the 'Autistic Genocide Clock' to highlight the possible abortion of foetuses with genotypes that, they argue, may potentially express both beneficial and impairing traits. Although autistic people may have very low empathy, this is often accompanied by areas of strength or talent (Baron-Cohen, 2011). Such variation in the gene pool is desirable for evolutionary reasons, the ARM contest.

A second major identity that has coalesced around ASD involves parents' groups that have come together to fund research into causes of ASD as well as research into interventions. They are committed to treatment and cure, and employ genetic and biological models to describe ASD, through organisations such as *Autism Speaks* (Bumiller, 2009). A number of such disease-specific support groups have wielded considerable influence over research and research funding. Rabeharisoa and Callon (2002), for example, describe the increasing influence of patients' associations in France. Rose writes of how as collectives such groups have tried to maximise their influence on the development of new science, technology and medical knowledge (2006, p.23). The ability of such groups to influence the research

agenda is part of the wider 'democratisation' of science. In the UK, the two major charities concerned with autism are the National Autistic Society (NAS) and Autism Speaks (in the UK renamed 'Autistica'). Both are predominantly driven by parents' interests.

What the new social health movements seem to share is a repudiation of 'victim' status and a push towards greater equality with those who are considered experts, clinicians, researchers or professionals who become involved after diagnosis. They demand a greater involvement in determining research priorities and policy decisions (Rabeharisoa, 2008). The power and credibility of their first person voice – the voice of affected individuals -is increasingly acknowledged by policymakers. *Autreat*, an ARM organisation, for example, were consulted by the parliamentary office of science and technology in the recent briefing paper on autism (D. Murray, Autreat, personal communication, September, 2009.)

Some of the most vocal critics of the ARM are non-autistic relatives, for example, those represented by the organisation *Families of Adults Affected by Asperger Syndrome*, who claim that it is predominantly high-functioning individuals who are opposed to treatment, and therefore the group is unrepresentative of the wider autistic community: those who have less ability to communicate are more likely to want or need treatment. Whether political groups really represent whom they claim to represent is a key issue. This is re-iterated by Sue Rubin, diagnosed with childhood autism, who argues that low-functioning people are just trying to get through the day without hurting, tapping, flailing, biting, or screaming, and a cure from her point of view would be wonderful (2005).

Many studies have shown that a child with ASD within the family causes stress for both parents and siblings as a consequence of their caregiving responsibilities, the child's cognitive impairment, and the need for long-term support (Gray 2006; Weiss 2002; Rivers & Stoneman 2003; Sharpley, Bitsika & Efremidas, 1997). Parent organisations characterise the movement's apparent lack of empathy for their plight in opposing a cure for ASD as typical of those with the condition. In response, some autistic rights activists have parodied the 'theory of mind' by claiming that 'neurotypicals' are insensitive to their perspectives, painting non-autism as a mental disorder characterized by lack of 'theory of other minds' (Slater, 2008).

Parental dilemmas about getting a diagnosis

If, as has been suggested, there is a large undiagnosed minority of children whose behaviour meets the diagnostic threshold for ASD, as indicated by research into other psychiatric disorders (Burns et al. 1995; Fergusson et al. 1993; Leaf et al. 1996; Ford et al., 2003), then it is worth considering whether parents are active contributors to the diagnostic process.

A survey by Howlin and Moorf (1997) looked at families who had obtained a diagnosis. Fewer than 10% of these children were diagnosed at initial presentation; another 10% of families were either told to return if their worries persisted, or that their child 'would grow out of it'. Many families reported that schools and other parents were the greatest help to them, not the medical community. Almost 20% alleged that they had to apply substantial pressure to get referrals or acquire them privately. It appears that some parents that seek diagnosis (and eventually get it) cannot easily obtain one.

Flisher and colleagues (1997) examined 'unmet need' where 'need' was defined in a policy document of the Medical Research Council (MRC) as 'where there is functional impairment …due to potentially remediable or preventable cause, for which there is accepted effective treatment'. They found approximately 17% of their sample had unmet need, measured in their terms as children who were assigned a research diagnosis but were not using services. Here, unmet need was associated with 'barriers' to service access such as parental concern that the child would be taken away, parents wanting to solve the problem themselves or a failure to cooperate. In this study and others, barriers mediated by parents also appear to be an obstacle to diagnosis.

Both the literature and policy guidelines, then, suggest parents play an active role in determining whether their child receives a diagnosis or not. As this is the case, it seems to be worth considering why parents may or may not decide to get a diagnosis for their child. The process of weighing up the benefits versus the disadvantages of receiving a diagnosis has been regarded as as a dilemma in the research literature (e.g. Norwich, 2008; 2009).

Norwich (2008; 2009) looked at professional views on the identification of special educational needs (SEN). The basic dilemma for these professionals was whether to suffer

potential drawbacks associated with SEN such as stigma, devaluation and rejection, or risk losing resources and opportunities that might be released by a formal identification. According to this research, professionals saw parents as experiencing dilemmas over their children with special educational needs, including those with ASD. However, this group shares some requirements with all other children and, of course, each child has unique individual needs distinct from others in the ASD group. The dilemma that parents consider, therefore, involves a balancing of requirements for their own child in these three dimensions.

In their study of parents of ADHD children, Hansen and Hansen (2006) also use the notion of dilemma to explain parents' decision-making processes when deciding whether to agree to a prescription of methylphenidate for their children. Again, drawbacks and benefits were weighed up, with present and future academic goals for children often cited as reasons to tolerate the medication's negative side effects.

Mansell and Morris (2004) also noted similar tensions in their survey of 55 parents of ASD diagnosed children. Parents had mixed feelings about the diagnosis noting both positive and negative effects. Over three quarters of the parents expressed relief, and felt they had a better understanding of their child's behaviour and access to practical help after diagnosis. However, the authors also noticed several negative consequences: parents became more worried about the future, and a substantial minority said they had been treated worse as a consequence of their children being diagnosed.

Clinicians also face dilemmas when giving diagnoses, especially where conditions are disputed and contentious. Woodward and colleagues (1995) found that 70% of doctors were reluctant to articulate a diagnosis of chronic fatigue syndrome (CFS), for example. Doctors felt nervous about both the uncertainty regarding the aetiology of the condition and were concerned that a diagnosis might become a self-fulfilling prophecy. In this study, patients, by contrast, highlighted the positive aspects of a singular coherent diagnosis and the benefits of having an explanation for their problems. Thus, labels sometimes lead to conflicts between patients looking for diagnosis and clinicians who fear that diagnosis might lead to worse outcomes. In this case the reluctance of doctors to diagnose may be well founded. Diagnosed CFS patients have a worse prognosis than fatigue syndrome

patients without such a label (Huibers & Wessely, 2006). The researchers explored the ways in which CFS patients perceived themselves, labelled their symptoms and appraised stressors, and concluded that diagnosis may perpetuate or exacerbate their symptoms. From the patients' points of view, however, finding a label that fits their condition can provide meaning, emotional relief and recognition.

Anita Ho (2004) writes about dilemmas over labelling children with learning disabilities. Her position is that labelling pathologises difference. Historically, society has a tendency to create a 'normal' group and to label others as 'deviant' (Minow, 1991, p.31). Ho contends that parents may not want to think of their child as being disabled: the stigma attached to such a label could cause others to treat their identified child as abnormal and inferior. Ho is also highly critical of what she sees as political and social reasons pressing for diagnosis. Several political and social pressures to diagnose children with a range of neurodevolopmental disorders (including ASD) have also been put forward elsewhere. These include:

- Aggressive pharma marketing (Conrad & Schneider, 1992)
- Mothers seeking absolution from blame for child's behavioural problems (Illich, 1999)
- Restrictions in funding in education, meaning diagnosis gives school additional finance (Kiger, 1985)
- Related to educational psychologists defining their territory (Slee, 1994)
- General increase in psychiatric labelling (Armstrong, 1993)

Riddick (2002) points out that labelling is sometimes treated as if it were 'good' or 'bad'. Some scholars have seen labelling as an unnecessary and wholly destructive process. However, there are many aspects to labelling, both positive and negative. Potential positive outcomes of labelling such as diagnosis and appropriate treatment and alteration to the environment, together with obtaining better resources through funding and legislation can be offset against potential negative outcomes. Quicke & Winter (1994) adopt an interactionist perspective to show that labelling can lead to discrimination or positive

outcomes depending on the type of label and its context, and the tensions between these effects.

Although ASD is still a stigmatized condition according to Gray (2002), Norwich and colleagues (2002) noted that the stigma of having a child with SEN has lessened considerably in recent times. In their qualitative research study, many educators especially thought that labels do not lead to devaluation. These findings are interpreted in the light of more positive social and educational images of disability in western society over the last decade. This is in part thanks to the work of the disability rights movement and more specifically the autism rights movement which has lead to the lessening of stigma of childhood ASD (Wolff, 2004). Norwich (2008) also found that in the UK, educators tended to question the validity of the parents' dilemma because of the perceived positive image of disability inside schools, while outsiders recognised tensions about the wider negative consequences of labels. School-outsiders such as parents would therefore suffer from dilemmas about diagnosis more than insiders.

In her very thoughtful analysis of a cross cultural study of ADHD, Malacrida (2004) notes a tradition of cultural resistance to labelling in the UK in contrast to the Canadian medicalised model. This reflects the influence of the anti-psychiatry movement in the UK and that of scholars like Szasz (1961) who argued that labels are inherently damaging. Malacrida points out that in the UK, money is also an issue as some parents may wish to avoid the time and energy which needs to be invested to get a statement by their Local Education Authority. It is not just the impact on carers that may lead them to seek a diagnosis. The decision may be prompted by a crisis such as a divorce or loss of support, or that someone else insists on it (Zola, 1973). In the case of ASD, this insistence may come from a teacher (D. Thomas, Special Educational Needs Co-ordinator, personal communication, July, 2009). The picture emerging is of a trade-off between potential positive and negative outcomes, as parents weigh up the likelihood of risks and benefits before making a decision to get a diagnosis. It is therefore necessary to look at both the positive and negative aspects of receiving a diagnosis from a parental point of view in more depth.

Functions of ASD diagnosis for parents

Mansell and Morris (2004) give a good account of the positive aspects of diagnosis for parents in their survey of reactions to a diagnosis of ASD. Their study took place in counties bordering London in the UK. They note that literature relating to parental reactions suggests that the process involves four stages: pre-diagnosis, diagnosis, post-diagnosis and a final stage of acceptance and adaptation. During the period prior to diagnosis, parents begin to notice that something is wrong. Howlin and Moorf (1997) found this period ranges from between one and ten years and is characterised by stressful family relationships, confusion about the child's behaviour, guilt and self-blame. Some of the direct benefits of diagnosis Mansell and Morris identified included an explanation for the behaviour of their child (which absolved parents from blame for their child's condition), access to practical help for the child, contact with other parents in similar situations, access to other professional sources of support, and increased sympathy from others who were aware of ASD.

In the Mansell and Morris study, the actions around the diagnosis and immediately afterwards were fairly typical of those for most areas in the UK. The diagnosis incorporated input from a multidisciplinary team, including a community paediatrician, a clinical psychologist, a child psychologist, speech and language therapist, Portage worker, worker from the local parents support group and child psychiatrist. An action plan followed immediately after the diagnosis, suggesting several other interventions for example Treatment and Education of Autistic and Related Communication Handicapped Children (Schopier, 1988); speech and language therapy and PECS (Bondy & Frost, 2001). In addition, the diagnosis led to a parent-support course provided by the NAS and contact with the local autistic trust, who managed a parent-run body; the local parents' support group. This group had a full-time worker who normally visited the family in the week after the diagnosis and provided an information pack containing advice on the nature of ASD, local and national information and contacts, local professionals, education needs, therapies, support for carers, benefits and grants, childminding and respite services as well as specialised activities and clubs. This group provided a full-time service to support parents with their needs.

Although there is much theory relating to labelling and stigmatisation, it may be that parents tend to put more weight on short-term gains when considering diagnosis. According to Rogers (2007) at the time of diagnosis parents do not often think of the label as a life-long tag that the child will need to negotiate as an adult, but as a tool to procure support, therapy and resources. Indeed, Zurifff (1996, p. 403) sees diagnosis of learning disability as an 'admission ticket' to the entitlement of opportunities and special accommodations. This is backed up by the notes from the 2004 Special Needs Co-ordinator (SENCO) forum in the UK which found that giving a label to a child's needs was an 'essential passport' to obtaining resources for help (SENCO Forum, 2004). The release of educational resources may be a big factor in convincing parents that diagnosis is required. Looking at ADHD, Minow (1991) comments that parents want their children diagnosed and treated differently in order to secure specialized help to overcome obstacles to academic success. The main interventions available to children who have received an ASD diagnosis are special education for the child, respite for the parents and home-based behavioural programmes.

A brief history of relevant education policy in the UK

The question of educational provision for children with ASD has been intensely debated since the condition was first recognised. Most experts now contend that a range of provision needs to be available to provide for the wide spectrum of needs. This fits in with the broader framework of education legislation which makes it clear that children with special educational needs should receive provisions which cater for them as individuals. According to a study commissioned by the NAS in the UK out of a sample of over 1,000, 50% of those diagnosed with ASD are in specialist schools and 50% are in mainstream schools (Barnard, Prior, & Potter, 2000).

The Warnock Report (1978) recognised a continuum of need that connected children requiring additional assistance in ordinary schools with those placed in special schools. It also recommended the move towards greater integration/inclusion of children with severe SEN in ordinary schools and more involvement of parents in decision-making. It recommended that parents should be regarded and welcomed as partners. When the report was written, about 2% of school age children had special needs to such a degree that they needed to be educated outside mainstream education.

The report was influenced by social models and medical and educational sociology and indicated a shift in culture from the biomedical model of learning disabilities to a more individual need and context-driven approach. It recommended that categories such as 'handicap', 'subnormality' and 'abnormality' should no longer be used. Those children that were termed 'educationally subnormal' were described as having 'learning difficulties'. The report stressed that the emphasis should shift to an understanding of what an individual child needs, in terms of their learning opportunities and provision to learn and develop. This was reflected in the recommendation to use a description of a child's *special educational needs*, rather than labelling a child. Since the report made its recommendations, Corbett and Norwich (1997) have noticed a sea change in education where parents have shifted from accepting expert opinions to making demands for their children.

Most of the recommendations of the Warnock report were made into law, but this took over two years to implement in practice (Rogers 2007). It led to a decline in the proportion of children in special schools (1.8% in 1983) to about 1.3% in 2000, where the percentage has remained. Children with SEN in England and Wales are now defined as having 'greater difficulty in learning than the majority of children', or as having 'a disability which either prevents or hinders him [or her] from making use of educational facilities of a kind generally provided for children of his age in schools within the area of the Local Education Authority' (Education Act, 1996, p.312). A series of policy documents over the following years were introduced, restructuring the education system. Mainstream schools were asked to provide education for children with SEN 'where possible'. The emphasis shifted from the idea that emotional social and behavioural difficulties were medical and therefore treatable to the individual needs of each child and their educational potential.

The merits of this policy of inclusion are debatable. Rogers (2007) and Benjamin (2002) argue that although children are included physically in a class, in many ways exclusion still occurs:

- Physically: children are often removed from the class for one-to-one work in an individual teaching unit.
- Intellectually: they often cannot understand the curriculum in the same way as peers.

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- Emotionally: social impairments can make it hard for children to sustain friendships.

Chrissie Rogers (2007) suggests focussing on *educational* needs may lead to a lack of support in social situations where ASD children need help most. This may lead to isolation in the playground. She further argues that the problems of teaching all abilities together are compounded by testing and examinations. Along with the National Curriculum, the UK government introduced school performance data which were compiled into league tables, with the onus on raising educational achievement. She further points out that if exam results are the only currency to buy your way into employment, this poses a problem for many with cognitive impairments. Competition between schools leads to a conflict between inclusion and academic excellence. As intellectually impaired children have less chance of attaining academic excellence, a culture of low esteem is created, unhelpful to parents, teachers and children that are never going to achieve unrealistic goals. Rogers contends that in contemporary society intelligence is overvalued. According to her, intellect is consistently esteemed above other equally worthy traits, such as compassion, loyalty and determination.

Today, in the English and Welsh educational systems, a child can be classed as having SEN at school action, school action plus or statement levels. If the child is classed in the school action plus or statement group, then the SENCO will provide a record in an annual school census of whether the primary and secondary needs of each child are on the autism spectrum or within other categories. This record is not necessarily related to the planning of provisions for that child; it is merely a census for information purposes. Other categories that the SENCO will consider include specific, moderate, severe, or profound and multiple learning difficulties, behavioural emotional and social difficulties as well as hearing or visual impairments.

If the child is thought to need a statement of special needs, a statutory assessment will be conducted by a range of professionals (medical, psychological, therapeutic professionals and teachers alongside parental advice, as the National Autism Plan recommends). Their advice will go to a panel who will decide whether to issue a statement, which outlines the goals for teaching the child and the child's needs. The task of the panel is often to act as

gatekeepers for additional provision. The panel requires a full medical, with the clinician giving their opinion of an appropriate diagnosis. If provisions are scarce and the number of places available at a special ASD unit is limited, a child may not be allocated the appropriate provision without a medical diagnosis.

The provision made and funded by education authorities varies from one LEA to another. The options for a preschool or school age child with ASD may be:

- Mainstream school

- A school for children with moderate or severe learning difficulties

- Other type of special school or unit (e.g. assessment unit; language unit)

- Specialist unit or school for autism run by the authority

- Specialist school run by an independent organisation

- Support from an outreach advisory and teaching service

- Home-based programmes (e.g. Lovaas; musical interaction; Option; PECS; Portage)

There is no clear evidence on the relative benefits of different types of school placement or on which groups of children with ASD might be best placed in a mainstream school, generic special school or a school with an ASD unit (Jordan et al., 1998). Reviews have suggested that intensive, sustained special education programs and behaviour therapy can help children acquire self-care, social, and job skills (Howlin, 2006). Myers and Johnson (2007) however, state there is no evidence to back up claims that intervention in the preschool years is essential. There is currently no cure for autism (Francis, 2005), although education and skills intervention for children have been shown to lead to an improvement in self-reported mental health of parents (Tonge et al., 2006).

In 1998, the UK government commissioned a review of educational interventions for children with ASD. This demonstrated the bewildering array of social communication and interaction approaches available. Evidence to support each of them is inconclusive and inconsistent (Jordan et al., 1998). Despite this, many interventions are in everyday educational use for children with ASD (Scottish Intercollegiate Guidelines Network, 2007). Which intervention or which combination would be best for any particular child remains unclear. As some families may have as many as twenty different professionals involved the need to receive unambiguous information is important. Feinberg and Vacca (2000) bemoan

this situation, where a wide range of specialists work for short periods with one child. Given that a core symptom of ASD is difficulty in social interaction, it seems the first step in any programme should be for the child to develop a trusting and comfortable relationship with specialists. This is going to prove impossible for autistic children where exposure to a bewildering array of unknown specialists may be a source of distress. Feinberg and Vacca suggest funding should instead be directed to train a central caring person to be responsible for the mediation of all provisions, programmes and interventions, allowing a trusting one-to-one relationship with the child to develop over many years. A teaching assistant would probably be best placed to perform this role in the UK.

Unfortunately the advice on effectiveness of various interventions depends on who is giving it (Francis, 2005). Critiques from the scientific establishment have pointed out that programme marketing may have a greater influence than evidence-based research (Stahmer, Collings, & Palinkas, 2005). Of the many therapies and interventions available, few are supported by rigorous randomised controlled trials.

ASD diagnosis is often framed as a crucial step to obtaining appropriate help, but it is not always clear what this help should achieve. If ASD is viewed as a disease, like cancer, or mental health conditions such as depression that have been conceptualized as disease, then appropriate help should mean treatment of the core symptoms of autism. The goal of treatment for autism, as outlined by the Committee on Children with Disabilities (2001), is to improve the overall functional status of the child by promoting the development of communication, social, adaptive, behavioural and academic skills as well as lessening maladaptive and repetitive behaviours. However, although some interventions aimed at ameliorating core symptoms such as social communication have had some promising results (Dawson et al., 2009; Landa et al., 2011), as yet, there is no established 'cure' for autism. Reviews have suggested the field does not yet have interventions that meet the present criteria for well-established or efficacious treatment (Francis, 2005; Warren et al., 2011).

Non-behavioural special education classes are the main service providers for most ASD children, with individual therapies, such as speech and language therapy often

supplementing classroom teaching (Smith, 1996). There are also several different specialist approaches that are currently used in UK schools.

Medications are often used to treat problems associated with ASD. More than half of U.S. children diagnosed with ASD are prescribed drugs. The most common drug classes prescribed are antidepressants, stimulants and antipsychotics (Oswald & Sonenklar, 2007). However, medications do not treat autism's core symptoms of social and communication impairments and attempts over the past decades to develop drugs that specifically improve social and communicative functioning have failed (Buitelaar, 2003). There are no long-term studies suggesting that pharmacological treatments alter core difficulties or outcomes in children with ASD.

Prognosis and outcomes for adults with ASD
Behavioural outcomes include the persistence of symptoms, occupational outcomes show how well autistic adults maintain steady jobs, and social outcomes describe the network of relationships that the individual maintains.

Above average IQ and development of speech by age 5 are the best indicators of a good prognosis. A study by Howlin, Goode, Hutton and Rutter looked at 68 individuals with an IQ over 50 (2004). A minority of adults achieved independence, most remained dependent on their families and support services. Few lived alone, had close friends or permanent employment. Stereotyped behaviours and poor communication frequently persisted into adulthood. However in this study 12% were fully independent, 10% had some friends and were generally in work but required some support, 19% had some independence but were generally living at home and needed considerable support and supervision in daily living. The remaining 46% needed specialist residential provision from facilities specializing in ASD with a high level of support, and 12% needed high-level hospital care. Another study, carried out by Autism and Developmental Disorders Educational Research in 2001, showed that in the UK 17% of higher functioning individuals with ASD lived semi-independent adult lives. It also reported that 30-50% of those diagnosed with ASD do not develop functional speech: with childhood autism, independent living is extremely rare. For the small minority, often with high functioning AS and savant skills, there are some stunning

success stories for adults (see Tammet, 2007 as an example), but these are the exception rather than the rule.

Because of the changes in diagnostic practice over recent years and increased availability of early intervention, however, Howlin and colleagues (2004) pointed out that their findings which relate to autistic adults may not be generalisable to recently diagnosed children. More recent epidemiological studies suggest that the majority of children have an IQ of more than 70, within the range that is considered to be normal (Fombonne et al., 2005) and most acquire some useful speech (Volkmar, Rhea, Klin, & Cohen, 2005). So the prognosis for the ASD group as a whole may have improved, but perhaps only as a result of changes to the inclusion criteria.

When considering factors influencing prognosis, stable traits such as development of speech and language and IQ may provide insight, but may not be particularly helpful for affected individuals and their families. Ruble and Dalrymple (1996) suggest it may be more useful in practice to look at interventions and changes to the education and care which can be altered to the benefit of the individual. This view is borne out by the experience of parents whose children are ASD adults and of professionals who work in adult services (Morgan, 1996), who report that significant progress is often made throughout adult life. When interviewing parents with children aged 18 in a follow-up study, Gray (2006) found that the majority of children had improved to the point that they were significantly easier to live with, even if they maintained many autistic symptoms. Factors that contribute to these changes include the nature of the environment created for the ASD adult, the opportunities offered and how sensitively the adult can be supported.

Coping with ASD
Bury defines 'coping' as the cognitive processes whereby the individual learns to tolerate or put up with the effects of illness (1991). It is well documented that giving a name to a medical condition enables patients to understand and come to terms with it (see Cooper, 2002; Hilbert, 1984; for examples of this). It is especially true in complex conditions like ASD where parents are able to then research the condition, make sense of the diagnosis and learn what can be done. This, according to Rogers (2007), enables them to regain control over the situation. The need to understand is highlighted by the parents in her study who

had all received a diagnosis for various conditions. All these parents reiterated that the label helped them to research the difficulty and feel on top of it. Tied to this is the ability to cope with their child's distressing behaviour.

A significant factor in helping parents cope is the degree and type of social support available including the use of professional services. In his Australian studies, Gray (1994, 2006) found that treatment services were given as the primary coping strategy at the time of diagnosis, although a follow-up study indicated that use of treatment services declined dramatically as the children aged. The local parent-run body was the most highly rated of all services according to Mansell and Morris's (2004) survey. This provided parents with a chance to talk to others in a similar situation, to exchange information about services, their child's condition and to set up social networks providing mutual support. It has been shown that if level of support meets expected levels it has very positive effects on parental mental health. When going through difficult periods social support acts as a buffer to stress and facilitates recovery from crisis (Oakley, 1993).

Midence and O'Neill (1999) carried out a pilot study of parents seeking a diagnosis of autism for their children. These parents expressed confusion about their children's behaviour prior to diagnosis and subsequent relief when the diagnosis was given. The authors saw diagnosis as a way to enable parents to accept the limitations imposed on their child by autism. Receiving a diagnosis also appears to have made adaptations to family life easier as well as fostering a 'more realistic' picture of the prognosis. This mirrors work by Singh (2004) where she describes the relief of mothers on receiving an ADHD diagnosis for their children.

Mothers and blame

Nikolas Rose (1999) has discussed how 'maladjustment' from bedwetting to juvenile delinquency has become a sign of something wrong in the emotional economy of the family. Although the 'refrigerator mother' theory of ASD has been replaced by genetic and cognitive causal models, ASD difficulties can still be interpreted by onlookers, teachers and other parents as 'bad parenting'. In particular, mothers are usually viewed as having the greatest responsibility for their child and they often feel considerable guilt for their child's disability (Anderson & Eifert, 1989). Most research concerning the parents of disabled

children consists of samples comprised mostly of mothers. Despite this, as Traustadottir (1991) reports, participants are often described as 'parents'. Traustadottir describes this as a problematic 'gender blind' description of research. Singh (2003) also points out that the category 'parents' is often euphemistic for 'mothers'.

Feminist scholars have linked the notion of blaming mothers to pervasive and idealised norms of mother and mothering behaviours (Mc.Donnell, 2001; Ladd-Taylor & Umansky 1998; O'Reilly, 2001). Avery (1999) suggests the feeling of guilt uncovered in many studies is in part due to parental investment in our culture's conflated ideologies in the areas of 'good parenting' and 'perfect' children'. Mothers in Rogers (2007) study felt that judgement and apportioning of blame came from other mothers. This notion exemplifies Foucault's notion of a community surveillance system in which every individual is a subject exerting a disciplinary gaze, as well as the object of the gaze. According to Foucault, this dual role leads to an internalisation of the gaze (1979).

Receiving the diagnosis is variously described in the literature both as a relief for parents (Midence & O'Neill, 1999) and in other cases as a terrible shock. Adapting to the diagnosis has been compared to grieving for a child, as it shatters hopes and dreams of normality (Bury, 1982). Rogers (2007) found that when a parent is faced with the diagnosis of a condition, she or he may be faced with shock, horror, or disappointment. According to her account, diagnosis can involve the process of loss and serve as a turning point towards the development of a new parental identity. An ASD diagnosis also legitimises invasions by education and health professionals. However, according to Rogers, because the desire to produce a well, adjusted 'normal' child is so great, mothers often welcome this intrusion.

In reality, of course, the archetypal mother-child ideal does not exist. Parker (1997) reports on the ambivalence inherent in mothering, that no mother finds it easy to accept that they both love and resent their children. Parker describes maternal ambivalence not only an anodyne condition of mixed feelings, but a complex and contradictory state of mind, shared variously by all mothers, in which loving and hating feelings exist side by side. In Singh's 2003 study, many mothers spontaneously talked about their feelings of self-blame and inadequacy. Fathers were often absent at diagnostic clinics and even felt diagnosis was not necessary. Again Singh interprets this in the light of the cultural (patriarchal) context which

demands ideal motherhood, and she analyses medicalisation as an act of self-preservation for mothers to avoid blame for unacceptable behaviours in their children.

This function of the diagnosis in absolving blame has been examined more in studies on ADHD than ASD. Biologically orientated researchers of ADHD argue that the tendency to associate poor parenting with the condition is unfair and inaccurate. Parents are not the cause of problem behaviours, they are instead biologically determined (Barkley, 1988). Whalen and Henker (1976) pointed out that when hyperactivity is attributed to inborn neurological factors, both the parents and the hyperactive child feel less guilt about the exhibited behaviour. Illich (1999) makes a similar point that diagnosis leads to a transference of blame from parenting to biological and medical factors. This function of transference of attribution led Hinton and Wolpert to describe ADHD as 'the label of forgiveness' (1998, p. 316).

The absolution of blame as a function of diagnosis, then, is sparked by the change in attribution. The cause of a child's ASD is no longer attributed to a parent, but becomes medicalised: biological or genetic in origin. In Weiner's theory of attribution (1974; 1979; 1985), the locus of attribution becomes both internal (within the child) and uncontrollable (not affected by the actions of the child). Weiner points out that this in itself creates a dilemma of reducing personal responsibility which inhibits the desire for personal change. At the same time, altruism and sympathy from others are promoted by uncontrollability. Smelter et al. (1996) also express concern that an ADHD diagnosis absolves the child from all responsibility for his or her behaviour in the school setting, and is actually becoming a desired diagnosis, giving children kudos.

Negative aspects of ASD diagnosis

Many overarching parental concerns when considering whether to have a child diagnosed stem from the sociological concept of labelling. Labelling theory was developed in part by Becker (1963). He argued that people who are labelled as deviant and treated as deviant become deviant. This sentence can be broken down into two, one that refers to the effects of self-labelling (self-fulfilling prophecy) and the other the differential treatment from society based on an individual's label (stigmatisation). Applied to ASD, the theory hypothesises that diagnosing and labelling a child as autistic will lead to shifts in

expectancy and attitudes of those in contact with the child, as well as altering the identity of the child, and this will serve to reinforce the 'abnormal' behaviour in question. Molloy and Vasil (2002, p. 664) argue that the diagnostic criteria ignore 'the role played by those doing the labeling in shaping and creating the condition'.

Labelling theory was often touted by social psychologists in the 60's and 70's. It influenced social policy and the practices of schools and asylums. Today, psychiatric training cautions professionals about the dangers and consequences of labelling patients too early, while educational research into the impact of streaming children has informed the development of the comprehensive system in Europe and the USA (Slattery, 2003).

Bogdan (1982) drew on work in semantics to suggest that people labelled 'mentally retarded' were covered with a 'cloak of incompetence' that is virtually impossible to discard. They thus become prisoners of the explicit and implicit meanings attached to these labels. Bogdan argued the label 'mentally retarded' created a barrier to understanding people on their own terms. It prevents others from seeing and treating people so defined as human beings with feelings, understandings and needs. Such work was instrumental in the replacement of derogatory terms with politically correct language over the last few decades which attempts to refer to disabled groups in a more positive way.

Edgerton (1967) pointed out that despite the enormous amount of literature in the field of cognitive impairment; nearly all of it was from the perspective of social workers, psychiatrists, psychologists and other medical specialists. In such circumstances, he elucidated, people with intellectual disabilities are only ever professionally 'known' in terms of their 'disabilities' or 'abnormalities'. This, then, informs the way that such people are perceived and treated; objectifying them. Goode was an anthropologist who attempted to get round this. He also argued that the focus on labels does not allow entry into the world of an intellectually disabled person. He attempted to eradicate the professional distance between himself and his subjects. He studied and developed a relationship with a severely intellectually impaired deaf/blind girl on her terms, for example (1984).

At first labelling theory, though influential, was fairly conceptual and lacked experimental evidence to back up its claims. In order to search for empirical findings that might support

its assertions, it is necessary to look at literature in psychology on stigmatisation, attribution theory and self-fulfilling prophecies.

Stigmatisation

This term originated from the ancient Greek word 'stigma'; a brand given to an animal or escaping slave. The word was extended to include any mark or sign for perceived deviation from the norm, and the bearer of the mark is thus defined as flawed, spoiled, and generally objectionable.

Stigma as a sociological concept was developed by Goffman in his pioneering book on prisoners *Notes on the Management of a Spoiled Identity* (1963) and has been applied to a wide range of settings, including the study of families of children with disabilities (Baxter, 1989; Gray, 1993; Farrugia, 2009). The parents of children with disabilities experience what Goffman defined as a 'courtesy stigma'. This is conceptualised as a stigma of affiliation that is applied to people who associate with stigmatised groups rather than through any quality of their own.

Gray (2002) looked specifically at stigma amongst a sample of parents who had children with high functioning autism. A majority of the parents had experienced stigma, especially mothers. Some mothers had been subjected to avoidance, hostile staring and rude comments. Other parents of children with disabilities – including childhood autism –also experience stigma (Farrugia, 2009; Baxter, 1989) or feel that they are stigmatised, even when it is difficult to ascertain whether this is really the case (Norvilitis, Scime, & Lee, 2002). Gray (1993) found that parents with younger or more seriously impaired autistic children reported themselves to be more stigmatised. Sabornie (1994) found that students with learning difficulties often express more loneliness and feel less integrated in school, compared to those without difficulties. Students reported being threatened, and even physically assaulted more frequently. Edgerton (1967, p.207-12) argued that the stigma of cognitive impairment leads to lower self-esteem, and, in order to maintain self-esteem, mildly impaired people must deny their condition. Stigmatisation does not only apply to children with low IQ. Many high functioning children in Gray's 2002 study experienced isolation or bullying at school and few had stable friendships with other children. One common effect of this stigma is social rejection for both the children and their parents. Gray

found parents' relationships with teachers and school administrators and even other parents were often strained.

The children in these studies were all diagnosed and it is hard to uncover to what extent it is the label that causes the stigma, or the behaviour of the child. Gray suggests that the normal physical appearance of ASD children combined with disruptive behaviour leads to greater stigma. Jussim, Palumbo, Chatman, Madon and Smith's' review of self-fulfilling prophecies and stigma only identified two studies looking specifically at childhood stigma in relation to diagnostic labelling (2000). In these experiments, boys were paired and one in each pair was led to believe that the other had an ADHD diagnosis. Boys perceived ADHD labelled targets as having more symptoms, and they talked less to them, were colder and gave them less credit in shared tasks. The ADHD targets felt that their partners were 'meaner' to them than the non-labelled targets did. This has interesting implications for ASD children, where social communication is a problem- indicating as it did that the diagnostic tag actually made social difficulties worse. However, a follow-up study showed that the effect of *actually* being diagnosed as ADHD had a far greater effect than being incorrectly labelled by experimental manipulation, even when the perceivers were unaware of the diagnostic labels. This was interpreted as showing that the actual behaviour of a boy had a far greater effect than a bogus label (Harris, Milich, Corbitt, Hoover, & Brady, 1992). Stigma is more likely to be inflicted because of a child's disruptive behaviour rather than because of a label per se. Of course, this work did not discount the possibility that the behaviour itself was partially caused by a self-fulfilling prophecy (SFP) from the diagnosis.

Farrugia (2009) analysed 12 parent interviews and concluded that a child's diagnosis with ASD is critical for parents to resist stigmatisation. Parents successfully resisted felt stigma by deploying neurological and biological models of ASD. Resistance to stigma was successful, he surmised, to the degree that medical constructions of ASD deployed by parents were accepted by others, notably those in power within institutions. Farrugia notes that parents wished they could dress their children in T-shirts proclaiming their autism. Such T-shirts have been produced by NAS in the UK, and Autism Speaks in the USA where they are in common use (Figure 1). Autism identity cards are also used to the same effect.

Figure 1: Autism T-shirt for young child.

Farrugia's analysis claims that courtesy stigma lessens for parents as others' attributions for their children's bad behaviour are switched from poor parenting to biological factors. Some have argued that inherently in this process, the child's identity may be 'spoiled' in Goffman's terms (1963), in order to protect the position of the parent. In a radical feminist analysis Singh (2003) describes the 'sacrifice' of the son to protect the mother.

Thus the work done by the diagnosis is in part to allow parents and others to deploy biomedical explanations. Grinker, the father of an autistic child, compared approaches to autism across cultures, as his family moved from Korea, to South Africa and India (2008). He found that the biomedical definition was a useful tool to explain his daughter's behaviour in order to deflect culturally situated definitions. For example, autistic symptoms might be attributed to demonic possession in some cultures. His work demonstrates that the deployment of the biomedical explanation by the parent is in part a response to the cultural context that the child inhabits.

In the Mansell and Morris (2004) survey some parents stated that their offspring had been treated worse *as a result of the diagnosis.* There is little evidence in the literature to show conclusively whether the diagnostic tag is a source of stigma in itself.

Another question that appears unanswered is whether a genetic diagnosis reduces or increases stigma. That is, as ASD has more recently been accepted as having biological, and primarily genetic causes, does this elicit more understanding attitudes and less stigmatisation? One can hypothesise that as a genetic or biological explanation can absolve parents of blame, then the stigma for parents and children should lessen. Weiner, Perry and Magnusson (1988) studied attributions for obesity and Acquired Immune Deficiency Syndrome (AIDS) and found that if conditions were perceived as uncontrollable they elicited more pity and less anger. However, endorsing biological factors as the root cause for schizophrenia and other mental illnesses has been associated with a greater desire for social distance from individuals with the condition, according to a review of this area (Read, Haslam, Sayce, & Davies, 2006). The sociological literature is full of examples of illnesses that are both biological and stigmatised (Nettleton, 2006).

Attribution theory

Attribution theory has encompassed work on depression, alcoholism, criminality and loneliness. Attributions are concerned with the way people attribute causality to outcomes. Whenever people encounter negative states or events, they search for causal explanations, and these may be classified as internal or external to a person, stable or unstable, controllable or uncontrollable, and global or specific. One of the most influential researchers in this area is the social psychologist Bernard Weiner who described these as the dimensions in which attributions take place. To understand these dimensions, an example is required: if a teacher ignores a child, this may be interpreted by the child in various ways. She may attribute it to the teacher having a headache, for example, which would be an external attribution as it is not directed at the recipient of the event, unstable as it unlikely to be repeated, uncontrollable and specific to that situation. Alternatively, the child might assume that stupid children never deserve to be acknowledged which would be an internal, stable, uncontrollable attribution. Such internal, stable, uncontrollable attributions have been associated with a state of 'learned helplessness'; a predisposing factor for depression (Seligman, 1975).

Uncontrollable self-attribution has also been shown to lead to withdrawal and lack of motivation (Wicker, Payne, & Morgan, 1983). A genetic predisposition to a condition is uncontrollable. It is possible that if a child has a clinical diagnosis of ASD, parents are

more likely to attribute autistic behaviours to genetic factors than they would in an undiagnosed child. This in turn might affect efforts to improve behaviours. Lavelle and Keogh (1980) found that if parents perceived their child's disability as internal to the child and stable, then expectations were low and intervention efforts were focussed on 'maintaining' rather than remedial. Where disability was perceived as internal but changeable, expectations were higher and intervention efforts were more vigorous.

A type of cognitive therapy, known as 'attributional retraining', is based on shifting the focus of attribution from uncontrollable dimensions such as ability - to controllable ones - such as effort. Attribution retraining programs have improved performance and persistence (Forsterling, 1985), as well as improving outcomes in writing (Hall et al.,2007) and academic achievement (Ruthig, Perry, Hall, & Hladkyj, 2004). Weiner's work has shown that achievement is linked to both ability and effort. A study by Barnhill (2001) on adolescents with AS revealed that if the teenagers attributed social failings to their own ability, they were more likely to suffer from depression.

Motivation is associated with the incentive to achieve (what a child can get) coupled with expectancy of success (the likelihood of getting it). Plenty of experimental work has taken place in educational psychology testing the effects of expectation. The expectation of success typically increases if a goal is attained, and decreases if it is not (Parsons & Ruble, 1977). A study by Stipek and Hoffman (1980) showed that boys with a history of low academic success in school had lower expectations for success on the task and tended to be more likely to attribute failure to lack of ability. Martinko, Weiner & Lord (1995) demonstrated that internal ascriptions lead to lower self-esteem for failure than external attributions.

Howlin and Rutter (1999) explained that whilst children with autism show many of the behavioural problems that normally developing children show, they may persist to a later age than is usual due to their delay. Parents often attribute these behaviours to the child's autism. This may affect how parents interact with their children. A study by Rubin and Mills (1990) concluded that parents who attribute social withdrawal to an uncontrollable trait within the child are less likely to attempt to modify the child's behaviour. Furthermore, in Mansell and Morris' 2004 survey, many parents saw their children make good progress

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and this lead them to question the original diagnosis which they felt meant that their child would not change.

Lack of ability is not an easy attribution for parents, and particularly mothers, to make. Literature from developmental psychology shows parents usually overestimate what their children can do; a kind of positivity bias. Mothers tend to give internal stable explanations for good behaviour, and external unstable ones for bad behaviour according to Dix (1993).

In the light of attribution theory, this serves a useful function in parenting, encouraging good behaviour and discouraging unwanted behaviour. According to developmental psychologists, parents who make optimistic attributions will be most likely to persist in helping their children overcome problems, as well as most likely to transmit a positive, development-enhancing pattern of attributions to the children (Neuman, 1997; Wagner & Phillips, 1992). Parental optimism may have a useful adaptive function.

If parents believe that autism has primarily genetic and/or environmental causes, this suggests that uncontrollable attributions are often made. The question is whether an ASD diagnosis is more likely to lead to these attributions for autistic traits. There is some evidence to suggest internal, uncontrollable and stable attributions may allow others to take a more sympathetic view, but also some evidence suggesting these may impact on a child's well-being and development.

Self-fulfilling prophecies

The notion of the self-fulfilling prophecy (SFP) was conceptualized by the sociologist Robert Merton in a 1957 work called *Social Theory and Social Structure*. The phenomenon occurs when a false definition of the situation evokes a new behaviour which makes the original false conception come true. Reviews of SFPs and behavioural conformation appeared in social psychology literature in the 1980s and 90s which highlighted the power of inaccurate beliefs to create erroneous realities. Placebo effects and false memory syndrome have been prominent examples (Jones, 1990). The SFP concept spawned experimental work in many diverse areas including market fluctuation and faith healing (Henshel, 1982). In education, teacher expectancies, streaming and self image have all been shown to have an influence.

Research has documented SFPs in both the lab and the real world. Rosenthal and Lawson (1964) told students who were training rats to run round a maze that their animals were either clever or stupid. Although rats were actually randomly assigned, the 'clever' rats learned the maze quicker. Students' beliefs about their rats were self-fulfilling. In Rosenthal and Jacobson's influential, much criticised yet highly cited 'Pygmalion' study (1966), the researchers posed as educational psychologists, went to a school and tested a class of pupils. They then told the teacher that 20% of these pupils were 'intellectual bloomers': potentially brighter than the others, despite these 20% actually being selected quite randomly. When the children were re-tested a year later, the 'bloomer' pupils really did do better in intelligence tests. Once an expectation is set, they argued, even if it isn't accurate, people tend to act in ways that are consistent with that expectation. The result is that the expectation comes true.

The work of Rosenthal and his colleagues provoked a flurry of work on SFPs, and in 1978 Rosenthal and Rubin reviewed 345 separate studies dealing with the phenomena, which confirmed its existence. The effects of SFPs have been noted in many settings, including in classroom amongst stigmatised groups (Jussim & Harber, 2005). The expectancy of worse outcomes for a target group has been shown to be self-fulfilling. Teacher perceptions were shown to predict achievement more strongly for low achievers than for high achievers (Madon et al, 1997). Rist (2000) showed how a preschool teacher placed children in reading groups that reflected their class backgrounds, and how subsequent teacher expectancy mediated learning. Another study showed how students who were more likely to have difficulties were assigned to groups less conducive for learning. These results question the utility of streaming by ability (Eder, 1981).

SFPs have also been shown to be self-mediating, that is the belief is about oneself rather than the expectation of others. An interesting experiment showing this effect was carried out by Steele and Aronson (1995). They gave two groups the same test. One group was told it tested linguistic ability, the other problem-solving ability. African Americans who perceived themselves as poorer linguistically did worse on the linguistic test than when they thought it was problem-solving exercise, an effect known as 'stereotype threat'. A

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similar effect has also been found in research using socioeconomic status (SES) rather than race (Croizet & Claire, 1998).

Teachers can also make false judgements based on labels. Fogel and Nelson (1983) found that disability labels could bias teachers' behavioural checklist scores. In their ingenious study, teachers who watched a video of a child and were told about their learning difficulties attributed more characteristics of cognitive impairment to the child than teachers who were not aware of the diagnostic label for the same child. In another neat experiment, Farina and colleagues (1971) told subjects that others believed they were mentally ill: even though in reality those perceivers have had no knowledge of their mental health status. Just believing other people thought they were mentally ill made them behave in ways that elicited more rejection from those perceivers.

The ASD label, then, could produce unconstructive perceptions and reduced expectations in parents, teachers, peers and even the children themselves, leading to self-fulfilling outcomes. Meta-analysis of all work on SFPs found effects real but small:- just 0.1-0.2 in terms of correlation or regression coefficients (Jussim et al., 2000). However, the size of the effect varied in different situations and contexts. Although SFPs are real, it can be concluded there are probably more powerful factors are at work in maintaining social stigmas.

Summary

Several potentially negative aspects of receiving a medical diagnosis, then, have been tentatively identified through the literature. Despite the many incentives to get a diagnosis, some parents do decide not to do so. Some of their reasons could be informed by labelling theory, others by more basic considerations. These include:

- Too costly, lack of time, energy.
- Invasion of privacy by numerous professionals.
- Reluctance to risk receiving a poor medical prognosis.
- Stigmatisation due to labelling.
- Potentially worse outcomes for the child due to a self-fulfilling prophecy.

If parents have fallen on the 'no' side of the diagnostic dilemma, however, by choice they are unlikely to leave an official trace.

Section 3: Aims, objectives and questions of study as originally intended

This section starts with a brief summary of the literature reviewed in the previous sections to clarify how the theoretical background led to the formulation of the initial research questions under study.

As outlined in the previous section, it is generally accepted within the medical literature, within organisations dedicated to the interests of those with ASD, in policy guidelines, and by most parents of autistic children that ASD is partially genetically determined and diagnosis should be made as early as possible to treat and manage the condition effectively.

In the UK, a medical diagnosis of ASD can act as a gate-keeping mechanism to release educational, social, professional and medical resources. The main interventions available for children who have received an ASD diagnosis are special education for the child, respite for parents and home-based behavioural programmes. Educational interventions are often mediated by a statement of special educational needs, drawn up ideally by a multidisciplinary team including a clinician who will issue the medical diagnosis. Educational policy has led to more inclusion in mainstream schools for children with special needs.

A diagnosis of ASD is viewed by some parents as a way to access resources, particularly support in education. In fact, the effectiveness of particular early educational interventions for ASD is inconclusive and inconsistent, according to Jordan, Jones and Murray (1998). Myers and Johnson (2007) found no evidence for claims that preschool intervention is essential. Nevertheless, reviews have concluded that there are enough positive outcomes to indicate that some form of intervention is warranted (Guralnick 1996; Tonge 2006). In the biomedical realm, ASD is conceptualized as a neurodevelopmental condition for which appropriate treatment can be obtained. Although most medical practitioners accept that ASD has biological, psychological and social causes, there is more extensive funding for studies aiming to uncover biomedical, particularly genetic and neurological causes, than other causal factors (Szpir, 2006).

Following from this biomedical perspective on ASD, it can be hypothesized, that diagnosis and the subsequent service contact and treatment this entails should lead to improved outcomes. However, although diagnosis is often framed as a crucial step to obtaining appropriate help, it is not always clear what this help should achieve. If ASD is viewed as a disease, like cancer, or a mental health condition such as depression that has been conceptualized as disease, then appropriate help should mean treatment of core symptoms for autism. As yet, there is no established 'cure' for autism. Reviews have suggested the field does not yet have interventions that meet the present criteria for well-established or efficacious treatment (Francis, 2005; Rogers, 1998).

The literature reviewed in the previous section from sociological and psychological traditions concerning stigmatization, SFPs and attribution theory, in contrast to the dominant biomedical discourse, suggest there may be some negative consequences to diagnostic labelling. This tradition of research is largely overlooked in the psychiatric and medical literature. One exception is a recent study by Sayal and colleagues (2010). These researchers conducted a randomized controlled trial where children who displayed high levels of inattention/hyperactivity were identified as having high probability of having ADHD to teachers. A second group, with equivalent symptoms, were not identified to teachers. The researchers found that children in the 'identified to teachers' group were twice as likely as the others to have high scores in inattention/hyperactivity at follow-up 5 years later. The authors discuss labelling theory and teacher expectancy as one possible reason for these outcomes, although another could be reporting bias. Despite the results of this study raising the question of the possible negative consequences of conspicuous identification, the authors still state 'the identification of these children by teachers and/or parents is the crucial first step in receiving appropriate help' (p. 462). The over-arching tenet, that identification/diagnosis is crucially important, seems to run deep through the psychiatry literature, even when anomalous results are apparent.

Gray looked specifically at stigma amongst a sample of parents of children with high functioning autism (2002) and classical autism (1993). A majority of the parents had experienced stigma, especially mothers. Other parents of children with disabilities – including low functioning autism –also experience stigma (Baxter, 1989; Voysey, 1972) or

feel that they are stigmatised (Norvilitis, Scime, & Lee, 2000, Farrugia, 2009). There is evidence to suggest that children themselves are also stigmatised (Sabornie, 1994). As all the children in these studies were diagnosed, it is hard to uncover to what extent it is the diagnostic label that causes the stigma, or the behaviour of the child.

Work on self-fulfilling prophecies has shown that the expectancy of worse outcomes for a target group can be self-fulfilling (Rosenthal & Rubin ,1978). Once told about a specific diagnosis, perceivers tend to interpret behaviour in the light of this diagnostic frame (Fogel & Nelson, 1983; Rosenhan, 1973). Self-fulfilling prophecies have also been shown to be mediated by individuals themselves rather than others' expectancies (Steele & Aronson, 1995; Crozet & Claire, 1998). But only two studies have been identified that looked at the effect of diagnostic labelling per se. (Jussim et al., 2000). These indicated that the diagnostic tag made social interaction more difficult, but that the effect of actual bad behaviour had a far greater influence than a bogus diagnostic tag (Harris et al., 1992). Overall the literature reports that the effects of self-fulfilling prophecies are real but weaker than other factors (Jussim et al., 2000).

It can also be hypothesized, then, that according to labelling theory, diagnosis might in some cases lead to *worse* outcomes. The ASD label could produce negative perceptions and reduced expectations in parents, teachers, peers and even the children themselves, leading to self fulfilling outcomes.

The central aim of the study as originally conceived was to examine whether diagnosis of ASD (and subsequent service contact) has an effect on behavioural and social outcomes.

There are very few empirical studies that attempt to disentangle the effect of diagnostic labelling from other forms of labelling. One problem for researchers is the difficulty in finding matched controls; undiagnosed groups with comparable behaviours (Angold, et al., 2000). In psychiatric research, prevalence literature indicates that there is a substantial minority of undiagnosed children whose behaviour meets diagnostic thresholds for psychiatric disorders (Ford et al., 2003; Goodman & Scott, 1997), but who have never received a diagnosis because their condition fails to have sufficient impact on their families (Rutter, 1989).

Several large, population-based surveys of child psychiatric disorders have provided data on the number of children who present behaviours that might justify a diagnosis. These studies were reviewed by Costello (2005) who found most have reported that some kind of psychiatric disorder is present in roughly 10-25% of children. Such studies imply that children with impairing psychopathology who have received a clinical diagnosis represent only a portion of the total number. For example, data from a large UK population based sample suggested only 25% of children with psychiatric disorders had been in contact with mental health services over the subsequent three years, and nearly half had no mental health related contact with public sector services at all (Ford, Hamilton, Meltzer, & Goodman, 2007). A recent study by Baron-Cohen et al. (2009) found that the ratio of ASD cases identified in the community (diagnosed) to numbers identified through research is about 3:2 in mainstream primary school children aged 5-9 years old. Another British study based in South East England screened a sample of undetected cases who were at risk and estimated prevalence to be over 1% in children aged 9–10 years old (Baird et al., 2006). Such studies suggest that not all of the large number of children displaying autistic type behaviours are identified as having difficulties. Not all children access specialist child health services and subsequently receive a diagnosis.

This thesis is not concerned with the true prevalence of ASD, rather with the overview of literature, it aimed to establish if there is a population of undiagnosed children whose behavioural profiles are comparable with those children clinically diagnosed with ASD. The overall objective of the study, as it was originally conceived, was to examine differences in outcome between children diagnosed with ASD, and those identified with severe autistic behaviours but no clinical diagnosis. Therefore the central hypothesis was:

> Diagnosis of ASD (and/or subsequent service contact) has an effect on behavioural and social outcomes, compared with an undiagnosed group who have comparable autistic type behaviours.

As outlined above, the core research aimed to examine whether diagnosing ASD and subsequent interventions led to an improvement or a deterioration of the condition over time, compared to an undiagnosed group who exhibit autistic type behaviours. Whilst

working towards the analysis to test the central hypothesis, the research also encompassed two subsidiary hypotheses:

1. There are children with autistic type behaviours at the same severity as those with a diagnosis who remain undiagnosed.

Given the inclusion of 'impact' in the diagnostic criteria, and with reference to the literature on access barriers and parents' surveys suggesting parents play an active role in determining whether children are diagnosed or not, an aim of this research was to ascertain if there are a group of children who would be diagnosed with ASD were they brought to the clinic. The second subsidiary hypothesis was related to this:

2. There are social and demographic or family-based factors in the children's backgrounds that may influence which of these children are diagnosed and which of them are not.

Parental perspectives on ASD diagnosis
It can be concluded from the literature reviewed that diagnosis has a number of important functions from a parental point of view. Some of those identified functions show that diagnosis seems to allow parents to:

- Identify, research and understand the condition enabling them to feel in control.
- Accept the autistic behaviour, and adapt to family life.
- Absolve them from blame.
- Access practical help such as respite care.
- Find social and emotional support through parent and professional groups.
- Access new educational pathways and resources for their children.
- Identify potential therapeutic interventions.

These functions seem to fall into two broad categories, those that directly benefit parents and those that facilitate improved outcomes for the child. The direct benefits of diagnosis for the parent include understanding the condition, accepting the behaviours, accessing

practical help/support and absolution. Tailored educational programmes and therapeutic interventions, on the other hand should lead to improved outcomes for the child. Obviously, these two categories are closely interdependent; direct benefits to parents almost certainly have a positive influence on children in terms of relieving stress within the family and improving relationships. Improved outcomes in ASD children would clearly be better for parents. Nevertheless these two categories are not the same thing, which begs the question; is what is best for the child always the same as what is best for the parent? Could there be tension between the needs of the child and that of the parent? In her study on ADHD and Ritalin, Singh (2003) believes this is the case and adopts a radical position describing the 'sacrifice' of the son to protect the mother. She therefore asks a variation of this question: who benefits more from Ritalin treatment, the child or the mother? The broadly inconclusive evidence on the merits of educational interventions and home-based therapies and treatments for ASD reinforce this type of question in the context of autism.

For parents of children with autism, some literature suggests the labelling theory issues outlined above are at least a cause for concern. Mansell and Morris's (2004) survey found that parents of autistic children expressed a range of negative reactions after diagnosis. The majority of parents stated that they had become more worried for their child's future, that it was difficult to know which problem behaviours were caused by the condition and which were not. A substantial minority also reported that their child had been treated worse as a consequence of the diagnosis, or in some sense stigmatised.

In tandem with looking at outcomes of children, the thesis aimed to explore parental perspectives on ASD diagnosis. The tensions between sociological and biomedical perspectives are reflected in the dilemmas faced by parents as they decide whether to seek formal identification of their child's difficulties (Norwich, 2008; 2009). One objective of the submitted study was to explore whether parents of children considering obtaining an ASD diagnosis for their child experienced similar dilemmas, or indeed, whether parents were active agents in obtaining an ASD diagnosis at all. The dilemma in this case can be encapsulated in terms of the tension between the perceived positive benefits and possible disadvantages of receiving an ASD diagnosis. The analysis of such dilemmas has been adopted by several other studies in the field (for example, Hansen & Hansen, 2004).

With reference to the literature on dilemmas of diagnosis, and tensions exposed in other qualitative studies, several research questions were formulated:

Are parents active agents in the process of obtaining a diagnosis of ASD for their child?

If so, are there dilemmas that parents consider when weighing up whether to act to pursue a diagnosis?

What triggers the parents to ask clinicians to make the diagnosis? Is this affected by personal or socio-economic factors in the situation of parents?

How do parents, whose children have received an ASD diagnosis, perceive the diagnosis; as helpful or stigmatising?

What does diagnosis actually do for these parents; what function does it serve?

The study that addressed these research questions is referred to in the next section and throughout this thesis as study 2. One objective of the study was a nuanced examination of whether parents of children with autistic type behaviours were active in determining whether a clinical ASD diagnosis was administered, and particularly why parents do *not* pursue services and subsequent diagnosis. Secondly, it sought to examine what function the ASD diagnosis performed for parents whose children had received one, and finally, how having an ASD diagnosis altered differences in position between parents whose children were diagnosed with ASD and parents of undiagnosed children who had been identified by professionals as on the spectrum.

The study which aimed to test the central and subsidiary hypotheses listed above, is referred to as study 1, or the main study throughout the thesis. A secondary objective of study 2 was to highlight areas of concern for parents, feeding back into the study design (selection of outcome variables of interest) of the main study.

Section 4: The methodology (as a set of principles)

i) Principles of methodology: Study 1

1. As Crawford and Vignoles (2010) point out, there is a pressing need to compare outcomes in children who have been identified with a comparable group of unlabelled children as they progress, as a means of evaluating likely effectiveness of provision. Angold et al. (2000) also discuss the need to compare outcomes in children who have been in contact with mental health services with matched controls. However, both sets of authors also discuss the difficulties in finding a signal to noise ratio in such an undertaking. It is important to distinguish between efficiency and effectiveness when considering the merits of service contact for children. Studies of the efficacy of a specific intervention explore its capacity to produce a desired size of an effect under ideal or optimal conditions, i.e. how much an intervention improves outcomes. The effectiveness of service contact, by contrast, shows the improvements in outcomes in real-world settings. Kelley, Nixon and Bickman (2000 p.466) describe effectiveness as 'allowing examination of such issues as service access, utilization, cost and quality, in addition to the issue of service impact (outcome evaluation)' and argue that more research on what works in real-world settings is urgently needed. This was the theoretical methodological approach of the outcomes study, as it was initially conceived, as advocated by Kelley, Nixon and Bickman (2000), Angold and colleagues (2000), and Crawford and Vignoles (2010). The study aimed to detect effectiveness of service contact triggered by diagnosis of autism spectrum disorders, (together with effects of diagnosis itself) through use of an appropriate control group.

2. As undiagnosed children with psychiatric disorders, and specifically autism spectrum disorders, are included in prevalence estimates taken from epidemiological population-wide studies, then it can be assumed that there are undiagnosed autistic children with symptomology comparable to those diagnosed.

3. In the prevalence literature, children may be given a 'research diagnosis' which is made according to measures taken by the epidemiological cohort of interest. This qualifies them as being included in prevalence estimates for ASD and other disorders. Therefore we can say that such children can be considered autistic but unidentified according to such studies (e.g. Baron-Cohen et al., 2009).

4. It should therefore be possible to identify a group of children who have comparable symptoms to those clinically diagnosed with ASD but remain unidentified in their communities, using a population-based cohort which has taken detailed measures of autistic traits. Quantitative techniques are most appropriate to test the central and subsidiary hypotheses.

5. If children who are not diagnosed are to be compared with children who are diagnosed, then the control group should meet clinical thresholds for each area of qualitative impairment necessary to ascribe an ASD diagnosis, specified in the diagnostic criteria for ASD, and severity of symptoms must be at comparable levels to the clinically diagnosed group.

6. Specific diagnostic criteria are particular to sub-conditions within the autism spectrum. According to ICD-10 criteria, to be ascribed a diagnosis of AS, children differ from children diagnosed with childhood autism in that they have no delay in language development. Measures of language development are not therefore suitable to predict AS diagnosis. In order to best match across symptoms, the sample should be split into the common sub-conditions within the autism spectrum.

7. If autistic type behaviours are used to identify the control group, if these are measured earlier than diagnosis, they may not be sensitive or specific enough to pick out a comparable undiagnosed group. Increasing variation in behaviour could account for differences in outcomes. If autistic behaviours are measured later than diagnosis, post-diagnostic bias could alter results i.e. diagnosis itself could have an influence on the behaviours measured. Measures of autistic type behaviours used to identify controls should therefore have been reported in the same age ranges as ASD diagnoses were ascribed.

8. If symptoms are comparable at baseline between diagnosed and undiagnosed children, then it may be possible to detect socio-demographic factors in the children's backgrounds that are predictive of whether an ASD diagnosis is applied or not.

9. If symptoms are comparable at baseline between diagnosed and undiagnosed children, and diagnosis was made at the same time as symptoms were measured, then analysis of outcomes some time afterwards should reveal any differences due to diagnosis and subsequent service contact. As one aim is to uncover any effects of diagnostic labelling/service contact, in order to better isolate such effects, undiagnosed children with extensive service contact should be excluded from the analyses.

10. As the study as it was initially conceived aimed to examine the possible effects of diagnostic labelling, any children diagnosed with alternative neurodevelopmental disorders should be also be excluded from the undiagnosed control group.

ii) Principles of methodology: Study 2

A number of exploratory research questions were drawn up as a result of the literature reviewed, at the outset of study 2. These were as follows:

Are parents active agents in the process of obtaining a diagnosis of ASD for their child?

If so, are there dilemmas that parents consider when weighing up whether to act to pursue a diagnosis?

What triggers parents to ask clinicians to make the diagnosis? Is this affected by personal or socio-economic factors in the situation of parents?

How do parents, whose children have received an ASD diagnosis, perceive the diagnosis; as helpful or stigmatising?

What does diagnosis actually do for these parents; what function does it serve?

The principles of the methodology referred to the formulation of the research questions. The principles of the methodology for study 2 were as follows:

1. The additional questions drawn from the knowledge base are not in the form of a testable concrete hypothesis as in study 1. Rather, they can be viewed as starting points for exploration of issues for parents around diagnosis. Qualitative methods are therefore appropriate to address these questions which provide an examination of the real issues for parents around diagnosis in everyday life.

2. The qualitative study should generate examples of the general relationship established by the statistical methods in study 1. The second study should be used to deepen the understanding of the issues around diagnosis for parents, in terms of processes behind trends seen in the main study; for example, reasons why so many children remain undiagnosed. Methodological triangulation should consequently be utilized in its broadest sense; quantitative methods showing data trends and qualitative methodology providing context and meaning to these trends (Silverman, 2006).

3. In order to complement findings from the main study, the second study should recruit two groups to reflect the methodology adopted, allowing comparisons between the experiences of one group of parents whose children have received an ASD diagnosis, and another group who, although they may suspect their children lie on the spectrum have not received such a diagnosis.

4. Data collection should take place at the convenience of participants, and in their own homes with no children present, in order to foster an undisturbed environment in which participants feel confident. A series of open-ended questions should be designed to allow parental narratives to unfold.

5. As Riessman (1993) points out, the authority of the researcher is itself an important factor in how participants respond. In order to establish a more neutral and open relationship between researcher and participant, the principal researcher should adopt a self-reflexive approach.

6. In order to minimise the effects of researcher bias, Hammersley and Atkinson (2000) point out that the researcher is an instrument through which qualitative data are interpreted. In order to minimise the effect of the individual researcher's stance, other researchers

should examine and comment on transcriptions, suggesting codes and themes. Concordant codes should arguably have greater reliability as accurate descriptions of the data. Armstrong, Gosling, Weinman, and Marteau (1997) illustrate how many qualitative researchers argue such inter-rater reliability is an important method for ensuring rigor.

7. It should not be assumed that there is a direct link between what participants disclose, and their lasting positions, rather the interview data are regarded as a snap-shot of their views on a particular situation at that time (Rogers, 2007), with the data also reflecting relations between the interviewer and participants under the circumstances of the interview (Silverman, 2006).

8. Data analysis should occur in stages, with further data collection informed by emerging themes in the first stage of data analysis. Rather than forcing the data into predetermined categories arising from the interview guide, thematic categories should be allowed to emerge from the data, using thematic analysis techniques as laid out by Braun and Clarke (2006). This approach to analysis is also informed by the grounded theory approach originally described by Glaser and Strauss (1967). 'In Vivo' terms from the interviews should be used wherever possible in the coding and written interpretation, in order to stick as closely as possible to parental accounts ('In Vivo' terms being the descriptive terms that used by participants themselves). Again, as far as possible, a bottom up data driven approach, using iterative analysis and theory building should be adopted.

9. Qualitative research is not a way of obtaining a true reading, rather a strategy to add breadth, depth and richness to the line of enquiry as outlined by Denzin and Lincoln (2000). The study adopts a critical realist epistemology. This means an understanding that there is a level of mental interpretation when reporting events, which seeks to understand reality which exists independently of the mind.

10. Interviews with parents of autistic children deal with many personal issues. Interviews should be conducted sensitively. All parents participating should be fully informed beforehand about what the study entails, and how their contribution might be used. After this, all participants should provide written informed consent.

11. All participants will be provided with any subsequent articles reporting this study, and invited to a presentation to discuss the issues with the researcher, after the submission of this thesis.

Section 5: Research methods used in the thesis

Research methods: study 1.

The research hypotheses were tested by secondary analysis of a large ongoing birth cohort study, the Avon Longitudinal Study of Parents and Children (ALSPAC). ALSPAC follows the health and development of over 14,000 children that had an expected date of delivery between April 1991 and December 1992, and were resident in the Avon area of South West England at the time of their birth. This study population had social and demographic characteristics in common with the 1991 UK national census (Golding, Pembrey, & Jones, 2001).

This particular cohort study was ideally suited to the requirements of the main study because it had taken detailed measures of autistic type behaviours throughout the lifetime of the children in the cohort, and it had access to independently validated ASD diagnoses for all children in the cohort by ICD-10 sub-category, up to age 12. In addition, very detailed data on socio-demographic factors in children's backgrounds were available, as were educational data on special educational provision. ALSPAC had also recorded multiple measures of outcome during early adolescence in behavioural, social, psychological and academic domains. The analysis was restricted to children in the ALSPAC cohort that survived to one year (*n*=13988).

Sample: children diagnosed with ASD

Within the ALSPAC cohort, 71 cases diagnosed with ASD (following a multidisciplinary assessment) were identified from their medical records, giving a prevalence of approximately 5 per 1,000 of clinically diagnosed cases. Diagnoses based on ICD-10 criteria were employed because this was the system most commonly used in the ALSPAC data. The children were all diagnosed when they were between 1.5 and 12.5 years old, and clinical records were reviewed by a consultant paediatrician to confirm diagnosis according to ICD-10 criteria. A detailed account of the methods used to identify the ALSPAC children diagnosed with ASD is given by Williams, Thomas, Sidebotham and Emond (2008). The diagnoses were made in the children's communities and assigned by clinicians who did not have access to any ALSPAC data.

Hypothesis, Article 1

The subsidiary hypothesis tested in article 1 was as follows:

> There are children with autistic type behaviours at the same severity as those with a diagnosis who remain undiagnosed.

Research Methods, Article 1

The children with a clinical ASD diagnosis were split into two sub-groups according to age and sub-category, as laid out in the methodological principles above: it was considered important to reduce post-diagnosis bias, as well as the possibility of wider variation in older children leading to a sub-clinical group. Thus for the purposes of this study, the traits considered were measured in the same age range as the diagnosis. This also minimised the problems of clinical presentation changing with age. It was therefore decided to split the diagnosed group into two.

The first group, termed the autism group (n=27), comprised those diagnosed with childhood autism or atypical autism between the ages of 2-4 years. Those with a diagnosis of AS were excluded. The second group, termed the AS group (n=30), were diagnosed with AS or atypical autism between the ages of 5-11 years; children with a diagnosis of childhood autism in this age-range were excluded. Cases of atypical autism or PDD-NOS, where diagnosis falls within the spectrum, but is uncertain in that it defies categorisation into any sub-condition comprised a small minority in each group. Children with ASD diagnoses assigned in other age-ranges were excluded from the analysis.

All autistic type behavioural measures taken by ALSPAC throughout the age-ranges of diagnosis of the children in the two groups were identified. These measures were all made by parental report, using questionnaires which were sent to each enrolled family by post at various time points. These questionnaires, from which the ALSPAC data on autistic type behaviours derived, were given to an experienced child psychiatrist, familiar with making diagnoses of ASD, and an educational psychologist. These two practitioners scored the ALSPAC traits in terms of how useful they thought individual variables would be in diagnosing autism or AS in the age ranges specified. Only traits that these practitioners considered useful in diagnosis of autism or AS were selected for subsequent analysis.

A child can only be diagnosed with autism if he or she presents a combination of behaviours from each area of the triad: impairments in social interaction, impairments in communication and those measuring restricted, repetitive or stereotyped patterns of behaviour. In order to establish a set of measures which in combination would fulfill the requirements for a diagnosis, it was necessary to consider behaviours from each behavioural domain of the triad separately.

Logistic regression was used to investigate the how strongly these various ALSPAC measures of social interaction, communication and repetitive or restricted behaviours were associated with a clinical diagnosis of autism or AS in the specified age ranges. Logistic regression was used since the outcome was a dichotomous variable (either having an ASD diagnosis or not). The 'predictor' or independent variables were the various measures of autistic type behaviour recorded by ALSPAC. To allow a comparison of the effect sizes for variables with different scales of measurement, the variables were standardised. Consequently the odds ratios from the analysis reflected the effect for a 1 standard deviation increase.

The predictor variables were assigned to 'blocks' according to which of the subcategories within the triad of impairments they represented. Thus, for example, the block for social impairment in autism was composed of measures of general sociability, a measure of avoiding eye contact or affection, and measurements of time spent alone and solitary behaviours. Details of all measures used throughout the entire set of analyses are given in the relevant articles in the main body of this thesis.

A series of backwards logistic regressions were performed on each block, or domain of impairment, for both the autism and AS groups. The analyses involved a three stage procedure where variables were assessed univariably, then multivariably, within each block, adjusting for interdependencies between behaviours within each domain. Only variables significant at 1% level at each stage were carried forward through to the next stage. This procedure allowed variables to be assessed with the maximum amount of data at each stage compared to traditional stepwise regression which limits cases to complete data across all variables. The direction of approximately half the variables was reversed so that a low score always predicted getting ASD diagnosis.

Peduzzi, Concato, Kemper, Holford & Feinstein (1996) showed that in logistic regression, the number in the sample with less common outcomes divided by the number of predictive variables should be at least ten to ensure reliability in the estimates of the regression coefficient. Nevertheless, in a review of logistic regression in medical literature Bagley, White, & Golomb (2001) note that in the 21 published logistic regression analyses they reviewed, the number of events per variable ranged between 2.4 and 25.7. In order to limit the number of predictive variables entering the regression, only a few measures rated most useful by the two clinicians were used as predictor variables. In practice, the final models (called composite traits) derived from such analyses involved only predictors thus maintaining the desired ratio.

A further complication in logistic regression is that sometimes estimates of effect sizes and significance may become imprecise with highly correlated variables. Consequently, the list of traits was screened to avoid these problems, for example, where the same measurement was taken twice within the same age range only one variable was used, for example.

Making the model

Once the measures of autistic type behaviour that were most strongly associated with a clinical diagnosis in each domain were established, a model, known as the 'composite trait' was created. This was composed of the measures most strongly associated with autism or AS diagnosis, from each area of the triad. The composite trait for autism was weighted according to ratios specified in the ICD-10 diagnostic criteria. According to ICD-10, there is no delay in language in AS. Therefore, only measures of social impairments (including conversational skill), repetitive behaviours and restricted interests were included in the composite trait constructed for AS. The composite scores were thus a measure of the extent to which the whole population of children displayed impaired functioning in autistic type behaviours.

The composite scores for both autism and AS were then broken into percentiles across the whole population to ascertain which range would include all the clinically diagnosed children. 100% of the children diagnosed with autism were captured in the top 2% of the composite autism trait, and 100% of children diagnosed with AS were captured in the top

4% of the composite AS trait. There were a number of undiagnosed children whose scores on the composite traits were also in this range. To check the model, its specificity and sensitivity in classifying diagnosed cases was examined, together with the statistical significance of the relationship between the model and the outcome, as well as goodness of fit.

Educational provision accessed by the clinically diagnosed group was examined and compared to that accessed by the undiagnosed children with comparable autistic behaviours; those identified by the model. This revealed whether diagnosed children and undiagnosed children who displayed autistic type behaviours were accessing educational services at age 11 or 12.

Subsidiary hypothesis and central hypothesis (Articles 2 and 3)

- There are social and demographic factors in the children's backgrounds that may influence which of these children are diagnosed and which of them are not.

- Diagnosis of ASD and/or subsequent service contact has an effect on behavioural and social outcomes, compared with an undiagnosed group who have comparable autistic type behaviours.

Before addressing hypothesis 2 and the central hypothesis, outcome measures of interest were selected according to reports of parents participating in study 2. Unfortunately, attrition in the dataset meant that only about half the families enrolled in the ALSPAC cohort were responding by the time the study children were 13. Therefore, it was apparent that the outcomes study would be underpowered if the sample was split by ASD sub-category (i.e. autism/AS) and age range as in the previous stage. It was necessary, therefore, to use the entire ASD sample, and repeat the method described above to create a composite 'ASD' trait. This consisted of autistic type behaviours measured when the children were preschoolers (aged 3-4). Again, this composite ASD trait was used to define the comparison group.

In practice, all the autistic type behaviours that were most strongly associated with childhood autism in the first study were also most strongly associated with ASD diagnosis, regardless of sub-category. A composite ASD trait that was composed of five measures of autistic type behaviour was created. It contained measures from each domain most strongly associated with clinical diagnosis of ASD. The number of traits included from each behavioural domain was again specified by thresholds required for diagnosis in the ICD-10 diagnostic criteria for autism. The only exception was that just one trait for communication difficulties was included to reflect differences in criteria between AS and childhood autism: the requirements for diagnosis of AS are the same as for autism, bar impairments in communication which may or may not occur. For a diagnosis of atypical autism, diagnostic criteria differ from autism in either of age at onset or of failure to fulfill all three domains of diagnostic criteria. Each of the behaviours that made up the composite trait were also given a weighting that reflected their relative power to predict ASD diagnosis. Article 2 describes the individual measures of autistic type behaviour in each domain of impairment that went into making up the composite ASD trait, together with the weightings that were assigned in the composite ASD trait.

The undiagnosed comparison group was defined as those children who had not been diagnosed with ASD, but who were reported as displaying ASD traits at age 3-4, those within this age-range with scores in the top 2 percentiles of the composite ASD score (*n*=142). The sensitivity of the trait in this percentile range in predicting ASD diagnosis was 68% and the specificity (the percentage of those correctly predicted without diagnosis) was 98.4 %. The sensitivity was substantially reduced by developing a generic ASD composite trait, as opposed to an AS or autism trait in limited age-ranges described above in the research methods for Article 1. However, this cut-off was chosen because it did capture the majority of the clinically diagnosed children.

To check the match of this comparison group with the diagnosed ASD children, mean scores of the composite trait were compared, for all of the measures of autistic type behaviours within the composite trait. A Levene's test was used to compare variance between samples, then where appropriate, a t-test of unequal variance was carried out to check the match between the undiagnosed comparison group and the group with ASD diagnosis.

Measures of background socio-demographic factors (Article 2)

Basic demographics such as gender, ethnic origin, age of mother at delivery and birth order were recorded by ALSPAC. A first born child was defined when there were no previous pregnancies that resulted in birth, as opposed to all children who were second or subsequent births. Several socioeconomic measures were also available. These were social class, measured by the occupational status of parents, maternal educational attainment, home ownership status, financial hardship and marital status. With family risk factors in mind, researchers at ALSPAC compiled a Family Adversity Index of stressors. These included measures of financial hardship, measures of maternal mental health, criminal activity, substance abuse and scales relating to partner cruelty and affection. A measure of depressive symptoms of mothers was also included. All these measures were derived from maternal report.

A chi squared test of association, and where appropriate, a Fishers exact test were applied in order to determine factors in children's social or economic backgrounds that were associated with diagnosis. First, demographic factors which predicted ASD diagnosis when compared with the undiagnosed (non-autistic) general population were considered. Second, using the undiagnosed comparison group, it was established if such social and demographic factors were associated with ASD diagnosis with the severity of autistic symptoms held constant at preschool age. Associations between socio-demographic factors and children displaying undiagnosed autistic type behaviours were also analysed. Analyses of variance were carried out where the social and demographic factors were recorded as normally distributed scores.

Outcomes study (Article 3)

Outcomes in adolescent behaviour, the development of prosocial behaviours and academic performance at age 12-16 were compared between the groups. One outcome measure of social skills, the Strengths and Difficulties Questionnaires (SDQ) prosocial score, had been measured repeatedly throughout the children's lives. The developmental trajectory of this measure of prosocial behaviour was therefore assessed in detail for each sample.

ALSPAC also holds educational data on all children attending state schools, (both mainstream and special schools), covering approximately 85% of children in the cohort. These data record whether children are identified as having some form of special educational needs within the academic year. Children listed at 'school action' level have an individual education plan which details tailored interventions provided by teachers and addressing their identified needs. Children at 'school action plus' have input from agencies outside the school, such as educational psychologists and health services. Schools whose children have a 'statement of special educational needs' are provided with extra funding for additional resources, usually a teaching assistant, to help these children function in the school environment.

Children who did receive intervention from health or educational services (according to educational data at age 11-12) but did not have an ASD diagnosis were initially discounted from the outcomes analysis. Thus, from the children who displayed autistic type symptoms as preschoolers, two dichotomous groups were established - one group comprising children who had received a clinical ASD diagnosis, the other group comprising children with autistic symptoms when young, but no support from additional health services at age 11-12. However the final analysis reported in this thesis in article 3 reverted to comparing the diagnosed children with the wider undiagnosed comparison group previously defined. This was in the light of comments from reviewers (see concluding chapter).

Social and behavioural outcome measures

A full list of outcome measures together with who reported them (either the child's parent, educational authority or the teenage children themselves), the items that made up these scales and at what age in the child's life they were reported is given in Article 3. These were selected either because they were measures of autistic type behaviours or they related to parental concerns reported in study 2.

Analysis of outcomes

To determine whether the children with ASD diagnoses differed significantly from the undiagnosed comparison group in terms of their other social and clinical behaviours as teenagers, t-test comparisons were made. A Levene's test was used to establish whether the groups had equal variance on the outcome measures. Where this was not the case, a t-test of

unequal variance was applied. Analysis of variance (ANOVA) was also performed in order to determine whether outcomes for the children with ASD diagnoses and those in the undiagnosed comparison group differed from those in the general non-autistic population. Where outcome measures were recorded as categorical variables, a chi squared test of association and Fischer's exact test was carried out as appropriate.

Given that IQ has been shown to be a major indicator of outcome for autistic adults (Howlin et al., 2004), further analyses co-varying for initial academic ability (derived from SATS results aged 4-5) as a proxy for IQ were conducted. Separate adjustment was also made for the total behavioural difficulties score taken from the SDQ at 47 months, which includes measures of hyperactivity, conduct problems, emotional difficulties and peer problems.

In part because of numerous potential confounding factors, and in part because of effects of attrition, the methods adopted failed to test the central hypothesis as initially planned. This failure is discussed in the concluding chapter.

With so many limitations to the study design described above, further analysis was conducted in an attempt to examine the impact of ASD diagnosis (and subsequent service contact) on prosocial behaviour. No comparison to a control group was made.

Hypothesis, Article 4
That an ASD diagnosis and subsequent service contact would lead to a change in the development of prosocial behaviour.

This was again a retrospective analysis using ALSPAC data on the group of children with clinical ASD diagnosis. Parents of children enrolled in the ALSPAC cohort had repeatedly reported on the prosocial behavior of their children at six time-points throughout the children's lives, (at 47, 81, 97, 115, 140 and 157 months respectively) using the SDQ (Goodman, 1997; 1999). This study examined the developmental trajectory of this measure of prosocial behaviour both before and after diagnosis of ASD.

A multi-factorial model was used to determine whether there was a significant change to the developmental trajectory of prosocial behaviour before or after diagnosis. Due to the

varying participation of children at each age of SDQ assessment, all analyses were adjusted for individual variation using fixed effect estimation. Thus variation in prosocial behaviors between individual children was taken into account in the model. Since prosocial scores tend to improve with age, analyses were also adjusted for age of assessment. SDQ scores were considered continuous and analysed by ANOVA.

Thus the development of social skills as a function of time before or after diagnosis was plotted and differences between the trajectory of the linear trend both before and after diagnosis were calculated.

A quarter of the children had been diagnosed, mostly with classical autism, before the first SDQ measures were taken. In these cases the data were only recorded after ASD diagnosis. To investigate the robustness of the adjustment for individual variation, the analysis was repeated, omitting children who did not have both 'before' *and* 'after' diagnosis scores.

In addition, the impact of diagnosis on the prosocial scores was investigated by considering the different trajectories in scores between groups of children diagnosed early in life and those diagnosed later: (age of diagnosis <80m, or 80+ m).

All research in study 1 was based on secondary analysis of the ALSPAC data. The original ASLPAC study was monitored by the ALSPAC Law and Ethics Committee. The ALSPAC ethical framework does not permit researchers to use information from ALSPAC to make contact with individual study members or their families. Parents who enrolled in the study were informed that their answers to ALSPAC questionnaires could be linked to their medical records for research purposes and were given the option to opt out of this process at any time. The material used is anonymous, confidential and is held in accordance with the Data Protection Act. In addition, ethical approval to perform the analyses was cleared by the University of Exeter Humanities and Social Science Ethics Committee.

Research Methods: Study 2.
Qualitative methods were employed, analysing semi-structured in-depth interviews with participants sampled from two groups. The first of these comprised parents whose children had already received an ASD diagnosis (*n*=9) and the second was made up of parents who

were not pursuing such an outcome, despite professional indication that their child might be on the spectrum (n=8).

It was necessary to recruit parents to the study who had undiagnosed 'autistic' children. Autism is defined in practise by its diagnosis. Children who had been judged to be on the autism spectrum by special educational needs coordinators at school, or educational psychologists, and whose parents had been informed of this, were identified. This was not to suggest that these professional opinions were 'correct', or that they were more valuable than opinions of parents themselves, rather it was a pragmatic move which was necessary to design a parameter for clear definition of the group. The strategy was to recruit parents of children whose autistic type behaviours had been recognized by either psychologists or educational professionals, but who decided not to follow the diagnostic route.

The first group, comprising of parents of undiagnosed children (n=8) were identified via educational psychologists, special needs co-ordinator teachers, or personal contacts. In all cases, either an educational psychologist, or special needs co-ordinator teacher at school had suggested that the child in question might have AS or autism.

The sample in the second group consisted of parents whose children had received an ASD diagnosis. These participants were recruited with the help of a local support group for parents whose children had been diagnosed with AS or with high functioning autism. Two participants who were not in the support group also volunteered to participate in the study.

Participants were interviewed using a semi-structured interview schedule (see Appendix, Article 5). Interviews ranged from one hour to two and a half hours in duration. Ethical approval for the study was given by the University of Exeter Humanities and Social Science Ethics Committee Ethics Committee. All interviews were tape recorded and transcribed. Details of the attributes of participants in this study that were recorded are given in Table 2.

Table 2: Details of attributes of participants in study 2

	Mother or father	Age of affected child	Sex of child	ICD-10 diagnosis of child	Type of School	Marital status [a]	Number of other children	Employment of participant
Participants whose child has ASD diagnosis								
1	mother	16	male	AS	special	partner	1	Part-time
2	mother	14	male	AS	mainstream	single	2	Part-time
3	mother	10	male	ASD	mainstream	partner	2	Ft mother [b]
4	mother	9	male	AS	mainstream	single	0	Part-time
5	mother	12	male	ASD/ADHD	special	single	2	Ft mother
6	mother	11	male	Autism	special	single	1	Part-time
7	mother	7	male	AS	mainstream	partner	1	Ft mother
8	mother	14	male	AS	special	single	1	Part-time
9	mother	11	male	AS	mainstream (private)	partner	0	Ft mother
Participants whose child is undiagnosed								
10	father	5	male	-	mainstream	partner	1	Full-time
11	mother	15	female	-	mainstream (private)	partner	2	Ft mother
12	mother	14	male	-	mainstream	partner	2	Part-time
13	mother	4	female	-	mainstream	partner	0	Part-time
14	father	5	female	-	mainstream	partner	1	Full-time
15	mother	5	male	-	mainstream	partner	1	Part-time
16	mother	5	female	-	mainstream	partner	1	Ft mother
17	mother	16	male		mainstream	partner	0	Ft mother

[a] Co-habiting with partner or co-habiting and married both given as 'partner'

[b] Full-time mother

Interviews took the form of a series of open questions allowing the narratives of parental experience of their children to unfold. The semi-structured interview schedule was developed as the study progressed. Semi-structured interviews allowed evaluation of specific topics whilst encouraging free responses to questions around these topics. The semi-structured format also aided the comparison between the two groups where similar questions were posed. Field notes were written up at the beginning and end of each interview and referred to throughout analysis, although the coding categories used 'In Vivo' terms for as long as possible to avoid impressionistic interpretation.

Although the sample size was small, the data were collected in two stages. Interviews were recorded over a period of one year and all took place in the participants' homes, some were recorded during the day and others in the evenings. Participants were able to devote themselves entirely to the interviews as prior arrangements were made regarding childcare.

Self-reflexivity

The researcher chose to disclose information about her own son's assessment and difficulties to participants. The researcher became aware of the emotional reaction to having a professional highlight her child's difference through her own lived experience. The researcher believed that her experience enhanced the research process as it led to a perceived level of shared understanding with parents. The choice of disclosure was influenced by the humanistic methodologies of Reason and Rowen (1981) where in-depth interviews allow interviewer and interviewee to become peers or even companions.

The transcripts from the initial stage of interviews were coded and analysed. The second stage of interviews allowed interesting perspectives that had developed into theory to be followed up and examined in more detail.

A modified form of the thematic analysis as set out by Braun and Clarke (2006) was adopted in analysis, incorporating elements of constant comparative method of analysis, as described by Strauss and Corbin (1998). These methods were deemed to be most appropriate as previous studies using grounded methods have yielded rich insights into the experiences of families. This approach was chosen as the specific aim of the second study was to systematically explore parents' accounts of their experiences.

Interviews with the two groups were analysed separately. The transcribed interviews were read through once, then significant statements and paragraphs were selected and categorised according to emerging themes in open coding. New meanings were compared with incidents in existing categories which were refined and adapted accordingly. Coding and development of themes continued in the cross comparison of interviews. In vivo terms from the interviews were used wherever possible in open coding and written interpretation, with higher level concepts abstracted from these in comparative analysis.

In generating the open codes, and constantly comparing them, the categories that were already determined were scrutinised and the conditions under which they were pronounced were considered. Secondly, the relationship of the category to other categories was examined. The diverse properties of categories were integrated by writing clear definitions for each category, encompassing all the properties of each category.

Finally, the categories emerging from the two groups (either parents of undiagnosed or diagnosed children) were cross-compared to explore underlying commonalities or differences. Differences were accounted for by single higher level concepts. Major modifications became fewer and fewer as new instances of a category were compared with the developing properties. The themes and concepts that appeared to be most salient according to the research questions were narrowed down. As a part of this process, the research interview schedule was adapted to a more focused set of questions for the second set of interviewees. Emerging themes were provisionally verified through this further data collection and analysis. Identification of negative cases also occurred at this stage, instances where codes did not fit into developing theory, and concepts were adapted accordingly. Thus a bottom-up data-driven approach, using iterative analysis and theory-building was adopted. This sat within the wider framework of the initial research questions that were informed by findings from existing literature.

Participants were not invited to comment on the analysis as part of the analytic process. This was because as well as analysing the content of what participants said, an attempt was made to analyse the function of the talk of the participants, i.e. commonalities in the ways they presented themselves and their children in order to draw attention to particular ways of

acting, according to particular agendas. Therefore the analysis of their talk was not necessarily always consonant with the participants' views of themselves, and as such their comments could have been counter-productive to the aims of analysis. The interpretation sought to understand the reality described by participants in a way that was meaningful to the participants, as well as providing an overview of the function of their talk situated within its social context. In this sense, the study adopted a critical realist perspective, assuming that the participants described and interpreted real events through their own interpretations.

Although the views of the two groups appeared fairly distinct, parents of undiagnosed children may potentially become the parents of diagnosed children in the future (or they may choose not to). Similarly the parents of diagnosed children were once the parents of undiagnosed children, and during interviews were able to recall whether they experienced the same feelings and conflicts at that time. This group of parents were also able to describe their post-hoc experience after the autism diagnosis. Thus analysis involved coding for process: described by Strauss and Corbin (1998) as looking at interaction and tracing it over time to see how it changes with context. In this study, the change in context was bought about by the ASD diagnosis and the new conditions the diagnosis exposed.

Interviews with parents whose children have ASD are potentially sensitive. This was particularly true for parents of children who had decided against diagnosis. All participants in the study were therefore fully informed about what it entailed and what their contribution would be, and interviews were conducted with caution, care and diplomacy. Participants were recruited in a non-pressurised fashion, giving them an opportunity to discuss their own feelings and beliefs about their views which was often experienced as a positive. Written consent was given prior to interview and all transcripts were anonymised in the analysis of the study, and pseudonyms were used in publications and this thesis. Relevant information such as gender was used to contextualise quotes, but names and other identifiers such as location were changed. The transcripts of interviews in their entirety remained confidential, only shared with the co-author of Article 5. At the start of each interview, the researcher began by briefly explaining details of the research project, and that any reference to the participant would be made anonymous. Participants were informed that they were able to stop the interview at any point, and were provided with a signed copy

of the consent form which gave contact details of the research supervisor and an autism advice organisation. Copies of the finished report will be sent to the parent participants and they will be invited to attend a dissemination session about the results of the project.

Section 6: The organisation of the thesis

In this submission, five articles are presented examining one theme: diagnosis of ASD in children. These were initially derived from the written reports of the various stages of the two studies described above, as they progressed. Articles 1-4 report on findings from the main study, and Article 5 reports the findings of the second qualitative study.

The first article deals with the identification of undiagnosed children with autistic type behaviours using the ALSPAC data. This was the initial attempt to define the control group for the subsequent outcomes study, and was published on initial presentation by *The Journal of Child Psychology and Psychiatry*. The second article involves the definition of the actual control group used in the outcomes study, and examines social and demographic factors that might influence whether a diagnosis is given in more detail. This has been published in electronic format ahead of the print version by *Social Psychiatry and Psychiatric Epidemiology*.

In addition to this, two more articles reporting on the outcomes study, which at the time of submission are under review, are presented. The first of these two articles, Article 3, represented the culmination of the work and examined behavioural and clinical outcomes for children diagnosed with ASD versus the outcomes in the control group. However, it failed to test the central hypothesis as it was conceived, due to limitations of the data which will be discussed in the concluding chapter. The hypothesis that Article 3 actually tested was that:

> Social and behavioural outcomes differ in the sample of children diagnosed with ASD to those in the undiagnosed sample with autistic type behaviours as preschoolers.

This was quite different from testing the effects of the diagnosis. This article was initially submitted to the *British Medical Journal* where it was reviewed and rejected. It was then revised and submitted to the *Journal of Child Psychology and Psychiatry* where it was also reviewed and major revisions were required. Revisions have been made in the light of reviewers' comments, and the article has been submitted for re-review. This is the current

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version submitted in this thesis. All the reviewers' comments, and the revisions that were made to articles as a result of these comments, are given in thesis Appendix 1. The process of revising the articles, and the implications to the aims of the thesis as it was initially conceived, are also discussed in the concluding chapter.

Article 4 aimed to give a more in-depth developmental picture, using multifactorial analysis to examine the development of prosocial behaviour in ALSPAC children diagnosed with ASD. Therefore Article 4 presented a truly longitudinal analysis of the data at hand. This article was initially submitted as a short report to the *British Journal of Psychiatry* where it was modified, resubmitted, and rejected. It was then revised, lengthened and submitted to the *Journal of Autism and Developmental Disorders* where it was reviewed and rejected, although the editors invited resubmission after revision. Major revisions were made in the light of reviewer's comments (Appendix 1) and the article has again been resubmitted to the *Journal of Autism and Developmental Disorders* for re-review. The resubmitted version is presented in this thesis.

Finally, Article 5 presents a report of the findings of the qualitative study. This article was submitted to *Clinical Child Psychology and Psychiatry*, where it was reviewed and minor revisions were requested. Revisions were made to this article, (the reviewers' comments, and the first author's responses are again given in thesis Appendix 1) and it was returned to *Clinical Child Psychology and Psychiatry*, where it has been accepted. The article can therefore be considered 'in press'.

Each article provides a brief literature review in a background or introductory section, and each gives the references used within. The bibliography of this introductory chapter and the concluding chapter is given at the end of the thesis. The first thesis Appendix gives the reviewers comments in full to the final three articles and the responses that were made by the candidate, exactly as submitted to the various journals. The second Appendix contains two additional published articles that were conceived and written during the period of registration, both of which are referred to in the text.

Details of the individual contributions of the co-authors to each of the articles are given after the contents and abbreviations pages, as specified by the requirements for thesis

submissions laid out by the University of Exeter. Analyses in study 1 were conducted with STATA and the initial syntax was written by Colin Steer. This was then modified by the first author (the candidate) as various analyses proceeded, with further help from Mr. Steer on several occasions. The exception to this was the analyses in Article 4. The first author (the candidate) wrote an initial version of the syntax of the first analysis in Article 4, but this was substantially modified and improved by Mr Steer. The STATA code used in Articles 1-4 in analysis is available from the candidate on request. The final section of the analysis reported in Article 4, which considered the different trajectories in scores between groups of children diagnosed early in life and those diagnosed later, was devised and developed in its entirety by Mr. Steer. The considerable contribution of Mr. Steer, who did not have any formal role in supervision of this work, together with that of the other co-authors, is highlighted in the Acknowledgements section.

Overall, although it was true that the central hypothesis as it was initially conceived was never tested satisfactorily, as the research progressed, the individual studies contributed a number of interesting findings. These contributions are summarized, together with an appraisal of the overall impact of the work, a detailed discussion of the limitations of the data, and an assessment of the broader theoretical and practical implications of the submitted work, in the final section.

Original research articles

'Article 1'

Russell, G., Ford, T., Steer, C., Golding, J. Identification of children with the same level of impairment as children on the autistic spectrum, and analysis of their service use. *Journal of Child Psychology and Psychiatry, 51*(3), 2010: 643–651

'Article 2'

Russell, G., Steer, C., Golding, J. Social and demographic factors that influence the diagnosis of autism spectrum disorders. *Social Psychology and Psychiatric Epidemiology* (On line early version :DOI 10.1007/s00127–010–0294-z)

Identification of children with the same level of impairment as children on the autistic spectrum, and analysis of their service use

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Background: Data from epidemiology have consistently highlighted a disparity between the true prevalence of childhood psychiatric disorders and their recognition as defined by receiving a clinical diagnosis. Few studies have looked specifically at the level of unidentified autistic spectrum disorder (ASD) in the population. **Method:** Logistic regression was used to determine the behavioural traits associated with receiving a diagnosis of ASD using data from the Avon Longitudinal Study of Parents and Children (ALSPAC). A composite score was derived to measure levels of autistic traits; undiagnosed children with scores matching those diagnosed with ASD were identified. Levels of educational provision beyond that provided by standard schooling were examined. **Results:** Fifty-five percent of children with autistic traits at the same levels as those who had an autism diagnosis had not been identified as needing extra support from education or specialised health services. Of those who were identified as having special needs, 37.5% had been formally diagnosed with an ASD. For children with impairment at the same level as that associated with Asperger's syndrome, 57% had no special provision at school, and were not accessing specialised health services. Twenty-six percent of those who did have special provision at school had an ASD diagnosis. **Conclusions:** The results suggest that there may be a substantial proportion of children on the autistic spectrum who are never identified by services. **Keywords:** Autism, pervasive developmental disorders, Asperger's syndrome, epidemiology, prevalence, child mental health.

Autistic spectrum disorders are conceptualised as consisting of a triad of impairments in social interactions and communication, as well as restricted interests and repetitive behaviour (Bailey, Phillips, & Rutter, 1996). According to the *International Classification of Diseases* (ICD-10), for a diagnosis to be made, children must display impairments in each area (WHO, 1992).

Costello's review of epidemiological studies (Costello, Egger, & Angold, 2005) suggests that impairing psychiatric disorder is present in roughly 3–22% of children. Such studies imply that only a minority of children with impairing psychopathology are in contact with services and thus have their difficulties recognised and their needs met (Ford, Hamilton, Meltzer, & Goodman, 2007).

A few studies have specifically examined the true prevalence of ASD in this manner. A study by Baron-Cohen et al. (2009) found that the ratio of known to unknown cases is about 3:2 in mainstream primary school children aged 5–9 years old. The South East of England Study screened a sample of children who were at risk and the minimum ASD prevalence was found to be over 1% (Baird et al., 2006). These studies suggest that few of the large number of children displaying functional and cognitive impair-

ments receive a clinical diagnosis. Howlin and Moorf (1997) reported that less than 10% of children diagnosed with ASD were given a diagnosis at initial presentation; another 10% were told either to return if their worries persisted, or that their child 'would grow out of it'. Almost 20% reported that they had to apply substantial pressure to get referrals or seek private assessments. So it appears that some parents who seek diagnosis cannot easily obtain one.

In a population-based study, Flisher et al. (1997) reported that approximately 17% had 'unmet need', defined as having impairing psychopathology and no reported service contact. Here, unmet need was associated with 'barriers' to service access such as parental concern that children would be taken away, parents wanting to solve the problem themselves or simply failing to cooperate. Barriers mediated by both parents and by service providers appear to be an obstacle to diagnosis. We frame our own study in terms of 'unmet need' – a lack of access to interventions provided by health services or by schools.

This secondary analysis of data from a longitudinal cohort uses autistic symptoms and individual behavioural traits to identify undiagnosed children who appear to match the diagnostic thresholds for ASD. Autistic behaviours in each behavioural domain of the triad are examined, thus breaking down the ASD phenotype into its individual parts. The hypothesis is that there are undiagnosed

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Conflict of interest statement: No conflicts declared.

children with unmet need, whose behavioural profiles match those who are clinically diagnosed on the autistic spectrum. In several studies children have been shown to be in contact with school-based services before any other service (Burns et al., 1995; Leaf et al., 1996). With teachers and educators usually providing the first port of call, access to educational services was also examined.

Methods

Ethical approval for the study was obtained from the ALSPAC law and ethics committee and the local research ethics committee.

Sample

The Avon Longitudinal Study of Parents and Children (ALSPAC) is an ongoing longitudinal cohort study. ALSPAC follows the health and development of children that had an expected date of delivery between April 1991 and December 1992, and were resident in the Avon area of South West England at the time of their birth. The analysis was restricted to children in the ALSPAC cohort that survived to one year (n = 13988). A total of 548 extra children have joined ALSPAC since age 7, and these were also included in the analysis. The final sample size for the current study was therefore 14,536. This study population had social and demographic characteristics in common with the 1991 UK national census (Golding, Pembrey, & Jones, 2001).

Measures

Within the ALSPAC cohort, 71 cases diagnosed with ASD following a multidisciplinary assessment were identified from medical records, giving a prevalence of approximately 5 per 1,000 of clinically diagnosed cases (Williams, Thomas, Sidebotham, & Emond, 2008). Diagnoses based on ICD-10 criteria were employed because this was the system most commonly used in the ALSPAC data. The children were all diagnosed when they were between 1.5 and 12.5 years old, and clinical records were reviewed by a consultant paediatrician to confirm diagnosis according to ICD-10 criteria. A detailed account of the methods used to identify the ALSPAC children diagnosed with ASD is given by Williams et al. The distribution for ages of diagnosis is shown in Figure 1.

As ALSPAC is designed to assess the environmental and genetic aetiologies of a variety of health and developmental outcomes in children, an assortment of measures of behaviour from across the triad have been recorded. Measurements of autistic traits were taken across the whole population throughout the study, both before and after any ASD diagnosis was assigned. In total, 44 measures with possible utility were identified, 26 traits for autism and 18 for Asperger's Syndrome (AS). These are listed with the ages at which they were measured in Tables 1 and 2.

In order to break down these measures into those which most expressly predict ASD, single answers to questions that matched the diagnostic criteria were entered into the regression as well as validated scales where appropriate. Some of the scales and modules available were designed for more generic use and were
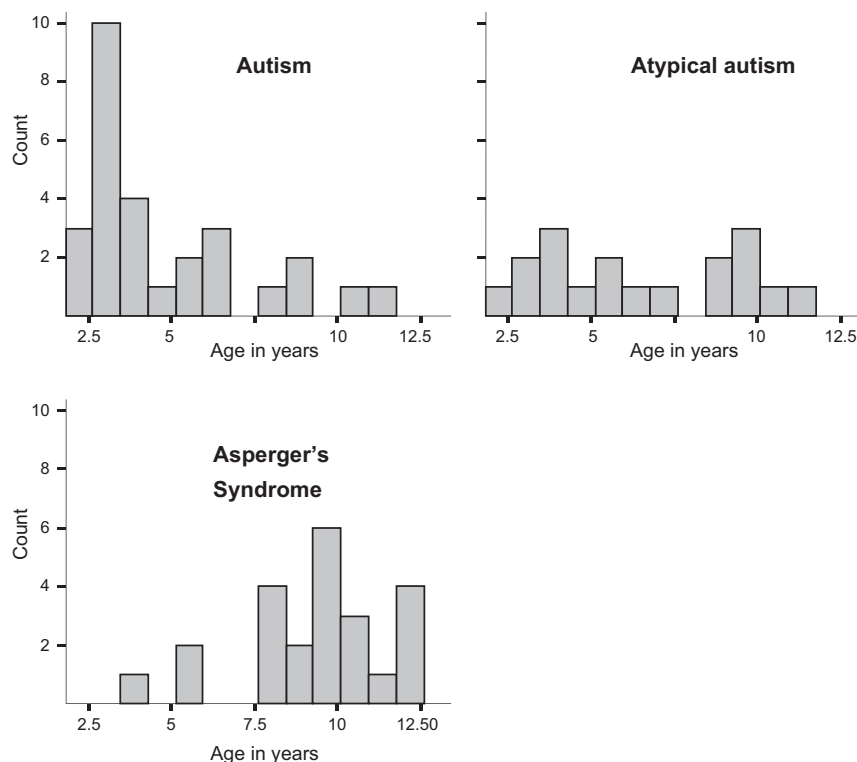

Figure 1 Frequency of each ASD condition by age of diagnosis

Table 1 Behavioural traits measured by ALSPAC between the ages of 2 and 4 showing which are most predictive of receiving a diagnosis of autism or atypical autism between the ages of 2 and 4. Blank fields denote no significant relationship with diagnosis at 1% level

Trait (variable name)	Age measured in months	Odds ratio [CI 95%] Unadjusted $p < .01$	Odds ratio [CI 95%] Adjusted for effect of interdependencies within domain $p < .01$
Impairments in social interaction		N = 8294–10181*	N = 8294
EAS sociability score	38	4.92 [3.34–7.26]	2.49 [1.53–4.02]
SDQ Prosocial score	47	13.68 [7.41–25.25]	11.45 [4.86–27.01]
Child gets on with other children	42	3.77 [2.71–5.23]	
Child avoids eye contact when talking	38	2.77 [2.07–3.70]	
Child is affectionate to family members	42	1.69 [1.25–2.28]	
Avoids eye contact when reunited with mum	42	1.34 [1.08–1.66]	
After separation child wants hug	30		
Child does things on his/her own	42	2.67 [2.09–3.41]	2.85 [1.67–4.84]
Child has at least one good friend	47	2.16 [1.71–2.72]	
Child prefers adults to children	47	1.93 [1.29–2.90]	
Impairments in communication		N = 8945–10207*	N = 8945
Child sings songs	38	2.16 [1.63–2.86]	
Child played looking after babies	30	4.16 [2.12–8.16]	
Enjoys pretend games	42	5.37 [3.43–8.41]	3.95 [2.43–6.40]
Child combines words	38	2.35[1.87–2.96]	
Number of words child can string together	38	1.78[1.60–1.98]	
Child stays mainly silent	38	2.62[2.12–3.24]	2.02 [1.62–2.53]
Child echoes what others say	38		
Child uses gestures to get what wants	38	5.35[2.97–9.62]	
Child talks with words in wrong order	38		
Intelligibility score	38	2.03[1.65–2.49]	
Restricted repetitive and stereotyped patterns of behaviour		N = 9881–10163*	N = 9881
Child is afraid of new things or new situations	42	2.20 [1.68–2.87]	2.04 [1.43–2.91]
Child is fussy or over particular	42		
Child often rocks head or body repeatedly	42	1.46 [1.30–1.64]	1.28 [1.10–1.47]
Child has other unusual repetitive behaviour	42	1.73 [1.51–1.98]	1.60 [1.37–1.87]
Frequency child rocks head or body	30	1.32 [1.09–1.59]	
Child has tic or twitch	42		

*Varies with number of complete scores per trait.

not autism specific; in this case, only those items measuring ASD criteria were included in the analysis. For example, the Development and Well-Being Assessment (DAWBA; Goodman, Ford, Richards, Gatward, & Meltzer, 2000) module on compulsions and obsessions included such ASD-specific items as '*child has arranged things in a particular way or so that they are symmetrical*' and '*child has repeated actions many times in a row*'. For other measures the relevant subscales were incorporated in the analysis in their full form. These were the Temperament subscale from the Emotionality, Activity and Sociability (EAS) questionnaire (Buss & Plomin, 1984), the Prosocial subscale of the Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997), the Social Cognition Disorders Checklist (SCDC; Skuse et al., 1997), and the Coherence, Conversational rapport, Conversational context, Stereotyped conversation, and Inappropriate initiation subscales of the Social Children's Communication Checklist (CCC; Bishop, 1998). Of the 44 predictor traits entered into the regression, 9 questions were specifically designed for ALSPAC questionnaires. All the data were derived from parental report.

ALSPAC also holds data from the Pupil Level Annual Schools Census (PLASC) for 2003/4 (when ALSPAC children were 11 or 12), which include all children attending state schools (both mainstream and special schools) and cover approximately 90% of children. PLASC data record whether children are identified as having some form of special educational needs (SEN) within this academic year. Children listed at '*school action*' level have an individual education plan that details tailored interventions addressing their identified needs. Children at '*school action plus*' have input from agencies outside the school, such as educational psychologists and health services. Children with a '*statement of special educational needs*' usually have extra funding for additional resources on a regular basis to help them function in the school environment. These data therefore provide a measure of whether children were accessing additional educational and other services.

Analysis

The children with a clinical ASD diagnosis were split into two sub-groups according to age and sub-category. The first group (n = 27) comprised those diagnosed with classical autism or atypical autism between the ages of 2 and 4 years, excluding those with a diagnosis of AS. The second group (n = 30) were diagnosed with AS or atypical autism between the ages of 5 and 11 years, excluding those with a diagnosis of

Table 2 Behavioural traits measured by ALSPAC between the ages of 2 and 4 showing which are most predictive of receiving a diagnosis of AS or atypical autism. Blank fields denote no significant relationship with diagnosis at 1% level

Trait (variable name)	Age measured (months)	Odds ratio [CI 95%] Unadjusted p < .01	Odds ratio [CI 95%] Adjusted for interdependencies of variables within domain p < .01
Impairments in social interaction		N = 8094–6802*	N = 6814
Child is rather solitary, tends to play alone.	115	3.45 [2.49–4.79]	
Child has at least one good friend	115	2.21 [1.79–2.73]	
SDQ prosocial score	115	2.70 [1.95–3.74]	
CCC stereotyped conversation score	115	3.69 [2.77–4.92]	
CCC conversational rapport score	115	2.78 [2.30–3.35]	
SCDC social cognition score	91	3.53 [2.71–4.60]	2.38 [1.71–3.31]
CCC inappropriate initiation score	115	2.93 [2.15–3.99]	
CCC coherence score	115	2.88 [2.39–3.46]	2.10 [1.64–2.69]
CCC use of conversational context	115	2.66 [2.88–4.60]	
SDQ prosocial	81	3.55 [2.54–4.94]	
Empathy subscale score	81	2.50 [2.06–3.04]	
DAWBA Social fears	91	1.81 [1.49–2.34]	
Restricted repetitive and stereotyped patterns of behaviour		N = 8073–6829*	N = 6428
Child has repeated actions many times in a row	128	1.31 [1.17–1.47]	
Child will arrange things in a certain way or so that they are symmetrical	128	1.58 [1.38–1.82]	
Child has repeatedly touch things or people in a particular way	128	1.49 [1.35–1.65]	1.47 [1.31–1.64]
Child has repeatedly counted to lucky numbers/ avoided unlucky numbers	91		
Child repeated arranges things in particular ways	91	1.74 [1.47–2.05]	1.55 [1.27–1.88]
Any other repetitive behaviours	128	1.65 [1.38–1.98]	

*varies with number of complete scores per trait

classical autism. Children with an ASD diagnosis assigned outside these two age ranges were excluded from the analysis.

Logistic regression was used to examine which measures of behaviour were associated with receiving a clinical diagnosis. The predictor variables were autistic behaviours or traits and the outcome variable was clinical diagnosis within each sub-category. To reduce the possibility of post-diagnostic bias (the diagnosis itself might lead to worsening or improvement of a behaviour), and to prevent the problem of entering behavioural traits measured in older childhood to predict early diagnosis, all the 'predictor' traits considered in the regressions were measured in the same age range as the diagnosis was made. In order to establish a set of traits that in combination would give the best correlation with diagnosis, we considered behaviours from each of the triad separately. To allow comparison of the effect sizes for traits with different scales of measurement, the traits were standardised. Consequently the odds ratios (ORs) from the analysis reflect the effect for a 1 standard deviation increase. The direction of measurement of approximately half of the traits was reversed so that a high score always reflected increasing impairment.

Backwards logistic regressions were performed on each domain of impairment in the triad separately. Only traits significant at the 1% level in univariate analysis were carried forward to the next multivariate analysis where traits were adjusted within each domain to account for interdependencies. Once the traits that were independently associated with a clinical diagnosis

in each domain were established, a composite trait was created composed of the most predictive traits ascertained across the triad. In order to produce a score that reflected the ICD-10 diagnostic criteria, the composite trait for autism was weighted with three impairments in social behaviour, two communication impairments and two types of repetitive behaviour. According to ICD-10, there is no delay in language in AS. Therefore, we included measures of social impairments (including conversational skill), repetitive behaviours and restricted interests only in the composite trait constructed for AS. The composite scores were thus a measure of the extent to which the whole population of children displayed impaired cognitive and social functioning. The composition of these scores is described fully in the Results section.

The composite scores for both autism and AS were then broken into percentiles across the whole population to ascertain which range would include all the clinically diagnosed children. There were a number of undiagnosed children whose scores on the composite traits were also in this range. Educational provision accessed by the clinically diagnosed group was examined and compared to that accessed by the undiagnosed children. This gave us an estimate of whether both children diagnosed with ASD and undiagnosed children who display equivalent levels of autistic traits were accessing additional educational or health services at age 11 or 12. These educational data acted therefore as a proxy for determining service use. A Fisher's exact test was performed to determine the relationship between certain demographic

factors and whether children received an ASD diagnosis or not.

Results

Autism sample

Table 1 shows the independent correlates of a clinical diagnosis for autism or atypical autism diagnosis between the ages of 2 and 4 years old for each behavioural domain of the triad.

The traits that were most strongly associated with diagnosis were fused in a composite autism trait to reflect the requirements of ICD-10 for autism diagnosis. The SDQ *Prosocial* scale accurately encompassed three traits of social impairment. In the other behavioural domains, *Stays mainly silent/Enjoys pretend games*: and *Unusual repetitive behaviour/ Afraid of new things* were most strongly associated with receiving a diagnosis. In the composite autism trait these measures were therefore fused with a weighting reflecting the emphasis of ICD-10 criteria (weighting = 3 to *Prosocial score*: 2 to *Stays mainly silent/Enjoys pretend games*: 2 to *Unusual repetitive behaviour/Afraid of new things*). As anticipated, the diagnosed sample have significantly higher scores in the composite autism trait than the general population (t = −17.661, df = 8123, p < .001). Figure 2 shows the distribution of the composite autism trait in the general population.

This model is a very good fit, with the composite trait explaining 65% of all the pseudo variance (the proportion of the log-likelihood) associated with the outcome of diagnosis, $\chi^2(1)$ = 134.36, N = 8125, ASD cases = 14, p < .001. All the scores for children in the diagnosed autistic sample fell within the top 2% of the autism composite score. The sensitivity of the composite autism score in predicting autism diagnosis within the top 2% is therefore 100%, and the specificity 98.3%. However, within this range, the positive predictive value of the composite autism
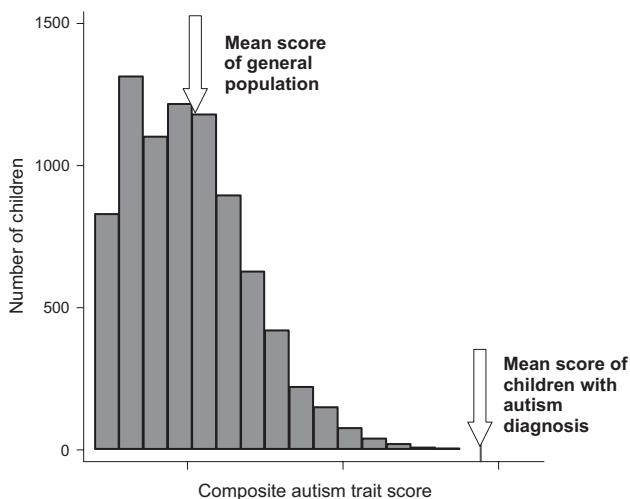

Figure 2 The distribution of the composite autism trait in the general population

score is still only 10.3%, as would be expected given the low prevalence of autism diagnoses. In comparison, looking at all ASD diagnosed children at all ages, this range of the composite trait has a sensitivity of just 53%, and again a specificity of 98.3%. For predicting children diagnosed with AS the sensitivity is 26%, indicating that the trait is far more specific in the early age range and for the autism diagnosis.

AS sample

Table 2 shows the corresponding odds ratios for diagnosis with AS or atypical autism using ALSPAC traits measured when the children were aged between 5 and 11 years old. All but one of the variables were strongly associated with receiving the diagnosis before the interdependencies were taken into account. The associations were weaker than those detected for autism at the younger age. After completing the multivariate analysis, only two measures of social impairment and two of restricted activities remained statistically significant.

According to the ICD-10 criteria, a diagnosis of AS requires qualitative traits in social impairment, and two in repetitive behaviours. The SCDC *Social cognition* scale accurately reflects two of the social impairment criteria. The CCC *Coherence* sub-scale described subtle impairments in conversation and lack of consideration for the listener, another trait of social impairment. The two traits associated with diagnosis from the repetitive behaviour domain, *Touches things or people repeatedly* and *Arranges things in particular ways*, were also included in the composite AS trait. Overall the composite AS trait contained ratios between domains of 3:2 to reflect the weightings between domains specified in ICD-10 (weighting = 2 to *social cognition score*, 1 to *coherence*, 2 to *Touches things or people repeatedly/Arranges things repeatedly in particular ways*).

The general population have significantly higher scores in the composite AS trait than those diagnosed with AS or atypical autism between the ages of 5 and 11 (t = 20.91, df = 5956, p <.001) [Figure 3].

The AS composite trait is a good fit; 44% of the variability in the outcome of diagnosis (pseudo variance) can be explained by the composite trait, $\chi^2(1)$ = 86.46, N = 5958, ASD = 14, p <.001. All the children with a clinical diagnosis in the AS sample scored within the top 5% of the AS composite trait score. The sensitivity of the composite AS trait in predicting diagnosis within the top 5% percent range is therefore 100%, and the specificity 95.4%. However, for this score and age range, the positive predictive value of the composite score is still only 4.9% for AS.

Educational provision

The composite traits indicated that there were undiagnosed children with autistic symptoms at the
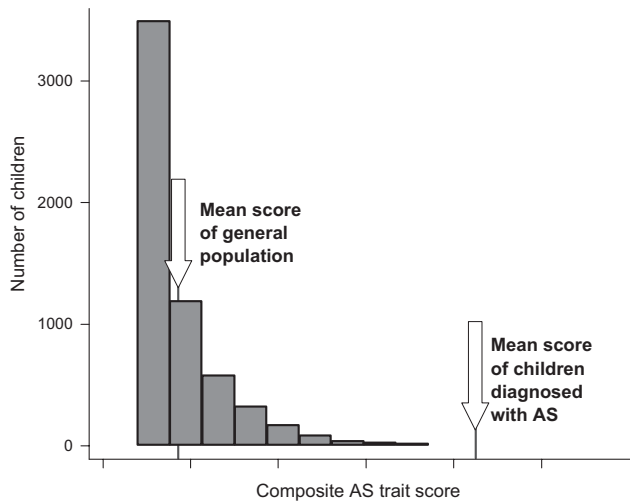
Figure 3 The distribution of composite AS trait in the general population

same severity as children with clinical diagnoses. Altogether there were 135 children in the top 2% of the autism composite trait; educational data were available for only 124 of them. There were 287 children who had scores in the top 5% of the AS trait. Data was available for 248 of these children. Both these figures include 23 children who were in the top range of both scores. Table 3 shows the breakdown of educational provision.

Undiagnosed children with behaviours at the same severity as those diagnosed with either autism or AS were much less likely to have any recognised special educational needs (Table 3). All of the children diagnosed with autism and 89% of those diagnosed with AS had a statement of special educational needs at school. However, 55% of children with autistic traits at the same levels as those who had an autism diagnosis and 57% of children with traits at same severity as those diagnosed with AS had not been identified as needing any special provision at school. Of all those who were identified as needing special provision at school, 37.5% of those with autistic symptoms had been diagnosed with an ASD, as had 26% of those with AS symptoms.

The majority of the children who were not diagnosed with ASD but who had received intervention at higher levels at school were identified by the educational data as having 'cognition and learning' difficulties (55%). In contrast, the majority of children with an ASD diagnosis had 'communication and interaction' difficulties listed as the primary reason for school intervention (72%).

There were no significant differences between the diagnosed and undiagnosed groups for maternal class, marital status, mother's educational attainment, child's ethnic background or home ownership status. However, girls were less likely to be diagnosed with AS and autism than boys $\chi^2(1) = 13.95$, $N = 435$, $p < .001$. The ratio of boys to girls in the undiagnosed samples was 2:1 compared to 10:1 in the diagnosed sample.

Discussion

Substantive findings

It is important to note that while our findings are fairly consistent with levels of unidentified ASD in the population that might be predicted from the existing literature, it would be more accurate to describe the children identified in terms of their impaired cognitive and social functioning as the composite traits used do not amount to a diagnostic assessment. However, the high-scoring composite traits encapsulated behaviours required for ASD diagnosis in the required ratios. For autism a child in the top 2% according to our measures is an asocial, mostly non-verbal child who does not indulge in pretend play, is afraid to try unfamiliar things and exhibits repetitive behaviours. Children scoring highly on the AS composite were reported to be children who lacked social skills and empathy, had difficulty in conversation and often repeated actions that had no obvious function. Furthermore, these traits highlighting qualitative impairments in the required domains were those that were most strongly predictive of getting a diagnosis in the sample.

Table 3 Number of children accessing each level of educational provision within each sub group of cohort. Reported at age 11 or 12, where complete data available

Level of school provision	Severity of impairment in top 2% *		Severity of impairment in top 5% **		No level of impairment specified	
	Autism diagnosis aged 2–4 years	No diagnosis	AS diagnosis aged 5–11 years	No diagnosis	Any ASD diagnosis	General population
No special provision	0	68	0	144	1	9851
School action	0	16	1	29	1	1236
School action plus	0	4	2	22	3	456
Statement of SEN	21	15	24	26	60	381
Total	21	103	27	221	65	11924

*In composite measure of autistic traits aged 2–4 years (composite autism trait).
**In composite measure of Asperger's syndrome traits measured 5–11 years (composite AS trait).

Thus, undiagnosed children with the highest levels of the composite traits displayed similar behavioural profiles to the diagnosed children. Some of the 'undiagnosed' children may be on the autistic spectrum in its wider sense, perhaps presenting sub-clinical levels of autistic traits such as those described by the broad autism phenotype (Lainhart et al., 2002), while others, particularly those in the top 2%, displayed impaired cognitive and social functioning that could lead to diagnosis were they brought to clinic. The results broadly support the findings of Baron-Cohen et al. (2009) who provide evidence that 'undiagnosed cases [of ASD] do indeed exist in the school age population'.

Our study reflects 'unmet need', described by Flisher et al. (1997) in terms of children who have functional impairments but are not receiving focused educational or health support services. As education generally provides the first level of services, any child who has not had any form of special provision at school by age 11 or 12 is unlikely to have been identified by any specialised service provider.

The use of this method as a screening tool either in schools or in primary health care would be problematic owing to the tendency to identify many children at risk who might not have the disorder after fuller assessment. Diagnosis relies on the experience and judgement of clinicians, and in the UK ideally from a multidisciplinary team, including the parents. Screening for ASD is not currently recommended in the UK because tools have not been fully validated (Williams & Brayne, 2006). The findings could be used to argue for expansion in child mental health services, by illustrating high levels of unmet need. However, service contact alone does not necessarily imply that a child's needs are being met. A wide-ranging review of educational and behavioural interventions for ASD concluded that there is inconclusive and inconsistent evidence about effective interventions (Jordan, Jones, & Murray, 1998). Ultimately, the wider issue of effectiveness of interventions for ASD must be addressed.

The ratio of boys to girls in the undiagnosed samples was 2:1, considerably lower than records of male:female ratios of autism in the general population which are recorded around 4:1 (Yeargin-Allsopp et al., 2003). This indicates there may be a bias towards diagnosing boys.

The SEN data suggest that the majority of the undiagnosed children who *were* identified at school were classed as having learning difficulties rather than social and communication difficulties. Although ICD-10 diagnoses other than ASD were not available for this study, this suggests that undiagnosed children who did have recognised needs were likely to be classified under alternative diagnoses to ASD.

The findings suggest that policy makers should look at barriers to accessing services in more detail. Studies have indicated that most children do not get referred for psychiatric disorders because families themselves are not concerned about symptoms (Burns et al., 1995; Fergusson, Horwood, & Lynskey, 1993; Leaf et al., 1996). No measures of impact on parents were available at the younger age, and in the older age range the influence of impact has yet to be assessed.

Barriers to service access include parents' perceptions that help might make matters worse, that they did not want a label, and that they considered their children's difficulties to be understandable given their situation (Ford, Goodman, & Meltzer, 2003). Financial issues, time pressures, mistrust of mental health services and uncertainty as to whether services exist have also been identified as barriers (Ford, Hamilton, Meltzer, & Goodman, 2008). Such reports, together with findings from this study and others highlight the need for more nuanced, in-depth studies of barriers to accessing services perhaps using qualitative methods.

Limitations

Missing data limited the size of the sample at every stage. It is not known to what extent the non-responders in the cohort would bias the results, although Wolke et al. (2009) indicated that despite drop-out in the ALSPAC cohort, the validity of regression models for behavioural disorders was only marginally affected. Educational data were not available for all children; some may have moved away or attended private schools.

There was a large gap between the measurement of the composite autism trait (age 2–4) and SEN data (age 11–12). This gave time for a potentially large improvement in behavioural profiles. However, in the diagnosed children we can see that all children were still statemented, and clinically the prognosis would be one of limited improvement.

In recent years, the prevalence of ASD has been increasing and this is largely attributed to changes in diagnostic practises (Fombonne, 2001). This autism sample was diagnosed in the mid-1990s and the AS sample late in the same decade or early in the next. These results do not therefore necessarily reflect the present level of identification among children with ASD.

Conclusion

This study demonstrates that there are a substantial number of undiagnosed children with difficulties of equivalent levels to those with clinical diagnosis of ASD, whose predicament is currently unrecognised, perhaps missing opportunities for appropriate intervention by education or health services.

Please note: Any queries (other than missing material) should be directed to the corresponding author for the article.

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Key points

- Epidemiological studies have consistently highlighted a disparity between the true prevalence of childhood psychiatric disorders, and specifically autistic spectrum disorders, and their recognition as defined by receiving a clinical diagnosis.
- This study shows that many undiagnosed children have behavioural problems and autistic traits at the same levels as children clinically diagnosed with ASD.
- More than half the children with autistic impairments at the same levels as those with an autism or Asperger's syndrome diagnosis are not identified as having additional needs in educational settings.

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ORIGINAL PAPER

Social and demographic factors that influence the diagnosis of autistic spectrum disorders

Ginny Russell · Colin Steer · Jean Golding

Abstract

Purpose Recent studies in epidemiology have highlighted the existence of children with autistic difficulties who remain undiagnosed. Other studies have identified 'access barriers' to clinics which include factors mediated by parents as well as health and education services. The purpose of this study was to examine whether social and demographic factors play a role in receiving a diagnosis of autistic spectrum disorder (ASD) independently of symptom severity.

Methods Retrospective secondary analysis of a longitudinal UK cohort study, namely, the Avon Longitudinal Study of Parents and Children (ALSPAC).

Results With the severity of autistic traits held constant, boys were more likely to receive an ASD diagnosis than girls. Younger mothers and mothers of first-born children were significantly less likely to have children diagnosed with ASD. Maternal depression before and around the time of their children's autistic difficulties was associated with lack of diagnosis.

Conclusions The study provides evidence that social as well as biological factors can influence whether children are brought to the clinic.

Keywords Pervasive developmental disorders · ALSPAC · Autism · Autistic spectrum disorders · Child psychiatry · Access barriers

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Introduction

Autistic spectrum disorders (ASD) are conceptualised as consisting of a triad of impairments in three domains of behaviour. These are in social interaction, in communication and in restricted interests and repetitive behaviours [1]. For a diagnosis of ASD to be made, children must display impairments in each area according to ICD-10 criteria [2]. Recently, estimates of the prevalence of ASD have increased dramatically, with several studies recording prevalence between 0.9 and 2.7% in areas of the UK, Japan, Sweden and the USA [3–8]. Studies reporting high prevalence rates often calculate results by including children who have been documented by researchers as having symptoms of ASD but who have never received a clinical diagnosis. A recent UK study established that a substantial number of undiagnosed children display the same levels of autistic symptoms as those with either an autism or an Asperger's syndrome (AS) diagnosis [9]. There has been a general trend of increasing prevalence of ASD over the last 50 years in the developed world [10], although results from another recent study suggested childhood levels of impaired social behaviour (a core symptom of ASD) may have levelled off or slightly decreased over the last decade in the UK [11].

Our study is not concerned with establishing the true prevalence of ASD. Rather, given that other studies have identified a large number of undiagnosed children who were included in prevalence estimates, we examine what distinguishes those children without diagnosis but with autistic traits at clinical levels from those who have received a formal ASD diagnosis in the clinic. Our hypothesis, then, was that there may be social and demographic factors that we can detect that might explain why some children are diagnosed and others are not.

There is an important distinction to be made between risk factors for autism and 'access barriers'. Whereas access barriers are socially determined factors which may prevent children with mental health difficulties from reaching the clinic [12], risk factors actually predispose individuals to developing a condition.

These differences may be hard to untangle in association studies that do not control for symptoms. Our secondary analysis of data from a longitudinal cohort used autistic symptoms to identify undiagnosed children whose behaviours matched the diagnostic thresholds for ASD. The association of social and demographic factors that might influence diagnosis was then compared between children clinically diagnosed with ASD ($n = 71$) and children in the undiagnosed comparison group ($n = 142$), as well as with the study population at large ($n = 13,981$). Controlling for severity of autistic traits offered a way of exposing factors independent of symptom severity that could predict ASD diagnosis. Such a methodology should expose access barriers, rather than risk factors which would predispose children to have autistic traits per se, whether diagnosed or not. The method used to identify the undiagnosed children was adapted from that of a previous study [9], which used similar techniques to identify children with autistic symptoms at comparable levels to children diagnosed with classical autism and AS.

Methods

Sample

The Avon Longitudinal Study of Parents and Children (ALSPAC) is an ongoing longitudinal cohort study based in South West England, following approximately 14,000 children. ALSPAC has taken detailed measures of the health and development of its study children throughout their lives, including an assortment of measures of behaviour from across the triad of impairments. The children were all born between 1991 and early 1993. Our analysis was restricted to children in the ALSPAC cohort that survived to 1 year ($n = 13,981$). The social and demographic factors of the ALSPAC cohort are representative of the overall population in the UK as measured by the 1991 UK national census [13]. Ethical approval for the study was granted by the ALSPAC Law and Ethics Committee and the Local Research Ethics Committee.

Children within the ALSPAC cohort with a clinical ASD diagnosis ($n = 71$) were identified independently from the parent study via their medical records. The 71 children were all formally diagnosed in clinical settings with either childhood autism, AS, or atypical autism according to ICD-10 criteria, before the age of 13 years.

Protocols concerning how the children's medical records were accessed and how formal clinical ASD diagnoses were identified from these records have been outlined in a previous publication [14]. The medical diagnoses were assigned by the children's clinicians who were blind to all measures taken by the ALSPAC cohort study.

In order to identify undiagnosed children who shared autistic symptoms early in their lives with those who were subsequently diagnosed, a range of autistic traits that had been independently assessed by ALSPAC were identified. As age at onset for atypical autism may occur later than for other sub categories of ASD, but must occur by age 3 according to ICD-10, autistic traits measured by ALSPAC between age 2.5 and 4 years were examined. In total, we identified 27 measures of impairment in social interaction, social communication, and repetitive behaviours or restricted interests that were measured by ALSPAC in this age range. We then used logistic regression to determine which predicted receiving the formal diagnosis of ASD in the clinic.

Measures of autistic behaviour included in regression

Two relevant subscales were incorporated in the analysis in their full form; the Temperament Subscale from the Emotionality, Activity and Sociability questionnaire [15], and the Prosocial subscale of the Strengths and Difficulties Questionnaire (SDQ) [16]. Some of the scales and modules available were designed for more generic use and were not autism specific; in this case, only those items measuring ASD criteria were included in the analysis. Items from the Revised Rutter Parent Scale for Preschool Children [17] and the Pre-School Activities Inventory [18] were included. Of the 27 predictor traits entered into logistic regression, 9 questions were specifically designed for ALSPAC questionnaires. All the data were derived from parental report. A full list of the measures of autistic traits that were entered into the regression is given in Table 1.

Analysis to establish comparison group

Logistic regression was used to examine which measures of autistic behaviour were most strongly associated with receiving a clinical diagnosis of ASD. In order to establish a set of traits which in combination would give the best correlation with diagnosis, we considered behaviours from each domain of the triad separately. To allow comparison of effect sizes for behaviours with different scales of measurement, the traits were standardised; consequently, the odds ratios (ORs) from the analysis reflect the effect for a 1 SD increase. The direction of measurement of approximately half of the traits was reversed so that a high score always reflected increasing autistic impairment.

Table 1 Behavioural traits measured by ALSPAC between the ages of 3 and 5 years showing which is most predictive of receiving a diagnosis of ASD

Trait (variable name)	Age measured in months	Odds ratio (CI 95%) Unadjusted $P < 0.01$	Odds ratio (CI 95%) Adjusted for effect of interdependencies within domain $P < 0.01$
Domain: Impairments in social interaction	$n = 9{,}532\text{–}10{,}218^{a}$		$n = 8{,}198$
EAS sociability score	38	3.11 (2.48–3.90)	
SDQ Prosocial score	47	5.66 (4.31–7.43)	3.77 (2.67–5.30)
Child gets on with other children	42	2.62 (2.19–3.14)	
Child avoids eye contact when talking	38	2.03 (1.73–2.39)	1.38 (1.10–1.72)
Child is affectionate to family members	42	1.61 (1.34–1.93)	1.41 (1.11–1.79)
Avoids eye contact when reunited with mum	42	1.35 (1.19–1.53)	1.29 (1.08–1.53)
After separation child wants hug	30		
Child does things on his/her own	42	2.35 (1.98–2.78)	2.12 (1.51–3.0)
Child has at least one good friend	47	1.89 (1.64–2.19)	1.78 (1.33–2.37)
Child prefers adults to children	47	1.90 (1.50–2.41)	1.58 (1.21–2.09)
Impairments in communication	$n = 9{,}615\text{–}10{,}243^{a}$		$n = 8{,}679$
Child sings songs	38	2.01 (1.71–2.35)	
Child played looking after babies	30	2.87 (2.07–3.99)	1.63 (1.14–2.34)
Enjoys pretend games	42	2.88 (2.40–3.44)	2.13 (1.72–2.64)
Child combines words	38	1.87 (1.66–2.10)	
Number of words child can string together	38	1.64 (1.52–1.76)	
Child stays mainly silent	38	1.97 (1.75–2.22)	2.02 (1.62–2.53)
Child echoes what others say	38		
Child uses gestures to get what wants	38	4.29 (3.16–5.81)	
Child has speech difficulty	42	2.11 (1.88–2.36)	2.42 (1.89–3.12)
Child talks with words in wrong order	38	1.52 (1.22–1.90)	
Intelligibility score	38	2.05 (1.83–2.30)	1.30 (1.073–1.57)
Restricted repetitive and stereotyped patterns of behaviour	$n = 9{,}881\text{–}10{,}200^{a}$		$n = 9{,}881$
Child is afraid of new things or new situations	42	1.77 (1.46–2.15)	1.64 (1.30–2.06)
Child is fussy or over particular	42		
Child often rocks head or body repeatedly	42	1.42 (1.31–1.54)	1.30 (1.19–1.43)
Child has tic or twitch	42		
Child has other unusual repetitive behaviour	30	1.57 (1.43–1.72)	1.43 (1.29–1.59)
Frequency child rocks head or body	42	1.40 (1.26–1.55)	

Blank fields denote no significant relationship with diagnosis at 1% level

[a] Varies with number of complete scores per trait. Any clinical ASD diagnosis ($n = 71$) was the outcome measure for logistic regression

Backwards logistic regressions were performed on each domain of impairment in turn. First, measures of social impairment were considered, second measures of communication and finally measures of repetitive behaviour and restricted interests. Traits significant at the 1% level in univariate analysis were carried forward to multivariate analysis where traits were adjusted within each behavioural domain to account for any interdependencies. Table 1 shows the results of this regression and which autistic traits measured between 2.5 and 4 years old were most strongly associated with clinical diagnosis of ASD.

The traits in each behavioural domain that were most strongly associated with a clinical diagnosis were the SDQ prosocial scale, the 'enjoys pretend games' score, the 'speech difficulties' score, and the 'displays unusual repetitive behaviour' score, and one item measuring how afraid a child was of new situations. An overall composite ASD trait was then constructed from these individual autistic traits. The number of traits we included from each behavioural domain was specified by thresholds required for diagnosis in the ICD-10 diagnostic criteria for autism (given in Table 2). The only exception was that we

included just one trait for communication difficulties to reflect differences in criteria between AS and childhood autism: the requirements for diagnosis of AS are the same as for autism, bar impairments in communication which may or may not occur. For a diagnosis of atypical autism, diagnostic criteria differ from autism in either age at onset or failure to fulfil all three domains of diagnostic criteria. The composite ASD trait therefore encompassed impairments that would result in diagnosis for all three common conditions on the autistic spectrum. Each of the behaviours that made up the composite trait was also given a weighting that reflected their power, relative to each other, to predict ASD diagnosis. The individual autistic traits in each domain of impairment that went into making up the composite ASD trait are described in detail, together with the weightings that were assigned in the composite ASD trait, in Table 2.

The ASD composite trait was a good fit, explaining 47% of all the variability (log likelihood) in the outcome of ASD diagnosis, $\chi^2 (1) = 288.76$, $N = 8{,}852$, $P < 0.0001$.

We defined the comparison group as those children who had not been diagnosed with ASD, but who were reported as displaying the most severe autistic traits, those with scores in the top 2 percentiles of the composite autism score ($n = 142$), in other words, children who were most likely to receive a diagnosis were they brought to the clinic. The sensitivity of the composite trait in this percentile range to predict an actual ASD diagnosis was 68% and the specificity was 98.4%. The undiagnosed comparison group was consequently composed of children who had autistic traits measured as preschoolers at the same levels of severity or worse than those with clinical ASD diagnosis in the ratios required for diagnosis of ASD.

According to our measures, then, as preschoolers, undiagnosed children in the comparison group displayed repetitive behaviours, exhibited speech difficulties, did not indulge in pretend play and were afraid to try unfamiliar things and lacked prosocial behaviours. We therefore describe this comparison group as 'children with severe autistic behaviours'.

Social and demographic factors that could influence diagnosis: measures

Basic demographics: gender, ethnic origin, age of mother at delivery and birth order were included for analysis. A firstborn child was defined as where there were no previous pregnancies that resulted in birth. Another recent UK study found socioeconomic disadvantage to be associated with childhood psychopathology [19]; therefore, several socioeconomic measures were also examined. These were social class, measured by the occupational status of parents, maternal educational attainment, home

ownership status, financial hardship and marital status. With family risk factors in mind, researchers at ALSPAC had previously compiled a Family Adversity Index of stressors [20]. These included measures of financial hardship, measures of maternal mental health, criminal activity, substance abuse and scales relating to partner cruelty and affection. The Crown Crisp Depression Index [21] was also included. All this information was derived from questionnaires sent to the mothers when their children were young. A complete list of measures together with the age of the children when they were reported is given in Tables 3 and 4.

We also examined whether respondents had ever had severe depression or other psychiatric problems at any point in their lives. Mothers reported this retrospectively when children in the cohort were 11 years old.

Analysis

A χ^2 test of association and, where appropriate, a Fishers exact test were applied to determine whether these factors in children's social or economic backgrounds were associated with ASD diagnosis. Analyses of variance (ANOVA) were carried out where the socio-demographic factors were recorded as normally distributed scores.

First, we established which demographic factors predicted ASD diagnosis when considering the general (non-autistic) population. Second, using the comparison group of undiagnosed children with autistic behaviours, we established whether such social and demographic factors were still associated with ASD diagnosis when the severity of autistic symptoms was comparable at preschool age. Finally, we considered whether there were any associations between these socio-demographic factors and the comparison group—children who displayed severe autistic traits but remained undiagnosed.

Results

Socioeconomic and fixed factors

As Table 3 illustrates, according to these data, ethnic origin, maternal class and mother's marital status did not significantly predict a child either having an ASD diagnosis or displaying severe autistic traits.

There were far more boys than girls in the diagnosed sample than in the general population, with ratios of 9:1. Boys were more likely to suffer from severe autistic traits, whether diagnosed with ASD or not. However, even with the severity of autistic traits held constant, boys were still significantly more likely to receive an ASD diagnosis than girls (Table 3).

Table 2 Requirements laid out in ICD-10 diagnostic criteria for research for autism and ALSPAC traits which were most predictive of diagnosis showing how they match the criteria

ICD-10 diagnostic criteria for research: diagnostic thresholds for autism Abnormal or impaired development in the following domains	ALSPAC trait most predictive of ASD diagnosis in domain (age of measurement, months)	How trait fulfils criteria	Weighting in composite score log (OR)[a]
A. One of the following three areas:	Enjoys pretend games (42 m)	This trait provides an exact match with the diagnostic criterion listed as number (3) (column 1)	0.73
(1) receptive or expressive language as used in social communication	This was a response by either parent to the single question *Does the child play imaginatively, enjoys 'pretend' games?* answered *Yes/Sometimes* or *No*. This trait was part of the Revised Rutter Parent Scale for Preschool Children		
(2) development of selective social attachments or of reciprocal social interaction			
(3) functional or symbolic play			
B. Social interaction, one of the following areas:	SDQ prosocial score (47 m)	Negative answers to all of these questions accurately capture impairments in reciprocal social interaction as described in criteria (2) and (3) of ICD-10 (column 1)	0.93
(1) failure adequately to use eye-to-eye gaze, facial expression, body posture and gesture to regulate social interaction	Composed of the following five items in the SDQ questionnaire,[b] which were rated by mothers as *Doesn't Apply/ Somewhat applies/Certainly applies*		
(2) failure to develop peer relationships involving a mutual sharing of interests, activities and emotions	*The child*		
	Is considerate of other people's feelings		
(3) A lack of socio-emotional reciprocity as shown by an impaired or deviant response to other people's emotions; or lack of modulation of behaviour according to social context, or a weak integration of social, emotional and communicative behaviours	*Shares readily with other children (treats, toys, pencils, etc.)*		
	Is helpful if someone is hurt, upset or feeling ill		
	Is kind to younger children		
	Often volunteers to help others (parents, teachers, other children)		
C. Communication, two of the following areas:	Speech difficulty (42 m)	Although very generic, this trait reflects criteria (1 & 2)	0.69
(1) a delay in, or total lack of development of spoken language not accompanied by compensation through the use of gesture	This was a response by either parent to the single question *Has your child got a speech difficulty?* This trait was part of the Revised Rutter Parent Scale for Preschool Children answered *Yes/ Sometimes* or *No*		
(2) failure to initiate or sustain conversational interchange (at whatever level of language skills are present)	*Only one trait included to reflect differences between diagnosis of AS and autism*		
(3) stereotyped and repetitive use of language or idiosyncratic use of words or phrases			
(4) abnormalities in pitch, stress, rate, rhythm and intonation of speech			

Table 2 continued

ICD-10 diagnostic criteria for research: diagnostic thresholds for autism Abnormal or impaired development in the following domains	ALSPAC trait most predictive of ASD diagnosis in domain (age of measurement, months)	How trait fulfils criteria	Weighting in composite score log (OR)[a]
D. Restricted, repetitive, and stereotyped patterns of behaviour, two of the following areas:	Afraid of new things or new situation (42 m)	This trait was considered as distress over changes in environment as described in diagnostic criterion (5)	0.21
(1) an encompassing preoccupation with one or more stereotyped and restricted patterns of interest; or one or more interests that are abnormal in their intensity although not abnormal in their content or focus	This was a response by either parent to the single question *Does your child tend to be afraid of new things or new situations?* This trait was again part of the Revised Rutter Parent Scale for Preschool Children and possible responses were *certainly true/sometimes true* and *not true*		
(2) compulsive adherence to specific, non-functional, routines or rituals			0.27
(3) stereotyped and repetitive motor mannerisms that involve either hand or finger flapping or twisting, or complex whole body movements	Displays other unusual repetitive behaviours (42 m)	This trait mirrored the diagnostic criterion (3) describing stereotyped and repetitive motor mannerisms	
(4) preoccupations with part-objects or non-functional elements of play materials	This trait originated from a question (c) designed by ALSPAC, namely, *How often does the child do the following: (a) repeatedly rocks head or body, (b) has tic or twitch, (c) any other unusual [repetitive] behaviour?*		
(5) distress over changes in small, non-functional, details of the environment	The measure was determined by response of *more than once a week/less than once a week* or *never*		

[a] The power of each trait to predict an ASD diagnosis, relative to other traits in the composite ASD score

Table 3 Association of social and demographic factors with ASD diagnosis and with severe but undiagnosed autistic traits

Demographic factor in child's background	Number of 'non-autistic' children (a)	Number of diagnosed children (b)	Pearson χ^2 (df) P (a) vs. (b)	Number of undiagnosed children (c) control group	Pearson χ^2 (df) P (b) vs. (c)	Pearson χ^2 (df) P (c) vs. (a)
Gender of child						
Male	7,301	64	43.31 (1)	111	4.63 (1)	41.52 (1)
Female	7,013	7	$P < 0.001$*	31	0.031*	$P < 0.001$*
Ethnic origin						
White	11,287	64	0.71 (1)	125	0.30 (1)	0.02 (1)
Black and ethnic minority	597	5	0.400	7	0.581	0.884
Age of mother at delivery						
25 or younger	4,237	9	13.10 (2)	43	8.11 (2)	1.13 (2)
26–30	5,411	30	0.001*	51	0.017*	0.569
31 or older	4,120	32		48		
Social class-maternal (measured during pregnancy)						
Professional	3,709	22	0.472	30	2.16 (2)	3.64 (2)
Non-manual	3,709	22	0.790	52	0.340	0.162
Manual	1,968	9		18		
Marital status (measured when child was 8 weeks old)						
Single	3,230	15	0.41 (1)	36	0.47 (1)	0.075 (1)
Married	9,652	54	0.524	102	0.494	0.785
Birth position of child						
First born	5,709	19	5.92 (1)	46	0.27 (1)	7.32 (1)
Subsequent	7,021	45	0.015*	92	0.606	0.007*
Mother's highest educational qualification						
None or vocational	3,660	18	1.05 (2)	51	3.17 (2)	6.45 (1)
School level	4,218	28	0.592	51	0.204	0.040*
Degree or higher	4,335	24		35		

(a) Children undiagnosed with ASD in population, without severe undiagnosed ASD traits limited to number that survived to one year and shown where data is recorded for children

(b) Children diagnosed with ASD between ages of 1–13 ($n = 71$)

(c) Children with severe autistic traits when aged 3–4 at the same severity or worse as those diagnosed with either AS, atypical autism or autism (top 2% of composite ASD score), without an ASD diagnosis ($n = 142$)

* Significant at 5% levels

The average age of mothers with an ASD diagnosis was 3 years higher than in the population generally: mean age at delivery for ASD cases = 30 years, while the mean age of the general population was 27 years; $F(1, 13,900) = 9.61$, $P = 0.002$. When controlling for autistic traits, mothers of diagnosed children also tended to be older at delivery than those in the undiagnosed comparison group. Mean age at delivery for mothers of children in the comparison group was 28 years, 2 years younger than mothers of children who subsequently obtained ASD diagnoses for their children, again very significant differences $F(1, 211) = 7.24$, $P = 0.008$. These results remain significant when maternal age at delivery is grouped by age ranges (Table 3). Mothers' age at delivery was associated to a lesser extent with children who display severe autistic-type difficulties at preschool age, $F(1, 13,979) = 3.34$, $P = 0.068$. So according to our data, although older motherhood strongly predicts ASD diagnosis, it does not strongly predict having autistic traits per se.

A first-born child was also less likely to receive an ASD diagnosis in this data set (Table 3), whereas diagnosis appears equally likely for second, third or subsequent siblings. There was no such association between second born children and ASD diagnosis, or third born: ($\chi^2(1) = 0.24$, $P = 0.626$; $\chi^2(1) = 0.02$, $P = 0.889$), when the likelihood of second- or third-born children is compared with the likelihood of subsequent children being diagnosed with ASD. So this effect apparently relates to whether the child is the first child or not, rather than the any other birth position in the family. In these data, second or subsequent

Table 4 Association of family adversity and mothers' mental health with ASD diagnosis and with severe undiagnosed traits

Demographic factor in child's background	Number of non-autistic children (a)	Number of diagnosed children (b)	Pearson χ^2 (df) P (a) vs. (b)	Number of undiagnosed children (c) control group	Pearson χ^2 (df) P (b) vs. (c)	Pearson χ^2 (df) P (c) vs. (a)
Family faces financial hardship (reported when child age 2–4)						
No	8,444	48	0.16 (1)	107	1.73 (1)	4.44 (1)
Yes	1,060	5	0.692	22	0.189	0.035*
Partner affection/cruelty (reported when child age 2–4)						
Affectionate/not cruel	6,971	42	0.59 (1)	93	0.18 (1)	0.19 (1)
Not affectionate/cruel	1,592	7	0.440	19	0.671	0.660
Psychopathology of mother–maternal mental health (when child age 2–4)						
Low	8,829	52	1.00 (1)	111	3.60 (1)	5.02 (1)
High	1,564	6	0.317	31	0.058	0.025*
Substance abuse (when child age 2–5)						
No	8,938	50	<0.00 (1)	126	0.25 (1)	0.94 (1)
Yes	1,469	8	0.944	16	0.618	0.332
Trouble with police (when child age 2–5)						
No	9,988	55	0.33 (1)	137	0.29	0.02
Yes	387	3	0.370	5	0.589	0.896
Home ownership status (when child was 8 weeks old)						
Owned	9,415	54	1.75 (2)	92	4.58 (2)	4.02 (2)
Rented privately	1,829	6	0.416	27	0.101	0.134
Rented council	1,578	8		20		
Mother had psychiatric problems or depression anytime during her life (when child aged 11)						
No	876	5	0.01 (1)	14	0.40 (1)	1.01 (1)
Yes	6,298	35	0.907	75	0.529	0.314

(a) Children undiagnosed with ASD in population, without severe undiagnosed ASD traits limited to number that survived to one year and shown where data is recorded for children

(b) Children diagnosed with ASD between ages of 1–13 ($n = 71$)

(c) Children with severe autistic traits when aged 3–4 at the same severity or worse as those diagnosed with either AS, atypical autism or autism (top 2% of composite ASD score), without an ASD diagnosis ($n = 142$)

* Significant at 5% levels

children were also more likely to be reported as displaying severe autistic-type difficulties (those with scores in top 2% of the ASD composite trait) whether diagnosed or not.

There was a small effect associating undiagnosed autistic traits with lower maternal educational attainment, replicating a previous finding in another UK cohort [3].

Measurements of family adversity and maternal mental health were also analysed to see if they predicted a child's ASD diagnosis (Table 4). Financial hardship, partner cruelty and affection, home ownership status, maternal mental health, substance abuse, maternal psychopathology, and being in trouble with the police showed no significant associations with a child receiving an ASD diagnosis. A greater proportion of children whose families suffered severe financial hardship displayed severe autistic behaviours, although this did not lead to diagnosis.

Mothers of children in the comparison group were more likely to suffer from depressive symptoms than those who (later) obtained an ASD diagnosis for their children. They had significantly higher scores as measured by the Crown Crisp Depression Index when the children were 21 months old; $F(1, 180) = 7.25$, $P = 0.008$. Mothers of young children who displayed severe autistic traits also reported slightly more symptoms of depression than those in the population at large, although this effect was far weaker—not significant at 5% levels; $F(1, 10,396) = 3.73$, $P = 0.053$. Maternal psychopathology when the children were aged 2–4 years was also associated with lack of ASD diagnosis (Table 4). However, when we considered whether mothers had experienced depression and/or psychiatric problems at any point during their lives as a whole (as reported when their children were 11 years of age), no associations were detected.

Discussion

Gender

We can conclude that there is a strong gender bias with a far higher proportion of boys than girls exhibiting autistic traits whether diagnosed or not. That there is a real and uneven distribution of ASD between genders is well established. Boys are more than four times more likely to have ASD according to many sources and are clearly more likely to suffer from these types of symptoms [22]. More interesting is our finding that even with symptom severity held constant, there is still a gender bias towards diagnosing boys. Our analysis suggests that an addition bias of identification occurs—girls are less likely to be identified with ASD even when their symptoms are equally severe. Other studies have put forward evidence for comparable gender biases in identification of children with special educational needs more generally [23, 24] as well as in attention-deficit hyperactivity disorder specifically [25]. A popular conception of ASD is often of a 'male' disorder [26]. Indeed, an influential psychological theory of ASD 'the extreme male brain theory' has been proposed [27]. Such understandings may have led to gender stereotyping by education professionals, clinicians and parents when identifying children with severe symptoms. Researchers have speculated that many girls with AS are never referred for diagnosis, and so are simply missing from statistics [28, 29].

Age of mother at delivery and birth position

We already knew that older motherhood would be associated with ASD diagnosis as a recent study had found this association using the same cohort [30]. What was new in this area was that age was still associated with diagnosis when controlling for severity of autistic traits. The association between maternal age and ASD diagnosis is stronger than that between maternal age and ASD traits per se. One possible interpretation is that older mothers are better at identifying their children's difficulties and have more confidence in bringing concerns to the clinic. Younger mothers may find it harder to identify problems. This finding may be related to the effect of birth position. Later motherhood is correlated with second or subsequent children. In the ALSPAC data, first-born children are less likely to receive a diagnosis, perhaps for similar reasons to those given above; the mother of a first-born child is less likely to have developed a standard of what 'normal' is like in her family. In this sense, the first-born child may act as a pioneer for identification of problems in siblings. However, another recent, much larger cohort study found that first-born children were more likely to be classified as having

ASD [31], suggesting 'stoppage' (a tendency for parents of one child with ASD not to have subsequent children) as one potential explanation. These inconsistent findings could be due to the limitations of our sample size as well as differences in methodologies.

Their study and several others studies also found correlations between older motherhood and children diagnosed with ASD [30, 32]. Interpretations generally encompass increased risk of birth complications with age and increasing genetic vulnerability. Such direct biological explanations could be tempered by the possibility that older, more experienced parents are more likely to be sensitised to problems. Also, parents with autistic traits (including sub-clinical traits) may have children later in life.

Maternal depression

Symptoms of maternal depression at 21 months were clearly associated with a lack of ASD diagnosis. There was no corresponding association when we considered whether mothers had psychiatric problems or depression at anytime during their lives, although other studies have reported increased risk of autism with parental history of any psychiatric disorder [33]. Our findings suggest that maternal depressive symptoms or psychopathology specifically around the time of a child's autistic difficulties might actually hinder diagnosis. We suggest that maternal depression may act as an 'access barrier' to clinical intervention, incapacitating mothers and thus preventing them from seeking clinical help for their children. Even if a mother seeks help, a health professional could explain her concern as a symptom of her depression.

Limitations

It is important to stress that the children who received an ASD diagnosis and the undiagnosed comparison group were not an exact match. We were not able to match on measures of IQ or a variety of other factors that may be important in clinical assessment of ASD. Diagnosis relies on the experience and judgement of clinicians, and in the UK ideally from a multidisciplinary team, including the parents. Children diagnosed with ASD are a very heterogeneous group, as the triad encompasses a wide range of behaviours, and clinical diagnostic practise is not standardised. This limitation should be taken into account when evaluating any study concerned with diagnosis of ASD. Furthermore, it was not possible to assess whether children in the control group had alternative diagnoses assigned to them, so the results relate specifically to diagnosis of ASD, although we were able to ascertain that by the time children

in the comparison group started secondary school more than half had not been recorded as having any special educational needs in their school records.

Missing data limited the size of the sample. It is not known to what extent the non-responders in the cohort would bias the results. Other studies have indicated that non-response is crucial when interpreting prevalence estimates of ASD [34], although a recent study utilising the ALSPAC indicated that despite drop-out in the cohort, the validity of regression models for behavioural disorders was only marginally affected [35]. In addition to this, the measures of autistic traits were subject to the same biases as any dataset derived from parental report. For example, it could be the case that mothers' depressive symptoms may have biased reporting of their children's behaviour.

Children in the control group were matched at preschool age. Approximately two-thirds of the children were diagnosed after age 4, so a significant improvement in autistic symptoms in the undiagnosed control group after starting school may have accounted for lack of diagnosis. Such differences in developmental trajectory could confound attempts to uncover which factors led to diagnosis as groups diverged. In order to clarify effects of such potentially improving developmental trajectories in later childhood, we also carried out additional analyses where the ASD sample was restricted to those children who were diagnosed before aged 7. Here, the ASD diagnoses were given to children over roughly in the same age range in which children in the comparison group shared symptom severity. In fact, all factors positively associated with ASD diagnosis remained associated even when the sample was limited in this way. This indicated a degree of reliability in the results.

Implications and future research

The study provides evidence that social as well as biological factors can influence which children are brought to the clinic. Another recent study in the USA also concluded that social factors have an influence, finding that children living very close to a child previously diagnosed with autism were more likely to be diagnosed with autism [36]. Understanding social factors that act as access barriers may provide useful insights for clinicians in practice. However, it is beyond the scope of this study to assess whether clinical diagnosis and subsequent interventions are valuable for children. Some autistic self-advocates argue for societal acceptance, rejecting treatment of all symptoms [37]. In order to engage with these wider debates, we intend to examine differences in social and clinical outcomes between children diagnosed with ASD and children in the comparison group as they reach adolescence.

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'Article 3'

Social and behavioural outcomes in children diagnosed with autism spectrum disorders: A longitudinal cohort study.

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Abbreviated title: Behavioural and social outcomes in children on the autism spectrum.

Abstract

Background

Our objective was to compare social and behavioural outcomes between children formally diagnosed with autism spectrum disorders and children who displayed autistic traits at preschool age, but remained undiagnosed as teenagers.

Method

A secondary analysis of data from a birth cohort study, the Avon Longitudinal Study of Parents and Children (n=13,944). Children clinically diagnosed with ASD were identified from their medical records (n=71). A comparison group, who displayed autistic traits at age 3-4, but without ASD diagnosis were also identified (n=142). Social and behavioural outcomes in adolescence were compared between the two groups.

Results

Children with ASD diagnoses were more impaired as teenagers that those in the comparison group on a range of measures of autistic-like behaviour. The developmental trajectory of prosocial behaviour shows differences between cases and comparison group increase dramatically in the preschool and early primary years, but after 6 years the trajectories were similar.

Conclusions

The divergence of the clinically diagnosed group and the non diagnosed group in measures of autistic-like behaviour increased with age. This study provides evidence that it may be difficult to distinguish preschool age children who exhibit autistic-like symptoms but improve, from those who go on to develop lifelong impairment.

Background

Autism spectrum disorders (ASD) are recognized as consisting of a triad of impairments, in social interaction, in communication and in social imagination- expressed as restricted interests and repetitive behaviours (Bailey, Phillips & Rutter, 1996). For a diagnosis of ASD to be made, children must display impairments in each behavioural domain of this triad, according to ICD-10 criteria (WHO, 1992). Early diagnosis of ASD, age 3-4, is frequently made in the developed world (Chakrabarti & Fombonne, 2005) reflecting the clinical trend to diagnose ASD younger (Fombonne, Misès, Jousselme, Fujiura, & Guralnick, 2005). In the UK, the National Screening Committee does not currently recommend routine screening of preschool children for ASD because tools have not been fully validated and interventions lack sufficient evidence for effectiveness (Williams & Brayne, 2006).

Studies reporting high prevalence rates for ASD often calculate results by including children who have been documented by researchers as having symptoms of ASD but have never received a formal diagnosis in their communities (Baird et al., 2006; Baron-Cohen et al., 2009). Several epidemiological studies encompassing all childhood neuropsychiatric disorders, reviewed by Costello and colleagues in 2005, suggest that many children with diagnoses made by researchers are not in contact with health services and their difficulties are never clinically diagnosed. Such studies of large populations comparing the numbers of children with psychiatric difficulties who are identified and those who remain clinically undetected have spanned forty years (Canino, Bird, Rubio-Stipec, & Bravo, 1995; Offord, 1995; Rutter, Tizard & Whitmore, 1976). Of those with psychiatric disorder measured in a large UK population based sample, only 25% had been in contact with mental health services over the subsequent three years (Ford, Hamilton, Meltzer, & Goodman, 2007).

Some studies have attempted to look at children's outcomes using unidentified control groups matched in symptom severity to those who have been identified (Angold, Costello, Burns, Erkanli & Farmer, 2000; Crawford & Vignoles, 2010). These researchers argue that using controls with undetected needs allowed them to examine progress of children with identified impairments more carefully, instead of simply comparing outcomes against the population at large, although they also point out the difficulties of matching cases and controls, and finding treatment signals amongst the noise of so many confounding factors. Kelley, Nixon and Bickman (2000) argue that there is a pressing need to compare outcomes in children who have been identified to a comparable group of undetected children as they progress, as a means of evaluating likely effectiveness of provision. For autistic adults, IQ has been found to be the best indicator of a good prognosis (Howlin, Goode, Hutton & Rutter, 2004).

We used prospectively collected data from a large longitudinal cohort; the Avon Longitudinal Study of Parents and Children (ALSPAC), to compare outcomes in adolescence of undiagnosed children with autistic-like behaviours with those who were given a clinical diagnosis. We tested the hypothesis that social and behavioural outcomes would differ in the community in a sample of children diagnosed with ASD, compared with an undiagnosed group who displayed autistic type symptoms at clinical levels when aged 3-4.

Methods

Ethical approval for the wider study was obtained from the ALSPAC Law and Ethics Committee and from the Exeter University Ethics Committee. Parent participants in the cohort were informed that their child's medical records would be accessed but were able to withdraw their consent for this at any stage throughout the ALSPAC study.

Sample

ALSPAC is an ongoing cohort study based in South West England, following approximately 14,000 children born between 1991 and 1993. It has taken detailed measures of the health and development of the study children throughout their lives, including repeated measures of behaviours symptomatic of autism. The social and demographic features of the ALSPAC cohort are representative of the overall population in the UK as measured by the 1991 UK national census (Golding, Pembury & Jones, 2001). Our analysis was restricted to children in the ALSPAC cohort that survived to one year (n=13,944).

Identification of children with clinical diagnosis of ASD was carried out by an independent review of medical records, which showed that 71 ALSPAC study children were formally diagnosed by a clinician with ASD following multi-disciplinary assessment by the time they were aged 12. The methods and protocols for identification of those clinically diagnosed with ASD are detailed elsewhere (Williams, Thomas, Sidebotham & Emond, 2008). These diagnoses were assigned outside the ALSPAC study, by a paediatrician or psychiatrist without access to ALSPAC information or data. Only those children who had been assigned a medical diagnosis (not those who were identified as autistic through educational data) were included. The children were all formally diagnosed with either childhood autism, Asperger's syndrome or atypical autism according to ICD-10 criteria. Children classified as PDD –NOS using DSM-IV standards were classified as 'atypical autism'; the corresponding ICD-10 classification.
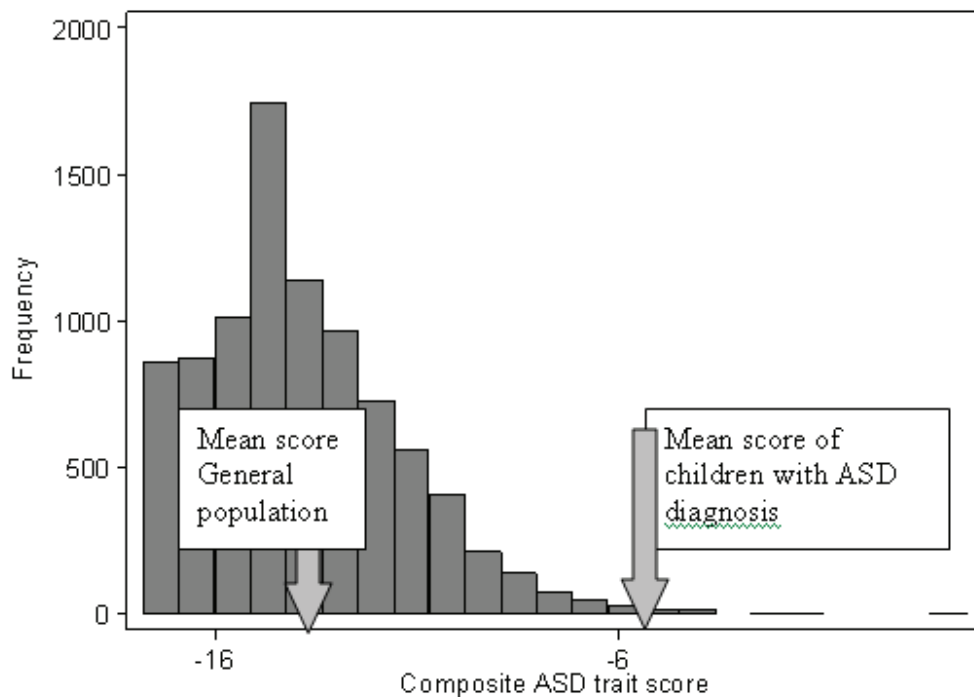
We then identified children with comparable autistic symptoms. ALSPAC had taken various repeated measures of autistic-like behaviours such as social skills, repetitive behaviours, empathy, eye contact, communication difficulties and measures of speech and language development. To establish a group of

undiagnosed children with autistic-like behaviours comparable with those with a formal ASD diagnosis, we created a 'composite ASD trait' score, which was our measure of autistic symptoms in the cohort. A detailed account of how this trait was constructed is published elsewhere (Russell, Steer & Golding, 2010). Briefly, 27 measures of autistic-like behaviour were recorded by ALSPAC when the children were between the ages of 3 and 4 years old. Backwards logistic regression analyses were performed on each domain of impairment in the triad: measures of social impairment, measures of communication, and measures of repetitive behaviour and restricted interests. Traits significant at the 1% level in univariate analysis were carried forward to multivariate analysis where traits were adjusted within each behavioural domain to account for any interdependencies. The traits were standardised, consequently the odds ratios (ORs) reflected the effect for a 1 standard deviation increment in increasing the odds of clinical diagnosis. The traits most closely associated with ASD diagnosis in each behavioural domain of the triad when accounting for interdependencies of other traits were: the prosocial subscale of the Strengths and Difficulties Questionnaire (SDQ) (Goodman, 1997), OR=3.77, 95% CI[2.67–5.30]; the 'enjoys pretend games' score OR=2.13, 95% CI[1.72–2.64], the 'speech difficulties' score OR=2.42, 95% CI[1.89–3.12], and the 'afraid of new situations' score OR=1.64, 95% CI [1.30–2.06], all from the Revised Rutter Parent Scale for Preschool Children (Elander & Rutter, 1996); and one measure designed by ALSPAC: the 'displays unusual repetitive behaviour' score OR=1.43 95% CI[1.29–1.59]. All these measures were recorded by parental report when the children were either 42 or 47 months old.

The 5 measures listed above were kept in the ratios that the ICD-10 criteria for autism specifies, necessary to diagnose ASD (WHO, 1992), within the 'composite ASD trait'. This was to ensure the symptomology of children with high scores on the composite trait matched the diagnostic criteria for ASD as closely as possible. The component measures were then weighted within the composite ASD trait

according to their relative power to predict ASD diagnosis. At ages 3-4 the children with high scores in

the composite ASD trait were reported as having poor social skills, speech difficulties and exhibited

repetitive behaviours. They did not indulge in pretend play, and were afraid of new situations or things.

The 'composite ASD score' was a good model accounting for 47% of all the variability (log likelihood)

in the outcome of ASD diagnosis, χ^2 (1) =288.76, N = 8852, p < .0001. The distribution of the composite

ASD trait is shown in Figure 1.

Figure 1: The distribution of the ASD composite trait in the general population used to define the comparison group, showing mean of general population (-13.82, 95%CI -13.87--13.77) and ASD diagnosed group (-5.59, 95%CI -6.93- -4.24).

The specificity of the top two percentiles of the composite ASD trait in predicting diagnosis was 98.4% and the sensitivity 68%. We therefore identified all children in the cohort without an ASD diagnosis, but with 'composite ASD scores' in the top 2% (*n*=142). This top scoring 2% was used as a cut off point because this range captured the majority of children with ASD diagnoses, and the undiagnosed children displayed autistic type symptoms at levels equivalent (or in some cases more severe) than the children who had been formally diagnosed with ASD. Researchers were blind to the identities of children in the cohort, so no feedback was given to parents at any stage. Two dichotomous groups with clear parameters were thus established - one group comprising children who had received a formal ASD diagnoses (*n*=71), the other group containing children with autistic symptoms aged 3-4 (children in the undiagnosed comparison group, *n*=142). In this way we distinguished a comparison group who displayed autistic-like behaviours but without formal labelling of ASD.

To check the match between the undiagnosed comparison group and the children with ASD diagnosis, a Levene's test was used to compare variance, and where appropriate, a t-test of unequal variance was carried out. Table 1 shows that at 5% levels, children (at age 3-4) who received ASD diagnoses did not significantly differ in autistic trait severity to those in the undiagnosed comparison group, not only in the 'composite ASD score' itself, but also on all of the individual measures of autistic-like behaviours that it contained.

Table 1: Comparison of mean scores of composite ASD trait and the individual autistic traits between samples.

Mean scores

Autistic traits	ASD diagnosis [1]	undiagnosed comparison group [2]	Non autistic study population [3]	[1] vs [2] (*n*) t *p*
Composite ASD trait	3.03	3.32	8.46	(192) 0.70 **.486**
Individual traits within composite score:				
SDQ prosocial	3.28	3.61	7.11	(196) 0.96 **.345**
Child has speech difficulty	1.49	1.33	2.83	(197) 1.23 **.223**
Enjoys pretend games	2.21	2.04	1.22	(197) 1.29 .199
Child has other [a] unusual repetitive behaviour	2.41	2.68	2.94	(196) 1.98 **.051**
Child is afraid of new things-and new situations	1.85	1.99	2.33	(197) 0.96 **.336**

[1]   Children with a clinical ASD diagnosis between age 1 and 12, (*n*=71)

[2]   Undiagnosed comparison group- children in the top 2% of composite autistic trait (severe autistic-type traits as preschoolers) but no ASD diagnosis, (*n*=142)

[3]   All other children in the study population (children other than those in above groups)

[a]   'other' than tic, twitch or rocking behaviours- (previous items in ALSPAC questionnaire)

Both the group of children diagnosed with ASD and those in the comparison group had significantly higher scores in all these traits than the rest of the 'non-autistic' study population as preschoolers, (all $p<.0001$). We did not attempt to match children on traits absent from ASD diagnostic criteria, for example children with ASD diagnosis had somewhat lower academic ability than those in the comparison group at age 4-5, $t(75) = 2.14$, $p = .035$, although baseline academic ability of both diagnosed and undiagnosed comparison groups was considerably poorer than that of the population as a whole on entry to school ($p<.001$ for both). Diagnosed children also displayed more behavioural difficulties overall as measured by the SDQ total difficulties score at age 3-4, $t(87) = 5.52$, $p<.001$. 72% of children in the comparison group received speech and language therapy before age 7 as opposed to 89% of those with ASD diagnoses.

Outcome measures

Selective attrition is a problem common to all longitudinal cohort studies and by the time the children had reached age 13, only around half of the families in the cohort were responding to any one measure. Wolke and colleagues (2009) found a higher proportion of single mothers dropped out of ALSPAC by the time their children were aged 8. Mothers who had larger families, no educational qualifications and financial difficulties were also more likely to drop out. It is not known how this would affect results, although Wolke et al. found aetiological models in ALSPAC were fairly robust to the effects of such attrition.

Outcome measures: Educational provision.

ALSPAC holds educational data on all children attending state schools, (both mainstream and special schools) covering approximately 85% of children in the cohort. These data record whether children are in

receipt of some form of special educational needs (SEN) support within the academic year. In the UK children listed at 'school action' level have an individual education plan which details tailored interventions provided by teachers addressing their identified needs. Children at 'school action plus' have input from agencies outside the school, such as educational psychologists and health services. At the highest level, a 'statement of special educational needs' will require assessment by a medical practitioner (for developmental disorder,) and a multidisciplinary team, including the parents. The school will then have a statutory duty to provide individualised support to the child specified in the statement; in the UK a mainstream or specialist school is normally granted additional funds to employ a teaching assistant to provide one to one educational support for an autistic child. We examined educational records for SEN classifications in the groups when the children were 11-12 years old.

Outcome measures: Social, emotional and academic.

Various measures of the teenagers' behaviour and academic performance were taken when the children were aged between 12 and 16. In tandem with this study, a complementary qualitative study was conducted, examining the experiences of parents with autistic children. The qualitative study revealed that parents of children with an ASD diagnosis were particularly concerned about educational attainment, levels of well being, friendships, self harm and aggression, bullying and school exclusion as their children became young adults (Russell & Norwich, *in press*). These concerns led to the selection of a variety of emotional and behavioural outcome variables of interest, as well as outcomes in autistic-type traits. A full list of these outcome measures together with who reported them (either the child's parent, educational authority or the teenage children themselves) and at what age in the child's life they were reported is given in Table 2.

The outcome measures included measures of social and communication impairment such as the prosocial score from the SDQ, the Social Communication Disorders Checklist (SCDC) (Skuse et al., 1983) and outcomes reported by the teenagers themselves, designed by the ALSPAC team. In the case of the SDQ questionnaire, which had been administered to parents at five time points throughout the children's lives, (at 47 months, 81 months, 115 months, 140 months and 157 months) it was also possible to scrutinize the developmental trajectories of the trait for the diagnosed and undiagnosed groups in detail. Outcome measures of repetitive behaviour and restricted interests included a ritualistic behaviour scale (Cronbach's alpha =0. 71) that we adapted from the Development and Well-Being Assessment, (DAWBA) (Goodman, Ford, Richards, Gatward, & Meltzer, 2000), in addition to further self reported items. We also included measures of mood such as the scale measuring depressive symptoms from the Short Moods and Feelings Questionnaire (SMFQ) (Angold, Costello, & Messer, 1995) and self reported scales of how happy teenagers were at school (Cronbach's alpha =0.81). A scale for coping with new situations was also constructed from existing responses to questions designed by UK Government Department for Children, Schools and Families, (Cronbach's alpha =0. 78) completed by the study children themselves when aged 15 or 16. Statutory Assessment Tests (SATS) documenting the children's academic ability were provided to ALSPAC by the UK Department of Education.

Analysis

To determine whether the children with ASD diagnoses differed significantly from the undiagnosed comparison group in terms of their social and behavioural outcomes as teenagers, t-test comparisons were made. A Levene's test was used to establish whether the groups had equal variance on the outcome measures. Where this was not the case, a t-test of unequal variance was applied. Analysis of variance (ANOVA) was also performed in order to determine whether outcomes for the children with ASD

diagnoses and those in the undiagnosed comparison group differed from those in the general non-autistic population. Where outcome measures were recorded as categorical variables, a chi squared test of association and Fischer's exact test were carried out as appropriate.

Given that IQ has been shown to be a major indicator of outcome for autistic adults, we decided to co-vary for initial academic ability (derived from school admissions tests results aged 4-5) as a proxy for IQ. Separate adjustment was also made for the total behavioural difficulties score taken from the SDQ at 47 months, which includes measures of hyperactivity, conduct problems, emotional difficulties and peer problems. The overall developmental trajectory of the SDQ prosocial score was examined for children in each group.

Results

Social, emotional and behavioural outcomes

Table 2: Differences between mean scores for social, emotional and behavioural outcomes in teenagers – children diagnosed with ASD vs undiagnosed comparison group. High scores reflect increased impairment or undesirable outcome where measure is marked +

age	report by	measure	Mean scores (Standard deviation)			n	T test [1] vs [2] Unadjusted scores	
			children with ASD diagnosis [1]	undiagnosed comparison group [2]	non- autistic population [3]		diff between means [95%CI]	P

Measures of social behaviour and communication

age	report by	measure	children with ASD diagnosis [1]	undiagnosed comparison group [2]	non- autistic population [3]	n	diff between means [95%CI]	P
13	parent	SDQ prosocial score	5.40 (2.7)	6.93 (1.9)	8.23(1.7)	105	1.5 [0.4-2.6]	**.008** [ab]
13	parent	+SCDC score	24.74(6.1)	17.26(5.8)	14.48(3.5)	108	7.4 [4.7-10.1]	**<.0001**[ab]
13	child	Time spent by themselves	8.86(2.5)	8.87(2.3)	9.11(1.8)	70	.01 [-0.6-0.6]	.993
13	child	Time spent with other young people	4.82(1.8)	6.33(1.7)	6.94(1.3)	74	1.5 [0.5-2.5]	**.004** [ab]
13	child	Time spent on computers	5.82(1.28)	6.09(1.58)	5.81(1.48)	74	0.3 [-0.6-1.1]	.532

Repetitive behaviour and restricted interests

age	report by	measure	children with ASD diagnosis [1]	undiagnosed comparison group [2]	non- autistic population [3]	n	diff between means [95%CI]	P
13	parent	Repetitive motor movements	9.21(3.2)	9.25(1.9)	9.91(1.9)	46	0.5[-1.51.5]	.949
13	parent	Repetitive noises/speech	6.53(2.8)	6.16 (1.9)	6.91(1.84)	47	0.4[-1.5-1.7]	.609 [b]
13	parent	+Ritualistic behaviour (DAWBA)	8.03(2.2)	7.11(0.7)	7.07(0.6)	104	0.9[0.5-1.8]	**.039** [a]
14	child	Restricted interests	3.26(0.7)	3.78(0.4)	3.85(0.4)	72	0.5[0.1-0.9]	**.014** [a]
15/16	child	+Copes with unexpected events	20.73(6.4)	16.36(4.6)	16.15(4.2)	66	4.4[1.0-7.7]	**.013** [a]

Measures of mood and feelings

age	report by	measure	children with ASD diagnosis [1]	undiagnosed comparison group [2]	non- autistic population [3]	n	diff between means [95%CI]	P
13	parent	SMFQ Depression Score	33.82(5.4)	35.76(3.7)	36.60(3.3)	106	1.9[-0.2-4.2]	.085 [ab]
13	parent	Self harm	2.79(0.5)	2.95(0.2)	2.98(0.2)	106	0.2[-0.4-0.4]	.105 [a]
13	parent	Displays aggressive behaviour /bullies others	1.17(0.3)	1.33(1.6)	1.13(0.8)	105	0.2[-0.4-0.7]	.570 [b]
13	parent	+Gets Bullied	1.68(0.6)	1.42(0.6)	1.22(0.5)	99	0.3[-0.6-.04]	.089 [ab]
13	parent	Difficulty controlling temper	1.96(0.6)	2.37(0.6)	2.52(0.6)	106	0.4[0.1-0.7]	**.019** [ab]
14	child	Happy at school	15.75(3.2)	17.06(2.5)	16.94(2.2)	44	1.3[-0.8-3.4]	.209
15/16	child	Amount skipped school	1.06(2.4)	1.17(0.6)	1.20(3.9)	75	0.1[-0.2-0.4]	.467
15/16	child	Aggressive behaviour (get in fights)	1.24(0.4) [a]	1.15(0.4)	1.15(0.4) [a]	64	0.09[-0.3-0.1]	.426
15/16	child	Bullied, called names	2.66(1.0)	3.26(0.8)	3.38(0.8)	64	0.59[0.4-1.2]	**.037** [a]

Educational attainment

age	report by	measure	children with ASD diagnosis [1]	undiagnosed comparison group [2]	non- autistic population [3]	n	diff between means [95%CI]	P
13/14	Dept. of education	English	23.72(7.8)	28.51(9.2)	32.80(9.2)	135	4.7[1.7-7.8]	**.002** [ab]
13/14		Maths	23.93(11.5)	32.04(12.65)	36.63(11.6)	135	8.1[3.8-12.4]	**.0004** [ab]
13/14		Science	22.02(10.6)	30.39(10.4)	34.11(9.1)	135	8.4[4.5-12.2]	**<.0001** [ab]

[1]   Children with a clinical ASD diagnosis between age 1 and 12, (*n*=71)

[2]   Undiagnosed comparison group- children in the top 2% of composite autistic trait (severe autistic traits as preschoolers) but no ASD diagnosis, (*n*=142)

[3]   All other children in the study population (children other than those in above groups)

a   Children with ASD diagnosis have significantly worse outcomes at 5% level than in general population [1] vs [3]

b   Children in comparison group diagnosis have significantly worse outcomes at 5% level than in general population [2] vs [3]

Results of social, emotional and behavioural outcomes in teenagers with an ASD diagnosis are given in Table 2. Social communication skills as measured by the SDQ and SCDC scores were significantly worse for diagnosed teenagers than in the undiagnosed comparison group. Nevertheless, teenagers reported that having close friends mattered to them just as much whether they were diagnosed or in the undiagnosed comparison group, (χ^2 =1.89, *p*=.168). In the ALSPAC population, 34 children were diagnosed with ASD between the ages of 1-6. We repeated the analysis restricting the diagnosed sample to these 34 children. This was in order to compare children who were diagnosed in roughly the same age ranges as the age range in which undiagnosed children had comparable autistic-like traits. Mean outcomes still diverged significantly between undiagnosed and diagnosed children in both SDQ, [*t* (14) =2.75, *p*=.015] and SCDC measures, [*t* (11) = 4.36, *p*=.013].

Teenagers with an ASD diagnosis were also reported as having more ritualistic behaviours than their undiagnosed counterparts, (Table 2) and reported themselves as having more restricted interests, although levels of more basic repetitive behaviours – motor movements and vocalisations-were similar.

Table 2 also shows that of the 10 measures of mood and personality that were compared, none differed significantly between the groups at the 1% level. Children with an ASD diagnosis did not skip school more, and were less likely to get into fights or display aggression. However, children with an ASD diagnosis reported that they were more likely to be bullied, and parents reported that the diagnosed children found it harder to control their tempers than children in the comparison group. None of the children in the study groups had been permanently excluded or suspended by age 16.

Co-varying for academic ability revealed that outcomes in most autistic-type behaviours (social cognition measured by the SCDC score, time spent with young people, coping with the unexpected and restricted interests) still differed significantly between diagnosed and undiagnosed comparison groups. However none of the significant differences between these groups in academic outcomes persisted. Adjustment for total behavioural difficulties at age 3-4 accounted for differences between the groups in all measures except autistic outcomes; that is, all measures of social communication and repetitive behaviours that had differed without this adjustment, remained significantly different (Table 3).

Table 3: Differences between mean scores for children diagnosed with ASD vs undiagnosed comparison group adjusted for baseline academic ability (aged 4-5) and total behavioural difficulties (aged 3-4). High scores reflect increased impairment or undesirable outcome where measure is marked +

			Adjusted for entry level academic ability (age 4-5). T test [2] vs [1]			Adjusted for baseline overall behavioural difficulties (SDQ total difficulties 47m). T test [1] vs [2]		
age	report by	measure	n	mean [2] [95% CI], mean [1] [95% CI]	p	n	mean [2] [95% CI], mean [1] [95% CI]	p
Measures of social behaviour and communication								
13	parent	SDQ prosocial score	66	7.0 [6.4-7.6], 6.0 [5.1-7.0]	.105 [ab]	105	6.8 [6.3-7.3], 5.6 [4.8 -6.4]	**.013**[ab]
13	parent	+SCDC score	70	16.6[15.0-18.1], 23.2[20.8-25.7]	**<.001** [ab]	107	18.3[17.0-19.5], 24.1[21.9-26.2]	**<.001**[ab]
13	child	Time spent by themselves	47	8.8 [8.0-9.6], 8.3[7.0-9.7]	.551	70	8.8 [8.1-9.5], 8.9 [7.7-10.1]	.931
13	child	Time spent with other young people	48	6.4 [5.9-7.0], 4.5 [3.7-5.4]	**.001** [a]	74	6.3 [5.8-6.7], 4.8[4.0-5.6]	**.003** [ab]
13	child	Time spent on computers	48	5.8[5.2-6.4], 6.0 [5.1-6.1]	.765	74	6.3 [5.8- 6.7], 5.8 [5.1- 6.5]	.307
Repetitive behaviour and restricted interests								
13	parent	Repetitive motor movements	25	9.7 [8.5-10.9], 8.4 [6.8-10.0]	.182 [a]	45	9.3 [8.3-10.3], 8.9[7.5-10.2]	.617
13	parent	Repetitive noises/speech	26	6.4 [5.3-7.5], 7.0 [5.3-8.6]	.573	46	6.2 [5.3-7.1], 6.4 [5.1-7.6]	.818
13	parent	+Ritualistic behaviour (DAWBA)	67	7.1[6.8-7.5], 7.4 [6.9-7.9]	.313 [a]	103	7.1 [6.8-7.5], 8.1[7.6-8.6]	**.003** [a]
14	child	Restricted interests	48	3.8 [3.6-3.9], 3.3 [3.0-3.6]	**.008** [a]	72	3.8 [3.6-3.9], 3.3 [3.0-3.5]	**.001** [a]
15/16	child	+Copes with unexpected events	45	16.6[14.3-18.9], 21.8[18.7- 24.9]	**.008** [a]	66	16.4[14.7-18.1], 20.7[18.2-23.2]	**.007** [a]
Measures of mood and feelings								
13	parent	SMFQ Depression Score	68	36.4 [35.2-37.6], 33.2[31.3-35.1]	**.007** [a]	106	35.7[34.7-36.7], 34.0 [32.4-35.6]	.084 [a]
13	parent	Self harm	67	3.0 [2.9-3.1], 2.8 [2.7-3.0]	.058 [a]	106	2.9 [2.9-3.0], 2.8 [2.7–3.0]	.083 [a]
13	parent	aggressive behaviour /bullies others	67	1.4 [1.0- 1.8], 1.2 [0.6-1.9]	.675	105	1.3 [1.0-1.7], 1.1 [0.7-1.7]	.515 [a]
13	parent	+Gets Bullied	63	1.4 [1.2- 1.6], 1.7 [1.4- 2.1]	.123 [a b]	99	1.4 [1.3-1.6], 1.7 [1.4- 1.9]	.135 [ab]
13	parent	Difficulty controlling temper	67	2.4 [2.2 -2.6], 2.0 [1.7- 2.3]	**.042**[a]	106	2.3[2.1 - 2.4], 2.0[1. 8- 2.3]	.068 [a]
14	child	Happy at school	29	17.2[16.0-18.4], 16.2[14.4- 18.1]	.354	43	16.8 [15.8-17.8], 15.8[13.8-17.8]	.361
15/16	child	Amount skipped school	49	1.1[1.0- 1.2], 1.1[0.9-1.3]	.769	75	1.2[1.0-1.4], 1.1[0.8- 1.3]	.358
15/16	child	Aggressive behaviour (get in fights)	44	1.1 [0.9-1.2], 1.1 [0.9-1.3]	.581	64	1.2 [1.0-1.3], 1.2 [1.0-1.4]	.532
15/16	child	Bullied, called names	43	3.3 [3.0- 3.7], 2.8 [2.4- 3.3]	.090 [a]	64	3.2 [2.9-3.5], 2.7 [2.3-3.1]	.071 [a]
Educational attainment								
13/14	Dept. of education	English	93	27[26- 30], 26 [23-30]	.439	125	28.3[26.4- 30.1], 25.5[22.4 -28.7]	.148[ab]
13/14		Maths	93	31.9[29.5-34.2], 29.1[25.1- 33.1]	.242	124	31.2[28.7- 33.7], 28.2 [23.9-32.5]	.246 [ab]
13/14		Science	93	29.1 [26.9-31.3] , 25.5 [21.7 29.2]	.096 [a]	124	29.7 [27.7-31.8], 26.4 [22.9-30.0]	.121 [ab]

[1]   Children with a clinical ASD diagnosis between age 1 and 12, (*n*=71)

[2]   Undiagnosed comparison group- children in the top 2% of composite autistic trait (severe autistic traits as preschoolers) but no ASD diagnosis, (*n*=142)

[3]   All other children in the study population (children other than those in above groups)

a   Children with ASD diagnosis have significantly worse outcomes at 5% level than in general   population [1] vs [3]

b   Children in comparison group diagnosis have significantly worse outcomes at 5% level than in general population [2] vs [3]

Overall, children who had received an ASD diagnosis were considerably more impaired as teenagers on a range of measures of autistic-like behaviour than those without diagnosis who displayed autistic-like behaviours when young. Other differences in outcome; 'non-autistic' difficulties at adolescence, were largely accounted for by adjusting for the children's entry level academic ability and other behavioural difficulties as preschoolers.

Special educational provision

Of the 142 children we identified above with autistic-like symptoms as preschoolers but no ASD diagnosis, 79 had had no special educational provision other than within-school provision and were not referred to any external agencies outside school when 11-12 years old (Table 4). These 'low-SEN' provision children were not listed as being referred to service providers such as educational psychologists, speech therapists, specialised mental health services or paediatricians as they began secondary school; the majority were therefore unlikely to have been diagnosed with any alternative behavioural disorder, although they may have received school based support of high quality. None of them had a diagnosis of ASD by age 12.

Table 4: Number of children with each level of additional school provision for special needs at age 11 or 12 for children in the ALSPAC cohort who display severe autistic traits as preschoolers (aged 3-4).

Level of additional school provision for special needs	ASD Diagnosis	Not diagnosed with ASD but severe ASD traits as preschoolers	Entire study population without or without ASD traits
	n=71	*n*=142	*n*=13944
None	1	63 [a]	9851
school action	0	16 [a]	1235
school action plus (external agency involvement)	3	2	456
statement of special needs	57	35	378
SEN status not listed	10	26	2024

[a] Low-SEN group: No special educational provision other than within school or external health referrals.

Of the 70 children with ASD diagnosis and high SEN provision, the majority (68%) were categorised as having 'communication' difficulties as their primary problem. Only 32% of the children in the undiagnosed group of 142 were identified with high SEN provision aged 11-12, and most of these (65%) were described as primarily having 'cognitive' difficulties.

Analysis of outcomes limiting the comparison group to the 79 children in the low-SEN provision group (identified in Table 4) showed that all measures reported as differing significantly between the diagnosed and comparison groups in Table 2 again differed significantly.

The developmental trajectory of prosocial behaviour

We compared the children's differing trajectories on the SDQ prosocial trait. The mean scores for each group at both the initial measurement at age 4 and the subsequent measurements taken throughout the children's lives are illustrated in Figure 2. Adjustments for the varying participation of children at different ages did not substantially change the mean scores. The figure shows how small non significant differences between the case and comparison groups increased dramatically in the preschool and early primary years. But after 6 years, the trajectories were similar. By the age of 13, SDQ prosocial scores and SCDC social communication scores in the undiagnosed sample were approaching those of the general population.

Figure 2: The developmental trajectory of SDQ prosocial trait in children by each sub group.

increasing scores = better prosocial skills.

Discussion

Substantive findings

Observational studies such as ours cannot determine causality, but we utilised a large contemporary

cohort, with a wealth of prospectively collected data, and linked health and educational records. A further

strength was the longitudinal design and repeated SDQ measures over a ten year period. Children who

had been diagnosed with ASD (all but one of whom received high special educational needs support aged

12) were more impaired in social communication and demonstrated more restricted interests and

ritualistic behaviours as teenagers than those without a diagnosis. There were few differences in moods and feelings of teenagers, but those teenagers with a diagnosis found it harder to control their tempers, and had poor academic outcomes when initial academic ability was not taken into account- suggesting perhaps non autistic behaviours such as disruptive behaviour and low academic ability may also lead to referral (Todman, Justice & Swanson, 1991). Previously we found few socio-economic differences between background factors of children with ASD diagnosis and the comparison group, (Russell et al. 2010). Unsurprisingly it is baseline academic ability (aged 4-5) that appears to be the primary cause of differences in academic outcomes. However baseline academic ability did not alter differences in autistic type outcomes such as social cognition or restricted interests. Similarly, non-autistic type behavioural difficulties in preschool children accounted for much of the difference in emotional outcomes, but not differences in autistic-type outcomes. Even adjusting for the influence of academic ability at baseline, and other types of behavioural difficulties, outcomes in measures of autistic symptoms were worse for diagnosed children compared to those in the comparison group.

As other studies have demonstrated that children with worse levels of impairment are more likely to reach specialist clinics (Angold et al., 2000) it is not unexpected that children diagnosed with ASD have worse outcomes - this suggests services were picking up the most impaired children. It is also possible that despite diagnosis and subsequent service contact there is less overall improvement in this group than those with comparable skills at preschool. On the other hand, it is not known what the trajectories of the diagnosed children would have looked like without additional service contact engendered by diagnosis- it is possible that diagnosed children may have exhibited further deterioration which intervention had ameliorated. Children with more severe difficulties may be more likely to receive a diagnosis, but

recognition may not guarantee access to effective intervention even where these exist (Feinberg & Vacca, 2000).

Limitations

Although our study attempted to control for preschool levels of behavioural impairment, some have argued that controls must also be matched in terms of pre-treatment clinical trajectory (Angold et al., 2000; Lambert & Bickman, 2004). It proved impossible to match ASD cases with the comparison group on all behavioural traits; ALSPAC was not set up to look at ASD therefore we did not have access to a well validated research tool for assessing ASD. However we argue that comparing outcomes in children with those who exhibited autistic-like behaviours aged 3-4, including social impairment, speech difficulties and repetitive behaviours provides a more subtle insight than simply comparing outcomes with non-autistic populations.

Another limitation is that parents or children themselves may over or under estimate symptoms. Furthermore, the exact nature of interventions given to children was not specified in our study. As a group, children with ASD diagnosis were likely to have received more individualised support within school, as well as from health /educational services but exact details of educational and health interventions were not available.

Despite such limitations, ours is the first study that we know of to look specifically at outcomes for children diagnosed with ASD in their communities that attempts to control for initial symptom severity. Our findings suggest clinical and educational services are picking up the most impaired children, as might be predicted (Angold et al., 2000). It is not clear from the data that differences in outcome are driven by

management, or reasons other than autism. It has been suggested that effects of conspicuous identification and remediation may selectively impede development (Scheff, 1974). It is beyond the scope of this article to ascertain if this is the case.

Clinical implications

Our findings contrast with clinical guidelines currently recommending early identification below 2 years of age (Crais, Watson, Baranek, & Reznick, 2006) and support studies that suggest the age at which ASD can be reliably diagnosed is unclear (Stone et al., 1999). Our results imply that behavioural traits increase in variability and range as children develop as demonstrated elsewhere (Landa, 2008). Thus it is difficult, for example, to distinguish between a two year old who cannot speak because s/he has an ASD from a two year old who cannot speak because he is a slow developer- autistic type symptoms in a 2-year-old will differ from more prototypic symptoms at age 5 (Charman et al., 2005; Charman & Baird, 2002). One recent study found approximately 30% of children diagnosed with ASD at age 2 failed to reach diagnostic criteria at age 4 (Turner & Stone, 2007). This group was associated with milder symptoms and younger diagnosis. Although our study provides no concrete evidence of any negative effect from diagnosis, perhaps a 'precautionary principle' could be applied.

Conclusion

There are various explanations of the findings. Children may be referred because of concerns including behaviours we did not measure. Early educational attainment and other behavioural difficulties did account for much of the difference between diagnosed and undiagnosed children in non-autistic outcome behaviours and academic achievement. It also seems likely that some of those exhibiting autistic-type behaviours at age 3 years may 'grow out' of the ASD features, while others do not. A third possibility is

that ASD diagnosis biases parental reports, or that diagnosis really could be disadvantageous, indeed, interaction between several of these factors may be at play.

The last ASD diagnoses in the data that were available were made in 2005, and since then age of diagnosis has fallen and the number of children ascribed an ASD diagnosis has increased. In the light of our findings we would advocate an examination of outcomes in a larger, more recent cohort, controlling for IQ and using repeated measures of autistic-like behaviour throughout childhood, so that issues of causality could be examined more carefully. Further innovation and research into how to support children with these incapacitating difficulties is important, regardless of whether a diagnostic label is applied or not.

Acknowledgements

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Key points

- Many children who have been documented by researchers as having symptoms of ASD have never received a clinical diagnosis.

- Children with ASD diagnoses had poorer outcomes than those with autistic symptoms as preschoolers but without clinical diagnosis. This suggests children with greater impairments are more likely to be referred to services as they mature.

- Amongst children who exhibit autistic type symptoms aged 3-4 it may be difficult to distinguish between those that will develop lifelong impairment and those that have improving prognosis.

'Article 4'

The development of prosocial behavior in children with autism spectrum disorders: Before and after diagnosis.

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Running head: **Development of social behavior in children diagnosed with ASD.**

Keywords: Social behavior**,** ALSPAC, strengths and difficulties, longitudinal cohort

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Abstract

Impaired social skills are core symptoms for children with an autism spectrum disorder (ASD). We plotted the developmental trajectory of the SDQ prosocial trait before and after diagnosis in a sample of 57 children who were enrolled in a longitudinal birth cohort study in the UK over a 10 year period. Multi factorial fixed effect modelling revealed that the developmental trajectory of this measure of prosocial behavior was not significantly altered by ASD diagnosis. Further analysis was conducted on 33 of these children who had both pre and post diagnosis information, and the same result obtained. Prosocial behaviors may be resistant to the typical services triggered by a clinical ASD diagnosis that have been delivered in the recent past.

Introduction

Impaired social skills are core symptoms for children with an autism spectrum disorder (Bailey, Phillips, & Rutter,1996; World Health Organisation, 1992). Individual symptoms of autism occur in the general population and appear not to associate highly (Happé, Ronald, & Plomin, 2006) without a clear demarcation separating pathological severity from common traits (Constantino & Todd, 2000). Researchers have argued that as core symptoms of autism spectrum disorders (ASD) may have distinct aetiologies (Ronald et al., 2006), it may be fruitful to focus on autistic symptoms separately (London, 2007).

Charman and colleagues (2005) have suggested a dimensional approach where changes in separate behavioral domains can be considered over time. Our study concentrates on the longitudinal development of one core autistic behavior; a measure of prosocial behavior. We analysed the developmental trajectory of this trait in children who were part of an ongoing UK birth cohort study; the Avon Longitudinal Study of Parents and Children (ALSPAC), using the Strengths and Difficulties Questionnaire prosocial score (Goodman, 1997, 1999). This has been shown to be strongly associated with an autism diagnosis in the ALSPAC cohort (Russell, Ford, Steer, & Golding, 2010), and was repeatedly recorded throughout the lives of the children in the study.

In Europe and the North American countries, a clinical diagnosis of ASD usually triggers access to many services for the child and his/her family. A clinical ASD diagnosis is often framed as a way for parents and children to access important health and educational services and other forms of support (e.g. Disability Resource Centre, 2011). Mansell and Morris (2004) provide a useful summary of typical

interventions and services that are adopted in the UK after an ASD diagnosis is applied. These include extra educational support in the classroom or special schooling, parents' support groups, speech and language therapists, educational psychologists, psychiatrists, respite care and focussed access to information such as books and academic journals. Special units and schools were rated as the most useful source of support.

International health guidelines recommend prompt identification and diagnosis of ASD so that parents and children can access such specialist services (e.g. Filipek et al., 2000). As many researchers have concluded that despite patchy evidence, early intervention for ASD is warranted (Guralnick, 1996; Howlin, 1998; Howlin, Magiati, & Charman, 2009), we would hope that children diagnosed with ASD and thus receiving additional help from health, education and other services might show improved functioning in core autistic symptoms, such as prosocial behaviors.

Angold and colleagues (2000) note the dearth of studies that have looked at the overall effectiveness of the parcel of services currently used for childhood disorders in community based settings, as opposed to efficacy studies looking at particular interventions including those using randomised controlled trials. Mesibov and Shea (2009) argue for a more flexible approach to interpreting evidence base for autism interventions: one that does not rely solely on randomised controlled trials. In effectiveness research, as Kelley, Nixon and Bickman point out (1999, p 471), the "intervention" under scrutiny is likely to be a complex package of several services, and this will make it more difficult to pinpoint the essential intervention components.

Historically, proponents of 'labeling theory' have argued that a clinical diagnosis might lead to worse outcomes (Scheff, 1974). Scheff suggested that diagnosis of mental disorders could lead to damaging preconceptions in others as well as self-fulfilling prophecies. Autistic children might 'inhabit the identities they have been ascribed' as Nadesan puts it (2005, p150). There is little, if any, empirical evidence to support such claims relating to autism. Sayal and colleagues (2010) found that children identified to teachers as having severe hyperactivity symptoms were rated as having worse outcomes several years later, compared to a group with equivalent symptoms who were not conspicuously identified. Because of this, we did not discount the possibility that prosocial behavior in children identified by a clinical ASD diagnosis might appear to deteriorate after identification.

Our hypothesis was therefore that an ASD diagnosis, and subsequent service contact would lead to a change (we hoped an improvement) in the development of prosocial behavior as children matured. Our aim, then, was to plot the developmental trajectory of autistic children's prosocial behavior both before and after the diagnosis had been applied, in order to determine whether additional service contact and support engendered by clinical diagnosis might lead to an improvement (or deterioration) in prosocial scores as reported by parents.

Wispé (1972) first proposed the term "prosocial," meaning to create an antonym to the term "antisocial". There are no studies, that we know of, following the developmental trajectory of children's prosocial behavior specifically for a sample of children diagnosed with ASD.

Methods

Sample

The Avon Longitudinal Study of Parents and Children (ALSPAC) is an ongoing cohort study based in South West England, following a cohort of over 14,000 children born between 1991 and 1993. The social and demographic features of the ALSPAC cohort were representative of the overall population in the UK as measured by the 1991 UK national census (Golding, Pembrey, & Jones, 2001). Demographic details of the children in the sample, compared to those in the UK population as a whole, are tabulated by the ALSPAC website together with detailed methodological information about the cohort and the measures taken throughout the lifetime of the children enrolled in the study (www.bristol.ac.uk/alspac). Our analysis was restricted to children in the ALSPAC cohort that survived to one year (n=13,988).

The medical records of 71 children who were part of this cohort showed that they had been formally diagnosed with ASD by the time they were aged 12. These records were checked independently of other measures taken by the cohort study by a research team that included an experienced pediatrician. Details of the methodology used to identify those children with clinical ASD diagnoses are described by Williams, Thomas, Sidebottom and Emond (2008). The clinical diagnoses recorded in the children's medical records were made in the children's communities, blind to the measures taken by ALSPAC, by a clinician, (either a child psychiatrist or community paediatrician,) with support from multidisciplinary assessment teams. The age range in which children were formally diagnosed with ASD is shown in Figure 1.

No data were available on IQ for individual children, but educational records at age 12 were obtained for 61 of the 71 children. These revealed that all but one with a clinical ASD diagnosis had support and

intervention at school tailored to the child's individual needs as well as external (i.e. outside school) contact with specialist health and/or educational services. The vast majority of these children (all but three) had a 'statement of special needs' at school by age 12. In the UK, this means the child's school has a statutory duty to provide individualised support to the child. The details of support are specified in the statement. In the UK, a mainstream or specialist school is normally granted additional funds to employ a teaching assistant to provide one to one educational support for an autistic child. However, other than this broad picture of a clinical diagnosis accompanied by specialist health and educational support, we did not have access to data on specific interventions that were administered to individual children. It is therefore beyond the scope of this study to ascertain the merits of particular interventions for ASD.

Measures

Parents of children enrolled in the ALSPAC cohort had repeatedly reported on the prosocial behavior of their children at six time points throughout the children's lives, (at 47, 81, 97, 115, 140 and 157 months respectively,) using the Strengths and Difficulties Questionnaire (SDQ) (Goodman, 1997; 1999). The prosocial subscale of the SDQ is composed of five items, namely:

The child ….

1. is considerate of other people's feelings;

2. shares readily with other children (treats, toys, pencils etc.);

3. is helpful if someone is hurt, upset or feeling ill;

4. is kind to younger children;

5. often volunteers to help others (parents, teachers, other children).

These items each were answered by parental report, either *not true/somewhat true* or *certainly true*.

Questionnaires were sent to each enrolled family by post. The SDQ questionnaires were nested within a

series of other questions on the health and development of the study child. Although the questionnaires were aimed at 'parents' (either parent was instructed to answer), it was the children's mothers who completed the questionnaires in over 90% of cases at each of the six time points for which SDQ scores were recorded.

The responses were used to produce a 'prosocial' score which together with the other scales in the SDQ have been widely used in research in childhood psychopathology. Children are scored 0 if the response is '*not true*', 1 if '*somewhat true*' or 2 if '*certainly true*'. Hence a score of 1 indicates very poor prosocial behavior and a score of 10 represents highly prosocial behavior. The mean prosocial scores for children in the ALSPAC population (*n*=4924) varied between 7.0 and 8.3 at each time point, but for the children diagnosed with ASD the mean prosocial scores were much lower, varying between 4 and 5.7 as the children matured. Muris, Meesters and Van den Berg (2003) note that the SDQ scores have good internal consistency, test-retest stability, and concurrent validity of the SDQ is good; that is, its scores correlate in a theoretically meaningful way with other measures of psychopathology.
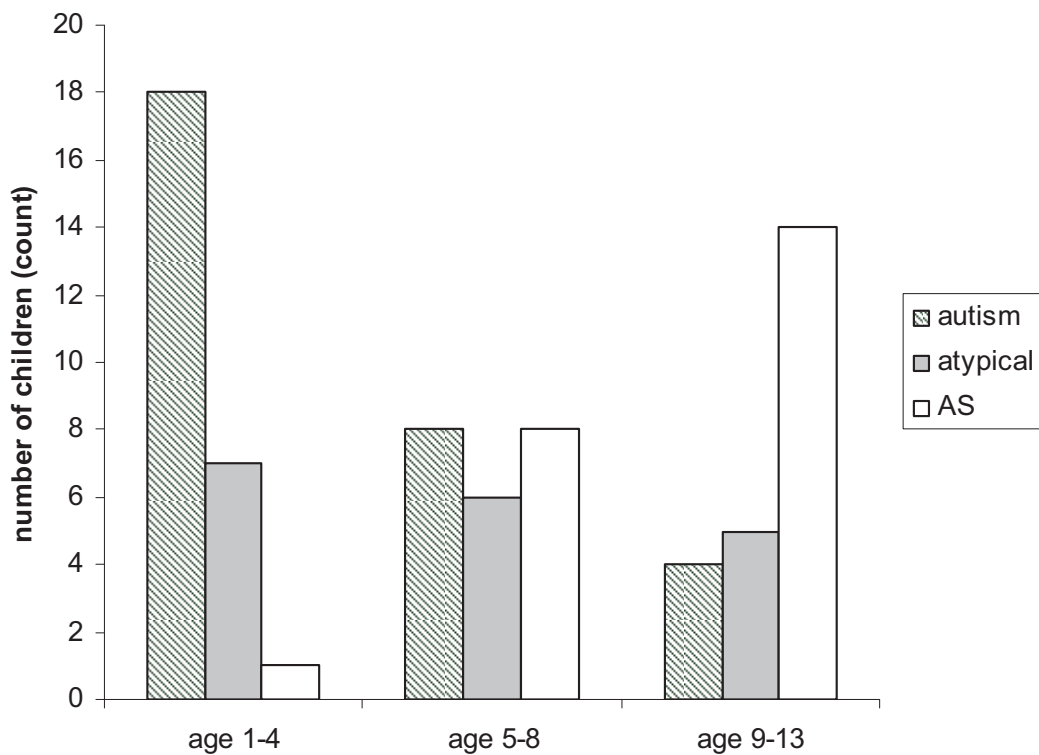
Analysis

Analysis examined the impact of diagnosis by calculating the difference between the age at SDQ assessment and the age of diagnosis. This was a retrospective analysis. Age difference was categorised into two year intervals with open-ended categories of more than four years before and more than six years after diagnosis.

We used a multi-factorial model to determine whether there was a significant change to the developmental trajectory of prosocial skills before or after diagnosis. Due to the varying participation of children at each age of SDQ assessment, all analyses were adjusted for individual variation using fixed

effect estimation. Thus variation in prosocial behaviors between children who were more severely impaired and those who were less impaired was taken into account in the model. A breakdown of the ALSPAC children who received a clinical ASD diagnosis into the diagnostic sub categories shows that the majority of children with classical childhood autism were diagnosed before starting school (mean age diagnosis = 4.8 years), whereas those diagnosed with Asperger's syndrome (AS) were generally identified much later (mean age diagnosis = 8.7 years) (Figure 1).

Figure 1: Sub condition of ASD by age of diagnosis in the ALSPAC cohort.

As prosocial scores tend to improve with age, analyses were also adjusted for age of assessment. SDQ scores were considered continuous and analysed by analysis of variance. We compared the development of social skills as a function of time before or after diagnosis: differences between the trajectory of the linear trend both before and after diagnosis.

About a quarter of the children had been diagnosed, mostly with classical autism, before the first SDQ measures were taken (Figure 1), in these cases the SDQ data were only recorded after ASD diagnosis. To investigate the robustness of the adjustment for individual variation, the analysis was repeated, omitting children who did not have both 'before' *and* 'after' diagnosis scores. This analysis may have greater statistical power, much as a paired repeat measure t-test is more powerful than an independent sample t test. However, this advantage may be offset, partially or totally, by the reduced sample size. In all, 57 children diagnosed with ASD had prosocial scores for at least one time point between 47 and 157m. Six measurements of the prosocial score were made in total, an average of 4.3 assessments were made by parental report and returned to ALSPAC for each of the diagnosed children out of the theoretical maximum of 6 assessments. When restricting to children with both pre and post diagnosis data, 33 children were available with an average of 5.0 assessments.

In addition, we investigated the impact of diagnosis on the prosocial scores by considering the different trajectories in scores between groups of children diagnosed early in life and those diagnosed later: (age of diagnosis <80m, or 80+ m). Here the hypothesis was that the difference between these groups would diminish as the impact of intervention took effect for the early group but subsequently would stabilise or
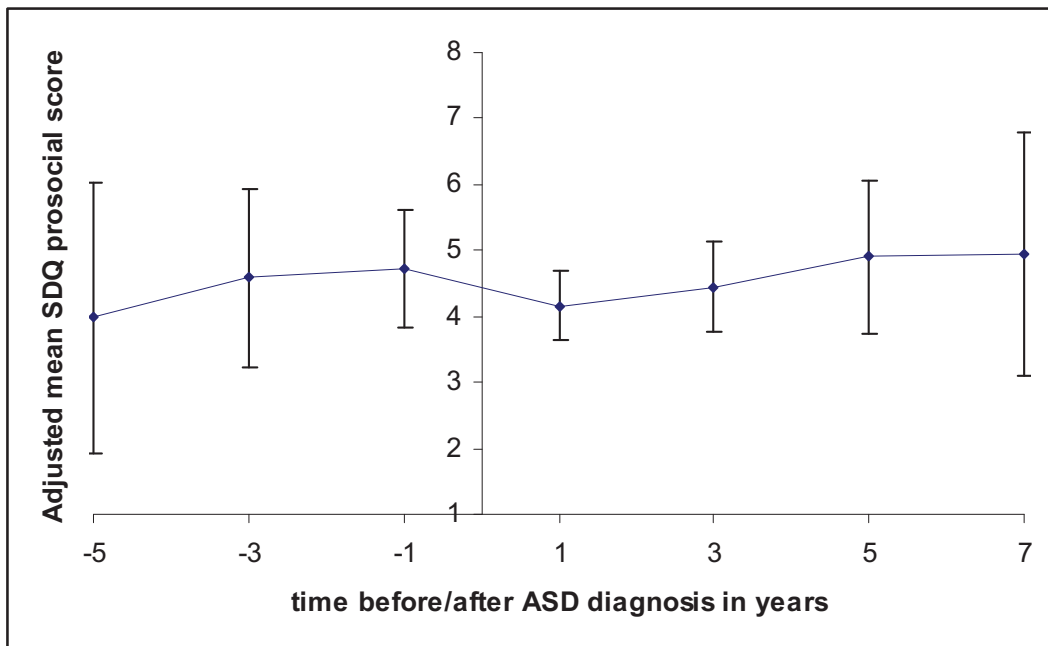
even increase as the later group benefitted from intervention. This would be reflected in an age of SDQ assessment by diagnosis group interaction.

Ethical approval for the wider study was obtained from the ALSPAC Law and Ethics Committee and the Local University Research Ethics Committee.

Results

The developmental trajectory of the SDQ as a function of time before and after ASD diagnosis is shown in Figure 2. The data for age of diagnosis spanned 10 years in total, and the SDQ questionnaires were administered over a 9 year period within this. The average age of the children at each data point in Figure 2 increased steadily from the first data point before diagnosis (average age when measured = 5 years) to the last data point after diagnosis (average age when measured = 11 years) as might be expected. Figure 2 therefore provides us with the signature of the diagnosis on the SDQ prosocial trait- how ASD diagnosis impacts the SDQ trait with time, with adjustment for improvement we see occurring naturally with age in this population, and crucially, adjustment for individual variation of prosocial behavior between children in this heterogeneous sample.

Figure 2: The developmental trajectory of the SDQ prosocial trait before and after ASD diagnosis: adjusted for improvement with age and variation in sample. (N at each data point varies between 24 and 49).
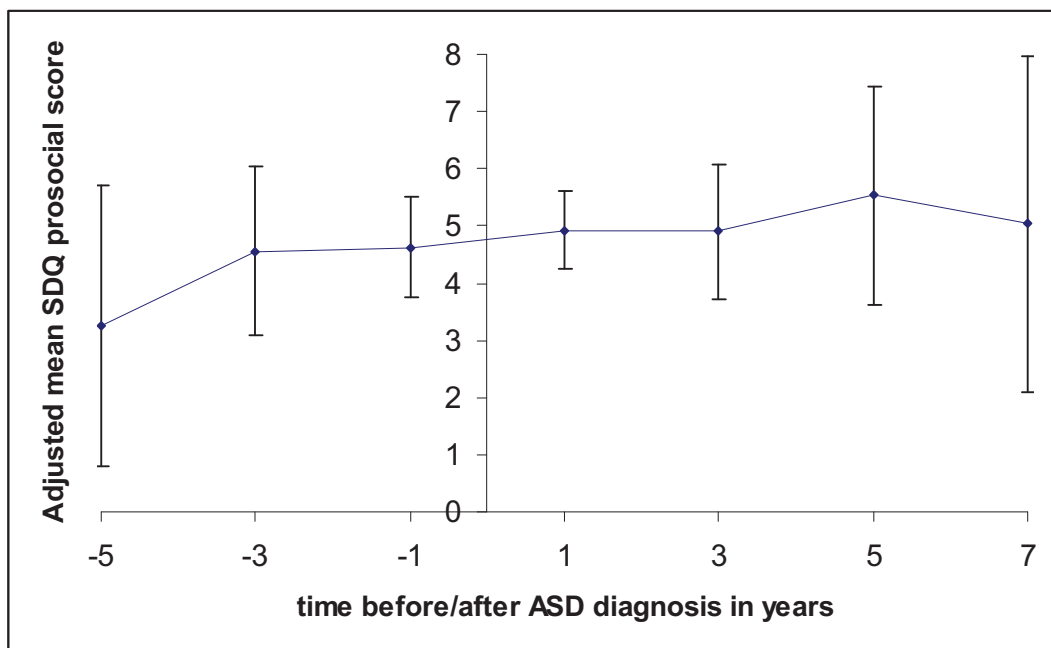

Multi factorial modelling showed that there is statistically no difference between the scores of children on the prosocial trait before or after diagnosis $F(1,177)=0.03$, $p=.857$. Furthermore, there was no evidence of different trajectories for the development of prosocial behavior before of after diagnosis, $F(1,182)=0.08$, $p=.778$. The change in scores before diagnosis was therefore similar to the change after.

Figure 3 illustrates the second analysis, which only considered children with scores both before and after diagnosis. This again shows the trajectory of the prosocial trait both before and after diagnosis, for this smaller group. Once more, the multi factorial model did not uncover any statistical difference between the

scores of children on the prosocial trait before or after diagnosis F(1,122)=0.43 p=.512, neither was the

trajectory of development of social skills significantly different before of after diagnosis in this sample,

F(1,125)=2.03 p=.156.

Figure 3: The developmental trajectory of the SDQ prosocial trait before and after ASD diagnosis: adjusted for improvement with age and variation in sample. (N at each data point varies between 36 and 12). Limited to children with before and after diagnosis scores.

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Both sets of analyses showed the same overall pattern of results, i.e. no difference in the average scores or

between the developmental trajectory of prosocial scores before or after diagnosis, indicating a degree of

reliability in the result.

According to these data, then, diagnosis and subsequent interventions do not appear to improve prosocial scores for children diagnosed with ASD as reported by parents over and above the improvements associated with age. We cannot dismiss the null hypothesis that diagnosis and subsequent service contact had no impact on the development of prosocial scores.

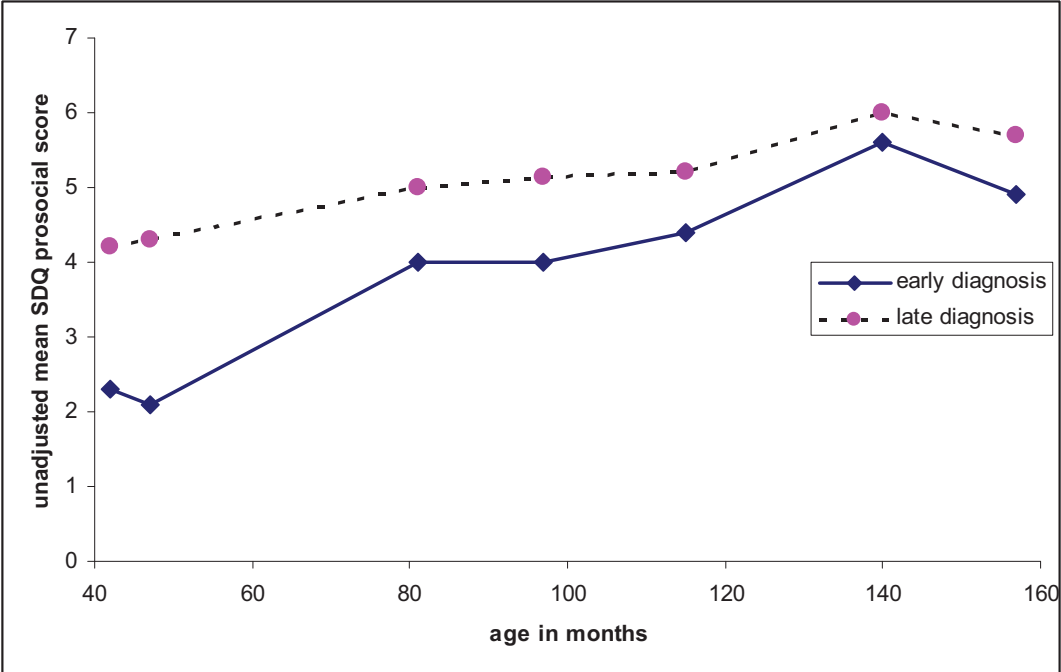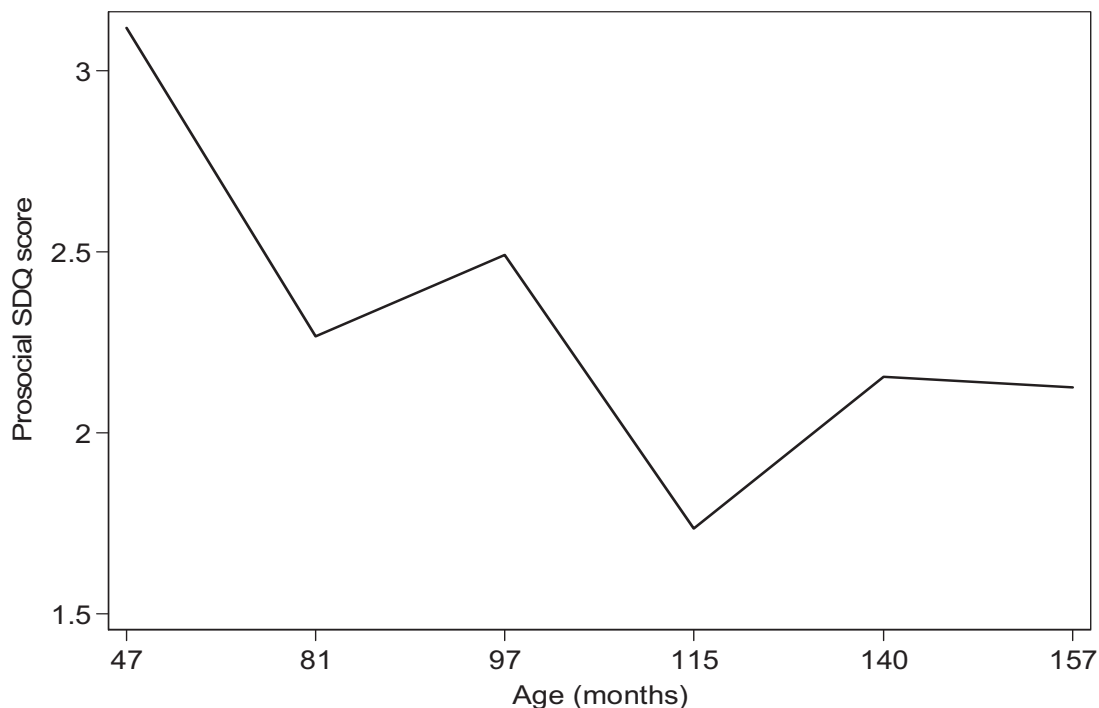*Figure 4: The trajectories of early and late diagnosed groups. Early age of diagnosis <80m, or late, 80+ m.*

Figure 5: The difference between trajectories of early and late diagnosed groups.

There were indications of a global trend. In both analyses, the lowest scores were observed for more than 4 years before diagnosis and a tendency for the highest scores to be achieved some years after diagnosis. These changes were equivalent to about one or two SDQ points. An analysis of early/late diagnosis groups supported this pattern (see Figures 4 and 5). Figure 4 illustrates the trajectories of mean prosocial scores in each group with age, (not adjusted for expected improvement with age), whereas Figure 5 depicts the differences between these groups. Although the difference in scores between these groups diminished by 1 SDQ point as the social skills of the early group improved there was a suggestion that the difference increased after 115m, however the age of SDQ assessment by diagnosis group was non-significant, $p=.439$. The median age of diagnosis for the late diagnosis group was 114m, which corresponds to the turning point. It is important to note, however that this effect was consistent with

random variation. In addition, these improvements, if they exist, appeared to start prior to diagnosis contrary to expectations. Whether the small sample size and the possibility of interventions not dependent of a formal diagnosis can explain these aspects of the results remains unclear.

Discussion

Children with autistic symptoms exhibit abnormal or impaired development in social interaction (WHO, 1992). Children with autistic symptoms do have impaired prosocial behaviors (Russell et al., 2010), but simply obtaining a diagnosis of ASD may not impact on the trajectory of these traits. Overall, this study provides no conclusive evidence either that prosocial behaviors are adversely affected by diagnosis or that diagnosis and subsequent interventions have led to an improvement in the behavior. Instead, the results indicate diagnosis and subsequent service contact may have had a very limited impact on the development of the prosocial trait.

Prosocial behavior may be difficult to remediate and change, but perhaps researchers have not yet identified appropriate interventions to develop these skills in children with ASD. Some promising recent studies have shown core social behaviors may be responsive to intervention (Dawson et al., 2009, Landa, Holman, O'Neill, & Stuart, 2011). The children in our study were diagnosed some time ago, (between 1994 and 2005) and so this study may not reflect the effect of current practice. Service contact may not have targeted prosocial behaviors, although as a core autism symptom, improving social development would fall into the definition of a goal of treatment. As outlined by the Committee on Children with Disabilities (2001), the aim of treatment for ASD is to improve the overall functional status of the child by promoting the development of communication, social, adaptive, behavioural and academic skills as well as lessening maladaptive and repetitive behaviours.

There is an important distinction in developmental psychology to make between traits and states. A trait is described as a more enduring characteristic than a state. A state is more transient, often triggering onset of specific symptoms. Bentall (2004) points out that psychological characteristics probably vary from being state like and changeable to being trait like and immutable. Our results might be interpreted as evidence that prosocial behaviors in autistic children are more stable; i.e. less state-like than trait-like. Such an interpretation would tend to point to biomedical, (neurological and genetic) underpinnings to ASD. This is in accord with the evidence for the high levels of heritability reported, (Szatmari & Jones, 2007) and mixed evidence for structural differences in the neurobiology of people with ASD (see Penn, 2006 for a review).

If a primary goals of treatment are to maximize the child's functional independence and quality of life by minimizing the impact of core autism spectrum disorder features, (Myers & Johnson, 2007), it is also possible that practitioners were simply not using interventions that were effectively treating prosocial behaviors. Because of this possibility, treatment guidelines are issued by national health bodies (in the UK, the National Institute of Clinical Excellence) that attempt to reflect the findings of efficacy studies. However there are often difficulties in transferring interventions that are efficacious in randomised trials on restricted samples with highly trained and closely supervised practitioners to routine clinical care (Weisz & Jenson, 2001). Neither is it clear whether clinical guidelines where they exist are unambiguous enough to affect practice (Kelley, Nixon & Bickman,1999).

Limitations

The small sample size was a limitation to generalizability and may have restricted the power to detect change in function.

In this sample, we knew that individualised provision took place at school (as the great majority of the diagnosed children had a statement of special needs) but exact details of overall interventions were unavailable. We did not know if the children who received more intervention than others had better prosocial behaviors, or if the type of intervention an individual child received influenced prosocial behavior. Instead the study is limited to an overview of impact of educational and health service contact in a broad sense. The prosocial behavior of individual children may have improved or deteriorated after contact with particular services, but these data do not illuminate this. They do, however provides a general picture of a group of children and the development over time of one particular aspect of behavior with regard to health and educational service contact, responding to calls for community based studies that track development over time (Angold et al., 2000).

Our study was a retrospective analysis; ALSPAC was not set up to look at ASD therefore we did not have access to scores from ASD validated screening tools such as the Autism Diagnostic Interview or Autism Diagnostic Observation Schedule. The SDQ was never specifically designed to detect ASD and this is a limit to our study, albeit the prosocial score is strongly associated with ASD diagnosis (Russell et al., 2010). The strongest aspect of the study lies in its longitudinal design and repeated measurement of prosocial behavior in children diagnosed with ASD over such a long (10 year) period.

Another UK study found that there were considerable delays (of several years) between time of first parental concern and a definitive ASD diagnosis being given (Howlin & Asgharian, 1999). Children may receive interventions before diagnosis is reached, limiting this studies ability to detect the effect of any intervention after diagnosis. This may explain the overall trend of improvement - a tentative finding as the improvement was also consistent with random variation. A recent randomised controlled trail for a promising behavioral intervention for ASD showed that outcomes were no better for core autism symptoms such as social communication, compared to a control group with treatment as usual, although secondary outcomes such as parent-child interactions were enhanced (Green et al., 2010). Diagnosis and access to services may have improved other areas of functioning and familial relationships whilst prosocial behaviors remain relatively resistant to intervention. Perhaps the general trend of small improvements in prosocial behaviors with time albeit the same before and after diagnosis reflect wider gains in factors such as these.

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'Article 5'

Dilemmas, diagnosis and de-stigmatisation: Parental perspectives on the diagnosis of autism spectrum disorders.

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Dilemmas, diagnosis and de-stigmatisation: Parental perspectives on the diagnosis of autism spectrum disorders.

Abstract

Recent studies have established that many children with autistic behaviours at clinical levels do not receive a formal diagnosis. Parental barriers to accessing mental health services more generally have also been examined. This study used qualitative methods to examine the influence of parents in pursuing a diagnosis of autism spectrum disorder (ASD). The aim was also to explore the function of ASD diagnosis for parents, and examine whether a diagnosis affected how they perceived ASD. Seventeen parents participated in in-depth semi-structured interviews. Interviews were transcribed and analysed using thematic and grounded theory approaches. Data analysis revealed the underlying dilemmas faced by parents when considering an ASD diagnosis, illustrating concerns over whether to retain the 'normal' status of the child or to 'normalise' the child through diagnosis and subsequent remediation. Parents who had received an ASD diagnosis for their children became proactive in trying to reduce stigmatisation of ASD more widely, and in some cases actively advocating ASD diagnosis to other parents. Thus their actions may make it more likely that others will opt for diagnosis in the future.

Keywords

Autism, autism spectrum disorders, diagnosis, dilemmas, parent-child relationships, stigma.

Background

Autism spectrum disorders (ASD) are considered to be a set of complex neurological developmental disorders which are defined in practice by their diagnosis. They are characterised by impairments in social interactions and communication, as well as restricted interests and repetitive behaviour (Bailey, Phillips, & Rutter, 1996). Estimates of the prevalence of ASD have increased dramatically over the last 30 years. Some recent studies have calculated high prevalence rates by including children who have been documented by researchers as having symptoms of ASD but have never received a diagnosis (Baird et al., 2006; Baron-Cohen et al., 2009). A recent UK study (Russell, Ford, Steer, & Golding, 2010) established that a substantial number of undiagnosed children display the same levels of autistic symptoms as those with either an autism or an Asperger's syndrome (AS) diagnosis.

Several studies have examined why children do not reach mental health services. Access barriers mediated by both parents and clinicians have been identified. For example, Howlin and Moorf (1997) found that in many cases parents may be pushing for ASD diagnosis but barriers are put up by health professionals. However in child psychiatry more generally, quantitative researchers have identified access barriers to diagnosis mediated by parents themselves (Flisher et al., 1997; Ford, Hamilton, Meltzer, & Goodman, 2008).

Studies that seek to understand parental influences have concluded that parents often face considerable dilemmas when making decisions relating to their children's behavioural difficulties (e.g. Hansen & Hansen, 2006). Educational professionals recognise tensions experienced by parents as they struggle with the identification of their children's special educational needs (Norwich, 2008). The basic dilemma for these parents is whether to suffer potential drawbacks such as stigma, devaluation and rejection, or risk losing resources and opportunities that might be released by a formal identification. Hodge (2005) found evidence that parents feel pressurised into accepting an ASD diagnosis in order to access services. Such parental dilemmas reflect longstanding debates between biomedical and sociological scholars and differing discourses concerning ASD.

Biomedical models of ASD hold that autism is a neurodevelopmental disorder with discoverable neurological and genetic origins. Prompt (early) and effective intervention for ASD is currently

considered to be an international health priority. On the other hand, social theorists, (Nadesan, 2005) autistic adults (Baggs, 2007) and clinicians (Timini, Gardner & McCabe, 2010) have argued that ASD is in part socially constructed, that society currently deems autistic behaviours abnormal and that a shift is required in norms and values. These arguments are influenced by the emerging social model of disability (Finkelstein, 1979; Oliver, 1997). The social model advocates a distinction between the physical deficit or 'impairment' and the 'disability' caused by attitudes and infrastructures of society. Critics of the social model have argued that over-emphasising the role of society denigrates the painful experiences of autistic children with debilitating impairments and the experiences of their carers.

The autistic rights movement draws from the social model, regarding autism as a differing cognitive style rather than a 'disorder', and calls for societal acceptance of 'neurodiversity': referring to the neurological variation within the brains of the human population. Other researchers such as Baron-Cohen et al. (2009) acknowledge the stigmatising potential of the ASD label and call for the term 'disorders' to be replaced with 'conditions'. Thus ASD becomes ASC, 'autism spectrum conditions' in the literature of this school.

In an Australian study, Farrugia (2009) notes the work done by parents in utilising a biomedical understanding of ASD to promote tolerance in others. Farrugia interprets this as a way for parents to lessen the stigma of their positions, albeit at the cost of a 'spoiled' identity of the child. Mansell and Morris (2004) found that parents became more worried about their child's future after an autism spectrum disorder diagnosis and a substantial minority reported some devaluation and stigmatising of their children after diagnosis.

Several other researchers have reported the narratives described by parents of autistic children, and children with intellectual disabilities. On the one hand, narrative threads running through accounts emphasise the joy that such a child can bring (Stainton & Besser, 1998), their strengths (Dura-Vila, Dein & Hodes, 2010), and the personal development and positive life experience of caring for such a child (King, Zwaigenbaum, King, Baxter, Rosenbaum, & Bates, 2006; Green, 2007). On the other hand, parental reports describe how such a child limits social and economic opportunities (Todd & Jones, 2003) and confers additional stress on the whole family (Runswick-Cole, 2007). Gray looked specifically at stigma amongst a sample of parents of children with high functioning autism (2002) and classical autism (1993). A majority of the parents had experienced stigma, especially mothers.

Stigma as a sociological concept, was developed by Goffman in his pioneering book *Notes on the Management of a Spoiled Identity* (1963) and has been applied to a wide range of settings, including the study of families of children with disabilities (Baxter, 1989; Voysey, 1972). Link and Phelan (2006) surmised that diagnostic labelling does exert an independent effect on the rejecting responses of the public. They describe a trade-off between treatment benefits and effects of stigma. Studies have shown positive aspects to diagnosis itself; ASD diagnosis as a way of helping parents to accept limitations (Midence & O'Neill, 1999) and to remain optimistic (Gray, 2006). Riddick (2002) reports that the majority of adults and children with dyslexia found the label helpful at a private level and Rogers (2007) found some were quite unqualified about the positive implications of having a definite diagnosis.

One difficulty with interpreting findings is that often samples are wholly composed of those with diagnoses, and do not include those who have resisted diagnosis or adopted a counter position. Landsman's study (2005) is an important exception. To correct this, we adopted in-depth qualitative methods, sampling from a group with a clinical diagnosis of ASD, as well as from a second group who had so far actively resisted or had not yet received a diagnostic tag in order to ascertain how parents themselves might actively influence which children with autistic symptoms receive an ASD diagnosis. We also sought to examine what function the ASD diagnosis performs for parents and whether the diagnosis might affect the conceptualisation of ASD itself.

Methods

Recruitment

We analysed semi-structured in-depth interviews with participants sampled from the two groups. Interviews lasted between one and three hours and took place in participants' homes. The first of these comprised eight parents who were not actively pursuing a diagnosis, despite professional indication that their child might be on the spectrum, and the second nine parents whose children had already received an ASD diagnosis. These diagnoses were all given when children were between 3 and 11 years old, and the age of the diagnosed children at the time of interview ranged from 7 to 16 years old. The time lag between when the diagnosis was assigned and the study interview conducted was highly variable within this small sample, ranging from 1-11 years.

In order to recruit parents to the study who had undiagnosed 'autistic' children, we identified children who had been thought to be on the autism spectrum by education or health professionals, and whose parents had been informed of this. This was not to suggest that these professional opinions were 'correct', or even that they were more valuable than opinions of parents themselves; rather it was a pragmatic move which was necessary to design a parameter for clear definition of the group.

This first group, comprising parents of undiagnosed children were identified via an educational psychologist, a special needs co-ordinator teacher, personal contacts or through the 'snowball' method. In all cases, either an educational psychologist or special needs co-ordinator teacher at school had suggested that the child in question might have AS or autism. For comparison we recruited parents whose children had a definite clinical diagnosis of ASD. All but one child was diagnosed with high functioning autism or AS. The remaining child was diagnosed with childhood autism. Six of these participants were recruited with the help of a local support group for parents, and three via word of mouth.

Data collection

All interviews were carried out by the first author, and took the form of a series of open questions [see Appendix] allowing the narratives of parental experience of children's lives to unfold. The semi-structured schedule was developed as the study proceeded. Field notes were written up at the beginning and end of each interview and referred to throughout analysis, although coding categories used in vivo terms (terms used by the parents themselves) for as long as possible to avoid impressionistic interpretation.

Ethical permission for the study was granted by the local University of Exeter ethics committee. Written consent was given prior to interview and all transcripts were anonymised in the analysis of the study. Names have been changed throughout to protect the identity of participants.

Analysis

A modified form of constant thematic analysis, as described by Braun and Clarke (2006), incorporating elements of comparative method of analysis as set out by Strauss and Corbin (1998), was adopted. These methods were deemed to be most appropriate as previous studies using grounded methods have yielded rich insights into the experiences of parents of children with disabilities (e.g. Harbourne, Wolpert, &

Clare, 2004; Kelly, 2005). This approach was chosen as the specific aim of the present study was to systematically explore parents' subjective accounts of their experiences.

Participants were not invited to comment on the analysis as part of the analytic process as we attempted to analyse the function of the talk of the participants, i.e. commonalities in the ways they presented themselves and their children in order to achieve particular agendas. Therefore the analysis of their talk was not necessarily always consonant with the participants' views of themselves, and as such their comments could have been counterproductive to the aims of analysis.

In total 17 parents were interviewed. Ryan and Runswick-Cole (2009) point out how the term 'parents' is euphemistic for 'mothers' in much research concerning parents of disabled children. This criticism could be leveled at our study; of those 17 volunteering to participate, only two fathers came forward. Participants all lived in the UK and hailed from a range of occupations and classes. Their children ranged from 5 to 16 years in age. Thirteen of the children were boys and four were girls, giving a gender ratio of 3.25:1, similar to studies that have reported on the prevalence of ASD.

Results

Parents of children whose autistic difficulties had been highlighted by professionals, but who had not obtained an ASD diagnosis are denoted in this text as (1), and parents of children with a clinical ASD diagnosis are denoted as (2).

Before diagnosis and resisting diagnosis

What the entire sample shared was that at some point a professional had highlighted their child's autistic behaviours. When discussing their reaction to this, almost all the participants recalled strong but mixed feelings. Often participants wanted professionals to point out their child's differences to vindicate their own concerns, whilst simultaneously wanting a denial of any impairment. Thus participants experienced psychological tensions.

> I wanted them to tell me that everything was fine and that we were imagining it, and you go through that disbelief thing where you pretend that everything's fine and they are going to say no, he's

fine…So by that time I'd gone through a process of wanting to know, not wanting to know, thinking it was autism, definitely, then thinking no, it's not, it's something else, it's just a speech difficulty.

Mother of diagnosed child (2)

She said, you know, I think there's something about Johnny that's not… I can't remember how she worded it, she was very diplomatic, it was like she was piercing my heart, you know? Because I knew it, I knew it, but I didn't want it to be, you know? I really struggled with it.

 Mother of undiagnosed child (1)

At this early stage these mothers did not necessarily face a conscious dilemma, which would have involved a mindful weighing up of pros and cons of having their child diagnosed. A more apt description was that they were ambivalent. A sense of inner conflict was clearly created by the tension between wanting to maintain the status of a 'normal' child; and recognising a difference. Other studies of childhood disabilities such as those of Rogers (2007) have exposed the parental emphasis on maintaining a child's normal status in the face of perceived threats. One mother in our study commented: "I believed it, I mean I didn't not believe it", revealing an often unconscious battle that left mothers in an "uncomfortable" place. To them the diagnosis itself was synonymous with the loss of normality, an official confirmation of difference; the label ASD. Adapting to such a diagnosis has been compared to the grief of bereavement, as it shatters hopes and dreams of normality (Bury, 1982). According to McLaughlin and Goodley however, reorientation after diagnosis is not the same as the biographical disruption described by Bury, as parents do not necessarily seek to 'resolve biographical disruption, to create a new sense of certainty' after clinical diagnosis (2008, p. 329).

Further evidence of the "devastating" impact of diagnosis was the shock and grief parents recalled on receiving the diagnosis: even when they had suspected ASD.

Devastated really, I know it sounds harsh but it's like I don't know nothing about this condition, I knew it was a condition people had for life – but to begin with I was devastated and for the first month or so I didn't even want to say the word. (2)

The parents of undiagnosed children (1) differed sharply from those who had been through diagnosis (2) in the language they used to describe diagnosis, frequently using terms that denoted a violation of the child. The process of diagnosis was referred to in pejorative terms such as to "pigeonhole" to "slap a label on" to "put him in their little boxes"; "he has to be branded" to "shove him in a box", to "stamp it on his head" , to have "some fat label stuck on her". One of these mothers described the ASD label as "a millstone that hangs round their necks".

This choice of language can again be interpreted as delineating a perceived threat to the 'normal' status of the child that the parents were defending. This was also exposed in descriptions of the diagnosis as "scary", "frightening" and "dangerous". The recruitment process also provided insights into the positions of each group. While it was extremely difficult to recruit the first group (1), the second group whose children were diagnosed (2) were keen to participate, with more parents volunteering to participate after hearing about the study through word of mouth than it was possible to include. When requests for help with recruitment were made, both professionals and parents stated that attempting to recruit parents of undiagnosed children into the study might harm their own relationships with parents of undiagnosed children. Bringing up the thorny subject of a child's autistic-type difficulties might be met with a "hostile" reaction, provoked by associating their child with autism. This suggests that our final sample (1) represented a sub-sample of parents of undiagnosed children – those prepared to talk.

When questioned further, mothers who were actively defending their children from diagnosis (1) were able to justify their actions, often adopting arguments of the anti-labelling discourse that have been articulated by social theorists in the past, expressed in their own words. For example they saw ASD as a lifelong tag that would lead to preconceptions and rejection by others, particularly in the classroom.

> I think if she was labelled autistic they just would pick up on the wrong end of autism from somewhere else, from our point of view, and think of her differently. (1)

Since its emergence in the 1960s 'labelling theory' has been influential in both psychology and sociology. The theory relating to mental illness was set out by Scheff (1974) who suggested that diagnosis could lead to damaging preconceptions in others as well as self-fulfilling prophecies. Autistic children might 'inhabit the identities they have been ascribed' as Nadesan puts it (2005, p150). A review noted that such effects do occur, but are weak in comparison to the effects engendered by actual behaviors (Jussim,

Palumbo, Chatman, Madon, & Smith, 2000). We did not attempt to ascertain whether any such negative aspects of labelling actually occurred (parents of diagnosed children were actually uniformly positive about the benefits of ASD diagnosis), rather, the ideas behind labelling theory form a discourse which was adopted by parents resisting diagnosis as justification for their decision. These ideas were articulated in their own words.

> And we just, you know, we just live in this world where people have stigmas about things and views about things and the way people are treated, I'm worried that he'll be discriminated against, or that it'll be seen as like a weakness or something. (1)

Fathers and mothers of undiagnosed children (1) often questioned the extension of the spectrum as a way of medicalising children who would previously have been regarded as "eccentric". Here they articulated the position that it is societal changes that have produced the categorisation of their children's behaviour as a disability. Molloy and Vasil (2002) argue that the category of AS effectively reduces the range of behaviour patterns that would count as 'normal' (though eccentric) and 'sentences the child to a lifetime of special needs and interventions' (p. 667). Overall, our data reveal that parents may shy away from diagnosis because they view ASD in a negative light as a stigmatising lifelong condition. Rogers (2007) discusses the concept of parental 'denial' in relation to receiving unwanted appraisal of a child's difficulties. Parents of diagnosed children were quick to pick up on this point:

> I think the biggest thing is partly parental denial, that they don't want anything to be wrong – that doesn't mean to say that they're doing anything wrong, it's just that I think that they… Like [her ex partner], just don't want anything to be wrong. And partly, then, as a result, if the school don't encourage it then the parents don't know where to go with it. ..it's about fear isn't it? It's a fear of the unknown. (2)

The data would demonstrate evidence of parental (1) 'denial', if ASD was defined by physical symptoms such as a blood test. However the negative impact of the condition is a necessary facet for its diagnosis (World Health Organisation, 1992). By definition, then, a child cannot be autistic unless carers consider a child's behaviour to have a significant negative impact on their lives. This negative impact must be great enough to overcome the perceived social stigma of ASD that has been portrayed. There may be differing

perspectives on whether a child's behaviour might have a negative impact on their wellbeing. This complicates the 'denial' of problems as described by others. Teachers might see a negative impact but parents might not, and a consensus of others might become a basis for attributing denial.
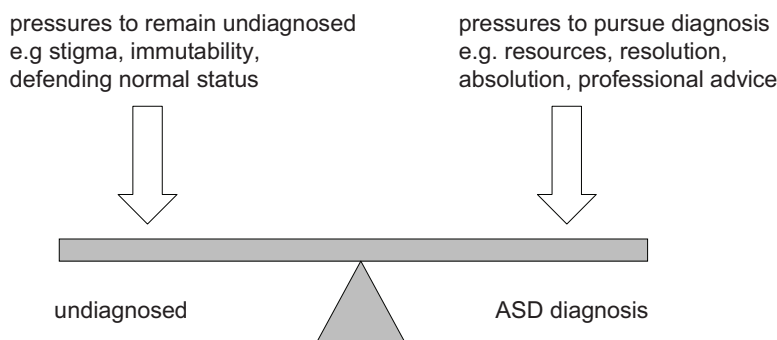
Parents who had not obtained an ASD diagnosis adopted anti-labelling discourses to support their position. Perhaps their children's difficulties were not impacting on their lives severely enough to justify opting in to the diagnostic process. Our data support the findings of Hodge (2005) detailing parents' reluctance to label their children.

On the point of diagnosis- the 'tipping point' and its aftermath

The initial dilemma of parents can be conceptualised as a balancing act with pressure on both sides (Figure 1). Parents resisting diagnosis (1) represented ASD as a frightening and stigmatising condition which causes damaging preconceptions. This represents a defended position- defending the normal status of the child. Parents who resisted diagnosis drew attention to the irrefutable nature of the diagnostic tag (preferring an informal transient label) and adopted anti-labelling discourse in order to justify their position.

One mother of an undiagnosed child (1) who had previously resisted diagnosis was on the point of changing her mind, responding in part to pressure from professionals. Soon after her interview this mother decided to get her son diagnosed with AS. Her position can be conceptualised as a point of tipping over from one position to another on a seesaw (Figure 1).

Figure 1: The dilemma of ASD diagnosis.

pressures to remain undiagnosed
e.g stigma, immutability,
defending normal status

pressures to pursue diagnosis
e.g. resources, resolution,
absolution, professional advice

undiagnosed

ASD diagnosis

On one side of the scales are pressures to get the diagnosis, on the other, pressure to resist. Although the perceived pressure and reasons to resist diagnosis have outweighed the perceived positive reasons to push for the diagnosis in the past, at the point of interview, the two sides seem fairly well balanced. This mother also described her predicament, in terms of 'weighing up' pros and cons of diagnosis. Other participants also framed their thinking as a process of 'balancing' arguments.

INT: Why do you think the label would be helpful, then? Or do you think it might not be?

Both, really. I just – having a label put on her will help me learn how to deal with it. I don't know whether I'm going about it in the right way and whether I'm making it worse. But then I think a label would also hold her back, once people know in her life that she's autistic or Asperger's or whatever, they'll think of her differently and treat her differently. I don't know. It's fors and againsts, really. (1)

This dilemma is vividly illustrated by the language used around diagnosis by the mother who was on the point of changing her mind, who adopted pejorative terms to describe labelling – "I'm not sure how much different it would be for them if they had that slapped on them" - as well as positive terms to describe the properties of the diagnosis in the same interview, for example its ability to "open up doors". Indeed the interview is interspersed with references to both positions including promoting an inclusive society without labelling children, wanting to distance her son from more severe forms of autism, and concerns

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about the perceived stigma and discrimination the naming of the condition could bring. However despite her stated reservations, she went on to give reasons why she should get a diagnosis. These pro-diagnosis arguments had been put to her by another participant and member of an AS social network who had a diagnosed son.

From a funding point of view apparently it will open up doors so that he can survive at college, and get the help he needs, make the best of it…..One thing that made me feel much more accepting about it was.. there was this lady, it was on Radio 4 and the lady was speaking about Aspergers, and her daughters have Aspergers… she was just talking about it in such an open and such a 'this is the way it is', 'this is what it is and this is how we're doing it and how we're coping', she was talking about it in such a kind of way like that I just thought, Oh actually, yes, maybe it is the right thing to do then, you know? (1)

The mother's attempt to bring the condition into the public eye by appearing on the radio led this participant to perceive a lessening of stigma. Such activity by parents employs the social model as it seeks to challenge the 'disabling' societal context to promote change.

A difference between the two groups was in the extent to which they disclosed the child's difficulties. The willingness of parents whose children had clinical ASD diagnoses (2) to talk to others generally about ASD was reflected by the ease with which they were recruited to this study. This echoes Bumiller's (2009) account of an 'anti-normalisation' strategy, whereby politically mobilised groups consciously bring previously unacceptable behaviours or taboo subjects into the public realm in order to make them acceptable. In contrast, parents of undiagnosed children were unlikely to discuss their children's behavioural problems with even their closest friends and families. A major concern was protecting their children from an unwanted autistic identity.

I'm still reluctant even to go to my closest family and friends and so this now is the first time we've spoken openly about it at all other than [her husband] – and trying to keep it all inside is actually quite, quite difficult. (1)

Additional factors, as well as the perceived lessening of stigma, contributed to the decision of the participant who changed tack after interview. Professional pressure on the participant had been renewed; diagnosis was framed as a way to seek academic support during a time of transition at school. A marital crisis was another factor bringing this mother to the point where she seems to tip over the balance from advocating the non-diagnosis arguments, to the pro-diagnosis counter arguments. Her concern for her own mental health is reflected in her admission that she was at a crisis point. Diagnosis therefore offered the possibility for a coping strategy.

Yes. No, I think I've got to [get the diagnosis] , I'm going to just do that now because I can't, I can't go on any longer. (1)

Overall, throughout the interview, this participant was torn between conflicting discourses. Here she describes the dilemma itself and the internal conflict it provoked:

Where my husband is very black and white I have masses of grey areas and I just can't, you know, I want to make the right decision. I mean I'll drive myself mad thinking about it. (1)

The triggers here for the decision to get a diagnosis show that despite wanting to be a good parent (the reason cited by all the parents as motivation for their actions), her needs mattered too, as well as her son's.

After diagnosis

All the mothers of diagnosed children (2) were enthusiastic about the benefits of diagnosis. Parents described naming the condition as a way of coming to terms with their child's behaviour, allowing them to research, manage and "understand" it. Bury defines 'coping' as the "cognitive processes whereby the individual learns to tolerate or put up with the effects of illness" (Bury, 1991).

For the sample of parents whose children had been diagnosed with ASD (2), inherent in the ASD label was the biological "explanation" for their child's behaviour; "I just think it's like his wiring's different." This explanation was repeatedly framed in neurological terms by parents with diagnosed children (2), for example; "he doesn't receive and send messages in the same way that we do". The invisible nature of the

impairment meant mothers in particular felt blamed for their child's behaviours. The sense of being blamed led parents "armed" with a diagnosis to proactively reject a 'naughty' label and deploy these biomedical accounts.

I think if he's playing up and we're not coping with him when we're out in public somewhere I think if you don't say anything people think they're misbehaving, can't the parents control them, but actually if have a quiet word with someone, say, 'I'm sorry, he's got Asperger's, he's just finding this situation a bit difficult', attitude completely changes. People are really different. I tend to say 'autism' to people though, 'he's got autism' because I think that people know autism but don't know Asperger's. (2)

Thus parents of diagnosed children successfully deflected accusations that their child's inappropriate behaviour was due to their bad parenting by using the ASD label. Explaining a child was 'autistic' effectively attributed the child's behaviour to integral biological, within-child factors. A similar process has been associated with ADHD, leading Hinton and Wolpert to describe ADHD as "the diagnosis of forgiveness" (1998, p.316).

Riddick (2002) reports that the dyslexia label is used to repel attributions of carelessness or laziness. This use of a label to deflect attribution to biomedical causes mirrors our findings. For parents, inherent in the ASD diagnosis was a neurological and genetic explanation for a child's behaviour. This was used to deflect blame from parents, as reported by Farrugia's study (2009). By contrast, in our study, parents of undiagnosed children (1) tended to reject biomedical explanations of their children's behaviours in favour of environmental explanations. For them a child labeled 'naughty' was preferable to an 'autistic' child.

When you apply perhaps a medical label it all seems to be, to the outside world it seems to be a lot more serious, a lot more scary- perhaps something to be frightened of, yes. Oh she's got a naughty child, oh well, he's just a little 5-year-old boy that's not really any problem – oh, her child's got such-and-such, then they start thinking, oh well, maybe I don't want my child to play with him.. Suddenly it becomes something very serious, something very scary and perhaps people don't want to be going anywhere near. (1)

Again, the official transformation effected by the diagnosis, an official loss of normality, is invoked. There is clear differentiation between the unthreatening temporary label or state of 'naughty' and the permanent trait of autistic. For the parents of undiagnosed children, this was one crucial distinction between the two labels.

One other function of ASD diagnosis was reflected in the way parents of diagnosed children (2) described it as "a key"; "a passport"; "a lever" and "an admission ticket". Diagnosis was also analogous to weaponry in their "fight", "battle" or "struggle" to secure resources for their children and themselves. These parents were clearly fighting to get their voices heard in order to advocate for the status of children as autistic and secure resources as they saw fit.

Many resources were accessed after the ASD diagnosis was applied. These included:

- Educational resources, specifically one to one support in class.

- Social resources e.g. access to support groups, holiday breaks.

- Health services e.g. mental health services.

- Access to information (mediated by naming the condition).

- Financial resources e.g. child benefits.

The study revealed that parents of children with an ASD diagnosis were particularly concerned about how their children would fare as they became young adults. Educational attainment, levels of well being, friendships, self harm, aggression, bullying and school exclusion were all areas of concern. These concerns reported by parents led to the selection of several emotional and behavioural variables of interest in a parallel study of outcomes for children with ASD. Our data suggest parents may be proactive agents in pursuing or avoiding ASD diagnosis. This is not to deny that real impairments exist. Nevertheless, differing discourses were deployed to justify desired outcomes, either promoting ASD diagnosis or avoiding diagnosis. Actions were justified in terms of doing one's best for a child; the 'good parenting

narrative' identified in other qualitative research concerning parents of children with disabilities (Silverman, 2006, p. 383). What 'good parenting' is considered to be, however, differs between cultures and times and, as evidenced by our study, from parent to parent.

Virtually all parents in the second group (2) experienced considerable delays between first concern and getting the formal diagnosis, often several years. This finding has been reported elsewhere (Howlin & Moorf, 1997). Some parents of younger children in the first group were less active in resistance to ASD diagnosis, rather they were adopting a 'wait and see' approach as their child underwent lengthy assessments.

Later stages after diagnosis: reconstructing ASD in a positive light

After diagnosis, parents in the second group described how they set about reconstructing their own ideas about ASD to cast it in a more positive light. Parents accepted biomedical understandings of ASD; that the condition was due to neurological differences in the brain, and further argued that such differences brought strengths, and even benefits. Parents sought to recast autism as both 'different' and 'valuable'. This enabled a re-evaluation of their own path; "travelling a different road" as one mother put it.

> It's like you've been given a ticket to Paris and then you find out you're in Paraguay. You suddenly become part of a parallel world with everybody else's. It's not the same – everybody else has got their children going along on one..[route]. –and you, suddenly, leave that place and start traveling a different road..
>
> INT: But generally there are areas of strength?
>
> Of enormous strength, and one of the things is he can think – well, not that he can think differently he just does think differently. (2)

One parent showed a children's picture book (Elder, 2005), designed to educate autistic children to regard themselves in a positive light, using famous 'autistic' people as role models. The book claims Newton, Einstein, Warhol and Kandinsky as on the spectrum.

These strategies serve to reconstruct what was once viewed as a negative label into a more positive one. They mirror the tactics of the autism rights movement, which also uses genetic and neurological models

of autism to claim for the right to 'neurodiversity'. These advocates argue that strengths in attention, focus and islets of ability, particularly in logic and science, are desirable genetic variations that ASD encompasses. So, like the autistic rights movement, parental understandings reflected aspects of both biomedical (ASD as a neurological difference) and social models of disability (agency to lessen societal disablement). In this context, the parents who were interviewed are part of a wider 'autistic culture' described by Bumiller (2009), attempting to shift negative attitudes within society to the more positive view of ASD as a condition bringing inherent strengths. The 'autistic advantage' as such strengths were recently dubbed by *New Scientist* (Wolman, 2010) were stressed and reiterated by mothers (2).

> Because he's defined as having autism, …the other thing that's turned it around for me is also looking at Harry and all the other children in the spectrum and how they think, and what they bring to the world, and how fantastic they are.   And it's like my friend, she was playing netball in Milltown, and one of the boys – I'm sure it was [autistic child of another participant] – was stood going, 'Losers! Losers!' at them, and she said he was doing it for virtually the whole hour that they were playing netball, and I said, 'Do you know, that boy is probably going to go on and design the most amazing Formula 1 cars, or computers, and you know, and he's going to be fantastic, and you just think that the way his mind works completely compensates for every other, almost to every other behaviour, and just that he's got so much to give, and I don't know that I could see that before. (2)

Parents of children diagnosed with autism have made positive adaptations in the form of changed world views concerning life and disability, and have come to appreciate the positive contributions made by children to society as a whole (King, et al., 2006). Thus the emergence of competing narratives in the literature, either describing children as a blessing or a burden is hardly surprising. Our data indicated that parents underwent a shift in world-view, from regarding receiving an ASD diagnosis as a calamitous event to appreciating the special competencies of their children, and in doing so, finding value in their own work as carers and advocates. Ryan and Runswick-Cole (2009) discuss the blurring of maternal position from advocate to activist. They consider that in a role where caring for a disabled child is widely socially devalued, mothers find that amongst their peer group their skills and experience have valued capital, thus within peer groups they are able to transform their status. A caveat to generalization of this interpretation is that in our study, parents of diagnosed children were largely recruited through one social

networking group. The group's membership, and consequent identity, may consist of parents who are more highly involved and motivated than the general population.

Riddick (2002) points out the tension between labels that can be seen to devalue or and labels that have been reclaimed as a positive assertion of difference such as 'gay' or 'black'. Both terms have been re-appropriated by the minority groups in question, a process of repositioning comparable to strategies of parents of ASD children (2). In a similar vein other groups and 'cultures' with perceived impairments are re-appropriating their terms to assert a positive difference such as 'deaf' (Skelton & Valentine, 2003) or 'dyslexic' (West, 1997).

Another facet of the transformative effect of diagnosis was parents' new ability to spot other children who they considered were in the spectrum. Once immersed in this particular aspect of life, they became sensitised to it.

> We were sat the other day having a meal and there was a family with a quite young lad, and he was chattering away to the parents and Harry and I just looked at each other and nodded. You kind of recognise it all the time. Watch things on television and say, 'That's Asperger's definitely', or autism. (2)

This ability had the effect of widening the spectrum, rendering other autistic children visible who were not previously seen. A recent US study (Liu, King, & Bearman, 2010) showed the effect of such social influences; children were much more likely to be diagnosed if living near other children diagnosed with ASD. Significantly, the two mothers of children with an ASD diagnosis who had autism-related jobs consistently recommended diagnosis to others whose children they viewed as autistic in their working capacities.

> Get the diagnosis! Because otherwise you don't know what you're dealing with, and actually neither do the professionals who are trying to help your child. (2)

To summarise, the views and positions of parents shifted and were transformed by the process of diagnosis. At first, all parents seemed to share misgivings about ASD as a negative "scary" term and were

keen to defend the normal status of their children. Those whose children subsequently received a diagnosis experienced considerable shock and grief at the loss of their normal child. However, after some time they reconstituted their vision of the condition, embracing a more positive view. The strategies of opening dialogue, recommending diagnosis to others and presenting the condition as different yet valuable served to improve the social status of their children as an identifiable group with ASD.
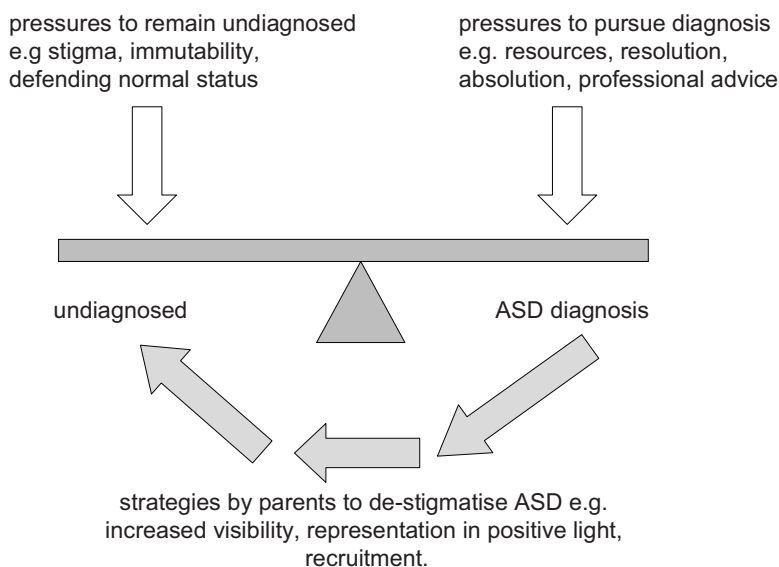
> You have a choice don't you? You either carry on or you let it make you sad forever, and you let it be debilitating and I chose to learn everything I could and to embrace everything I could and to accept that this was the way my son was. It took a long time but I have done that now and accepted it for many years. So that's what I did. …He likes his things, he likes his certain toys, he likes his certain clothes. If his needs are met, he's as happy as anybody else, and I think it's really important to keep hold of that. He can make a big contribution. I can see that now. (2)

In this way, parents, particularly mothers, reframed their children's autism using 'different but valuable' narratives. As a result of this process their world views shifted, affording self-actualisation. Ryan and Runswick-Cole (2009) argue that all parents advocate for their children, but for parents of children with ASD this activity becomes heightened, bordering and blurring the lines between advocacy and activism. They describe how mothers of autistic children act to raise awareness of autism in their local communities and beyond. McLaughlin and Goodley (2008, p.330) use the term 'strategic agency' to describe how parents work with whatever narratives are available to benefit their disabled children, be they biomedical or social. In our study we saw instances of such strategic agency being adopted by parents in order to de-stigmatise the concept of autism not only in the eyes of others, but also for themselves.

Discussion

The data highlight two differing strategies adopted by parents; either to retain the 'normal' status of their child or to 'normalise' their child by identification of ASD and subsequent remediation. In tandem with this second strategy came an effort to change the attitude of society itself, to reframe and de-stigmatise ASD. This resonates with the work of Norwich (2007; 2008) who describes how educational professionals who see the value of identifying disability in pupils go on to resolve the dilemmas by seeking to promote positive images of disability. The deliberate silence of parents of undiagnosed children suggests that counter arguments may be less effectively communicated in mainstream society.

Figure 2: Parental dilemmas and strategies to de-stigmatise ASD once diagnosed.

Parents, autistic adults and support services may collude in redefining ASD as both different and foreign, special and valuable. Indeed the phrase 'special needs' was coined to capture the positive aspects, to deflect focus on deficits but to capture requirements of provision. Our findings support the conclusions of Mansell and Morris (2004) who reviewed literature relating to parental reactions to ASD diagnosis and suggested that the process involves four stages: pre-diagnosis, diagnosis, post-diagnosis and a final stage of acceptance and adaptation. Our findings also suggest that parents may go through such distinct stages as their views transform with time. What is novel is our finding of a more proactive agency of parents in seeking to de-stigmatise the condition and recruit others to their point of view. Initially, they may resist diagnosis, attempting to hold on to the 'normal' status of the child. Later after a period of loss (of the child's 'normal' status) and adjustment, parents seek to reposition ASD in a more positive light. Thus parental experience proceeds in stages.

The dilemmas of parents may be analysed in terms of cognitive dissonance theory (Festinger, 1957) which has been influential in social psychology for over half a century. Cognitive dissonance can be defined as the 'uncomfortable' feeling caused by holding two contradictory views simultaneously, as described by parents of undiagnosed children in our data. The theory of cognitive dissonance proposes that people will act to reduce dissonance by changing their attitudes, beliefs, and behaviours, or by justifying or rationalising them. For parents of children who receive a diagnosis the dissonance motivation can be encapsulated as concern that the formal clinical ASD diagnosis makes a child 'autistic' (where 'autistic' has a negative connotation). This according to Kunda (1990) makes a person search for 'cognitions' where this cannot be true; hence the development of the increasingly powerful 'different and special' discourse. For the parents adopting this discourse, 'autistic' is no longer a pejorative term, and parents set about recruiting others to this viewpoint.

Malacrida's study (1990) of children with ADHD highlighted cultural differences. She points out that Europeans are less likely than North Americans to accept medical labels for their children's behaviour. Our small samples may therefore not be representative of wider populations. Recruitment efforts show that parents of children without ASD diagnoses were a subgroup of a wider section of parents resisting diagnosis or possibly unaware of potential problems. Nevertheless, the phenomena that we picked up have also emerged in comparable studies of childhood disabilities: despite the socio-cultural constraints associated with caring for a child with complex needs, studies have demonstrated that most mothers who have children with these types of diagnoses consistently perceive valuable benefits in having a child with a disability. Children portrayed in a positive light, a light which seems determined by the cultural constraints in which they operate. For example, in Dura-Vila, Dein and Hodes' study (2010), for example, religious parents interpreted a disabled child as having sacred meanings. The 'autistic advantage' described by the parents in our study may be a secular version of the same process of parental optimism. This optimism in parents is well documented and may serve not only to help them cope but also to have an adaptive function to help their children. Research has shown that the expression of positive beliefs that a child has good cognitive abilities and that these are under the child's control, leads to better outcomes (Miller, 1995).

Ryan and Runswick-Cole (2009) point out that in attempting to 'normalise' their child, parents of children who have a diagnosis adopt activism which is underpinned by a medical model of disability.

However, parents in our study seem to have adopted both medical and social models of disability in strategic ways, with the ultimate goal of repositioning their children as well as themselves. Parents do not appear tied to a particular model; rather, they adopt whatever discourse is required to secure the best outcome as they see it. These strategies adopted by parents with diagnosed children together illustrate an underlying trend towards actively de-stigmatising ASD. By challenging a disabling society the social model is also adopted. Such arguments and strategies of parents of children with diagnoses (2) are deployed to lessen pressures on parents of undiagnosed others to retain the undiagnosed status of children (1) (Figure 2).

According to Haidt (2001), all groups that share a social identity will adopt positions that tacitly influence others. But here there is a crucial difference highlighted by our data. Whereas parents with diagnosed children (2) are more likely to talk about their position in the public arena as it serves their purpose, those resisting ASD (1) are likely to keep it a private matter, defending their child from a 'dangerous' threat. Parents of children diagnosed with ASD may self-organize rather than remaining isolated. Hence their potential power to put their points across is enhanced by their ability to speak publicly and collectively.

Through more widespread communication and greater recognition of ASD in the community, parents of children who are diagnosed may have a greater influence on the balance of decision-making than those who may have private reservations and resist diagnosis. The feedback effect illustrated in Figure 2 may act to extend the diagnosis to more children, so these data outline how one psycho-social process may be a factor in persuading other parents to opt for diagnosis, potentially leading to an extension of the spectrum. Parental de-stigmatisation of childhood ASD may therefore act to increase prevalence of diagnosed autism spectrum conditions.

Acknowledgements

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Appendix:

Interview schedule:

Who lives in your household?

Number of children

Are you working?

What is the name of your affected child?

How old are they?

How old were they when you first noticed a difference?

Tell me your story: your experience concerning your child.

Is it a difficult thing to discuss? /Who do you speak to?

Can you tell me your feelings about get him/her diagnosed?

Why diagnose? /Why not- diagnose ?

Is the difference more pronounced at home or school? Easier to deal with?

What sort of behaviours concern you?

Did the school first notice a difference?

What do you know about the term 'autism/Asperger's syndrome'

What do you feel other people think?

Is there an issue with having a label? If so, why?

Have you ever thought about what might have caused the behaviour? What are your thoughts on that?

What is your greatest concern for the future?

Concluding chapter

The concluding chapter is divided into four sections. In the first, the contribution, merits and limits of each individual article will be reviewed, in relation to diagnosis of ASD, the ways in which the articles engage with the key themes in the introduction, and how each article addresses the original aims, objectives and research questions of the thesis. In this section the revisions to articles 3, 4 and 5 are also discussed in the light of the reviewers' comments, the initial objectives and the existing literature. Details of all the revisions made to the submitted articles, and all the reviewers' comments, are given in thesis Appendix 1.

In section 2 an overall evaluation is made of the strengths and limitations of the mixed methods design. The merits and limits of the both quantitative and qualitative approaches are considered, and how these worked together, including the use of secondary data.

In section 3 the overall contribution to knowledge of the thesis as a whole includes a reappraisal of the biomedical and sociological perspectives on ASD diagnosis described in the introduction. There is also some speculation of the impact the body of work could potentially make, situated in the current social and cultural context, and its contribution to understanding of the aetiology of ASD.

Finally, in the last section, potential directions for future research will be discussed, followed by some concluding remarks.

As all of the articles have already been reviewed by anonymous reviewers, sections of their comments are included, where illustrative of wider points. Unfortunately the identities of these reviewers are unknown, so informed consent could not be obtained. However, the contribution of these anonymous reviewers is recognized in the 'Acknowledgements' section.

Section 1: Contribution to knowledge of individual articles, their merits and limitations

i) Article 1: Contribution, merits and limits

The first article, Article 1, defined the methodology that underpinned two subsequent studies that used quantitative techniques (Articles 2 and 3). The methodology itself was novel in that it cross-compared two discrete datasets, both concerning the same cohort of children; the Avon Longitudinal Study of Parents and Children (ALSPAC). These two independent data sources were the medical records of the children in the ALSPAC study, and the measures taken by ALSPAC itself.

The regression of the ALSPAC data to predict the outcome of ASD diagnosis demonstrated which autistic traits were most closely associated with the diagnosis of ASD. Impairments in prosocial behaviour appeared to be the most salient trait. The cross comparison of these two datasets established that there were children in the cohort who had behavioural symptoms similar to and at comparable levels with, those with a clinical ASD diagnosis. Article 1 provided local evidence to indicate that there may be many children in the UK, and possibly worldwide, who display autistic traits at clinical levels, but who remain undiagnosed. Of course the generalisation of the finding would be more robust if the study was repeated and it was confirmed in other cohort studies.

The findings of Article 1 concurred with those of epidemiological studies that have highlighted a disparity between the prevalence of childhood psychiatric disorders recorded by researchers and specifically autistic spectrum disorders and their recognition as defined by receiving a clinical diagnosis (e.g. Baron-Cohen et al., 2009; Honda, Shimizu, Misumi, Niimi, & Ohashi, 1996; Kadesjö, Gillberg, & Hagberg, 1999; Yeargin-Allsopp et al., 2003). The findings of this tradition of work formed a key theme in the thesis.

The research reported in Article 1 tested the first subsidiary hypothesis given in section 4 of the opening chapter:

1. There are children with autistic-type behaviours at the same severity as those with a diagnosis who remain undiagnosed.

The literature reviewed has demonstrated ASD to be a spectrum, with the sub-clinical phenotype (the broad autism phenotype) extending into the 'normal' population. The literature indicates that an arbitrary cut-off separates the pathological from the non-pathological for ASD. Also, given the inclusion of 'impact' in the diagnostic criteria, plus the literature on access barriers and parents' surveys suggesting parents play an active role in determining whether children are diagnosed or not, an aim of this research was to identify an undetected group of children who would be diagnosed with ASD were they bought to the clinic, as outlined in the introduction, section 3.

Article 1 argued that some of the undiagnosed children that were identified could be considered to be 'on the spectrum':

> Some of the 'undiagnosed' children may be on the autistic spectrum in its wider sense, perhaps presenting sub-clinical levels of autistic traits such as those described by the broad autism phenotype (Lainhart et al., 2002), while others, particularly those in the top 2%, displayed impaired cognitive and social functioning that could lead to diagnosis were they brought to clinic (Russell, Ford, Steer & Golding, 2010, p.649).

However, this had not been tested in any way. This served to expose a major limitation in the data: that ALSPAC had never attempted to assign a research diagnosis of ASD to its cohort, and therefore the definition of the undiagnosed control group relied on a series of measures, that were not a standardized or validated tool for identifying ASD such as the Autism Diagnostic Observation Schedule (ADOS) or the Autism Diagnostic Interview (ADI). As discussed in the introduction (p. 34), in the prevalence literature, children may be given a 'research diagnosis' which is made according to measures taken by the epidemiological cohort of interest. This qualifies them as being included in prevalence estimates for ASD and other disorders. Therefore we can say that such children can be considered autistic but unidentified according to such studies (e.g. Baron-Cohen et al., 2009). However the criteria used by such studies to identify undetected children are usually validated, unlike than in Articles 1-3. It was not possible to claim that the children in the two groups were equally likely to receive a diagnosis in the age ranges compared, even if all social and demographic factors that were examined had been equal, as there may have been many other concerns which the ALSPAC data had not recorded. Nevertheless the composite autism trait can also be defended, as it did not leave out key features of ASD- in fact it encompassed all the

qualitative impairments specified in ICD-10 necessary to make a diagnosis of ASD, and these were included in ratios specified by the diagnostic criteria. Even with these ratios, though, the measure of autism or AS in the population as a whole had a fairly low positive predictive value, as evidenced by the large number of undiagnosed children who were exposed. Thus the definition of the undiagnosed comparison group was not ideal, but it was the best match that could be achieved within the confines of the secondary data available.

The second main finding from Article 1 was gleaned through cross-comparing data on educational support with the children that did, or did not, have an ASD diagnosis. Over half the children with autistic-like traits but without an ASD diagnosis were unsupported by additional health and educational services by the time they were aged 11 or 12. This finding broadly concurred with results of others studies reviewed in the introduction that indicate only a proportion of children who display symptoms of childhood disorders reach health and educational services.

Despite its obvious limitations, this study prompted one anonymous reviewer to comment:

> This is a potentially very important study on an issue of both theoretical and practical concern. It provides evidence for current debates on the value and validity of current diagnostic systems and on the criteria for educational support in UK schools. Although based on UK data, the study has wider significance, since such debates are current internationally.

And the second:

> This in my view is an important paper. The findings suggest that there are a number of children with symptom severity equivalent to diagnosed autism and Asperger syndrome approximately half of whom have some form of special provision at school.

Interest lay not only in what the contribution of the articles had been, but in how they were subsequently deployed to frame ongoing debates about diagnosis and identification of ASD. In fact Article 1 provoked an editorial in its issue of *Journal of Child Psychology and Psychiatry* which described its impact thus:

Russell and colleagues provide vignettes of the typical 'undiagnosed' child. For autism 'an asocial mostly non-verbal child who does not indulge in pretend play, is afraid of unfamiliar things and exhibits repetitive behaviours'. For Asperger syndrome 'a child who lacks social skills and empathy, has difficulty in conversation and often repeats actions that have no obvious function.' If the needs of children such as these are apparently hidden to the professionals who work with them, what chance for children with less pervasive difficulties, such as specific language difficulties or dyslexia? According to recent data collected by our group, some 7.6% of children in a nationally representative sample of 1200 children enter secondary school with a reading age below 8 years; yet only 15% of these very poor readers have a Statement of Special Educational Needs (Snowling, 2010, p. 642).

Here and elsewhere (the article prompted a press release and widespread media interest), the findings were cited as evidence to call for closer monitoring of children, and increased identification of, and intervention with, difficulties. This was not the intention behind the article, for within it we clearly stated that:

> The findings could be used to argue for expansion in child mental health services, by illustrating high levels of unmet need. However, service contact alone does not necessarily imply that a child's needs are being met. A wide ranging review of educational and behavioural interventions for ASD concluded that there is inconsistent and inconclusive evidence about effective interventions ..... Ultimately the wider issue of effectiveness of interventions for ASD must be addressed (Russell et al., 2010, p.649).

ii) Article 2: Contribution, merits and limits
The second article followed on from the first, examining in more depth the social and demographic factors that might influence whether a child received an ASD diagnosis or not. However, at the same time as embarking on what was to become Article 2, the outcomes study (the research reported in Article 3) was initiated. At this point it became clear that only about half of parents in ALSPAC were responding to any one questionnaire measure by the time when their children were teenagers.

Thus the sample sizes with the split samples defined in Article 1 were too small to be viable for the analysis of outcomes. This was a severe setback. The sample had been split to encompass:

- AS or atypical autism diagnosed children and children with AS-type behaviours who were not diagnosed with any ASD
- Childhood autism or atypical autism diagnosed children and children with similar impairments who were also undiagnosed

The reasons why the sample had been split in Article 1 into the AS/atypical sub sample and an autism/atypical sub sample, are outlined in principles 6 and 7 in the methodology section. Firstly, the sample had been split because ASD is such a heterogeneous condition; within the range of behaviours that are classified as ASD there is a huge variation in the severity of symptoms. The type of symptoms presented can also differ enormously as demonstrated in Table 1 in the introductory section of this thesis.

The slight differences in diagnostic criteria also provided a good reason to split the sample; the composition of the composite autism and AS traits could then be adjusted according to the exact requirements that were specified in ICD-10. The top 2% of the composite autism trait, for example, was a very good fit, with the composite trait explaining 65% of all the pseudo variance (the proportion of the log-likelihood) associated with the outcome of diagnosis. All the scores for children in the diagnosed autistic sample fell within the top 2% of the autism composite score. The sensitivity of the composite autism score in predicting autism diagnosis within the top 2% was therefore 100%. Unfortunately, as mentioned, attrition in the dataset meant that the outcomes study (Article 3), would be underpowered if the sample was split by category and age range as in Article 1. It was necessary, therefore, to fuse the ASD sample, to repeat the method of Article 1 to create a composite 'ASD' trait. This trait showed a similar distribution to that derived from the autism and AS sub-samples (Figure 1, Article 3).

When the samples were fused to produce a composite ASD trait (rather than separate composite AS or composite autism traits), the sensitivity dropped to 68%, and the model only explained 47% of the variability in outcome of ASD diagnosis. This shows how splitting the sample produced a composite trait that more accurately reflected factors related to diagnosis of each sub-condition on the spectrum.

Despite this further limitation, the second article, where the comparison group for Articles 2 and 6 was defined, was well received. The article aimed to detect socio-demographic and family factors in the

children's' backgrounds that were associated with diagnosis, or lack of diagnosis, as outlined in principle 8, testing the second subsidiary hypothesis that:

2. There are social and demographic or family based factors in the children's backgrounds that may influence why some of these children are diagnosed and others are not.

Results presented in Article 2 showed that with the severity of autistic traits held constant, boys were more likely to receive an ASD diagnosis than girls. Younger mothers and mothers of first born children were significantly less likely to have children diagnosed with ASD. Maternal depression before and around the time of children's autistic difficulties was associated with lack of diagnosis. The limitation concerning causality was compensated for by repeating the analyses using a sub-sample who were diagnosed early (ages 2–6). All the reported effects were still observed. Perhaps the most interesting of these findings (to its readership) was that in the ALSPAC population, even with comparable severity of symptomology, boys were more likely to receive an ASD diagnosis than girls. Indeed several researchers had speculated that this is the case in the past (Ehlers & Gillberg, 1993; Wing, 1981) although the literature review conducted for the introductory section to the thesis uncovered no empirical evidence. It is possible that this article may be cited by researchers and stakeholders who have previously suspected gender biases in the identification of ASD, to back up their theories. A prominent expert in ASD in Australia, Tony Attwood, and also Prof. Judith Gould, who is conducting a review of literature for the National Autistic Society both requested copies of this article.

'This is an important paper' stated one reviewer, and continued

The finding that girls are less likely to receive an ASD diagnosis independent of the severity of autistic traits is particularly important as there has previously been debate about a gender bias in diagnosis and the "extreme male brain" proposal. This paper may assist in raising awareness amongst health professionals of females with ASD.

Article 2 interpreted this finding in the light of the popular conception of ASD as a 'male' disorder (Murray, 2008). As outlined in the introduction, Baron-Cohen, (2002) suggests autistic people can systematize, but are unable to empathize with other people, or deal with events and stimuli generated

externally. Recent work by the Cambridge group has suggested that high levels of foetal testosterone may be linked to the development of autistic traits (Ingudomnukul et al., 2007). This has been interpreted as concurring with Baron-Cohen's well known 'extreme male brain' theory of autism. Such understandings may lead to gender stereotyping by education professionals, clinicians and parents when identifying and referring children for diagnosis, resulting in a possible gender bias. Autistic behaviours may be interpreted in boys as autism, but the same behaviours may be interpreted as autism in girls less often. This could partially explain the results presented.

Whatever the reasons for the results obtained, they provide evidence that differences in the demographic and family context of the child can influence whether children are diagnosed with ASD or not. Because of the limitations discussed above, caution must be exercised in interpreting results. Nevertheless, Article 2 highlights a component to the diagnosis of ASD that exists in relation to society. This relates to the discussion of normative values in the introductory section. In the case of the interpretation of the gender bias for ASD diagnosis, what is normal is that ASD is 'male'. This is not to deny that real differences exist; as already noted epidemiological studies have shown the male to female ratio for autism is reported as averaging 4:1, although the most recent estimate from a population based survey on ASD as a whole in the UK is 9:1 (National Health Servoce Information Centre, 2009). There is no doubt that boys are more likely to exhibit autistic-like symptoms. But although real differences exist, there may also be a difference in interpretation of symptoms.

This reading of the data took a critical realist position in that it was assumed mental interpretation is used to describe reality (Bhaskar, 1998; Sayer, 1992). The work was therefore in the tradition of other studies reviewed earlier that have uncovered discrepancies in the prevalence of ASD as a function of regional and cultural variations.

iii) Article 3: Contribution, merits and limits
The initial objective of the research reported in Article 3 was to test the central hypothesis given in section 3 of the introduction; that

> Diagnosis of ASD (and/or subsequent service contact) has an effect on behavioural and social outcomes, compared with an undiagnosed group who have comparable autistic-type behaviours.

To provide enough statistical power the research utilised the same (fused ASD) groups as Article 2. According to the principles of the methodology, it had been seen as important to use sub-samples where diagnosis had been applied in the same age ranges in which the comparison group had exhibited similar levels of autistic behaviour. This was to give the study more power to address debates about causality. If symptoms had been measured earlier than ASD diagnosis occurred, then they might have naturally improved in the non diagnosed children and deteriorated in other children leading to ASD diagnosis (Scenario 1). If symptoms were measured after diagnosis took place, biases from diagnosis itself (the effects we were trying to detect) might have confounded results (Scenario 2). Therefore it was deemed essential to split the sample so that autistic symptoms were measured around the same age as ASD diagnosis. This was in addition to reasons for splitting the sample by ASD sub-classification given above.

Because the ASD diagnoses had been assigned over a ten year period, it was necessary to define a comparison group who shared symptom severity before most of the diagnoses were applied, i.e. Scenario 1 described above. This lack of matching over time had dire consequences for testing the initial central hypothesis. Inferences about the nature of causality that could be made in Article 3 were limited. Was deterioration in performance that was recorded due to diagnosis or was diagnosis due to deterioration in performance? Because principles 6 and 7 were violated, the hypothesis that was actually tested was

> Social and behavioural outcomes differ in the sample of children diagnosed with ASD to those in the undiagnosed sample with autistic-type behaviours when young.

This was quite different from testing the effects of the diagnosis.

One co-author on Article 3, a child psychiatrist, suggested that the attempts to control were like 'comparing apples and pears' and that the result of the study was evidence that children with more pervasive autistic difficulties were more likely to be diagnosed. 'Not news' she commented. There appeared to be reluctance amongst co-authors, especially those with clinical training, to even question whether ASD diagnosis and intervention might have any negative impact, as was the suggestion by the social theorists and those who have studied the various aspects of labelling theory reviewed in the

introduction. Perhaps this reflected an understanding of ASD that was more closely aligned to the biomedical perspective.

Thwarted by not having a close enough match between cases and controls across time, the contribution to knowledge of this article was partially restricted to a consideration of factors (other than autistic behaviour) that could prompt diagnosis: disruptive behaviour, and low academic ability emerged as potential triggers. This concurs with the findings of other studies, previously reviewed, that have also suggested that children's problems are more likely to be addressed if they present externalising behaviours. As the literature reviewed demonstrated that obtaining educational support and resources was a strong motivating factor for parents in securing ASD diagnosis, indeed diagnosis has been framed by educational professional in these terms, then it follows that poor academic outcomes might also provide the spur to seek help.

The research resulting in Article 3 initially attempted to emulate studies that assess outcomes using real world settings, de-emphasising specific treatments. Kelley, Nixon and Bickman (2000, p.466) decribe effectiveness research as 'allowing examination of such issues as service access, utilisation, cost and quality in addition to the issue of service impact' and this was the type of study that the quantitative research presented in this submission originally aimed to be.

Initially, then, the quantitative study was an attempt to look at the possible benefits or negative consequences of ASD diagnosis on outcomes in children. This research was informed by tensions between the biomedical perspective where ASD is regarded as a medical condition to be diagnosed then treated, and the sociological and psychological research that centred on labelling theory, stigmatization, SFPs and attribution theory. But this aim of the research, to which the studies reported in Articles 1 and 3 were also directed, failed because of limitations in the data and arguably, inherent flaws in the methodology.

Because an objective of the outcomes study was to examine the benefits of SEN provision, children (at 'statement' or 'school action plus' levels) were initially excluded from the undiagnosed group. It was argued that this group (who had no external referrals outside school according to their educational records at age 11/12) were unlikely to have received alternative diagnoses, and as level of SEN provision is fairly

stable over time in the UK (Department for Children Schools and Families, 2009). For the same reason, it was argued that these children were likely to have received low-SEN provision throughout their primary school careers. The idea was to establish two dichotomous groups, one with ASD diagnosis and high-SEN provision (*n*=71, all but one with statements, age 11/12), and the other with no diagnosis and low-SEN provision (*n*=79), in order to better isolate the effectiveness of SEN provision for the diagnosed children.

This set of assumptions led one reviewer to question whether the data had been checked carefully enough:

> Did the children have absolutely no referrals even as preschoolers since they all had speech problems?

This comment led to a major reanalysis; the definition of the group with low-SEN provision relied on educational categorization at age 11-12. This was only taken at one time point. It was assumed that this would reflect the history of SEN provision for each child. Although this was probably broadly true, for individual children this was not the case. There was no access to educational records at younger primary school ages but, in response to the above comment, arrangement was made to access educational data from two years earlier (at age 9-10) and it was evident that 3 children in the low-SEN provision undiagnosed group of 79 were moved from 'statement' to 'no SEN provision' between age 9 and 11. Also, when the study children were seven years old, parents reported whether their child had *ever* been given speech and language therapy. Of the parents of the 79 children in the low-SEN provision group, the majority of children (34 out of 60 who reported) had received some form of speech and language therapy when young (this compared to 41 of the 47 children with an ASD diagnosis whose parents responded).

So it was clear that many of the low-SEN provision undiagnosed children had probably received various interventions when young, albeit as a group probably less service contact than those who were formally diagnosed. Other comments made by the second reviewer related to interventions:

> The introduction has a heavy focus on services for children with autism spectrum disorders but I could not understand the purpose of this focus.

> This paper does not contribute in any way to the literature on intervention for ASD and this seems a misplaced discussion.
>
> A more appropriate focus would be on research in other areas comparing clinically identified and undetected groups in epidemiological populations.

The comments of reviewers and external examiners implied that the article can not tell us anything about effectiveness of interventions, rather the focus should be on divergent development of behaviours between two groups. It was decided at this point to redefine the comparison group as simply those with autistic-like symptoms when young, but no ASD diagnosis, regardless of SEN support. This meant that the 37 children who were not diagnosed with ASD but were given some form of SEN were included in all the subsequent analyses. In fact this made no difference to whether outcomes significantly differed between the groups, (Table 2, Article 3). It did mean confidence intervals were tighter because of the larger sample size.

The figures were redrawn with the changed parameters of the comparison group, but again although the numbers changed, the significance levels of the results reported were unaffected. It also meant the groups were slightly better matched in the baseline measures reported. With reference to the initial results, using the low-high SEN provision split, limiting the comparison group to those with low-SEN age 11/12 provided no evidence that high-SEN provision for children diagnosed with ASD was particularly effective. This concurred with the findings of Crawford and Vignoles (2010). On the other hand, neither could it be concluded services were ineffective as the comparison could have been of children on differing trajectories due to initial factors that were not accounted for. Also, given that many of those in the low-SEN provision group had received speech and language therapy, perhaps the therapy had been effective for this sub-group of children, and this resulted in improving trajectories. Warren et al. (2011) have called for more attention to be paid to identifying which interventions may work for which sub-groups of ASD children. The decision was taken to redefine the control group as outlined above was contested by Colin Steer, who argued that the initial design should be maintained as it provided the rationale for the study.

Although inconclusive concerning the debates about diagnosis and subsequent remediation around which the work was initially conceived, Article 3 did have something to contribute in terms of raising questions

about the age at which children can be reliably diagnosed. As the cases and controls were well matched across a range of autistic measures as preschoolers, but diverged as they matured, the results provided evidence that it could be difficult to distinguish at young ages between a child who may have lifelong impairments and a one who is a 'late developer'. This case is a further illustration of how children who share autistic symptoms at young ages may have differing developmental trajectories, as in the Romanian orphan study (Rutter et al., 1999). However there are various possible explanations for the poor outcomes as one reviewer of this article noted:

> A diagnosis of ASD will have been made on the basis of history and observation of persistent traits and behaviours (not recorded here) specific to autism excluding other diagnoses. The children were compared at time point 1 on the traits alone, for which there may have been other explanations, including lack of persistence and pervasiveness, and not on other factors such as developmental delay or behavior problems which would have helped to answer the question about a bias in those referred. It is certainly possible that children are referred because of a multiplicity of concerns and this accounts for the different outcome, also possible that those diagnosed with ASD at age 2 years 'grow out' of the ASD features as was found by Stone or that they had a different explanation for the traits or that ASD diagnosis is a disadvantage!

The reviewer went on to add
> The warning about care in too young a diagnostic insistence is important at a time when all the emphasis seems to be on early intervention.

It is hoped the study will be published and used as evidence to counter the arguments for earlier and earlier ASD diagnosis as made by autism charities and other researchers (e.g. Crais, Watson, Baranek, & Reznick, 2006; Filipek et al., 1999). Article 3 concluded that the 'precautionary principle' (Science and Environmental Health Network, 2000) which states that preventive measures should be taken (or no action taken) in the face of scientific uncertainty about risk, should perhaps be applied when assigning ASD diagnoses to very young children.

Overall the changes in design prompted by comments from reviewers meant that the initial rationale for the study was undermined. One final comment from a reviewer then became hard to defend:

The authors discuss the problem of not having matched groups, but I cannot understand why they would want to undertake a matched analysis.

As has been explained, the reasons were similar to those of Angold, Kelley, Nixon Bickman, Crawford et al.; to better try and isolate effect of diagnosis/service contact. Although this has not been possible with so many confounders in the study, the divergence of the two groups is nevertheless of interest.

iv) Article 4: Contribution, merits and limits

The analysis presented in Article 4 was conceived in reaction to the limited conclusions of Article 3. After the failure of the design to test the central hypothesis as initially conceived, Article 4 sought to establish developmental trajectories before and after ASD diagnosis was made. The aim of the work was to try and assess likely effectiveness of service provision triggered by ASD diagnosis itself, and again to see if there were negative consequences of diagnosis in terms of outcome. Therefore Article 4 represented another attempt to re-engage with the biomedical versus sociological arguments outlined in the introductory chapter.

Specifically, the work engaged with the study of Mansell and Morris (2004) who reported that a range of service provision becomes available both from health education and other sources, after an ASD diagnosis is applied. Other sources reviewed in the first chapter suggest ASD diagnosis is advocated by a range of health, education and parent bodies because it allows access to appropriate treatment and support.

There was only one relevant outcome measure that was repeatedly taken throughout the lives of the children in the ALSPAC cohort, and this was the SDQ questionnaire. The analysis was limited to one outcome: the prosocial score, because it had been associated with autism and ASD diagnosis in the cohort (Articles 1 & 2). Longitudinal assessment of the developmental trajectory of prosocial behaviour over ten years suggested diagnosis and resulting service contact may not have led to any net improvement of children's prosocial abilities.

The objective of the study was both to assess the effectiveness of the general package of educational provisions, health services, support etc. that may be accessed after ASD diagnosis, on improving prosocial behaviour, through identifying a developmental trajectory of children's prosocial behaviour before and after diagnosis. However this was not at all clear to the first reviewer who commented:

> Given the current structure of the introduction, it is unclear if the focus of the article is on the effectiveness of the array of educational intervention programs and home therapies or on identifying a developmental trajectory of children's social behavior (as described later in the paper).

One limitation of the article, given its stated objective, was in the lack of details on the nature and type of service provision. A recurring complaint from reviewers was that unknown were:

a) what intervention services children were receiving;

b) whether these interventions were specifically designed to enhance socialization;

and c) how long children were enrolled in these types of services. Did all children receive intervention or only a portion of children?

All of these points were relevant; unfortunately the sample size was just too small to look in any more detail at types of provision, dose size and lengths of time of various provisions. The only question that could be meaningfully addressed was whether service contact triggered by ASD diagnosis had an effect on prosocial behaviour.

A further point made by the first reviewer of this work was that:

Until it is clear that these skills were targeted by efficacious interventions with a strong evidence-base, it is premature to conclude these symptoms may be resistant to intervention.

Aside from dismissing the results altogether due to their limits, there are two possible interpretations of the results, first, that interventions targeted at prosocial behaviours were not being used at the time of the study, and second that despite additional provision aimed at improving social skills engendered by ASD diagnosis, prosocial behaviours may be resistant to change. It was this latter point that was 'premature' to make according to this reviewer. In response to this comment, various claims were qualified in the article; for example it was made clear that the claim was not that prosocial behaviours *are* resistant, rather they *may* be resistant, and only at the time of the study, in other words results do not necessarily reflect current practice.

The points in the wider context of the literature reviewed in the introduction are:

1. ASD is considered to be a medical condition, a medical diagnosis is assigned and this must be assigned by a clinician.

2. ASD is considered to be a medical condition and diagnosis is framed as a way to access effective treatment by various health bodies, such as the NIH.

3. As outlined by the American Academy of Pediatrics (AAP) treatment should target core autism symptoms; promoting the development of communication, social, adaptive, behavioural and academic skills as well as lessening maladaptive and repetitive behaviours.

4. Decades of intervention research have not turned up robust evidence for effective interventions of core symptoms according to reviews (Francis, 2005; Jordan et al., 1998; Myers & Johnson, 2007; Warren et al 2011).

5. ASD diagnosis releases a variety of health and educational provisions ('treatment').

6. The study reported in Article 4 uncovered no differences in the trajectory of prosocial behaviour before and after diagnosis.

When, then, will it not be 'premature' to even suggest that prosocial behaviour may be resistant to service provision? Confusingly, the reviewer immediately went on to apparently contradict him/herself (emphasis added):

> *Indeed, socialization and prosocial behavior are difficult to remediate and change*, but perhaps, researchers have not yet identified appropriate interventions to develop these skills in children with ASD. I encourage the authors to consider the implications of their findings on intervention research as well as access to services after families receive an autism spectrum diagnosis.

The point about whether provisions/treatments were targeted at social skills seems rather circular. If effective treatments to improve autistic treatments to improve autistic children's skills were available, would they be in widespread use? For example if there was a drug with no side-effects that meant that

autistic children could socialize easily in an appropriate manner, with no social anxiety, would it be in general use? Presumably, given point 2 (above), and the continuing search for interventions aimed at ameliorating social deficits (see Charman, 2011) the answer is yes. The fact that effective interventions for core symptoms have yet to be established is most likely the reason provisions were not specifically designed to enhance socialization. If such provisions had been available, they would be in use.

The article in its current form concluded that additional service contact engendered by ASD diagnosis *at the time the study was carried out may* have been ineffective *if* (as outlined by AAPs definition of treatment) it was targeted at (pro)social behaviour (emphasis added). So a number of qualifiers were added to the article in the light of reviewers' comments. A further qualifier was that service contact could have occurred many months or even years before diagnosis as several studies have exposed long delays from point of first concern to time of actual ASD diagnosis.

v) Article 5: Contributions, merits and limits

Article 5 was a direct attempt to address the research questions formulated in the introductory chapter, which were:

Are parents active agents in the process of obtaining a diagnosis of ASD for their child?

If so, are there dilemmas that parents consider when weighing up whether to act to pursue a diagnosis?

What triggers the parents to ask clinicians to make the diagnosis? Is this affected by personal or socio-economic factors in the situation of parents?

How do parents, whose children have received an ASD diagnosis, perceive the diagnosis as helpful or stigmatising?

What does diagnosis actually do for these parents; what function does it serve?

A major obstacle to the generalisability of findings from the qualitative research was the small sample size, making it more appropriate to describe this research as a pilot study designed to generate questions of interest for the future. Even though the findings from this study have been presented throughout this thesis and in article 5, as if widely generalisable, this major caveat should be borne in mind throughout when interpreting this research.

First, it did indeed appear that parents were often very proactive in determining whether their children should receive a diagnosis of ASD. For example, one mother stated:

> So I could, now, I still believe I could play it any way I wanted to. So if I wanted right now to go and get Aiden diagnosed...I could.
> (Mother of undiagnosed child)

Article 5 highlighted how parents have a sophisticated understanding of labels and will balance the perceived benefits and possible risks of applying a diagnostic label in order to facilitate what they perceive to be the best outcome for a child, replicating similar findings in other studies (Rogers, 2007). The analysis went further than these studies, showing that where some parents may prefer a transient informal label, others may gain more from a medical diagnostic tag of ASD, and these positions may shift with time and circumstance. Parental network groups exist both advocating (e.g. nas.org.uk) and rejecting (e.g. ablechild.org) diagnostic labels for childhood disorders such as autism. This echoes Epstein's (1995) discussion of strategies used by politically motivated health identity groups, and here parents can be considered as part of wider 'autistic culture' (Silverman, 2008) and act to improve the social standing of their children.

There was a general willingness on the part of parents whose children had formal ASD diagnoses to talk about ASD in relation to their children. This echoes Bumiller's account (2009) of an 'anti-normalisation' strategy: whereby politically mobilized groups consciously bring a previously unacceptable behaviour or taboo subject into the public realm in order to make it acceptable. For example this strategy is adopted by 'lactivists' who encourage more women to breast feed in public thus making a taboo behaviour more visible; normalising it and lessening the stigma of performing the act. Bumiller (2009) reports on how the

autism right movement attempts to raise the visibility of previously unacceptable behaviours, describing such tactics as 'anti-normalization' strategies.

In contrast parents of undiagnosed children were unlikely to discuss their children's behavioural problems with even their closest friends and families. A major concern was protecting their children from an unwanted autistic identity. Table 1 reports major trends showing difference between the two groups.

Table 1- Summary of trends in differences between the two group and their positions.

Dominant position on..	**Mothers of undiagnosed children (1)**	**Mothers of diagnosed children (2)**
ASD diagnosis	Aim to keep 'normal' identity intact	Loss of normal child- 'grieving'
Justification	Justify position: adopt sociological discourse	Justify diagnosis: adopt both biomedical discourse and social model
Responsibility	Blamed for child's behaviour, but 'state' is maintained as temporary	Blamed but biomedical model provides weapon to counter this, behaviour becomes 'trait' like
Recruitment	Hard- hostility and defence	Easy- in interest to engage
Disclosure of child's difficulties	To no-one	To many
Preferred labels	Naughty	Autistic
View of ASD	As weakness/frightening	As bringing strengths together with weaknesses.
Language to describe diagnosis	'a millstone around their necks', 'pigeonhole' , 'put him in their little boxes' , 'he has to be branded' 'some fat label stuck on her'	'a key' 'a passport' 'a lever'

Although not as original as the first two studies in a methodological sense, as many other studies have adopted grounded methods to analyze the parental experience of childhood disability (see for examples, Harbourne, Wolpert, & Clare, 2004; Kelly, 2005), Article 5 did have a fairly distinctive sample. The recruitment of parents with non-diagnosed children (where their children had been identified as possibly on the spectrum by education or health professionals) meant that the study encompassed an important perspective which is often neglected in research as patients who do not reach the clinic are hard to recruit. In all the literature surveyed, only one other study, by Landsman (2005), was found that had recruited a similar sample (parents of undiagnosed children with disabilities). It was the difference between the perspectives of the parents of undiagnosed and diagnosed children that led to the substantive findings from the study.

Article 5 reported how parents without diagnosis experienced considerable dilemmas, and utilised anti-labelling discourse to support their positions. This was a discourse shaped by the social psychological and sociological literature reviewed in the introduction, on SFPs and stigmatization in particular. These ideas were articulated in their own words to justify their positions. However labelling theory, although a useful background, was not tested in any sense by any of the research undertaken. It could not be ascertained if any negative aspects of labelling did occur, rather the ideas behind it formed a discourse which was adopted by parents resisting diagnosis. This was clarified in the revised version in the light of reviewers' comments (see thesis Appendix 1).

Parents whose children had a formal ASD diagnosis, on the other hand, were not only unqualified in their advocacy of ASD diagnosis, but deployed strategies to actively improve the status of children with ASD as a group, within society as a whole. Thus it was argued that their actions may make it more likely that others will opt for diagnosis in the future. This reappraisal and reframing of ASD in a more positive light was interpreted as a form of parental optimism, as discussed earlier.

The sample size in Article 5 was small, therefore it was tentatively concluded that findings might illustrate one psycho-social process – the active strategies adopted by parents of children with ASD diagnoses – which could have contributed to the rising prevalence of ASD over the past 30 years. Such strategies could be deployed to influence the balance of pressures in the eyes of parents of undiagnosed

others (Figure 2, Article 5). Of course an all encompassing explanation for the increasing prevalence of ASD would almost certainly include a complex web of interactions including social, cultural, historical, and possibly environmental/biological factors.

The strategies of parents of diagnosed children as a group were reminiscent of the social model of disability in that parents sought to de-stigmatise ASD as a named condition by changing the reaction of the 'disabling' society. In other words, they fought against their children's disablement by raising awareness and framing ASD in a more positive light. Despite this utilization of the social model, parents still regarded ASD as having biological (neurological) causes. This echoes the work of the ARM, where neurological differences are re-conceptualized as beneficial, and society's lack of tolerance is overtly challenged (see introduction, section 2, p. 40-43). In this sense the parents in our study were part of the emerging 'autistic culture' described by Silverman (2008). Rather than passively accepting that the diagnostic label was stigmatizing, they took action in various ways to transform its connotations. This is consonant with Farrugia's (2009) perspective, where stigma is not a fixed mark, but a property of societal attitude that can be altered with time and effort.

Returning to the initial research questions, some parents did speak about potential triggers that led them to seek ASD diagnosis. In particular one participant who later started the process of reassessment of her son, in order to get him diagnosed, described how a marital crisis, her perceived need to secure academic support for her son, and renewed professional pressure had convinced her this might be a good idea. She also described how she believed children with AS diagnoses were less stigmatised than in the past, due to increasing awareness. However such findings, although concurring with literature reviewed were too local to generalise.

One function of ASD diagnosis was reflected in the way parents of diagnosed children described it as 'a passport' to secure resources. Such resources included educational resources, specifically one to one support in class, social resources e.g. access to support groups, health services e.g. mental health services, access to information (mediated by naming the condition as discussed) and financial resources e.g. child benefits. This list was very similar to the resources listed as potentially beneficial to parents in the earlier literature review, suggesting that service access and motivations were similar in this group to those

uncovered by previous studies. These reports illustrated the function that ASD diagnosis actually performed for the parents enrolled in the study.
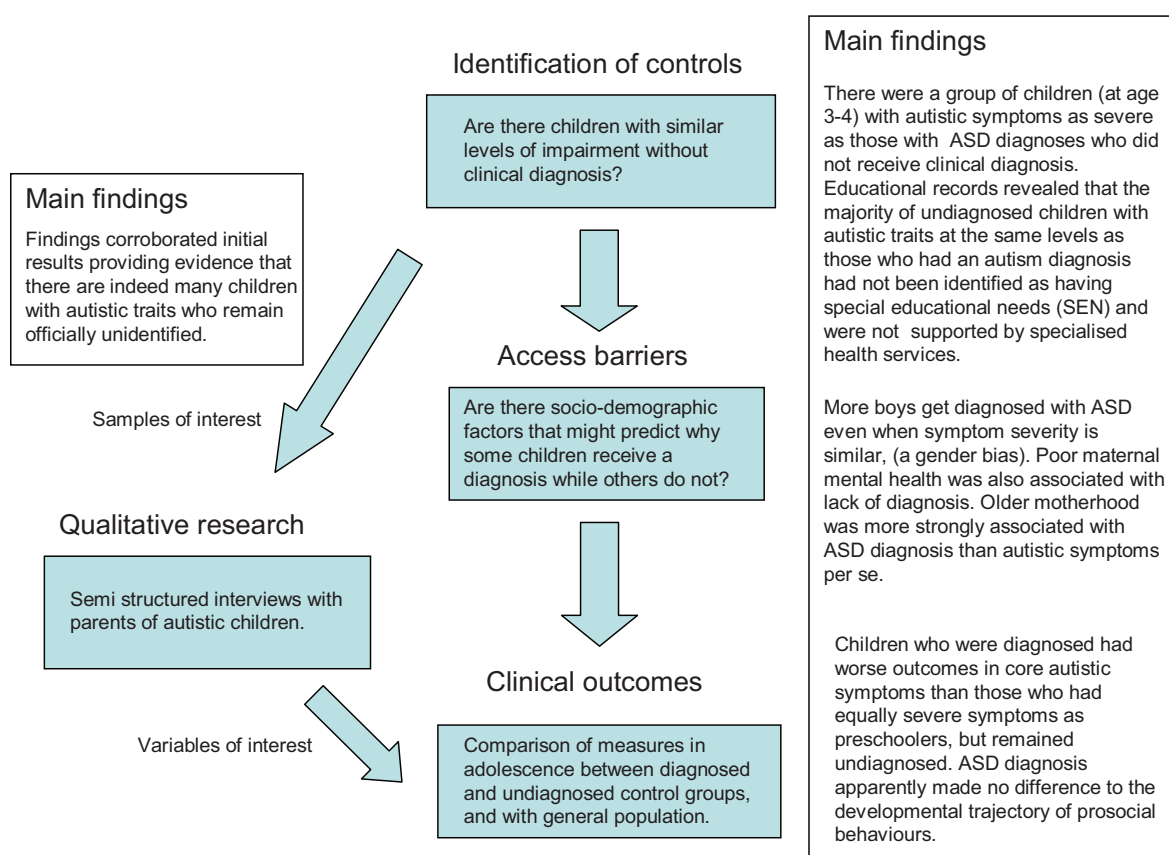
A second function of diagnosis was to reattribute causes of autistic-type behaviours to biological factors. Parents of diagnosed children deployed a neurological model of ASD to successfully deflect accusations that their child's inappropriate behaviour was due to their bad parenting. Explaining a child was 'autistic' effectively attributed the child's behaviour to integral biological, within-child factors. As discussed in the introduction, a similar process has been associated with both ASD and ADHD, leading Hinton and Wolpert (1998) to describe ADHD as 'the diagnosis of forgiveness'.

Various revisions to the paper were made in response to the reviewers' comments and these are documented in thesis Appendix 1. The most useful of these were references to the work of Sara Ryan, who has written extensively about the advocacy role of parents of autistic children, and is herself the parent of an autistic child. A meeting between Dr Ryan and the author subsequently took place. Dr Ryan is one of a loose alliance of medical sociologists who are also parents of disabled children. The literature by these authors provided a window on the conflicting narratives reported by various researchers with parents either as viewing their children as 'gift-like' or 'tragic'. On the one hand, narrative threads running through accounts emphasise the joy that such a child can bring (Stainton & Besser, 1998), their strengths (Dura-Vila, Dein & Hodes, 2010), and the personal development and positive life experience of caring for such a child (King et al., 2006). On the other hand, parental reports describe how such a child limits social and economic opportunities (Todd & Jones, 2003) and confers additional stress on the whole family (Runswick-Cole, 2007). Article 5 concluded such apparently conflicting narratives were hardly surprising given that parents appeared to adjust their view after ASD diagnosis according to circumstance and time. Perhaps the ambivalence of mothers toward their children, described by Parker in 1997, where mothers both love and resent their children, is heightened in the case of a child with autistic-like behaviours. Children with any type of impairment are more likely (than non-impaired children) to be cared for primarily by mothers rather than fathers (Cohen & Petrescu-Prahova, 2006).

Section 2: Merits and limits of the quantitative and qualitative approaches

In the previous section, some of the strengths and limitations of each article were presented. In this section, the strengths and limits of the mixed methods design as a whole will be considered in more depth. A schematic diagram of the overall research design is given in Figure 1.

Figure 1: Overall research design

In several ways, the two studies provided complementary strengths; however, each method also exposed fundamental flaws in the epistemic interpretation made by the other.

Silverman (2006) describes the benefits and possible uses of mixing quantitative and qualitative methods. Quantitative methods may be used to establish samples of interest for qualitative research; and qualitative

work is used to explore questionnaire design, for example when piloting a diagnostic tool. Quantitative data may also be nested within a wider qualitative study; a larger study size in study 2, for example, may have allowed some statistical analysis of attributes of parents presented in Table 2 of the thesis introduction, or cross-comparison with themes emerging from the analysis.

In the overall design, quantitative and qualitative research were combined, beginning with the first stage of the quantitative study in order to establish the broad contours of the research questions. In study 1, quantitative methods were used to establish the phenomena at hand (the existence of undiagnosed children who display autistic behaviours) and were used to generate subsequent research questions for the qualitative study, for example *why are there so many undiagnosed children with impairments at the same levels as diagnosed children on the spectrum? Why have these children not been diagnosed?* Thus the samples required for the qualitative work were also identified (parents of undiagnosed children with autistic behaviours compared with parents of children with an ASD diagnosis). The subsequent qualitative work was able to add breadth, depth and richness to the quantitative line of inquiry. Silverman (2006, p.293) points to the value of qualitative data in these circumstances, adding 'there are areas of social reality that statistics cannot measure'.

The existence of a group of undiagnosed children who in both the qualitative and quantitative studies appear to be on the spectrum, in its broadest sense, is an instance of different approaches which focus on the same phenomena. In this sense the two studies corroborate each other and together provide strong evidence that there are indeed many children with autistic traits who remain officially unidentified and unsupported by services.

The qualitative study revealed that parents of children with an ASD diagnosis were particularly concerned about educational attainment, levels of well being, friendships, self harm and aggression, bullying and school exclusion- all these outcomes, in fact, as their children became young adults. Thus the findings of this qualitative research, study 2, provided the rationale behind selecting these particular items for analysis in the outcomes analysis of study 1.
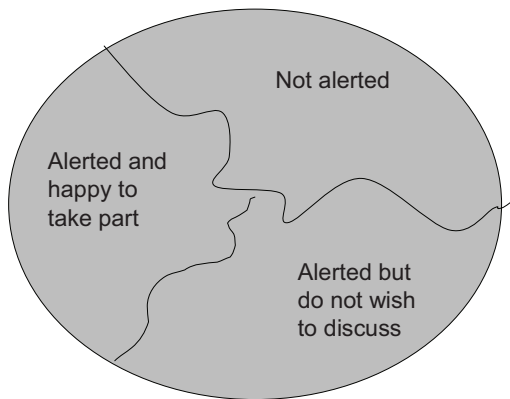
Methodological triangulation was consequently utilised in its broadest sense; quantitative methods showed data trends and qualitative methodology provided context and meaning to these trends. Methods

were mixed in a way that providing complementary strengths, with qualitative research and quantitative research both guiding and informing the other.

However, mixing methods also threw up several issues that were not easy to resolve. Although the one line of inquiry was supposed to feed into the other, this and other studies have shown that two such distinct sets of data cannot easily be compared. For example, Brannan (2004) noted how the form in which a question is asked alters the response that is given. In his research, he set out to explore why mothers of young children return to work, or not. Originally, this was conceived as a questionnaire survey, with measures of socio-economics status, mothers' educational attainment, age and other factors included. Brennan found that the questionnaire item about fathers' involvement (paternal contribution to child care) generally elicited positive ratings. Consequently this did not emerge as a salient factor in whether young mothers returned to work on not. When Brennan interviewed young mothers, however, data revealed it was the role fathers played in childcare that was of crucial importance in how supported mothers felt, enabling them to go back to work. This was an instance where global questions about a husband's participation in quantitative surveys elicited positive responses, but in depth face-to-face interviews elicited particular complaints and negative responses.

Another reason data cannot be directly cross-compared between the two studies was that the samples were not equivalent. Although the qualitative study sought to establish a sample which was parallel to that of the quantitative study; namely a sample of parents whose children had received an ASD diagnosis, together with a group who were 'on the spectrum' but had not received a diagnosis; the groups were established in completely dissimilar ways. The method used to recruit the children in the comparison group in study 2; via educational psychologists and SENCOs' assessments, was nothing like the method used to identify children in study 1. The suggestion by professionals and parents that recruitment might impeach their own relationships with parents of undiagnosed children, suggests that the sample in study 2 represented a sub-sample of parents of undiagnosed children, those prepared to talk, as illustrated in Figure 2. Even within this very small sample some parents were more actively resisting diagnosis, whereas others were ambivalent about the assessment process, adopting a 'wait and see' approach until assessment, often a very lengthy process, was completed.

Figure 2: Schematic diagram of the population of parents whose children display autistic symptoms but remain undiagnosed. Dividing lines do not represent estimated proportions.

To summarise, the overall design of the qualitative study attempted to mirror the quantitative methodology by identifying comparable samples in order to add 'depth' to the line of inquiry as advocated by Silverman. The reality was that the differing nature of the methods used meant that the samples were not composed of the same population of parents, and the diverging form of the inquiry meant information obtained was often of a different nature.

Findings from study 1 also exposed fundamental flaws in the epistemic assumptions made when reporting study 2, and vice versa. In study 2, the qualitative data apparently provided evidence that parents were attempting to present their children's behaviour in certain ways according to what they wished to achieve. For example, in some cases parents presented similar behaviours as autistic or as not autistic; perhaps according to whether they wanted to promote their child as having autism or whether they wanted to block a diagnosis for their child:

> She can do this thing where if you try and talk to her she won't look at you. I don't particularly think she's being autistic, I think she's trying not to make eye contact. I think she did that at school quite a bit.
> (Mother of undiagnosed child)

It's obvious- because he doesn't make eye contact- his diagnosis is screaming out at me you know.
(Mother of diagnosed child)

Of course it is not possible to state that the behaviours described were equivalent, this depends on many other factors, and whether they could be deemed 'autistic' the persistence, the motivation behind the behaviour etc., factors that mothers were well placed to judge. In other words it is not possible to establish whether mothers were reporting real differences from the quotes above, or whether there really was some element of maternal 'interpretation' involved. Because of this, some analysis of the motivations and meanings of individual accounts took place, akin to a critical realist analysis. Behaviour, it was suggested, was perhaps 'framed' in a certain light, or exaggerated by parents to get the desired outcome for the child. In some cases such representation was explicitly described. One mother reported on her attendance at the psychological assessment of her child thus:

From my point of view the whole assessment was endless questions they asked me -it's very difficult to answer them completely honestly if you actually don't want your child to be diagnosed as autistic. I think this is semi-subconscious and this is analyzing it looking backwards, I didn't sit there thinking, 'I'm going to fake this'. Because otherwise I wouldn't have gone. But I think that I probably wasn't as truthful as I could've been.
(Mother of undiagnosed child)

These data suggest that in some cases, a situated interpretation of children's behaviour may have occurred at an unconscious level. If this occurred, it throws into question the accuracy of the data from the parental report measures made in the cohort study measuring children's behaviour. It is possible the agendas of parents themselves may have biased reporting of their children's difficulties according to how they wished their children's behaviour to be perceived.

Because ALSPAC is such a large study and answers to questionnaires are confidential and anonymous, proponents of these type of longitudinal cohorts would argue that as parents have nothing to gain from being inaccurate in their descriptions of their children: there is no implication from how they describe their children, these types of biases are minimal. Nevertheless, if biases in reporting are unconscious they might be picked up in questionnaire data. What study 2 revealed was that some parents pushed to have

their children diagnosed, whereas other were active in protecting their children from this. Other studies have shown there are systematic biases dependent on *who* is reporting, for example, teachers give children's behaviour different ratings in to parents (Papageorgiou, Kalyva, Dafoulis, & Vostanis, 2009).

It was not just the qualitative research that exposed the limitations of the quantitative methodology, the reverse was also true. The ALSPAC data revealed biases in retrospective reporting that threw the validity of data collected using qualitative methods into question. For example, in ALSPAC, speech difficulties were reported by parents both when children were toddlers and when children were teenagers. When the children were teenagers parents were asked to report retrospectively whether their children had any speech difficulties when they were toddlers.

In the diagnosed and comparison groups, the proportion of parents who reported that their children had speech difficulties was roughly the same for preschool children who subsequently received an ASD diagnosis as for controls who did not, χ^2 (3)=1.87, *p*=0.599. So, roughly the same proportion reported speech difficulties in cases and controls, around 80% in each group. However when a similar question was retrospectively answered: that is, parents were asked whether their teenage children had speech difficulties *when they were toddlers*, less than half of the same group of parents in the comparison group recalled there having been any speech problems when their children had been toddlers. Because of this effect of retrospective reporting significant differences did occur between the two groups χ^2 (1) = 10.15, *p*=.001. Apparently, then, parents in the control/comparison group, whose children had fewer ongoing problems, 'forgot' the difficulties they reported when their children were toddlers.

The results described above challenged the validity of qualitative data again in terms of it being an accurate reflection of real events, which employed retrospective reporting. The qualitative data were gleaned from interviews with parents conducted several years after an ASD diagnosis was made, or a professional had suggested that the child might be on the spectrum. Thus, findings from one set of results undermined the other as a 'true reading' of reality. In both methodologies, the idea of a 'true reading' is fundamentally flawed. It can be seen from the findings discussed above that parents interpret reality in different ways, as indeed researchers interpret data in different ways (Kuhn, 1996, p.63).

The merits and limitations of using secondary data

The main limitation of using secondary data, as has become apparent, rests in the incapacity to design a study specifically tailored to answer precise questions. Instead, the process involves thinking about the data available, then considering how it can be used to answer an interesting question: 'data mining' in a sense. The limitations discussed in the sections on individual articles, highlighting how the sample size became too small to uphold methodological principles as the study progressed, and the lack of specificity in the ASD measures used, were prime examples of this limitation. Data availability was key. What has actually been recorded in any particular cohort study is dependent on who was studying what at the time the cohort questionnaires were designed. If a researcher received a grant to explore asthma during the set up of questionnaire design, there may be very detailed information on asthma, but little on repetitive behaviour. Different psychometric tools fall in and out of fashion. This problem is further exposed when attempting to use data from more than one country, there is a lack of standardisation in measures used. Recently, moves to standardise data sources, particularly in Europe have been made, for example the European Longitudinal Study of Pregnancy and Childhood which has data sources from over seven participating countries. There are obvious advantages to using secondary data which are worth recording. These include the vast amounts of data available, from sources that are increasingly open access. Most cohorts provide data that is already data gathered, coded, and cleaned. An enormous amount of time and expense is spared if researchers are able to address questions effectively without designing and implementing new studies from scratch, and cohorts together with other pre-existing sources of data drawn from a variety of countries, continue to address questions of global significance in biomedicine and beyond.

A further question is whether secondary data were adequate to test the central hypothesis as initially conceived. The analysis of secondary data did not provide any concrete evidence that diagnostic labelling led to better or worse outcomes. But would the methodology have tested the hypothesis adequately had all the data required been available, or was the methodology itself fundamentally flawed? Crawford and Vignoles (2010) argue that there is a pressing public policy need to compare the performance of children who receive specialist educational provision with a control group with similar difficulties who do not. They discuss the 'classic evaluation problem', namely it is difficult to establish an appropriate control group. Angold and colleagues (2000) and Lambert and Bickman (2004) also discuss this problem with reference to child psychopathology. Theoretically, according to these authors, there is no intrinsic

problem with assessing outcomes by using matched controls, in fact it is deemed essential to adopt such a design and assess findings in tandem with other forms of evidence, e.g. randomized controlled trials (RCTs).

In the medical literature and in systematic reviews RCTs are considered the 'gold standard' when considering the efficacy of various interventions for medical conditions, including childhood neurodevelopmental disorders such as ASD. However RCTs are often carried out under highly controlled conditions and do not show how effectively interventions may be implemented in 'real world' settings, or how they interact with other specific interventions, forms of service provision, reactions of others etc. that may be engendered by the intervention under scrutiny as it is applied in practice.

The philosopher of science Nancy Cartwright (2008) cites the example of bicycle helmets to point out limits of RCT methods. Case-control studies suggest that cyclists wearing helmets have fewer head injuries than cyclists not wearing helmets. Conversely, longitudinal analysis in states that have passed helmet laws do not show a clear decrease in numbers of head injuries after helmet laws have been implemented, and in some cases there is an increase in head injuries. Cartwright elucidates that although RCTs illustrate what happens under research conditions, once the helmet law is implemented state-wide, helmets become a common sight, and driver behaviour is altered; drivers become blasé about cyclists with helmets giving less space to cyclists, and cyclists take more risks (a 'false sense of security' phenomenon). Cartwright (2007) argues that RCTs can therefore never reveal the full extent of causality when interventions are implemented in practice.

On diagnosis or identification of ASD, a package of educational provisions, including one to one support and/or placement in a special school, together with support for families, specific therapies, and additional health care services are released. In addition, these services are combined with the recognition of the condition and the differential treatment of the child that this may bring. So ASD diagnosis does not always engender specific interventions or medical 'treatments' like the medical diagnosis of asthma, for example, rather it leads to a complex package of services. In education these may be used to relieve pressure on mainstream teachers (Blatchford, Bassett, Brown, & Webster, 2009), parents, and to provide some adaptive teaching. In effectiveness research as recommended by Kelley, Nixon and Bickman

(2000), it is virtually impossible to determine what the effect of each individual component within the whole package of services might be.

A further limitation of secondary data is that there is never going to be a perfect match between cases and controls. Consequently, critics can always point to inadequate matching as a flaw in the methods. A second inherent criticism is in terms of the time lag between results and current practice; any longitudinal study will necessarily be looking into the recent past, and again critics may argue this will not reflect the effect of current practice. Nevertheless this thesis argues, there needs to be some assessment in real world settings of the package of services currently in use to support autistic children, using a matched control group, rather than relying solely on RCTs for evidence of efficacy of specific interventions. Both methods have strengths and both have limitations.

In ASD research the question of effectiveness of services seems particularly important when, as discussed in the introduction, policy documents and autism charities stress the importance of (early) ASD diagnosis because it leads to appropriate (helpful) action. This is accompanied by an emphasis on early diagnosis and funding for research to deliver diagnostic tools which identify ASD at ever younger ages. Often publications reporting such efforts note the 'importance of early identification and treatment', for example Bryson, Zwaigenbaum, McDermott, Rombough, & Brian, (2007, p.731). These authors cite three studies as evidence for the statement above, none of which presents conclusive evidence as to which approach may be best, and one of which concludes 'the field does not yet have a treatment that meets the present criteria for well-established or probably efficacious treatment' (Rogers, 1998, p. 167). As various reviews have noted, most recently a systematic review of early interventions for ASD in the May 2011 edition of *Pediatrics* (Warren et al., 2011), there is little evidence for positive effects of pharmaceutical or behavioural interventions for ASD. Furthermore, little is known about which sub-groups of children best respond to various treatments.

It is worth reflecting on what treatments for ASD are trying to achieve. According to the AAP, the aim is to improve core symptoms, in other words, the aim is to treat 'autism'. In a recent editorial of the *Journal of Child Psychology and Psychiatry* (2011), the autism researcher Tony Charman reviews the 'new wave' (p.22) of intervention studies that focus specifically on enhancing social communication outcomes. Some of these have had promising results, (Dawson et al., 2009; Landa et al., 2011) however, Charman notes

that the improvements were in proximal behaviours, rather than autism severity, and studies that have measured outcomes using the ADOS, have not seen ADOS measures ameliorated (Dawson et al., 2009; Green et al., 2010).

The discussion above relates to behavioural interventions for ASD, a subject, as reviewers commented, outside the scope of the research presented in this thesis. Many parents (as Article 5, and other sources reviewed in the introduction to this thesis) suggest, may see educational provision as a crucial reason to secure an ASD diagnosis. What, then does educational provision provide in relation to social deficits? It is not possible to pinpoint the exact influence of any type of provision from the results presented in Article 4, although overall prosocial behaviour appeared to be unaffected by diagnosis in the group under study. In Article 3 a low-SEN provision group performed significantly better in the measures of social communication available than the diagnosed high-SEN provision group, even when the diagnosed sample was limited to children diagnosed early, before aged 6, so autistic like behaviours were matched in around the same age ranges (3-4) that diagnoses were applied (aged 1-6). These studies together provided weak evidence that SEN provision might not be effective in improving prosocial behaviours and other social behaviours as measured by the SCDC, for example. However the limitations of the method and different interpretations possible do not exclude the possibility that differences in outcome were a result of divergent developmental trajectories rather than differences in SEN provision. Indeed, as already noted, the fact that some children in the control group received speech and language therapy when young might be seen as evidence that this therapy is successful for some children.

The weak, though inconclusive, evidence of the limited effect of SEN provision engendered by diagnosis might be interpreted in the light of Chrissie Rogers' (2007) criticisms of SEN provision as focussing on supporting educational, i.e. academic, outcomes whilst neglecting the needs of children who have difficulty socialising in the playground. Tamsin Ford, a child psychiatrist comments on the need for support at school for any autistic child who 'might function perfectly in the classroom, but you find in a social unstructured situation they are completely lost. ….in terms of their capacity to develop, their social skills are such are dipped below their peers, without intervention that is going to have a major impact of their life trajectory' (T. Ford, personal communication, September 2009). Although touched upon, ultimately, the question of effectiveness of such support for social deficits in school as currently practiced is beyond the scope of this thesis.

Section 3: Reappraisal of the biomedical and sociological perspectives on ASD

This section will briefly review what is meant by 'biomedical' and 'sociological' perspectives. First, the biomedical model of medicine rests on the assumption that the body can be repaired like a machine; so it adopts a mechanical metaphor. It is reductionist in that explanations of disease concentrate on biological causes and neglect social and psychological factors. Biomedical understandings of ASD originated from Kraepelin's classification of mental disorders as discussed in the introduction (p. 14-17). These systems of classification were underpinned by the belief that mental disorders were like disease in that each disorder shared a distinct aetiology. The biomedical model portrays ASD as 'treatable'; health bodies like the United States National Institute of Health prioritise 'treatment' of ASD.

There are various 'sociological' perspectives on ASD reviewed in the introduction. First, the social model of ASD separates disability (engendered by a discriminatory society), from biological (neurological/genetic) impairment. Second, social theorists have argued that diagnostic labelling itself may have unintended negative effects on those who are labelled. Third, social constructionists maintain that ASD as a category has developed in reaction to a changing social and political context. The extension of the autism spectrum and younger diagnosis is viewed as part of the wider process of 'medicalisation'. This term describes the process by which non-medical problems become understood as disorders.

Both the undiluted biomedical model and the radical social construct perspectives are easy to criticize in the light of the submitted work and the various sources of literature reviewed. Firstly, the nature of ASD contradicts major assumptions of the biomedical model as described above. There is no definitive biological test for ASD, such as a blood test. ASD is behaviorally defined. In most cases the exact aetiology of ASD is uncertain. Recent reviews in autism research (Happé et al., 2006) suggest it may be time to give up on a single causal explanation for autism; instead many different causal pathways may lead to the same symptoms. Most 'causes' are best thought of as risk factors that increase the likelihood of a disorder without guaranteeing it will occur.

A further criticism of adopting a purely biomedical perspective on ASD is there is no 'cure' for ASD, and arguably, not even any 'treatments' for core ASD symptoms which are scientifically supported (Charman,

2011). Given the above, should ASD be considered as a medical condition at all? This is the question posed by radical constructionists such as Timini (2010), who argues the medical concept of ASD is invalid. But ASD, and particularly childhood autism have been shown to be highly heritable (Muhle et al., 2006). The brains of autistic people are anatomically and physiologically different to those of the general population (Penn, 2006). Work in this area has provided strong evidence for real biological causes i.e. neurological deficits and genetic predispositions to ASD.

What has this thesis contributed to such sociological/biomedical understandings? The research presented in Articles 1, 2 and 5 clearly emphasizes social influences in the diagnosis of ASD. This, to a social theorist, would provide evidence that ASD is to some extent constructed, in that ASD diagnosis apparently occurs more often under certain social and demographic circumstances. According to the findings presented here, such factors may include the mental health of mothers, gender of children and parental attitude. Educational and school based factors such as academic progress also emerged as potential triggers. Social constructivists point out that the prevalence of ASD has risen at the same time as society's emphasis on raising performance standards in social communication, intellect and language skills in children (Klotz, 2004). There is much literature about the damage this has caused those who struggle to learn and behave according to dominant norms (e.g. Dockrell, Peacey, & Lunt, 2002).

In the UK, once diagnosed, 'treatment' for ASD is under the jurisdiction of both health and educational services. Clinicians, psychologists both in educational and health services, child health workers, teachers and specialized therapists are all involved. Some tensions between the aims and needs of health and educational services are apparent. Ironically, after the 1981 Education Act emphasized the importance of identifying needs of individual children, clinicians who may prefer to give a functional description of an individual child's strengths and difficulties may be encouraged by colleagues in education to apply simple diagnostic labels in order to release educational resources (A. Emond, child paediatrician, personal communication, January, 2010).

The thesis as a whole draws attention to social processes that may underlie ASD diagnosis yet also highlights biomedical underpinnings. Results from Article 4 point to resistance of impairments in prosocial behaviour to service provision triggered by diagnosis in the recent past. As discussed in Article 4, and this conclusion, the continuing spotlight on finding effective therapies illustrates how despite

decades of research, undisputed and truly successful treatments for social deficits have yet to be developed. Perhaps this effort indicates how resistant these behaviours may be to intervention. This might be interpreted as evidence that such traits are stable i.e. more trait-like than state-like (Article 4). Such an interpretation would perhaps tend to point to biomedical, (neurological and genetic) underpinnings to ASD.

One contribution of the research rests with its evidence to sustain an understanding of ASD as biological *and* social in origin. The body of work as a whole therefore supports conceptions of ASD that utilise the bio-psycho-social model.

Morton and Frith (1995) outlined a more interactionist approach where different levels of explanation – biological, cognitive, behavioural – each interact with the environment. However, despite acknowledging the role of the environment in the developmental process, Frith (1999) went on to conceptualise neurodevelopmental disorders as existing from birth, and explicitly states that their 'origins' are biological, concluding there is evidence for a 'genetic' basis and a 'brain' basis.

More recently, Hulme and Snowling (2009) incorporate a systems approach into their aetiological model for childhood disorders. Recent advances in systems biology have shown that the environment of the cell affects gene expression and protein synthesis at molecular levels within cellular metabolic pathways. Thus environmental influences can alter 'core' biology: for example Mack and Mack (1992) describe how tweaking rats' whiskers changes gene expression in the sensory cortex. In systems theory, genetic influences are conceptualised more like a set of piano keys where the notes are played or not played, played slowly or quickly, and there is enormous variation in the music produced even with the same basic set of keys. Hulme and Snowling therefore describe how difference is thought of as *both* biological *and* as a product of the social world. Clearly, there are biological differences between children with and without ASD, but that as well as considering how the biological affects the social (e.g. social behaviour), it is also plausible that the social may affect the biological. The bio-psycho-social model supported by this thesis assumes that ASD is the label given to an atypical clustering of functional characteristics (with negative impacts) that arise from the interaction of genetic and environmental factors though a developmental process and whose expression and interpretation also depends on the social context.

The second 'sociological' perspective considered (outlined in the introduction) stems from labelling theory; the idea that diagnostic labelling might have unforeseen negative consequences for children. Although testing the premise of labelling theory was the initial objective, in the final analysis, this thesis provides no evidence to support the concerns of labelling theorists regarding ASD. However, literature reviewed in the introduction provides empirical evidence for expectancy effects and stigmatization which could have potentially damaging consequences. It is beyond the scope of this thesis to ascertain if such concerns outweigh the valuable benefits of service provision engendered by diagnosis, as advocated by most health and education recommendations.

Research reported in Article 5 did demonstrate that the possibility of negative impact of diagnosis is of *concern* to parents. Indeed, some parents adopted the various discourses of labelling theory and social constructionism to justify resistance to ASD diagnosis. In these terms, parental understandings were far more nuanced than the bold ideas laid out by Scheff (1974) concerning labellling. For example, parents saw some labels as more stigmatizing than others, a tacit understanding, of a hierarchy of labels and conditions. 'Autism' was perceived as more damaging than 'Asperger's Syndrome'. 'ASD' generally was viewed as more damaging than 'pragmatic language disorder', or 'speech difficulty'. Parents without ASD diagnoses discussed a number of potentially negative consequences of labelling together with potential benefits. Parents (where a professional had expressed an opinion that their child was had AS or was autistic) did experience dilemmas over diagnosis as predicted by the literature review and they were often actively involved in pursuing the desired outcome for their child. Although this thesis provided no empirical evidence to support the claims of labelling theorists regarding ASD, it did reveal this theoretical stance to have continuing reach and influence on parental perceptions.

The final 'sociological' perspective on ASD that has been reviewed is the social model of disability. One function that ASD diagnosis has for parents is to provide legitimacy to the impairment by invoking its biological origins to those who are skeptical of the construct (Article 5). On a political level, a label may allow an individual to prove the validity of their impairment, and form focused networks, or act as individuals to challenge the society that is perceived as discriminatory. In this way a largely biomedical understanding of ASD *impairment* served parents with diagnosed children to press for a reduction in disablement by wider society. Parental strategies serve to reconstruct what was once viewed as a negative label into a more positive one. Landsman's work (2005) is primarily a discussion of whether parents

utilize social or biomedical and genetic models of disability. She finds they use both, as qualitative research presented here also suggests, according to what position is being justified or what outcome targeted at a particular time.

Possible deployment of work (if any)

The summary of the contribution made by each individual article in section 1 of this chapter only partially addresses the question of the contribution made by the body of work as a coherent whole. When taken as a whole, the impact of these articles must be assessed in the context of current debates in autism science and social science, psychology, sociology and medicine. Their impact and contribution, if any at all, will be partly determined by the wider social, historical and political context.

Any 'contribution to knowledge' is itself historically and culturally determined. Jasanoff (2004) argues against the concept of science as a straightforward vehicle for accumulation of knowledge, instead conceptualizing knowledge generation as a process of co-production, in which a piece of knowledge is co-produced at many time-points by researchers, together with those who disseminate findings e.g. the media, those who influence modes of dissemination e.g. publishers, and those who interpret it (the audience). Perhaps a more salient question in this interpretative tradition is 'how (if at all) will the body of work provide insights that will be interpreted or deployed by different actors in the future?' This is especially pertinent when considering 'knowledge' about mental health. It is therefore hard to position the contribution of the work as a whole without the brief overview of the changing status of 'knowledge' about ASD as reviewed in the introduction.

The brief review highlighted the differing agendas of groups concerned with ASD. How would these groups be likely to react to the submitted body of work? First, charities such as *Autistica* might argue that the earlier work highlights the extent of the problem of unidentified children; that many children may be missing valuable services and support. If there are children with autistic symptoms who do not reach the clinics it is easy to make the assumption that these children are prevented from accessing valuable services and consequently reaching their potential. It is possible the body of work will be used, particularly by those who support early identification, to justify more diagnosis or to promote optimum social conditions for diagnosis to occur.

Some medical researchers might also stake this claim as in much medical literature 'access barriers' to clinics, such as those described by Flischer and colleagues (1997), seem to be implicitly assumed to be a bad thing. The question for such biomedical researchers is 'how can we bring more children to the clinic?' But as Feinberg and Vacca (2000) point out, when considering policies on childhood ASD there is often an 'erroneous belief that the more time professionals spend with autistic children, the more likely the child is to have a positive outcome' (2000, p.135). A similar point was made in the psychiatry literature by Henggeller and colleagues in 1994: 'the dearth of clinical outcome data in the services research literature raises the frightening possibility that evidence of increased access and variety of services may be construed as a proxy for quality and effectiveness of clinical services rendered' (Henggeler, Schoenwald, Pickrel, Rowland, & Santos, 1994, p.230). Lauchlin and Boyle (2007) provide a useful summary of arguments and counter arguments for the use of labels in educational settings. They note how educators often presume that a label equals more money for teaching assistants or that a label equals placement at special school, without sufficient consideration of how resources are targeted in tackling children's specific difficulties. 'While labels may be indicative of educational problems', they write, 'it does not necessarily follow that they suggest relevant solutions' (p.37).

Groups such as the ARM, however, might have different ideas about how this research could have an impact. As anti-cure, they might emphasise that service provision for ASD released by diagnosis do not 'treat' social deficits (Articles 3 & 4 contributed to a lack of clarity as to whether service contact is effective in ameliorating core symptoms) and that 'treating' core symptoms may not be desirable. Such autistic self-advocates might position the work to argue that the continuing search for treatments of core ASD symptoms is misguided. A further group, researchers involved in developing specific interventions, might argue that the body of work serves to highlight how effective interventions have not yet been translated into practice, so the search for interventions that are effective, as well as their translation into the real world, must be intensified.

These speculations as to how different groups might engage with the work are conjecture. One further way the work may be positioned is to at least raise a question about the merits of early diagnosis.

The scientific and medical literature centres on how reliably an early diagnosis can be made, and the accuracy of screening checklists as tools. There are very few if any references to literature questioning the

merits of an early diagnosis of ASD per se. The value of early diagnosis would be hard to dispute if service contact/ provisions triggered by diagnosis were clearly effective in ameliorating symptoms for all children, and were welcomed unreservedly by the autism community. However the body of work submitted raises the issue of why there is such a push to diagnose early, when it is not obvious children with or without lifelong impairment are clearly distinguishable at early ages.

Clinical implications

The work has clinical implications, raising awareness of the social constraints within which clinicians assign ASD diagnosis and raising the question of what age an ASD diagnosis can be reliably assigned.

Given the heterogeneous nature of autism spectrum disorders and the 'anti-cure' stance of the ARM, the question of whether diagnosis is always useful must be addressed both in terms of the effectiveness of service provision for particular children, and whether all interventions are desirable in terms of their impact on autistic individuals themselves.

Amongst parents whose children have never received an ASD diagnosis, all were unreservedly positive about it. This finding, together with the literature reviewed in the introduction, indicates that diagnosis and identification may be of enormous benefit to parents. Parents were able to list a number of practical ways in which ASD diagnosis had aided them, listed in Article 5 and collated in the introduction. Parents themselves prefer a clear diagnostic label as opposed to functional descriptions of a child's strengths and difficulties as indicated by Brogan and Knussen (2003).

Nobody disputes that symptoms of autism can be distressing for both children and their families. Diagnosis of ASD, then, seems to incur obvious benefits to parents, in terms of their understanding, and access to respite care, resources and interventions, and attribution for their child's behaviour. A wider question raised by this research is whether what is of advantage to parents is always of equal benefit to children.

The school of thought in psychology that encompasses family systems theory regards the family as a holistic unit and relationships as an important factor in psychological health, whatever of the origin of the problem. They argue that regardless of whether the clients consider problems to be 'within-child' or

'family' issues, involving families in solutions is often beneficial (Friesen & Koroloff, 1990). In this model, benefits to parents would necessarily confer benefits to children. There is no doubt that ASD diagnosis is helpful to support wider family functioning, especially in the short term.

The work has highlighted why children may remain unidentified and undiagnosed with ASD; why some children may slip through diagnostic nets. The findings suggest that maternal depressive symptoms specifically around the time of a child's autistic difficulties might actually hinder diagnosis. Clinicians may become wary of dismissing concerns about children as a symptom of depression. An awareness that girls may be less likely to be assigned an ASD diagnosis than boys may also provide a useful insight. Finally, understanding dilemmas that parents face when professionals from other fields suggest their child is on the spectrum will help clinicians to work better with this group in practice.

The picture is complicated as the thesis cannot enlighten clinicians as to whether there may be cases where ASD diagnosis is ill advised. As the cases and controls were well matched across a range of autistic measures as preschoolers, but diverged as they matured, results provided evidence of how difficult it is to distinguish at young ages between a child who may have lifelong impairments and a one who is a 'late developer'. Article 3 concluded that the 'precautionary principle' (Science and Environmental Health Network, 2000) which states that preventive measures should be taken (or no action taken) in the face of scientific uncertainty about risk, should perhaps be applied when assigning ASD diagnoses to very young children. This is because there remains some debate between sociological and biomedical perspectives, and uncertainty about the exact effects of diagnosis on a child's outcomes.

Ultimately, clinicians must assess children on a case by case basis. Frith (1999) points to the importance of clinical intuition in assessment. This raises the question of how to strike a balance between expert intuitions which may be subjective and therefore subject to bias, and standardisation of diagnostic tools and categories which seek to minimise the clinical judgment of experienced practitioners.

Rosenhan's classic experiment of 1973 was concerned with the 'stickiness' of psychiatric diagnosis. Once labelled 'schizophrenic', for example, a patient would be termed 'schizophrenic' for life. The conceptual issue of whether ASD should be considered a lifetime diagnosis is discussed by Tony Charman in 2003 who notes:

Once an individual has met criteria at one point in their development, should they be considered 'a case' (for the purposes of scientific investigation) throughout their lifespan, whatever the improvement in symptoms over time? (Charman, 2003, p.14)

Given the findings presented here, and more rigorous studies such as those of Turner and Stone (2007), perhaps a sensible policy would be to apply a 'time-limited' ASD diagnosis for younger children, one that elapsed after a few years, and was automatically reviewed by a multidisciplinary team. Such a policy could prove costly and time consuming, but would prevent misdiagnosis in later childhood or adherence of a label that becomes inappropriate.

Section 4: Suggestions for future research following on from submitted work

1. Adapting the method to other childhood disorders

The central hypothesis of this thesis, which was that diagnosis itself leads to a change in symptom expression, is not ASD specific. Many of the same arguments can be applied to other childhood disorders, notably ADHD. Future research could extend the research methodology to other childhood disorders, but use a cohort with more power to detect effects. Such a project would involve a series of research questions/hypotheses concerning other childhood conditions, similar to those identified at previously:

> Are there a large group of children which the research team rate as having child psychiatric disorders, (such as ADHD) but remain undiagnosed? If so why?

> Do these results differ between cohorts? What are the differences in outcomes between those with diagnosis and controls?

Such a study would involve a comparison of outcomes between diagnosed and undiagnosed children using longitudinal data and age of diagnosis, in an attempt to decipher the effectiveness of interventions for child psychiatric disorders. This would also involve investigation of longitudinal data on some interventions such as methylphenidate use, speech and language therapy, and family therapy. It would also be interesting to compare measures of impact on families between diagnosed and undiagnosed children. Cross-cultural comparison of two cohorts with similar measures would yield further insights, although in cross-cultural comparisons it is difficult to assess what is a real difference and what is due a context of reporting (Obel et al., 2004).

2. Developmental trajectory of behaviours by ASD sub-group

One factor that became salient throughout the course of this study was the heterogeneous nature of the ASD group. When reviewing evidence for the effectiveness of service provision it appears some provisions are effective for some children, but a difficulty for clinicians and educational service providers is to distinguish what provisions may be most beneficial to which children. Warren and colleague's recent review (2011) also drew attention to the dearth of research on the needs and outcomes of sub-groups of children rather than ASD children as a uniform group. The findings here suggest autistic-like behaviours

of children diagnosed with ASD don't improve substantially, but there may be differences by sub-category, level of functioning, age of diagnosis, or gender that could not be detected due to lack of resolution. The literature reviewed also highlights the considerable overlap with other conditions and childhood disorders. A useful study, given a large enough cohort, would be to examine differing developmental trajectories of various SDQ measures, for sub-groups within the ASD diagnosed group, whilst taking into account age of diagnosis. Simply examining sub-populations by gender, and estimating the prevalence of ASD within each sub-condition would also be straightforward, and could lead to cross-cultural comparison.

3. A study with adults diagnosed with ASD assessing their perspectives on interventions and autistic identity

Although much research has been done concerning parents of children with autism and other childhood disorders and conditions (e.g. Gray, 1993; Green, 2007;. King et al., 2006; Woodgate, Ateah, & Secco, 2008), only a tiny proportion of research takes the views of autistic people themselves into account. This lack of voice has been a constant criticism from sociologists and others (e.g. Huws & Jones, 2008). Bagatell's ethnographic research on 'autistic culture' is an exception (2010), but does not address the question of participants' experiences of specific interventions. The UK Department of Health has stated that patients themselves should be increasingly involved in decision making and research so as to be advisors in own their treatment and care, and particularly in the shaping of child-specific services (2004). No studies encountered have addressed how autistic adults perceive utility of diagnosis and subsequent interventions for ASD as well as what function these diagnoses serve for affected individuals. Research questions could include:

Do autistic adults feel that interventions should always be sought? Under what conditions do they feel interventions are justified?

What interventions did autistic adults undergo as children? Do they perceive them as having been successful and/or beneficial?

Is ASD reconfigured to have positive connotations? Are neurological and biomedical conceptualisations operated to argue a case for rights?

How do autistic adults understand their own symptoms, and do they see them as having value? Are there tensions between parental perspectives and those of their grown up children, both within families and between generations?

How would autistic people themselves design interventions for others, particularly children?

The internet could be utilised as a research tool as often social face-to-face difficulties are problematic for this group. Adults with AS, for example, have been successful in designing and developing virtual environments for research (Parsons, Mitchell, & Leonard, 2004). Qualitative research of this nature could be useful in informing future policy and would concur with stated aims of inclusion of patients in health policy.

Section 5: Concluding remarks

The body of work as a whole has been subject to Bentall's first law of research: 'By the time an experiment is completed the researcher will know how it should have been done properly.' (2004, p.6). It was not possible to test the original hypothesis because of limitations of the data. Perhaps it could be done via data from another large cohort study with a large sample, access to medical records and independent records of autistic behaviours taken throughout the lives of children enrolled. If possible the independent data on autistic behaviours would involve a well validated research tool for assessing ASD. Then there would be a complete range of outcome measures in adolescence and beyond. Whether such a cohort study exists may be a question for the future.

Meanwhile, much has been learnt from engaging in the research submitted. Perhaps its endeavour is best encapsulated by the old adage that it is not the arriving at an end point that counts, but rather the journey itself.

Acknowledgements

Thesis Appendix 1: Reviewers' comments and responses to reviewers

(as submitted to journals)

Article 3

Dear Sir/Madam,

I am writing to resubmit the article attached. We have detailed our responses to the editor and reviewers comments below, and would be grateful if editors could pass these responses to reviewers.

Editors' comments

I would draw your attention in particular to the issues raised by both reviewers about the need to consider other possible reasons for the 'baseline' differences between your two main study groups - and the need to covary for indicators of these in your analyses.

We have reported the results of co-varying in table 4 and in the text (described below). As table 4 which reports this is basically an extension of table 3, editors may prefer to have table 3 extended and merged with table 4 to save space. This issue can be discussed if this article is accepted.

Referee: 1

Of the 142, 79 had never been referred to service providers but did have special educational needs (all of them?) but the other 63 had apparently received both.

1a) Of the 142, 63 had no identified SEN at school at age 11-12, 16 had within school support, although this was at the lowest level, and did not entail any support to additional educational of or health services. Another 37 were referred to external (non-school) services at age 11-12, ie specialist health/education, and 35 of these had statements. We have made this clear in table 1.

The suggestion is that there was an equivalence of ASD traits in the preschool years but differing trajectories for reasons other than autism. However it is not possible to state this from the data available.

1b) We agree with this comment.

We have clarified this was not our suggestion by adding to the sentence 'it is not clear from the data that differences in outcome are driven by management, or reasons other than autism.'

A diagnosis of ASD will have been made on the basis of history and observation of persistent traits and behaviours (not recorded here) specific to autism excluding other diagnoses. The children were compared at time point 1 on the traits alone, for which there may have been other explanations, including lack of persistence and pervasiveness, and not on other factors such as developmental delay or behavior problems which would have helped to answer the question about a bias in those referred. It is certainly possible that children are referred because of a multiplicity of concerns and this accounts for the different outcome, also possible that those diagnosed with ASD at age 2 years 'grow out' of the ASD features as was found by Stone or that they had a different explanation for the traits or that ASD diagnosis is a disadvantage!.

Yes- exactly.

The warning about care in too young a diagnostic insistence is important at a time when all the emphasis seems to be on early intervention.

Despite difficulties in distinguishing between these various explanations for the data, nevertheless, this is an interesting paper.

1c)

To make clear that there are various explanations for the data we have added a paragraph at the start of the conclusion, 'There are various possible explanations of the findings. Children may be referred because of a multiplicity of concerns including behaviours we were unable to measure, and these could account for the different outcomes. It is also possible that those exhibiting autistic-type behaviours in the preschool years may 'grow out' of ASD features. A third possibility is that ASD diagnosis biased reporting, or that diagnosis itself really could be disadvantageous.'

I hope you don't mind us paraphrasing a little. We have also rewritten paragraph 1 and 2 in the discussion to reflect this.

What would have been a great addition would have been something about the high trait score undiagnosed group who had external referral and additional special needs

1d) We have now included these children in the comparison group- see 1h) below.

At first, we did take a look at the high SEN group- the 37 with statements or external referrals age 12 who didn't have ASD diagnosis.

This high SEN group were academically as poor as the ASD group but worse than the rest. In social skills and restricted/repetitive behaviours they were often poorer to the population at large, but had similar outcomes to both undxed/ASD subgroups.

In measures of mood and feeling they differed from other groups depending on trait- eg more depressive than the population at large, less likely to self harm than ASD cases, more bullied than undxed group- however it is very hard to interpret as this is a small group (even smaller than 37 at outcome measures) and we don't know the nature of their difficulties. In the end, given reviewers comments about misplaced discussion relating to SEN/intervention we decided to include them in the comparison group-see 1 h) below.

and something more from the rich ALSPAC data about other features of the children preschool modeled into their long term trajectories.

1 e) Given that IQ has been shown by Howlin and colleagues (2004) to be a major indicator of outcome for autistic adults, we decided to co-vary for initial academic ability as a proxy for IQ. Separate adjustment was also made for the total behavioural difficulties score taken from the SDQ at 47 months, which includes measures of hyperactivity, conduct problems, emotional difficulties and peer problems.

Co-varying for academic ability revealed that outcomes in some autistic-type behaviours (social cognition measured by the SCDC score, time spent with young people, coping with the unexpected and restricted interests) still differed significantly. However the significant differences between the sub groups in academic outcomes did not persist. Adjustment for total behavioural difficulties age 3-4 accounted for differences between the groups in all measures except autistic outcomes; that is, all measures of social communication and repetitive behaviours that had differed without this adjustment, remained significantly different.

Unsurprisingly it is baseline academic ability that appears to be the primary cause of differences in academic outcomes. However baseline academic ability did not alter differences in autistic-type outcomes such as social cognition or restricted interests. Similarly non-autistic-type behavioural difficulties in preschool children accounted for much of the difference in emotional outcomes, but not differences in autistic-type outcomes. Even adjusting for the influence of academic ability at baseline, and other types of behavioural difficulties, outcomes in measures of autistic symptoms were worse for diagnosed children compared to those in the comparison group.

We have reported this co-varying in the analysis in the methods section, the results (para 3 results & table 4), and the discussion (end of para on Substantive findings.) table 4 displays the means of groups (rather than difference in means) so the direction of difference is apparent.

Specific features:
Background. First para fine but para 2 seems off the point.
Some explanation of SEN etc is needed for wider world audience.

1 f) We have deleted much of para 2, and given an explanation of SEN in the methods section under 'Outcome measures: Educational provision.'

Para 3—use of the word 'clinical'—the difference is between a local clinical and research clinical diagnosis

1 g) We have replaced this confusing term with 'a formal diagnosis in their communities'.

Page 8—did the 79 children have absolutely no referrals even as preschoolers since they all had speech problems?
1 h) This comment has led to a major reanalysis!
The definition of the group with low SEN relied on educational categorization at age 11-12. This was only taken at one time point. Although we assumed that this would reflect the history of SEN for each child, although probably broadly true, for individual children this was not the case. We did not have access to educational records at younger primary school ages but, we were able to arrange to access

educational data from two years earlier (at age 9-10) and we saw that 3 children in the low SEN group of 79 were moved from statement to low SEN between age 9 and 11. Also, when the study children were seven years old, parents reported whether their child had *ever* been given speech and language therapy. Of the parents of the 79 children in the low SEN group, the majority of children (34 out of 60 who reported) had received some form of speech and language therapy when young (this compared to 41 of the 47 children with an ASD diagnosis whose parents responded).

So although we were originally trying to define a dichotomous group with low SEN, vs a diagnosed high SEN group, in order to better detect the effect of diagnosis/ special education provision, it was clear that many of the low-SEN undiagnosed children had probably received various interventions when young, albeit as a group less service contact than those who were formally diagnosed. Given the comments of reviewers that the paper can not tell us anything about effectiveness of interventions, rather the focus should be on divergent development of behaviours between two groups, we decided at this point to redefine the comparison group as simply those with autistic-like symptoms when young, but no ASD diagnosis, regardless of SEN support. This meant that all the 142 children who were not diagnosed with ASD but were given some form of SEN were included in all the subsequent analyses. In fact this made no difference to whether outcomes significantly differed between the groups, (table 3) but we could be more confident because of the larger sample size. We also redrew the figures with the changed parameters of the comparison group, but again although the numbers changed, the significance levels of the results we reported were unaffected. It also meant the groups were slightly better matched in the baseline measures reported in table 1.

Nevertheless the differences in special educational needs provision and speech and language therapy between these two groups and the population at large was still of interest, therefore this has been included in the measures section and in the first paragraph of the results section. In the redefined comparison group, 72% of undiagnosed children received speech and language therapy before age 7 as opposed to 89% of those with ASD diagnoses.

And any idea what the special educational needs were for?

1 i) Of the 70 children with ASD diagnosis and high SEN, 60 children had school records, of these the majority (41) were categorised as having 'communication' difficulties as their primary problem. There were 37 children in the undiagnosed group of 142 with high SEN and most (24) were described as primarily having 'cognitive' difficulties. We have added this information to the results section.
.

Limitations clearly stated

Referee: 2
COMMENTS TO AUTHORS
Clinical and social outcomes in children diagnosed with autistic spectrum disorders: a longitudinal cohort study.

This paper uses a proxy measure of autism spectrum disorders to identify children in the general population ALSPAC cohort, to classify those wit and without a clinical diagnosis and to compare the social and emotional/behavioural outcomes in those two groups, and also compared to the general ALSPAC population, at age 13. This paper is potentially extremely interesting and could shed light on a number of important questions regarding autism spectrum disorders. However, there are two very significant problems in the way the paper is currently drafted that I would like to call to the authors' attention. The first is that the measure constructed is considered a proxy measure of autism spectrum disorders without much critique of how that measure is constructed. None of the items/variables employed to define the group are measures that have been developed specifically to identify individuals with autism spectrum disorders and it would appear all are "convenience" measures. Like all clinical diagnoses, the diagnosis of ASDs requires not only the use of an ASD- specific history and observation but also clinical interpretation of the findings from these measures.

Furthermore, although the sensitivity is high, the specificity is low and more importantly, the measure lacks good face validity, because it leaves out many key features of ASD. However, the way in which the paper is written appears to make the assumption that the two groups (those with a clinical diagnosis and those without) would be equally likely to have fulfilled an autism spectrum disorder diagnosis at age 3-4. However, this has not been tested in any way. While I appreciate the fact that ALSPAC did not attempt to diagnose ASD at age 3-4, the study needs to handle the limitations of the measures in a way that

reflects these limitations. This is discussed below.

2 a) We are aware of this limitation in the data and we need to make it clear within the paper that we are not claiming that the children in the two groups were not equally likely to receive a diagnosis. However we must defend our composite autism trait- we were at great pains to ensure it did not leave out key features of ASD- in fact it encompassed all the qualitative impairments specified in ICD-10 necessary to make a diagnosis of ASD, and these were included in ratios specified by the diagnostic criteria, (so 2 impairments in repetitive behaviours/restricted interests, one in social communication etc.) In fact these ratios and a description of exactly how they fulfil the diagnostic criteria is included table 2 of the previous (2010) paper, where we describe the development of the trait and how it relates to the diagnostic criteria for autism in detail. Nevertheless we accepted the composite measure is in no sense a validated instrument, and we have drawn attention to this in the limitations section.

The second main concern I have is the absence of a theoretical perspective on the questions asked in the analyses presented in the paper. Hence, ALSPAC has numerous measures and a range of different ages and it is far from clear why the authors have selected the measures presented here for comparison at age 13. While some of the measures are clearly about autistic-like behaviours, others seem to have been selected in a more random fashion. This may well not be the case, but there has been inadequate justification of the selection of individual items, both because of their content and the failure to use scale scores rather than individual items. Hence, a more tightly argued case about the key questions to be addressed in the current paper would be helpful.

2 b) In fact this study was conducted in tandem with a complementary qualitative study of parents' concerns relating to ASD diagnosis. The qualitative study revealed that parents of children with an ASD diagnosis were particularly concerned about educational attainment, levels of well being, friendships, self harm and aggression, bullying and school exclusion- all these outcomes, in fact, as their children became young adults. The findings of this complementary study provided the rationale behind selecting these particular items for analysis. We didn't previously include a reference to this as it has not been published. However an article that describes the qualitative study, which includes this information about the parental concerns which drove our selection of outcome variables, is now accepted subject to submitted revisions so we hope it should be accepted for publication shortly. In the meantime we have explained the rationale

and have cited this study as unpublished material in the section 'Outcome measures: Social, emotional and academic.'

From the Abstract onwards, I would advocate that the authors use the phrase "autistic-like behaviour", rather than "autistic behaviour".

2 c) OK, we have done this.

This relates to the first concern I have highlighted above. Focusing on the Abstract, last sentence of the Results, and also included subsequently in the body of the paper, I did not understand the final sentence "over 9 years considered, ASD diagnosed children fell further behind children in the comparison group in terms of academic performance by an average of 0.07 SD (95% C I – 0.62 to 0.49). This would appear to be a non significant difference and I cannot see why the authors have focused on this.

2 d) We focussed on this result because one co-author was particularly interested in this finding. However as you say we can't be confident that those diagnosed did not improve academically relative to those in the comparison group, therefore we have deleted it from the abstract. The apparent discrepancy in results was due to taking baseline academic ability scores into account in the regression you quoted above. As we have now co-varied for all outcomes for baseline academic ability, including academic outcomes, this non significant result is dealt with in table 4, and reported in results with other outcomes- there is no need to report the regression in the text.

In the first sentence of the Conclusions, "variation in measures of autistic behaviour apparently increases with age" I cannot understand quite what the point of this comment is and I think the authors could have a more focused and helpful sentence here. I suspect the authors are referring to the divergence of the clinically diagnosed group and the non diagnosed group but I am not sure that the best way of conceptualizing this is in terms of increased variation.

2 e) Another good point. This is exactly what we meant. We have therefore replaced the term variation with 'The divergence of the clinically diagnosed group and the non diagnosed group in measures of autistic-like behaviour increases with age.'

The Introduction has a heavy focus on services for children with autism spectrum disorders but I could not understand the purpose of this focus.

This paper does not contribute in any way to the literature on intervention for ASD and this seems a misplaced discussion.
A more appropriate focus would be on research in other areas comparing clinically identified and undetected groups in epidemiological populations.-

2 f) Yes, we have deleted the references to intervention as detailed above. As the first reviewer suggested the first para was fine we have some of it intact. We have replaced the paragraph on interventions (paragraph 2 in the introduction) with a paragraph on undetected groups in epidemiological populations. We have also reframed the discussion as outlined above, and deleted the reference to intervention in the key points section. This comment also led to our decision to include 142 children in the comparison group rather than a low/high SEN group.

By the way, I think it is now generally agreed that the correct phrase is "autism spectrum disorders" rather "autistic spectrum disorders".

2 g) We weren't aware of this until it was pointed out! We have now replaced the term throughout.

On page 4, lines 34 onwards, the authors suggest that studies reporting prevalence rates for ASD include children who meet the symptom criteria but do not have a clinical diagnosis. This is the strategy used in all epidemiological samples for any psychiatric disorder and is nonspecific to ASDs. The sentence should be rephrased to clarify this.
2 h) The studies we mentioned are ASD specific, however there are many other studies of childhood psychiatric disorder using this technique- we have added to this paragraph to elucidate: 2nd paragraph of the background section.

In the Methods, under Sample (bottom of page 5) the authors need to clarify how they have handled those children who have received a diagnosis of PDD-NOS.

2 i) Children classified as PDD –NOS using DSM-IV standards were classified as 'atypical autism'; the corresponding ICD-10 classification. We have added this point.

In justifying their ASD composite score and the cut off groups, the authors provide a figure (Figure 1) showing the mean of the general population and the children with an ASD diagnosis. For both of these groups, it would also be helpful to have the 95% confidence intervals placed in that figure.

2 j) We have now included this in the figure caption.

Turning to the Results, the authors refer to the "clinical outcomes". Can I suggest that "mental health" or "emotional/behavioural" might be more appropriate than "clinical" as no clinical assessments have been undertaken in these analyses.

2 k) We have replaced 'clinical' with 'emotional and behavioural' to describe these outcomes.

A difficulty for me is that the authors have undertaken no comparisons of the diagnosed and undetected groups at baseline. While it is reassuring that the groups are similar in the autism domains, it would not be surprising to find that they differ from each other in respect to a range of other characteristics, including their other emotional behavioural problems, developmental level as well as social and demographic characteristics of the families. Without having examined these it is really quite difficult to conceptualize the analyses that come later. There are many studies identifying a range of differences between diagnosed and undiagnosed groups and these may well be important in understanding the differential outcomes.

2 m) Differences in the background social and demographic characteristics of the families of two groups were reported in a previous article – we have referred to this
In the first paragraph of the substantive findings section. Regarding co-varying for other problems at baseline, we have now included this –as described, see 1e) above. It is true that children were not matched at baseline on non-autistic measures. There is now a discussion of this in the paragraph after the *'Insert table 1 here' caption.*

On page 12, line 18, the sentence commencing "we repeated the analysis restricting the diagnosed sample to children aged 1-6….." is confusing. I understood what the authors meant but it needs rewriting for clarity.

2 n) This confusing sentence has been rewritten.
  The presentation of the results in Table 3 is confusing. In particular, the use of the a and b superscript notation did not make for easy interpretation of the findings and this may have been aggravated by the fact that I think the statement given next to the b footnote is inaccurate.

Interpretation would be more easily made by a further column that indicated the significant differences in the three comparisons.

2 o) We have tried it adding two columns with the figures for this and it overcomplicates the results presented, especially with the addition of the further co-varying table. As we want to focus in tables 3/4 on the differences between the diagnosed and comparison group, we have stuck to the superscript, but we agree it was hard to interpret, so we have given the superscript its own 'column' to make it easier to interpret. We have altered the statements next to the b and a footnotes so they are consistent and easy to understand.

This brings me to my concern about the measures included in Table 3. As indicated above, for many of these I cannot see the theoretical basis on which they have been selected.

See 2b above

Table 3 also only included the educational attainment findings for age 13. I found it difficult to understand the results presented in the text about academic performance on page 13 where reference was also made to change over time but where little was presented in relation to the statistical findings. As stated before in relation to the Abstract, it would appear that the authors are mainly reporting a difference with a clearly non significant set of confidence intervals and this is somewhat puzzling.

See 2 d above.

In the Discussion, the authors once again come back to their points about interventions and I still think that this is outside the remit of this paper.

2p) We have deleted the major paragraph on intervention from the discussion, the key points and the introduction. This had also led to the redefinition of the comparison group without the 'low-SEN' parameter, as discussed above. This has been very a helpful review that has helped us to clarify a few things about our own paper.

In relation to the discussion on limitations, the authors haven't highlighted the areas that I think are most problematic, mainly the absence of an autism specific measure to classify the two autistic-like groups.

2 q) ALSPAC was not set up to look at ASD therefore we did not have access to involve a well validated research tool for assessing ASD such as ADI or ADOS. We have added this point to the limitations.

The authors discuss the problem of not having matched groups, but I cannot understand why they would want to undertake a matched analysis.
2 r) Our reasons similar to those of Angold, Kelley, Bickman, Crawford etc.- to better try and isolate effect of specialist service contact. Although this has not been possible with so many confounders in the current study, the divergence of the two groups is nevertheless of interest. We have added a paragraph concerning the rationale for this type of work, and its inherent problems, to the introduction.

As I have indicated above characterizing the baseline differences in the two groups would be helpful and then analyses could also be undertaken co-varying for any important base line characteristics that vary between the two groups.
2s) See 1e above.

While the authors highlight that a limitation is the retention rate (and gives reference to a paper that suggests this may not be problematic for examining correlates as apposed to prevalence rates), there is no

information about selective attrition and how it affects the groups. This should have been included early on.

2t) Selective attrition is a problem common to all longitudinal cohort studies. In response to this reviewer comment we examined various social and demographic factors to look for selective attrition, however as these didn't appear to affect the respondents/non respondents in the autism/comparison groups. As a result we have described the selective drop out in the cohort as a whole that was brought to light by Wolke et al in more detail. This is now reported earlier- in the methods section in the first paragraph in the section 'outcome measures'.

Thank you for your comments. It is really gratifying to have such informed and interested reviewers engaging with the work. We feel the article is improved and appreciate your time.

Article 4

Dear Sir/Madam,

As we have extensively revised the manuscript in the light of the first set of reviews. For your information only (I realise that you may not require this) I have detailed the revisions that have been made (a-x) in the light of reviewers comments (1-20) below.

Best wishes,
Ginny Russell (on behalf of the authors).

First Reviewer's comments
1) Given the current structure of the introduction, it is unclear if the focus of the article is on the effectiveness of the array of educational intervention programs and home therapies or on identifying a developmental trajectory of children's social behavior (as described later in the paper). A better discussion of these topics in the introduction may elucidate the current problem the article is addressing. The purpose of this study should also be clearly stated.

a) We agree with this comment, therefore, the introduction has been rewritten and refocused on the issue at hand: passages relating to intervention research have been dropped. The purpose of this study has been clearly stated.
:
2) While I understand these data were drawn from the ALSPAC, it might be helpful for readers to gain a better understanding of the children in this study. The manuscript states that "the social and demographic features of the ALSPAC cohort were representative of the overall population in the UK as measured by the 1991 UK national census." It might be helpful for readers unfamiliar with the ALSPAC to have a table with these demographics. If available, it would be helpful to provide additional information on the sample including children's mean age, percentage of males, IQ or

developmental quotient, if they are attending school - how many are

enrolled in inclusive classrooms as opposed to self-contained classrooms, etc.

b) This information and a series of tables outlining these demograhics is on the ALSPAC website- a link is provided- there is so much information, and we are short of space, a link to the website seems the most expeditious way of pointing the reader unfamiliar with the cohort to this information.

3) The authors report that medical records of the 71 children showed that children had a formal diagnosis of ASD. Did the records note how children were diagnosed (using what measures, the ADOS, ADI-R, SCQ, etc.) and by whom (schools, clinicians, psychologists, etc.)?#

c) The clinical diagnoses recorded in the children's medical records were made in the children's communities, blind to the measures taken by ALSPAC, by a clinician, (either a child psychiatrist or community paediatrician,) with support from multidisciplinary assessment teams. It is not known what diagnostic tools were available. This information is now given in the sample section.

4) Is there a breakdown in children who were diagnosed with autism as compared to children diagnosed with Asperger syndrome?

d) Yes, in figure 1

5) In regard to the SDQ, the authors report that questionnaires were administered by post. What was the return rate for these surveys?

e) This information is given in the end of the second last paragraph in the analysis section, it has been slightly rewritten to make it clearer: 'In all, 57 children diagnosed with ASD had prosocial scores for at least one time point between 47 and 157m. Six measurements of the prosocial score were made in total, an average of 4.3 assessments were made by parental report and returned to ALSPAC for each of diagnosed child out of the theoretical maximum of 6. When restricting to children with both pre and post diagnosis data, 33 children were available with an average of 5.0 assessments.'

6) Were there incentives for parents to complete the survey and stay in the study across time?

f) No financial incentives were used but, parents were encouraged to participate. See ALSPAC website as cited .

7) What percentage of the questionnaires was completed by mothers and fathers?

g) This is a really good point. Although the questionnaires were aimed at 'parents'(either parent was free to answer), it was the children's mothers who completed the questionnaires in over 90% of cases for each time point in which the ALSPAC questionnaires were sent out. We have explained this in the measures part of the methods section.

8) Was the SDQ also given to other raters such as teachers or aides/behavioral support? If available, it may be interesting to cross validate parent and teacher (when the children are school-age) reports.

h) The SDQ was administered to teachers at two time points. We did not use these data because of known discrepancies- reported elsewhere in the SDQ literature- between teacher and parent SDQ ratings in the population at large. There were not enough data to repeat the analyses solely using teacher ratings.

9) Were additional measures of children's socialization or Theory of Mind used? If so, it might be interesting to determine if children with greater theory of mind were more prosocial.

i) Unfortunately TOM measures such as Sally Anne test scores were not administered during ALSPAC.

10) A major limitation is the lack of details of overall interventions. If the information can be retroactively obtained, it would be interesting to know a) what intervention services children were receiving; b) whether these interventions were specifically designed to enhance socialization; and c) how long children were enrolled in these types of services. Did all children receive intervention or only a portion of children? This may be an interesting variable to control for in the model. Given the number of children diagnosed with autism and Asperger

syndrome and whether cognitive functioning sores are available, it may be interesting to test to see if there were differences based on children's level of functioning.

j) All but four children received intervention at individualised level at school, as well as a clinical diagnosis which we could consider as 'service contact'. This is clarified in the last paragraph of the 'sample' section in the methods. As the model was virtually over parametized in the second analysis, further splitting by AS/autism or intervention type was not feasible in this study. The suggestions would be an interesting area for further research.

11) While this study addresses an often neglected area of great concern, the conclusion as presently written may not effectively address the variety of interesting questions posed by these findings. The authors suggest that "this core symptom of ASD may be resistant to intervention." This point would be enhanced by further understanding (in the method section) what types of intervention services children received and what the direct targeted skills were. If this information cannot be obtained, it should be noted extensively in the limitations section. Until it is clear that these skills were targeted by efficacious interventions with a strong evidence-base, it is premature to conclude these symptoms may be resistant to intervention. Indeed, socialization and prosocial behavior are difficult to remediate and change, but perhaps, researchers have not yet identified appropriate interventions to develop these skills in children with ASD. I encourage the authors to consider the implications of their findings on intervention research as well as access to services after families receive an autism spectrum diagnosis.

k) The first paragraph of the conclusion has been rewritten to make this point clear. We agree there are two possible conclusions to these results- however we do are not concluding that social behaviours ARE resistant, rather they MAY be resistant. We do not think this is unreasonable in the light of these results. In terms of broad picture of interventions in use by services-'service contact' being ineffective on social skills, but also in terms of whether social skills were targeted in the first place.

Second Reviewer
12) The hypothesis presented for the current study seems to be at odds with
the majority of literature discussed in the introduction, i.e. Dawson (2010)
concludes that interventions are not particularly effective; Spence &

Thurm (2010) note the difficulty of detecting change when testing interventions for ASD where children have an assortment of symptoms. It is unclear why the authors therefore predict that prosocial behaviour is likely to improve as a result of ASD diagnosis. The hypothesis is not justified.

l) We now have fully explained the rationale for our hypothesis in the methods section. We have taken out the section on lack of evidence for interventions effective on social impairment and replaced with literature that led us to formulate the hypothesis.

13) * Lack of sensitivity in the methodology: There are only 5 items from which a forced choice answer (3 options) must be provided.

m) The SDQ is a well validated and internationally used scale- we have no concerns about its sensitivity as we are not claiming it is a diagnostic screening tool, rather just a measure of prosocial behaviour.

14) * As can be seen in Figures 2 and 3, the sample is extremely heterogeneous. Variation in symptom severity between participants appears to be ignored.

o) Figures 2 and 3 do not illustrate heterogeneity- we think you must mean fig. 1? We adjusted for variation between children's scores. This was detailed in the methods section. We did not ignore individual variation at all.

15) * On page 8, the authors note that they assume that "response to diagnosis would be the same at age 2 as at aged 12". This seems an unwise assumption to make.

p) Why? We thought it was reasonable given that we were looking at impact of service contact per se. for any child and we adjusted for variation in scores between individual children. Particularly as the variation in ongoing scores between children who were more severely impaired in this measure and those who were less impaired was taken into account.   We have rewritten this to make it clearer.

16) * There is only pre and post diagnosis information for 33 participants (not 57 as implied in the abstract), the resulting statistical analyses are under powered.

q) The statistical limits of the second analysis are referred to in the discussion. We present two analyses- one of 57 children, and a second of 33 children. We have altered the abstract to reflect this.

Reviewer 3.

I recommend that the authors temper several claims:

17) This paper draws on the premise that SDQ scores should improve because children in the sample had improved access to intervention after they received their diagnosis. A paper by Mansell and Morris (2004) was cited to evidence that children in this study received a variety of intervention services as a result of their diagnosis. I strongly urge the authors to better support this claim with additional analysis. It would be good to know if the children who received more intervention than others had better prosocial behaviors. Additionally, different types of intervention will most likely affect the development of prosocial behaviors. A variety of intervention services are referenced in the paper (e.g., respite, educational services, speech therapy). It would be nice to know if the type of intervention a child receives influences the prosocial behaviors s/he exhibits. Furthermore, it would be nice to know if intervention dose influences SDQ scores.

r) Unfortunately, due to lack of data resolution, this is beyond the scope of this paper- we have altered the methods and discussion sections to make it absolutely clear what was within the scope of the research to determine.

18) "This study provides no conclusive evidence either that prosocial behaviors are adversely affected by diagnosis." Need to more explicitly clarify that ASD does affect prosocial behaviors, but that simply obtaining the label of ASD may not.

s) We have clarified this in the discussion. In fact we have paraphrased this reviewer in the discussion.

19) "The study suggests this core symptom of ASD may be resistant to intervention." A study by Green and colleagues was cited. First, there are methodologically rigorous studies that report improving core autism deficits and these should be cited.

t) Yes we have added these in the discussion.

20) Second, the prosocial behavior construct you opperationalize is very different from the social communication behavior Green and colleagues targeted in their intervention.

u) Green et al has now been cited to flag up the improved secondary symptoms they found, eg. Improved parent-child relationships, rather than as example of intervention targeted at social-communication.

21) Third, most interventions received by your population probably never claimed to target core ASD symptom(s).

v) This is now explicitly covered in the discussion- above.

22) ABSTRACT: -Need an introductory sentence.
w) We have added this

23) INTRODUCTION:-Include background information regarding your prosocial construct.

x) The details are included in the measures section

24) -Reference studies that show prosocial behaviors are responsive to intervention in children with ASD. (These do exist, though in your introduction you make it sound like there are not any).

y) These are mentioned in the discussion- the introduction is rewritten in the light of comments from reviewer 1.

25) METHODS:-"Questionnaires were administered by post." Please explain what that means. Who administered the SDQ? -Did the same parent answer the SDQ at each time point?

z) We have changed this line in the measure section to clarify. We have added the lines: 'Questionnaires were sent to each enrolled family by post. The SDQ questionnaires were nested within a series of questions on the health and development of the study child. Parents enrolled in the study were asked to fill in the questionnaires and return them to ALSPAC'

The second point is dealt with in the comment above i.e. it was the child's mother 90% of the time.

26) Were the parents answering a series of other questionnaires at the same time?
i) Yes ALSPAC is a longstanding study that has resulted in over 1000 research papers. There are many other forms of data being taken, including genetic, measurements, edaucational, health and many other questionnaires. – see above.

27) Explain the meaning behind the prosocial trait score (e.g., what does a score of 1 on your graph mean?)

ii) We have added this information to the measures section: 'Children were scored 0 if the response was 'not true', 1 if 'somewhat true' or 2 if 'certainly true'. Hence a score of 1 indicates very poor prosocial behavior and a score of 10 represents highly prosocial behavior. The mean prosocial scores at each time point for children varied in the ALSPAC population (n=4924) lay between 7.0 and 8.3, but for the children diagnosed with ASD the mean prosocial scores varied at each time point between 4 and 5.7.'

28) Justify the use of the SDQ by citing other studies that have used the SDQ
with children who have ASD. It would be great to know that this instrument
is valid with a broad spectrum of individuals with ASD. (Might even be
good to include information about the use of the SDQ with the ASD population in
your introduction).

iii) The only study we know looking specifically at ASD and SDQ is the Russell et al one which we have
included in the introduction, and how the term prosocial was derived. We have clarified in the discussion
that this was a retrospective analysis- in other words, the ALSPAC study, nor the SDQ was never set up
to look specifically at ASD – as such ALSPAC is not using ASD screening tools such as ADI- the SDQ
just contains one measure which is strongly associated with ASD diagnosis, it isn't specifically designed
with ASD in mind. We hope we have made this clear—we have written into the limitations section.

29) RESULTS:
 -It is good that statistical limitations are acknowledged. Put these in
 your discussion.

iv) OK- have done this
30)The sentence starting with "the median age for the late diagnosis group"
(page 11) should be in your discussion.

v) We feel this is reporting a result- so it is better placed in the results section.

Figures:
31) 4: Perhaps the y-axis should be scaled the same way as figures 2 and 3
 (maximum value of 8)?

v) We have slightly different scales to improve the resolution in the graph.

32) Fig 4a: A) Should be renamed figure 5,

vi) Have done this

33) Label the y-axis,

vii) Have done this

34) provide a more detailed interpretation of this figure in the text.

viii) This has been covered in the results section.

35) DISCUSION: -You need to acknowledge the limitations of the SDQ measure. This questionnaire is brief, parent-report, and other more psychometrically valid measures are commonly used to measure the prosocial behaviors of children with ASD.

ix) We have added a section on SDQ to the limitations section- see above.

36) Need to more prominently explain this statement: "Practitioners were simply not using treatments that are effectively treating prosocial behaviors." This fact most likely explains a large portion of your results.

x) We have added more discussion to this section.

37) -Include a future directions paragraph.

We have now drafted a paragraph suggesting further work, however as the revised article is over the 4500 word limit although well under the 40 page limit, we have not included it.

Thanks so much to all the reviewers for their insightful comments!

Article 5

Dear CCPP,

As advised, I am returning the revised article 'Dilemmas, diagnosis and de-stigmatisation: parental perspectives on autistic spectrum disorders' for your consideration The article has been revised in the light of the reviewers comments which were very helpful. I have detailed all the revisions which have been made in response to these comments below, and highlighted them in the article text in red, I have also highlighted the responses to comments in red, below.

Response to Reviewer One comments:

>>Although there is a discussion later on in the paper about how autism is understood – as a disorder, or condition or as 'neuro-diversity' - I think it would be helpful to include a fuller discussion of this complex and interesting issue at the start of the paper. The work of Timini on AS and ADHD might be useful here.

Understandings of autism as discussed by Timini and others are dealt with in a new paragraph which has been inserted on page 4 para 2. Para 1, page 4 lines 1-4 have been further modified to introduce this issue at the start.

1. Throughout the paper hints at the label of autism as a passport to services. There is evidence to suggest (Hodge, 2005) that parents feel pressured into accepting diagnoses in order to access services. I think it would be helpful if this point were made more clearly in the paper. Furthermore, Hodge's paper engages in an extended discussion of parents' reluctance to label their children which the authors may find useful for the current paper, especially as they claim this is an area which has received little research attention.

The work of Hodge was missed by the authors and we are grateful for this useful pointer. Hodge's work has been incorporated into the introduction on p.3 para 3, lines 9-12 and also in the analysis on p.11 para 4, lines 3-4

2. The authors might also find it useful to look at McLaughlin et al (2008) which challenges

'bereavement models' of understanding of parenting disabled children

McLaughlin and Goodley's criticisms of Bury's notion of biographical disruption are made explicit on on p.9 para 1, lines 4 -6.

3. The authors might also want to read Ryan and Runswick-Cole (2009) which also explores the extended advocacy role of parents of children on the autism spectrum.

This article is of great interest and relevance to us and reference to it has been made throughout the article, referencing gender blind research on pages p7, para 4, lines 1-4, p20, para 5, lines 3-end and p.21 para 1, blurring between advocacy and activism. A meeting between the first author and Dr Sara Ryan has also taken place thanks to this useful pointer.

4. I also wonder if the authors could make clear how long the parents who had a diagnosis for their children had had that information.
5.

This information was available and been inserted on page 6, para 1, lines 4-8, and also p.7 , para 4, lines 5-6.

Just a couple of small points
- p11, para 2, line 3 change the capital on This- adjusted
- p12 para 2 line 5 – there is a direct quote with no page number- added
- p314 – the interviewer leads the participant, the authors might want to think about whether to include this extract or perhaps to make clear the interview rationale in the methods section.

This leading part of the quote relates to a part of the conversation established earlier (the participant feeling 'uncomfortable') and has been deleted as it is reported elsewhere; the second part of the quote concerning influence of another participant was again established in an earlier part of the interview, and this point is outlined in the text on p12, para3, lines 1-2, instead of given as a quote.

Referee Two: Response to comments

1. A general proof read from a critical friend is needed as some sentences were grammatically incorrect or a bit clunky.

A colleague who is the copy editor of a well respected journal has proof read and this manuscript, and corrections to clunky English made throughout.

2. Also the reader is lead to believe the research is about high functioning autism/ AS. This needs to be reflected in the title.

This is a very good point. In fact only one of the children in the diagnosed sample was not high functioning. Without a label, it was not possible to ascertain whether undiagnosed children were high functioning. The text has been changed to explain this p.6, para 3, lines 5-8. However on careful reflection we decided not to change the title as not all the sample were high functioning or AS. So changes to the text to clarify, not changes to the title.

3. In the discussion about social construction the social and medical model need to be mapped out and critically engaged with. This is due to the fact that the authors suggest that some argue that ASD is socially constructed. This is a huge statement and even though it is backed up with research needs further analysis. Look at the work of Prof. Carol Thomas' work 'sociologies of disability' regarding impairment effects, so regardless of the social construction of disability every day difficulties of an impairment impact on a persons life. Or Prof. Tom Shakespeare's work regarding his critical engagement with the social model. But it cannot be overlooked that the social model (see Prof. Mike Oliver's work) has been hugely influential. The paper would have benefitted from this discussion (and then this work would have aided the critical narrative discussion in the findings sections).

An introduction to the social model and the critiques that have been made (such as those by Thomas and Shakespeare) has been included on p4, para 1, lines 7-13. This is also related to the previous discussion of the various conceptualisations of ASD and links into it. These introductory sections are then used to inform the analysis of findings on p.13, last lines, first line of p.14, and p17, pa 2, lines 3-6, and p.18, 3, 6-8, and p23, pa3, lines 3-9 as suggested.

4. Then after this 'Labelling Theory' is dropped in so to speak but left with little further analysis. It is mentioned again in the analysis but this needed to be a thread if it was a large part of the literature (which the paper implies engaging with the likes of Goffman and Gray). Also the authors say it was developed by Scheff (1974) but this probably needs to be rethought as Becker in 1963 and later in 1973 (where a chapter called 'labelling theory revisited' was added) discussed labelling and although he did not want it to be seen as a theory it was picked up as such. So yes there was quite a lot of work going on in the 1960s and 70s, especially in the USA around labelling.

Labelling theory is not a thread that informs the analysis in the sense that we are interested in whether the negative aspects of labelling occur; rather, the ideas behind it form a discourse which is adopted by parents resisting diagnosis. To clarify this, we have moved the section on labelling theory from the introduction to the appropriate place in the analysis of finding section page 10, para 4. The passage has also been rewritten to clarify that its relevance within the article and to make clear the contribution of Scheff's work, re: the reviewers comment about Scheff/Becker.

5. I would like to see the authors just say what 'in-mates' are as not all audiences might know.
 This term has been deleted p.5, para 1 line 1- (no highlight as term deleted)

6. Again, the paper makes some assumptions that ASD is socially constructed (or at least it seems to be the case). The reader needs clarification on this.

No, we are suggesting some others view ASD as socially constructed see page 4, para 1. We are suggesting parents may be active agents in perusing or avoiding ASD diagnosis. This is not to deny that real impairments exist. We have clarified this on p.21, para3, lines 1-4.

7. I am not too sure why in qualitative research 'n = number' is used?
This has been altered to express sample numbers in prose throughout the methods section, for example p.6 para 3 lines 8-9.

8. Page 6: it is NVivo not In Vivo!

No, 'in vivo' is a term in qualitative methodology that means using the words of the interviewees in coding rather than abstracted concepts. Nvivo is analytical software. This is clarified on p.6, last line.

9. The methods section is a little pedestrian. I would like to see this edited in a sophisticated way. That way it would leave space for more critically engagement.

We have edited and rewritten the methods section throughout.

10. The sentence 'their affected children' could be read as a little offensive.
We have deleted the word 'affected'

11. On this note also I would warrant some caution over suggesting that some might see ASD people as eccentric historically.

We are not suggesting that autistic people are eccentric ourselves but were reporting on what interviews said to us at this point- this is how they viewed the rise in prevalence of ASD

12. Page 20 'on the spectrum' needs to be in ''. It is also said further on in the paper.
We have not corrected this as 'on' or 'in' it appears to be correct.

13. In the discussion ASD children are mentioned as being 'gift like'. The tragic versus 'gift – treasures' narratives need to be thrashed out.

Researchers have reported narratives of parents that have described having a child with autism as a gift in some accounts or as a tragedy in others. The literature that encapsulates some of these two perspectives have now been described in the introduction on page 3 last 3 lines; p.5 & para 1, lines 1-5; and the reasons behind this apparent discrepancy are discussed in the analysis on page p.19, para 1, lines 4-16.

14. Social Identity Theory is parachuted in but not critically engaged within the paper.

We have deleted the section on Social Identity Theory and reduced to a brief reference to Haidt's work on page 23, para 4, lines 1-2 as we agree it does not relate to theory anywhere earlier in the paper.

15. On the whole once the theory and literature have been tightened and critically discussed the findings should then be edited and tightened to reflect these discussions

We have edited the findings section, e.g. p16, para1, lines 4-6 and the changes we have made throughout have answered this general criticism. We would like to thank this reviewer for the useful references to other literature that were supplied, which we have incorporated as mentioned throughout the text.

These above are our responses to all the reviewer's comments. I would like to point out that I have used English spellings throughout the article. Please do let me know if you need any further changes.

I look forward to hearing from you,

With best wishes,
Ginny Russell

Appendix 2: Additional articles first authored by candidate

On following pages (as originally pdf files)

A qualitative analysis of lay beliefs about the aetiology and prevalence of autistic spectrum disorders

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Abstract

Introduction   There has been a dramatic increase in the prevalence of autistic spectrum disorders (ASD) in the last 20 years. The reasons for this are disputed. The consensus among epidemiologists and other experts is that greater case load is due to changes in diagnostic practice rather than reflecting changing aetiological factors leading to a true increase in incidence. We set out to examine lay views concerning the aetiology and prevalence of ASD and whether they conflict with or support this consensus position.

Methods   Over 100 unsolicited communications (letters e mails and several telephone calls) were received by a UK epidemiological study of ASD. We carried out a qualitative analysis of all correspondence in order to examine spontaneously expressed lay beliefs about the prevalence and aetiology of ASD.

Results   The majority of correspondents suggested theories about environmental causes of ASD. This study demonstrates the strength of lay belief that the true incidence of autism is rising, and this is due to risks from modern technologies and changing lifestyles.

Conclusion   This study based on unsolicited data highlights the contrast between lay explanations of increasing prevalence and the consensus opinion of medical experts. It also demonstrates how many people in direct contact with ASD have important information to share.

Introduction

There have been far more diagnoses of autistic spectrum disorders (ASD) in recent years with the US Department of Education showing an astounding 556% increase in the number of children being treated between 1991 and 1997 (Stokstad 2001). In a 2005 review, Rutter writes: 'the true incidence of autism spectrum disorders is likely to be within the range of 30–60 cases per 10,000, a huge increase over the original estimate 40 years ago of 4 per 10,000' (Rutter 2005). According to Muhle and colleagues (2004), ASD now has a greater prevalence in children than that of cancer or Downs syndrome. There is no doubt that the reported prevalence has increased spectacularly, and particularly over the last 20 years. The question is, why?

Influential work by Eric Fombonne (2001) suggested that the change is not a true reflection of more children with ASD in the population now than in the past, but is the result of the ASD diagnosis being given to more children. Three main reasons have been cited: the extension of the spectrum in recent times to include milder conditions like Aspergers syndrome, the extension of diagnosis to younger children, and increased awareness of ASD by parents and clinicians.

'Diagnostic substitution' may provide a partial explanation. Barbaresi and colleagues (2005) found that increased rates of diagnosis followed increases in funding for special educational programmes. Where resources are being directed towards ASD, greater incidence is associated with declines in other diagnostic categories indicating that clinicians prefer to label children with ASD in order to allow them access to greater resources (Shattuck 2006). If this is occurring, children who in the past would probably have been diagnosed as having a specific learning disability or a psychiatric disorder, or not diagnosed at all, are recorded as cases of ASD.

The view that the increased volume of ASD cases is due to changes in diagnostic practice has become the consensus position. As Howlin (2006) states: 'Recent increases in rates of diagnosis reflect greater awareness of autism spectrum disorders among professionals, together with widespread improvements in diagnostic practice'. We have found that this position conflicts with the opinions of many people in contact with autistic children. Individual parents, for example, have published their view that the true incidence of ASD has increased; i.e. there are actually more children now who display ASD. For example activists like Blaxill (2004) argue that large increases in numbers cannot be attributable solely to changes in diagnostic criteria or improvements in case ascertainment.

The aetiology for most cases of ASD is unknown. The idea that ASD is caused by a genetic predisposition triggered by an environmental insult is common among researchers, with proximate causes understood as risk factors making it more likely that children will develop the condition. Reviews report that many risk factors, predominantly genetic but also environmental, are associated with ASD but the effects of each are poorly established (Newschaffer *et al.* 2007). Gray suggests that uncertainty about aetiology has created a vacuum in which speculation about possible causes may flourish. Risks that have been implicated by parents include prenatal, postnatal and perinatal factors (Gray 1995). In particular, active groups have mobilized around concern over vaccines. In the UK, the measles mumps and rubella (MMR) vaccine has been a focus for activists despite clear evidence showing that the vaccine is not associated with ASD (Rutter 2005). In the USA, the use of the preservative thiomersal, which contains mercury, in many vaccines has been cited by activist groups.

In 2007 an annual review of public health concluded that the question of whether incidence of ASD really has increased remains unanswered (Newschaffer *et al.* 2007). Szpir (2006) described a 'furor' over this issue and claimed it has divided scientists and the public alike, but provided no evidence to back up this claim. Here our study seeks to examine how an unsolic-ited sample of members of the public views the increasing prevalence of ASD, and whether their explanations differ from the consensus medical position.

Methods

In 2004 a UK epidemiological study announced via a press release that it had been funded to examine the environmental causes of ASD. This release, describing a 'study to look at environmental causes of autism', provoked worldwide media interest with articles appearing in both the national and international press, and sparked interviews with the director of the study on national radio and local television. Some UK media outlets reinterpreted the study as focusing on MMR. For example the headline 'Funding U-turn on MMR jab study' was published by the Daily Mail (8 July 2004).

A total of 105 unsolicited letters, emails and several telephone calls were received in response to this media attention. The majority of letters and calls were from the UK with a small proportion from North America and other industrialized nations, all written between 2004 and 2006. The sample was self-selecting, from a population that had access to the Internet, television or newspapers. We analysed the content of the letters in order to examine the spontaneously expressed lay beliefs about the prevalence of the condition.

Our study had the advantage that data were neither solicited nor was data collection influenced by the presence of any researcher. This has been shown to lead participants to edit and adapt their responses (Hammersley & Atkinson 1995).

Names, locations and any other identifying features of correspondents were changed. The letters were read by two researchers, then transcribed and analysed. Significant statements and paragraphs from the letters were categorized according to themes and codes developing in the data in open coding, by the first author. A second researcher (second author) then analysed and coded a further set of letters, and the two sets of data were brought together to compare and cross code. The themes in the remaining letters were developed and codes were abstracted, linked and compared with the original categories. Thus a bottom up data-driven approach, using iterative analysis and theory building, was adopted. Theoretical saturation was reached at the 60th letter, with no new codes being identified A third researcher (third author) reviewed the overall coding, analysis and interpretation, providing a further validity check.

Results

The sample was a mixed group of relatives of people with autism, individuals self-described as having autism, and

professionals and clinicians with first-hand experience of ASD (Fig. 1). The overwhelming majority of the letters and telephone calls (81%) put forward the correspondents' theories and hypotheses about the causes of ASD. Of these 96% suggested potential environmental causes of the disorder. Roughly two-thirds of this correspondence (62 letters in total) put forward causes other than vaccinations; only 16 letters mentioned MMR. Over 40 different environmental factors were offered as potential causes, the majority of which (all but three) invoked modern technologies or a changing way of life (Table 1).

The vast majority of correspondents identified manmade risks; either due to medical technologies, modern lifestyles or industrial risks. Furthermore, these risks were described as associated with the latest adoption and applications of technologies so their effects are perceived as recent. This latent unease with present-day life and scientific progress was described by the sociologist Ulrich Beck (1992) as 'insecurities of the contemporary spirit' and forms a repeating theme:

> Letter 60: Our body and brains are not infinitely capable of absorbing ever increasing amounts of drugs, vaccines etc.
> Letter 45: I think this proves conclusively that colourings, preservatives and additives are playing a major factor on how the brain develops while in the womb between the fifteenth and seventeenth week of pregnancy.
> Letter 87: I have been very concerned about the amount of foetal monitoring. The rise in hyperactivity in children and in autism goes hand in hand with the rise in foetal monitoring.
> Letter 19: I hope I am barking up the wrong tree but I am still concerned at the number of children who have come to me in the past ten years with parents who are in some way associated with nuclear power stations.

The risks of modern societies as described by Beck are frequently invisible, and are known or 'made visible' through scientific means (e.g. measurement). In this sense science can be seen to cause, identify and respond to risks. This understanding of the relationship of scientific knowledge to risks of the modern world is reflected in the correspondence. Although correspondents frequently suggested that the technological applications of modernity cause the risk of ASD (the problem), they also looked to the science of epidemiology to define and address them (the solution). Correspondents requested the epidemiological study to test or validate their theories rather than insisting that they were true, including in many cases requests to adopt specific methods:

> Letter 62: We moved near a mobile phone mast and had two severely autistic children. In your research on autism will you ask people if they lived near a mast during pregnancy?
> Letter 9: Even when CO is detected it is almost impossible for our victims to find someone to test the appliance and the house for CO and therefore no evidence is found of CO . . . However, possibly CO or suspected CO was recorded during this original research in which case it would be of great interest to us.
> Letter 12: Has your longitudinal study shown up any dietary differences in the incidence and or the severity of autism? and is there any room in the new work to undertake some dietary manipulation work, and the imposition of A2 milk in situations where there are significant problems with autistic behavior?

The correspondents argued that there has been a real increase in incidence of ASD. Associated with this rise as hazards caused by modern technologies and lifestyles. The majority of the correspondence thus put forward theories and hypotheses about
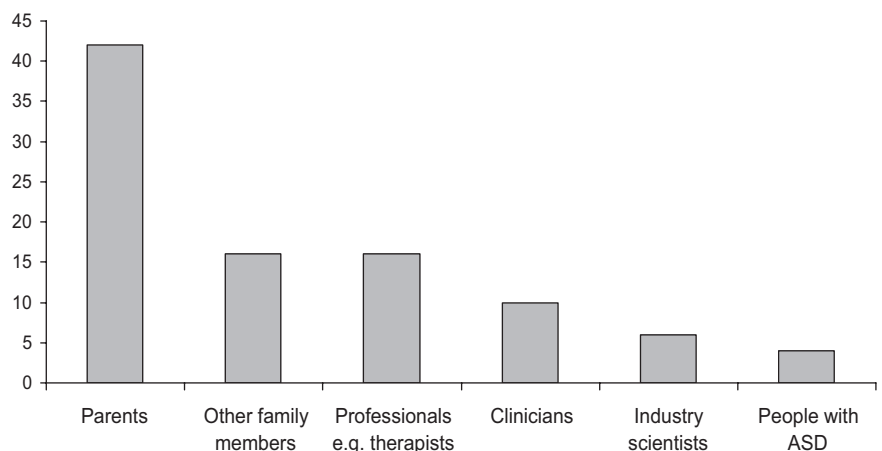

Figure 1. Frequency of relationship to autistic spectrum disorders (ASD) where given (80% of sample).

Table 1. Range of environmental causes given as possible risk factors for ASD

Causes of risk	
1. **Medical technologies**	
	Ultrasound scans
	Baby induced
	Early cord clamping/cord wrapped around baby's neck
	Respiratory distress
	Caesarean section
	Birth trauma
	Foetal stress due to medical intervention
Related to drugs/toxins during pregnancy	Rhogam shots
	Contraceptive pill
	Steroids
	Antihistamines
	DES (to prevent miscarriage)
	High levels of mercury due to dental mothers fillings
Related to vaccines	Time of day of vaccination
	Lack of aspiration when vaccine administered
	MMR
	Mercury due to thiomersal
	Vulnerability to injections when teething
	Polio vaccine
	Egg products in vaccines
	Pain of injection
	DPT/toxins
2. **Changing lifestyle**	
	Working mother leads to stress during pregnancy
	Amount of alcohol drunk during pregnancy
	Indoor air quality/time indoors
	Overstimulation by cot toys
	Too much television
Related to diet	Lack of cod liver oil
	Food additives/aspartame
	Disaccharides and starches
	Food preservatives
	Genetic origin of cows milk due to intensive animal breeding
	Gluten in diet
3. **Unavoidable technologies**	
	Low level radiation, from VDUs
	Carbon monoxide exposure
	Father works in nuclear power stations/ exposure to radioactivity
	Exposure to chemicals
	Living near mobile phone mast/ exposure to low level radiation
	Moulds from indoor environments
	Air pollution
	Pollutants in water
4. **Biological factors**	
	Previous miscarriage or bleeding during pregnancy
	Dry birth (no amniotic fluid)
	Child being born after twins

ASD, autistic spectrum disorder; MMR, measles mumps and rubella; DPT, vaccines to immunize against diphtheria, pertussis (whooping cough) and tetanus; VDU, visual display unit.

environmental causes of ASD. Increasing incidence of ASD due to more contact with modern environmental triggers was a recurring topic.

> Letter 52: I have been amazed at the increased incidence of autism – and pondered about the causes as have other people . . . since I left in 1995 something has happened – an explosion. The autistic did not exist in quantity pre-1995- so bearing in mind children enter schools at five years old – something changed around 1990 onwards. . . . Something definitely changed in the 1990s and which has persisted. I don't think it can all be down to better detection of autism.
>
> Letter 64: I also believe that the increasing rates of autism in our population . . . are due to the ever increasing levels of sucrose consumption in our diet.
>
> Letter 78: I hope that one of the factors being considered is the exposure of the foetus to ultrasound examinations in pregnancy. It does seem that the rise in incidence in autism corresponds to the rise in use of ultrasound.
>
> Letter 74: It has recently come to my attention that there are those who believe there is a correlationship between the rise in autistic spectrum disorders and the practice of early umbilical cord clamping . . . As a midwife I find this very disturbing as this has been my practice and that of my colleagues. As a precautionary measure, I now leave the cord longer before cutting it in order that the neonate might receive possible 50% more of its blood supply from the placenta.

In many cases the juxtaposition with the medical consensus is made explicit.

> Letter 75: My colleagues and I were increasingly concerned about the growing number of children with autistic spectrum disorders. We did not think that the increase was solely due to improved diagnosis . . .

Discussion

The overwhelming majority of respondents in the unsolicited sample in this study suggested that autism is caused by environmental factors. Although media reports described the epidemiological study as looking for environmental causes, the number of unsolicited letters espousing the idea of an environmental trigger demonstrates the high level of concern in this lay sample who have direct contact with ASD. Given the extent of media coverage of MMR as a potential cause of ASD it is

perhaps surprising that such an enormous range of other theories emerged. The correspondence also highlights a sharp disconnection between lay beliefs and the expert consensus explaining the increasing prevalence of ASD. The explanations given by lay correspondents focus on new hazards introduced by medical and technological advances and the lifestyles of modern society, with such hazards being responsible for the 'explosion' (Letter 52) or 'epidemic' (Letter 13) of numbers of cases of autism.

Most of the environmental risk factors proposed in the letters were already included in the scientific programme of the epidemiology autism research grant. Some other ideas were categorized as 'plausible' by the epidemiologists and indeed went on to provoke original research. Ultrasound and carbon monoxide poisoning were both examined as a direct consequence of the letters, for example. The correspondence allowed epidemiology to reap the benefit of the knowledge and experience of those with direct contact and differing relationships to autistic disorders.

Calnan (1987) highlights the logic and integrity of lay beliefs born of their social context, while Beck warns against removing the human and emotional aspects from scientific inquiry. Science should consider what is culturally significant he argues: 'Social movements raise questions that are not answered by the risk technicians at all, and the technicians answer questions which miss the point of what was really asked and what feeds public anxiety' (Beck 1992). Correspondents did not lack understanding of the 'correct' biomedical interpretation of the situation. As Greenfield and colleagues (1987) have pointed out, lay explanatory models cannot be viewed as misconceptions because they contradict current biomedical explanations. Rather, lay explanations put forward other arguments. In time these views may become more accepted, or they may not. Today, it is unclear whether true incidence has increased, and the exact aetiology of ASD remains uncertain in most cases.

Collins and Evans (2007) argue that legitimacy afforded in the western world to 'lay expertise' through consultations at policy levels has gone too far. They argue for the recognition of the elevated value of consensus medical expertise expressing a preference for norms and cultures of evidence based scientific research. Nevertheless it is important for medicine to acknowledge and be informed by the 'situated knowledge' described by Haraway (1991) as locatable critical knowledge based on the experience of actors. Perhaps a third way can be found, where lay voices neither dominate the agenda nor are ignored. Lay views can embellish paediatric expertise and provide valuable insights. In our study the correspondence contained information that epidemiologists were not aware of and upon which they were able to capitalize. Seeing through the eyes of those at the 'coal-face' – relatives, affected individuals, a range of clinicians – also provided them with emotional and cultural perspectives on their work.

Szpir claims this issue – of increasing prevalence of ASD and the reasons behind it – has divided scientists as well as the public. It seems in the light of our study, much lay opinion lies on one side of the debate and most expert opinion on the other. This appears to be an instance where the professional consensus (or at least dominant view) is that the increased incidence is an artefact of changes in the diagnostic arena, while the correspondents' appear to take the increased incidence as a fact. The professional view here accepts that scientific understandings and opinions change – in other words, that 'facts' can be constructed and relative. The public view, as expressed here, is to argue for the 'fact' status of increasing incidence. The environmental aetiology because of modernization is also consonant with this belief in increasing incidence. Correspondents were not anti-science, however. They showed their faith in the legitimacy of science as an epistemic authority with their requests to test their ideas about the reasons for the increase. According to them modern life and medical technologies bring new risks and this is what underlies the increasing prevalence of autism.

Key messages

- The number of children with autistic spectrum disorders worldwide has increased in absolute terms according to lay opinion.
- Many lay people believe this increasing incidence of autistic spectrum disorders is due to increased exposure to new environmental, medical and technological hazards, including vaccinations.
- The position of lay opinion contrasts with consensus expert medical opinion where changes in diagnostic practice are perceived as responsible for increases in prevalence.
- Lay opinions may become accepted in time – and cannot be dismissed as misperceptions. They may provide useful insights for researchers and paediatricians alike.

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Looking beyond risk: A study of lay epidemiology of childhood disorders

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Davison, Frankel, and Davey Smith (1989) conceptualised lay epidemiology as the process of interpreting health risks through considering non-traditional sources of information drawn from personal networks and from the public arena. We analysed 100 unsolicited letters received by professional epidemiologists studying the causes of autistic spectrum disorders (ASD). The correspondents sourced their ideas from an interaction between personal, social, media, and scientific sources, as well as conducting their own studies. Thus their correspondence represented a form of lay epidemiology in action, and as such provides a useful pointer to how 'lay expertise' may be harnessed in practice. The vast majority of the letters put forward theories and hypotheses about hazards related to early twenty-first century society, either medical technologies, unavoidable by-products of technology, or risks created by modern lifestyles. Given the conclusions of previous studies a surprisingly small minority put forward causes related to vaccines. This disparity is discussed. The findings shed light on the nature of public trust of institutions involved in the definition of risk. They underline the strength of public concern not just about vaccines, but more widely about risks of modern society and technology causing childhood disorders.

Keywords: autism; Autistic Spectrum Disorders; lay epidemiology; expertise; childhood disorders

Introduction

The medical fields of nosology, aetiology, and epidemiology all 'have identifiable counterparts in the thoughts and activities of people outside the formal medical community' according to Davison, Davey Smith, and Frankel (1991). They define lay epidemiology from a sociological perspective as the process of interpreting health risks through considering non-traditional sources of information drawn from personal networks and from the public arena (Davison *et al.* 1989). Whereas in traditional epidemiology the focus is on those causes which exert the largest effect, in lay epidemiology, without the overview of a large amount of data, the emphasis shifts to personal situations drawing on a wide range of sources to identify perceived causes. In his seminal text *Risk Society*, Beck (1992) outlines how risks brought about by industrialisation have entered the public consciousness. He argues that benefits of modern technology are accompanied by many dangers and many of these risks are unseen. It is the science, he suggests, often in the form of epidemiology, that defines these risks, rendering them visible.

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Research in the UK on public understanding of risk factors for autism has frequently concentrated on the parental and public resistance to vaccination; specifically the measles, mumps, and rubella (MMR) vaccine after the much publicised report by Wakefield *et al.* (1998) suggested a link between autism, Crohn's disease, and the MMR vaccine. Little work has been carried out on the public's conceptions of other risk factors, although some studies address parental understandings of ASD aetiology. Such studies often had a clinical rationale and have been directed toward improving compliance with clinician recommendations (Furnham and Buck 2003) and at enabling genetic counsellors and parents to communicate more effectively (Mercer *et al.* 2006).

According to social scientists such as Wynne (1980) and others, trust in scientific institutions influences the uptake and understanding of scientific messages about risk. Publics are able to critically evaluate sources of risk information, and this process of trust has been studied with reference to resistance to MMR (Hobson-West 2003, 2007, Brownlie and Howson 2005). In Casiday's (2007) analysis of interviews about MMR, parents experience uncertainties and parental dilemmas as they 'weigh up' risks, both in terms of likelihood of developing conditions but also the gravity of outcomes, with autism seen as a lifelong disability. Here and elsewhere sources of information about risks are evaluated for trustworthiness when parents consider who to believe. Studies have suggested that conditions to promote trust include personal familiarity (Brownlie and Howson 2005) and an acknowledgement of uncertainties by sources (Marris *et al.* 2001). Kee and Knox (1970) evaluated the dimensional nature of trust: it is not a question of trusting or not trusting an institution, but the *degree* to which it is trusted. Further, the degree of trust is dependent on a person's perceptions of the motives and competence of the institution, as well as their own personal experience, disposition, social identity, and situational factors. Wynne points out that people may publicly profess to trust an institution when in private they hold a different view, and also draws attention to the reluctance of establishment science to take heed of lay expertise (1995).

Over the last half century, biomedical and popular understandings of the root cause of autism have undergone a shift in emphasis from a psychiatric disorder caused by cold mothering to a genetic and biological disease (Nadesan 2005). A common model is that an environmental trigger leads to expression of a genetic predisposition. Both Gray (1995) and Furnham and Buck (2003) showed that most parents of children with autism accepted a genetic cause or congenital damage as an explanation for the aetiology of their child's autism.

Three major social identities concerned with autism and ASD have emerged, each with its own agenda and organisations. First, parents' groups came together to fund research into causes of ASD as well as research into interventions. They are committed to treatment and cure, and employ genetic and biological models to describe ASD, through organisations such as 'Autism Speaks' (described from a social science perspective by Bumiller 2009). A number of such disease specific supporter groups have wielded considerable influence over research and research funding. For example, Rabeharisoa and Callon (2002) describe the increasing influence of patients' associations in France. 'As collectives' writes Bumiller, 'they have tried to maximise their influence on the development of new science, technology and medical knowledge'. The ability of such groups to influence the research agenda is part of the wider 'democratisation' of science. This takes several forms, including

greater public engagement, publics having increasing access to scientific advisory bodies, departments and institutions, and lay critiques of scientific methodologies and results. The issues of democratisation are discussed in more detail by Kleinman (1998).

A second active group is the autistic rights movement (ARM), a collection of self advocacy groups formed by autistic adults who use genetic and neurological models of autism to claim the right to 'neurodiversity'. The ARM argues that autistic people bring with them many desirable traits, such as close attention to detail and islets of ability in areas such as logic, science, and information technology. In contrast to the position of parent-led treatment-oriented groups they are anti-cure, acknowledging and elucidating their differences (see Baggs 2007 for a powerful film by an autistic woman giving an account of this position).

Third, it is largely parents who have come together to form activist groups centred around the possibility of environmental causes of ASD, notably vaccines. In the UK, widespread resistance to the MMR vaccine has been a focus for activists, despite evidence showing that the vaccine is not associated with ASD (Rutter 2005). In the USA, the use of the preservative thiomersal, which contains mercury, in many vaccines has been cited by activist groups, known as the 'Mercury Moms'. According to Bumiller (2009) they are marginalised because their 'advocacy is seen as dangerous because it could lead to widespread rejection of vaccinations that could prevent resurgence of deadly diseases'. The concept of lay epidemiology has usually been deployed to explain lay resistance to new public health measures (see Bury 1994) However the MMR vaccine was well established, instead it was the Wakefield *et al.* (1998) article that provided the spur for lay resistance.

There is a growing social science literature on autism, reviewed recently by Silverman (2008). Increasing attention has been given to the ways ideas about autism are represented and reinforced through popular culture, autistic autobiographies, medical literature, self help books, support groups, and through the treatment and intervention industries (Murray 2008). Bumiller points out that autism activism and group identity seems well described by the notion of 'biosociality' (Rabinow 2005). This term describes the growth of social identities associated with inherited medical conditions. The internet has been influential in allowing parents of children affected by specific rare disorders, as well as those affected themselves, to create virtual communities even when they are geographically dispersed.

Work in cultural studies such as that by Singh *et al.* (2007) has highlighted media biases in selectively communicating information. They noted that 41% of published research on autism focused on brain and behaviour research, but only around 10% of newspaper stories in the USA, the UK, and Canada dealt with this, and nearly half dealt with environmental causes, particularly sceptical reporting of the MMR evidence. Parents of children with regressive autism who were diagnosed after the MMR scandal tended to recall onset as being shortly after MMR more frequently than parents whose children were diagnosed beforehand (Andrews *et al.* 2002), showing that the scandal had engendered recall bias in these parents. The MMR controversy had a considerable impact on parents (Hobson-West 2003, 2007). In the UK, public fears about the MMR vaccination led to a decline in vaccination rates and an increase in the incidence of measles in 2006.

Vaccine activists often cite the science that back up their claims. For example DeSoto and Hitlan (2007) are often cited by the 'mercury moms', as their study

found mercury levels to be associated with autism diagnosis. Other studies have not supported this association, although thiomersal has now been withdrawn from use in vaccines in the USA. Philosophers of science have described the selective use of science to support agendas and back up claims (Hauskeller 2005).

Many environmental factors have been implicated as risk factors for ASD by epidemiologists. A comprehensive review reported recently that identified effects are often weak and findings are often not replicated (Newschaffer *et al.* 2007). There is no one clear aetiological path for the development of the condition, more likely many different underlying causes that converge in the expression of similar behaviours (Happé *et al.* 2006). Specific biological causes are also poorly established: scans and autopsies have shown that several brain structures have been affected in individuals with autism but none of these findings have been consistently replicated, according to another broad review (Benayed *et al.* 2005). As no simple genetic or neurological explanation has emerged, some social scientists and researchers are concerned that genetic studies have been given priority over work on environmental hazards (Herbert and Silverman 2003, Szpir 2006). Both genetic and environmental causes of autism remain uncertain within biomedical communities, and this knowledge vacuum has, in various ways, been transmitted to the public.

Calls by policy advisors such as Watterson (1994) and others for greater engagement of health institutions with lay epidemiology reflect the general policy trend to encourage more public engagement with science. Engagement tends to be framed as public attitude research, societal response to scientific activity/discovery, or public engagement 'events' rather than how 'publics' might *themselves* initiate engagement with scientific knowledge production. In medicine, there is a dearth of studies that treat parents or publics as 'active' participants, rather than the 'subjects' of medical research. Social science research that does fall into this approach has focused on social movements; patients involved with advocacy groups, as part of their social identity. We present an analysis of unsolicited communications with an epidemiological research programme by members of the interested public: spontaneous forms of public engagement with an institution concerned with the definition of risk. In this study of unsolicited public communication we also sought to examine the public's conceptions of risk factors.

ASD represents a useful case study because, like a number of other disorders, the aetiology is uncertain, likely to be complex, and is contested both within and outside academic scientific circles. The sample is distinctive because it represents not just parents but a range of individuals who are committed to a specific topic of interest and have pursued that topic extensively.

Methods

In 2004 a major grant was awarded to a well-established longitudinal cohort study of children in the UK to examine environmental causes of ASD. A press release was issued by the organisations concerned announcing the launch of this research. This release provoked worldwide media interest, with articles appearing in national and international media outlets, and led to interviews with the director of the study on national radio and local television. Exactly 100 unsolicited letters and emails were received by the study in response to this media attention, and these were the subject of our analysis. Such forms of unsolicited narratives may provide a rich

source of data for analysis in health research (Jones 2000, Robinson 2001). The director of the epidemiological research suggested an analysis of this unsolicited correspondence might be revealing.

The majority of letters were received between 2004 and 2006 and were from the UK, with a small proportion from the USA, Canada, and other Western countries. The sample was self-selecting, from a population that had access to the internet, television, or newspapers. All letters, calls, and emails were included in the analysis, with the exception of those written by participants in the cohort study itself. The unsolicited letters represent a unique source of data as they were not pre-selected on the basis of any limiting criteria imposed by researchers. They represent a selection of interested members of the public who have encountered information about a particular instance of scientific practice. The content of the letters reflected the interests and intent of their producers, unlike the 'co-produced' nature of data from traditional interview sources (Riessman 1993, Hammersley and Atkinson 1994). However it must be noted that the sample was not representative of 'the public' as it was taken from a self selecting group who had a stake in the disorder. Caution must therefore be exercised when generalising from these results.

We noted, where given, whether correspondents were active members of parent groups, self advocates, or any details about their relationship to ASD. A modified form of the thematic analysis described by Braun and Clarke (2006) was used to analyse the dataset. The letters were read in their original form by two researchers. We asked, what reasons for contacting the study do correspondents give, what motivations? What kinds of social action do the communications represent? How do correspondents represent themselves in relation to epidemiology? Who do correspondents trust to define risks according to their communications?

Significant statements and paragraphs were selected from the letters and categorised according to emerging codes. Themes were then reviewed and cross checked to assess whether the relationships between themes accurately reflected the information and meanings in the overall dataset. Finally, themes were defined and refined, to identify the essence of each theme. Replies to all the letters were sent thanking correspondents and indicating whether their suggestions could inform the epidemiological research.

Results

The analysis was limited in that we did not have access to all the media reports which prompted the response. Although the press release described the purpose of the epidemiological study as 'to look at the role of environmental risks in development of autism', of those media reports we did have access to, we found some reinterpreted the study as focusing on MMR. For example, in July 2004 the headlines 'Experts to study "perils" of MMR jab' (*Yorkshire Evening Post*), 'Scientists probe claims of MMR jab link to autism' (*Birmingham Evening Mail*), 'Autism probe into MMR link' (*The Journal*, Newcastle), and 'Funding U-turn on MMR jab study' (*Daily Mail*) were published. This reinterpretation mirrored the findings of Singh *et al.* (2007), who noted the discrepancy between the emphasis of scientific publications and the focus of press coverage which often concentrated on sceptical reporting of the MMR evidence.

This pattern of reporting of autism science reflects the extent to which autism has become a site of social movement action.

Suggested risk factors

A total of 84% of the letters put forward the correspondents' theories and hypotheses about risk factors for ASD. A small portion of the unsolicited mail was concerned with job requests and specific enquires about the cohort study. Of those concerning risk factors, 96% suggested that risks caused by environmental hazards might trigger the disorder. Table 1 shows environmental risk factors suggested by correspondents as potential triggers of ASD.

Beliefs about the aetiology of ASD as found in previous studies (e.g. Gray 1995) were primarily biomedical and biological, with environmental prenatal, perinatal, and postnatal factors well represented. The writers generally adopt a realist approach, assuming there is an answer 'in nature' to the question of what causes autism and that it is simply a matter of asking the right questions. That is, they assume the condition of autism is a real biological or neurological difference isomorphic with nature, and that there is a discoverable 'cause'.

Although MMR was mentioned by a minority of the correspondents (16 in total), a wide range of more than 40 alternative environmental triggers, ranging from prenatal drugs or diet to ambient triggers such as radiation or exposure to chemicals, were proposed. Thus we found MMR (and vaccines more generally), were cited as a cause of ASD much less often than might be predicted from the reinterpretation by the media discussed above and existing literature. This emphasises the influence of mobilised anti-vaccine groups (Hobson-West 2007) and widespread resistance to MMR vaccination by parents in the UK (Brownlie and Howson 2005).

What virtually all the theories had in common were their origins in man-made risks, products of technologies and lifestyles of the late twentieth/early twenty-first century. We were able to classify all but three of the risk factors proposed into three sub categories. First, those caused by new medical technologies (e.g. ultrasound, drugs to prevent miscarriage); second, those caused by ambient effects of modern technologies (e.g. radiation from mobile phone masts, carbon monoxide pollution); and third, those caused by diet and lifestyle (e.g. too much sucrose, gluten, additives, origins of milks, or too much television). Thus almost all the theories related to perceived hazards about technology. Many of the theories proposed by correspondents reflected reports of associations found in the scientific literature. However we were unable to find any scientific literature pertaining to more than 10 of the risk factors cited by correspondents. Some of these apparently underresearched theories were deemed 'plausible' by epidemiologists, although data was not available, while in two cases (carbon monoxide poisoning and ultrasound) the correspondence provoked original research.

The risks of modernisation as described by our correspondents are invisible risks as described by Beck (1992) in terms of the scientific knowledge about them; they can be changed, magnified, or reduced within that knowledge. The invisible nature of such risks leads to more speculation. Beck describes how people 'reflexively' interact with society in a growing awareness of risk. Epidemiology, the definition of risk, is in a sense a reflection and critique of modern society itself, and this makes epidemiology a political issue as risks come about as a result of man-made threats and economic pressures (Beck 1999).

Table 1. Environmental risk factors suggested by correspondents as potential triggers of ASD.

PRENATAL FACTORS	
	Ultrasound scans
	Miscarriage/bleeding
	Foetal stress/stress to mother
	Born after twins
Diet during pregnancy	Food additives/aspartame
	Alcohol drunk
	Pollutants in water
Vaccines/drugs during pregnancy	RhoGAM shots
	Contraceptive pill
	Steroids
	DES (drug to prevent miscarriage)
	Antihistamines
Ambient	Father works in nuclear power stations/ exposure to radioactivity
	Exposure to chemicals
	Living near mobile phone mast/exposure to radiation
	Moulds from indoor environments
	Air pollution
PERINATAL FACTORS	
Medical interventions	Baby induced
	Early cord clamping
	Respiratory distress
	Caesarean section
Other complications	Birth trauma
	Dry birth (no amniotic fluid)
	Cord wrapped around baby's neck
POSTNATAL FACTORS	
Diet	Disaccharides and starches
	Food preservatives
	Genetic origin of cow milks
	Gluten in diet
	Lack of cod liver oil
Vaccines	Time of day of vaccination
	Lack of aspiration (pulling back on the syringe plunger prior to injection).
	MMR
	Mercury due to thiomersal
	Vulnerability to injections when teething
	Polio vaccine
	Egg products in vaccines
	Pain of injection
	DPT/toxins
Ambient/other	Low level radiation e.g. VDUs
	Carbon monoxide exposure
	Indoor air quality
	Over stimulation by cot toys
	Too much television

A proportion of the letters (approximately 10%) mentioned genetics or family history, putting forward a model where ASD may be the result of genetic predisposition triggered by one or several environmental risk factors. Letters frequently made links with other diseases and disorders:

> Educator (teacher): I have also seen a number of autistics and children with Aspergers who also get labels of ADHD, DCD and dyslexia.

> Parent: I understood autism to be part of a spectrum of disorder along with ADHD, Aspergers and Dyslexia ... will you be investigating possible genetic links?

The reason correspondents included other conditions, particularly childhood disorders such as ADHD, dyslexia, dyspraxia, specific learning difficulties, and developmental coordination disorder (DCD), as well as medical conditions such as bowel disease and asthma, was not to question the current understanding of nosology and the classification system, rather to suggest that these other childhood conditions might have similar underlying environmental causes to autism, 'a complex web of associations' as one correspondent put it. Thus in the minds of the correspondents, a wide range of environmental hazards could potentially be causing a range of other childhood disorders, and these were hazards caused by late twentieth/early twenty-first century technologies and society:

> Parent: I have four children and had the RhoGAM injection both during my pregnancy and after my pregnancy ... My oldest daughter is a well behaved child, does well in school however has ADD and is on medication. My second child ... has some major psyche/neuro issues ... his behaviour is horrible and he is on major psyche/neuro drugs and has been in and out of institutions and doctors for several years ... my fourth child ... has Retts syndrome ... She has seizures, she does not walk or talk, she is now 101/2 and functions at the three month old level.

The make up of the sample

A total of 83% of the letters described the correspondents' relationship to autism or ASD, in particular the basis upon which their knowledge of autism was founded and the reason they 'should be taken seriously' by the epidemiologists (particularly by the research director). Of those that described their relationship to ASD, most were parents or grandparents of autistic children or had family members with autism or related conditions; some were professionals in contact with autistic individuals, such as teachers, speech therapists, advisors, alternative practitioners, and midwives; others were doctors, scientists, or scientific researchers; and a minority were on the spectrum or suffering from similar conditions themselves. Figure 1 shows the breakdown of the sample in terms of their relationship to ASD where it was given.

The letter-writers saw their expertise, although not necessarily 'scientific', as being nonetheless credible and valid, reflecting a view of expertise described by Evans and Collins (2007) as social fluency within particular communities. There were many claims to expertise in this wider sense; extensive experience caring for a child with autism, years in clinical practice, personal experience of the symptoms of autism, or experience as a head teacher at a special school for children with social, emotional, and behavioural difficulties, were some examples:

> Health professional (retired therapist): I am now retired, but worked for over 30 years as a speech and language therapist for the NHS.
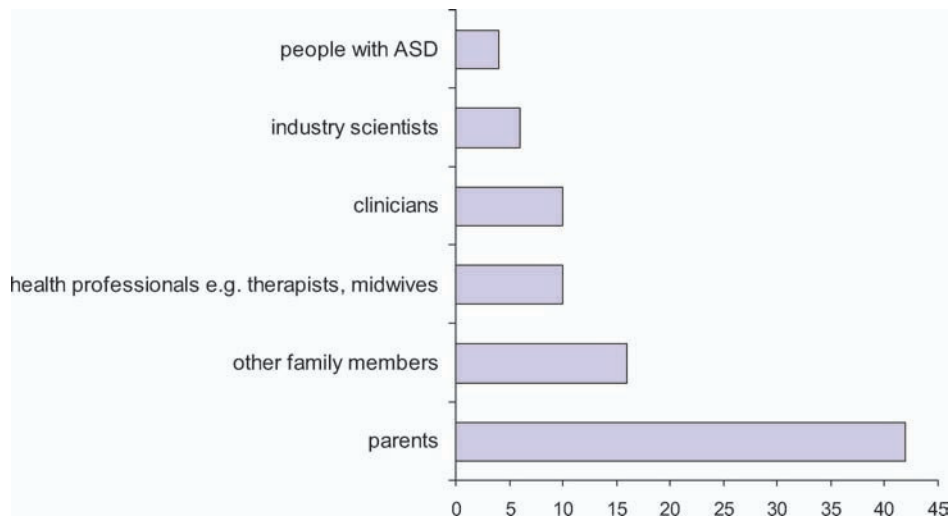
Figure 1.   Distribution of sample and relationship to ASD where given.

> Person with ASD, learning disabilities professional: I would like it [if the epidemiologist considered his theory] since I've spent ten years researching and making sense of my own situation ... [I am] now a job coach for learning disabled people, specialising in autism.

Epstein (2010) notes that boundaries between lay people and experts have become 'fuzzy': health activism seems increasingly less suited to explanatory models that presume an opposition between experts and lay challengers. Our sample is well described by Epstein's notion of a 'hybrid' group. Here a *hybrid* sample is one that draws from across a wide spectrum of expertise; what the hybrid sample shares in this case is an interest in and a relationship to autistic spectrum disorders. The correspondents represent their expertise in ways that they perceive would command the attention of the epidemiologists. Some were self-taught lay-experts who had educated themselves in science as the basis for their own theory development:

> Person with ASD: I say this only to add to my credibility that if I dedicate a year of my life to intensively studying an area of medicine that I have attained sufficient understanding to be able to write a paper published in a peer-reviewed journal then what I say should be taken seriously.

This strategy is reminiscent of that described by Epstein (1995) in his analysis of AIDS activists who educated themselves in the scientific basis of their subject area and then used scientific language to demand consideration by the biomedical researchers whom they addressed. Almost all of our sample felt that that they possessed expertise relating to ASD but this was expertise from the perspective of different disciplines and communities to epidemiology. This expertise, scientific, professional, and personal, was frequently displayed in order to prove their credentials, the reason that epidemiologists should heed their words. Epidemiologists and publics talk across an 'expert–lay divide' according to Wynne (1996). Lay epidemiology here extended into a process of reaching across these cultures allowing the culture of traditional epidemiology to reap the benefits of lay expertise.

Sources of information: from experiential to the scientific

Correspondents cited a wide range of sources of information and data, ranging from simple observation and their own experience, to extensive reviews of information on the internet, to conducting their own research studies. Thus the letters proposing risk factors clearly represent an expression of lay epidemiology coined by Davison *et al.* (1989): experiential sources were often interwoven with more traditional scientific approaches to data sources:

> Person with ASD: I have also spent an enormous amount of my time over the years reading thousands of postings on the internet ... and by reading these I learnt a great deal more about the disease than has been published in medical literature.

The relative influences of each type of information source on lay epidemiology have been considered elsewhere (Hunt and Emslie 2001). In this dataset many correspondents had also carried out their own research and designed their own studies. These were of particular interest because these often had quantitative methodologies reflecting epidemiological techniques or qualitative methods reminiscent of those used in social sciences. Within these studies a variety of sources of data were utilised and an array of methodologies were adopted:

> Parent: Hello, this is my little study I did on mothers who were 'injected' with mercury while pregnant ... [goes on to list 16 names, 17 including herself; gives details of injections containing thiomersal during pregnancy, and brief descriptions of developmental problems of children.]

Some correspondents had published these studies in peer-reviewed journals, others had utilised their own experiences to formulate their own questions. In many cases it was difficult to distinguish among these studies which were 'science' and which were 'non-science'. Science appears here relationally defined by its context of production or dissemination. Despite this, the desire was communicated for traditional epidemiology to quantify risks and provide clear epistemic authority:

> Parent: I desperately want a clinical study looking at children who have reportedly regressed ... We have to have studies into children who have suffered regressive autism otherwise we will never know the answers ... Prof. X I plead with you to look at these children ... Please Prof. X please help my children and thousands like them and find the answers.

The studies carried out by the correspondents themselves give the impression of a group of people highly motivated by their interest in this specific topic. The time and effort put into these studies conveys a sense that the authors were aware, perhaps unconsciously, of the increasing 'democratisation' and growing participatory culture of science.

Hobson-West (2007) refers to a 'discourse of ignorance' adopted by anti-vaccine groups when challenging government representations of risk. She notes how risk is reframed by vaccine activists according to their own agendas. Instead of dismissing risk statistics out of hand, they questioned whether previous methodologies had really looked at salient factors. This was also true in the current dataset, and there

were many requests to adopt particular methodologies in order to reveal hitherto unforeseen risks:

> Industry scientist: Is there any room in the new work to undertake some dietary manipulation work, and the imposition of A2 milk in situations where there are significant problems with autistic behavior?

Despite having conducted their own studies and in many cases having published their own results, a self-proclaimed 'uncertainty' permeated the correspondence in this study:

> Health professional (database collator): I have spent the last two and a half years validating the computerized medical records of a dental practice in London … by the time I had a feeling about this and started to gather figures, but I left for my present position and so do not have enough data to make a qualified scientific comment. I worked on about 2000–2300 records, not all children.

Explicit in some letters and implicit in the rest was the request to test proposed risk factors for associations with autism. This was the rationale for writing. This underlines the assumption that ASD is biological, that its cause will be discovered. Uncertainty is recognised but is framed as a temporary phase that may be overcome by more research. Here traditional epidemiology, as represented by a large, funded, institutionally located and authoritative study, will have a greater 'voice' and impact than the writers themselves as lay epidemiologists. The authority accorded to the study and trust in the power of institutional epidemiology to define risks was reflected in the fact that 30% of correspondents also offered their data, themselves, and even their children as research subjects: 'you are welcome to include my children in your studies'. Although the dataset as a whole voiced their trust in the epidemiologists, a minority of correspondents, those writing about the vaccines around which parent groups have mobilised, expressed a deep mistrust and scepticism of the institution of 'mainstream science', and a feeling of being ignored and marginalised:

> Parent (active member of parent anti vaccine group): This is just a waste of public money and I'm sure many others will agree with me, how the hell are we supposed to take this seriously if all science does is skirt around the truth to protect the pharmaceuticals, it's a disgrace!!

These results illustrate Brownlie and Howson's (2005) framing of trust as a complex relational practice within particular socio-economic contexts, in this case the context of mobilised anti-vaccine campaigning. Wynne (1996) suggests that given the uncertainty about risk, questioning the trustworthiness of particular institutions is a rational strategy. According to Hobson-West (2003), health literature pointing out relative risks is ineffective because parents mistrust the sources of such information. The mistrust of institutional, and particularly government, science by the parent activists in our sample produced an uncomfortable tension in some letters, where establishment science was viewed as untrustworthy but the epidemiological study (itself an exemplar of established scientific practice) is viewed as a force to legitimate and define risks, being able to 'settle the issue'. Nelkin (1975) alludes to this paradox, in that technical knowledge is widely regarded as a source of power and as such scientists are both

indispensable and suspect. This duality, encompassing both scepticism and ultimate faith in deliverance, is neatly summarised within one letter:

> Relationship to ASD not given: Many doctors do not report adverse reactions because the ones the parents talk about are 'rare reactions' in the drug manufacturers leaflets and doctors do not know of these reactions. They only know what the government leaflets say ... Our children are sick and getting worse. They are in a lot of pain and are being ignored by the medical profession. More studies WILL be coming out soon which will prove the link.

Here a conception of science as having been wrong and inadequate in the past, but clear and useful if carried out in the way suggested by the correspondents, is presented.

Discussion

The correspondents were a highly heterogeneous group, united by their common concern about autism. Our sample of letters cannot be described as an attempt by a uniform group of disempowered people to oppose the scientific consensus from the outside. Experts such as clinicians, scientists, and researchers in industry were represented. Many correspondents were parents, had family members with autism, or were on the spectrum themselves. Others were professionals with an interest, such as teachers, speech therapists, special needs coordinators, alternative practitioners, and midwives. Some were knowledgeable and educated about scientific arguments in the field, others were not. As we have written elsewhere (Russell *et al.* 2009), correspondents appeared more convinced that the actual incidence of autism is rising than is reflected in the specialist literature, which emphasises increased awareness and refined diagnostic practices. The response to media coverage was an instance of lay epidemiology in reaction to uncertainty. This was the uncertainty about causes of autism and lack of health advice on how to avoid it, fostering the deluge of correspondence concerning lay theories. In this sense, lay epidemiology seems a focused attempt to fill the vacuum that exists in biomedical understandings about causes of ASD.

Our first finding is the strength of public concern with potential risks of modern lifestyles and new technologies, not just for ASD but childhood developmental disorders more generally. This latent unease may in part explain the lack of focus on MMR and vaccines in the dataset as a whole. Three other reasons may partially explain this result. First, our sample only captured a proportion of parents, and a minority of these explicitly identified as part of anti-vaccine groups. The sample of letters also encompassed professionals and scientists who may have been less likely to be concerned by vaccines. Second, our sample comprised those who were responding to press and media reports of the epidemiological research which sometimes focused on MMR vaccines. Many correspondents were therefore aware that the epidemiologists were already looking at vaccines as risk factors, and had no need to suggest that the study should test this association. Third, our study revealed an inherent trust in institutional epidemiology. Those parents most likely to question risk statistics generated by traditional epidemiology are also those who resist vaccination (Hobson-West 2007). This group therefore may not have been motivated to influence the direction of traditional epidemiological research.

Trust

Trust in traditional epidemiology was highlighted: concerned citizens accorded authority to the recognised epidemiological institution to delineate the risks associated with ASD. However, mistrust of other scientific and governmental institutions was expressed by a minority of correspondents, revealing trust to be a 'complex relational practice that operates at a number of levels including the individual, interpersonal, institutional and socio-political' (Brownlie and Howson 2005). Further, although correspondents emphasised their expertise within their own realms, they stressed their own ignorance, uncertainties, and doubts, despite the extent of their own lay epidemiological research and studies.

Brownlie and Howson (2005) argue that the 'leap of faith' inherent in trust is partly based on relations of familiarity. Responding to press and broadcast media does not imply a great deal of familiarity, although perhaps television appearances by the director of the epidemiological study did breed some familiarity and consequently trust. Our findings on this point are therefore inconclusive. Current uncertainties in the aetiology of ASD were also acknowledged in broadcasts where the research director professed to be 'open minded'. Marris *et al.* (2001) suggest that if uncertainties are denied by scientific institutions, the institutions are less likely to engender public trust. Here we have seen the question of who is trusted to define health risks and under what circumstances depends on trust in institutions and the authority they are accorded. This appears dependent on both social and political circumstances.

The communications with the study followed from understandings or perceptions of epidemiology as a flexible activity, distinguished from their own work by the authority given its claims. The correspondents frequently represent what they are doing as 'scientific' and by some as 'science', but not as 'facts'. Thus the 'discourse of uncertainty' is adopted. 'Facts' are claims that are recognised to have authority; it matters who makes the claims. We cannot say that parents, professionals, and those in contact with ASD were not concerned about MMR and vaccines. Rather, a much wider spectrum of concerns emerged. This sample underlined general concerns about modern technology, of which MMR is one example. Brownlie and Howson (2005) also note that concern about MMR in their study could be seen as part of more generalised risk anxiety over new technology.

Was the controversy stirred by the Wakefield *et al.* (1998) paper an event waiting to happen, reflecting as it did the underlying unease of concerned citizens with potential risks posed by modernity? Would any suggestion of association between a technological hazard and autism or other childhood neurodevelopmental conditions have been seized on, in an expression of this underlying distrust? Could we envisage the same scenario and media furore if epidemiologists had found an association with ultrasound and ASD: parents refusing routine scans, for example? In fact MMR was a particularly potent case as a potential risk factor for autism as it is administered at about 15 months, while the typical onset of regressive autism, where children may progressively lose their communication and social skills, is around 24 months. MMR was therefore likely to be a persuasive candidate for parents highly disturbed by and searching for causes of their children's regressive autism. But the strength of feeling evidenced by the letters demonstrates latent nervousness not just with vaccines, but unease with the perceived hazards from our technological society generally, and their effect not just on autism but on a wide range of childhood neurological conditions. On this evidence, and as Hobson-West

(2007) points out, future examples of resistance to health technologies and modernisation can be anticipated.

Lay epidemiology in action

The unsolicited correspondence was written by those who had a specific interest in identifying whether particular environmental risk factors are associated with ASD. Their knowledge was born from a combination of personal experience, media, and scientific sources. Certainly this was 'lay epidemiology' as defined by Davison *et al.* (1989) but in reaching across lay/expert boundaries the correspondents went further than this.

Risks were formulated from lay sources: 'personal networks and public arena, as well as from formal and informal evidence arising from other sources such as television and magazines' (Davison *et al.* 1989, p. 428). But these risks were brought to the attention of epidemiology, and their insights, moreover, allowed epidemiology to take advantage of expertise of those with direct and personal knowledge of ASD by incorporating the ideas into the research design.

Most correspondents to the study represented themselves as possessing some form of expertise (experiential, credential, or in some instances scientific) with regard to autism that justified engagement with the study or provided a basis on which their observations might be taken seriously by the scientists. This suggests a broad and flexible understanding of 'expertise' relevant to epidemiology and the definition of risk. Rather than understanding expertise in terms of training, Evans and Collins (2007, p. 620) suggest that 'knowledge is acquired by socialisation, so expertise is acquired through a prolonged period of interaction with relevant communities and is revealed through the quality of those interactions'. Expertise, from this perspective, can be understood as a continuum of knowledge states reflecting the quality of social fluency within a relevant specialist community.

Calls for greater public engagement with health research are emanating from a range of sources, including public funding bodies. Although there is significant action by social advocacy groups with regard to the aetiology of autism, our study highlights the level of *private* activity and mobilisation. Such activity may become increasingly important with the use of information and communication technologies that facilitate direct communication with scientific projects. Together such challenges may modify traditional ideas of epidemiology and have the potential to remake professional boundaries and alter the practice of research. They present challenges also to the structures of power and inequality as they have the effect of transforming power relationships between expert and 'lay' actors.

Watterson (1994) argues that the very process of involvement of lay epidemiology would ensure greater public confidence in definition of risk, but that instances of good practice where epidemiologists help communities do their own epidemiology are rare. Casiday (2007) suggests that the 'challenge for doctors and scientists is to find ways of taking seriously these experiences while interpreting other types of evidence'. Brown (1997) lists hazards and disease uncovered by lay epidemiologists and suggests that professional epidemiologists could usefully appropriate the data provided by them. This seems optimistic as professional epidemiologists employ strict methodological techniques, the boundaries that the lay epidemiologists prefer, often restricting data to that gathered from an initial cohort. Our study does show one aspect of how lay epidemiology

may function in practice, not in increasing the number of data points but in offering *insights* from the front line of a condition which could surely catalyse fruitful research. In our study the letters themselves succeeded in inspiring and shaping study design, a neat demonstration of lay epidemiology's success in reaching across the boundaries between communities.

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