Latent Structure of the Autism Phenotype

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Certification

I, Rachel Grove, certify that this thesis is my own work and has not been submitted for a higher degree to any other university or institution. While Rachel Grove is the primary author for each of the manuscripts contained in this thesis, multiple authors are also listed. Rachel Grove was responsible for the preparation of each of the papers. The contributions of each author are specified below.

Chapters two, three and four. As principal author Rachel Grove was responsible for the overall study design, data analysis, interpretation of results and preparation of the manuscript for publication. Andrew Baillie provided guidance on the application and interpretation of the data analysis. Carrie Allison and Simon Baron-Cohen provided the data that was used in the manuscript and reviewed the manuscript for publication. Rosa Hoekstra provided input into the study design, interpretation of results and reviewed the manuscript for publication.

Chapter five. Ilona Roth and Rosa Hoekstra developed the Special Interest Motivation Scale as part of their extensive survey of special interests in adults on the autism spectrum (Roth et al., 2013; Roth et al., 2014 in preparation), and provided the data for analysis. Rachel Grove was responsible for the overall study design, data analysis, interpretation of results, and preparation of the manuscript for publication. Rosa Hoekstra contributed to the data analytic strategy. Both Ilona Roth and Rosa Hoekstra provided input into the study design, interpretation of results and reviewed the manuscript for publication.

Data utilised in this thesis was obtained from two existing data sets. Therefore, Rachel Grove was not responsible for designing the data collection methods and the measures included in the data collection phase. Chapters two, three and four contain data provided by Simon Baron-Cohen and Carrie Allison at the Autism Research Centre (ARC) at Cambridge University in the United Kingdom. This data set contains individuals with autism, parents and general population controls. Data was collected online via two volunteer webpages at the ARC and the Department of Psychology. The collection of online data enabled a large sample (n = 1034) to be obtained containing comparable numbers of individuals with ASC (n = 363), parents (n = 439) and controls (n = 232).

The second data set was provided by Rosa Hoekstra and Ilona Roth from the Open University in the United Kingdom. This data set also contains individuals with autism (n = 158), parents (n = 185) and controls (n = 267). Data was collected online through the Open University's Biomedical Online Research Network web portal.

The use of large data sets is critical for implementing latent structural statistical techniques. These data sets provided a unique opportunity to evaluate the latent structure of the autism phenotype within large samples of individuals with autism, parents and controls. The use of these existing data sets enabled an assessment of whether the latent structure of the autism phenotype differs across these three groups.

This thesis is presented in non-traditional research thesis by publication format as outlined and recommended by the Macquarie University Higher Degree Research Office. It is comprised of six chapters consisting of four individual papers prepared for publication and an overall introduction and discussion. As a result of this structure the main themes and literature review components are repeated across chapters.

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Thesis Summary

The Empathising-Systemising (E-S) theory of autism argues that the persistent deficits in communication and social interaction in autism spectrum conditions (ASC)[1] can be accounted for by an impairment in empathy, whilst the repetitive behaviours and narrow interests can be explained by a strong drive to understand and derive rules about a system; namely systemising. Autistic traits are thought to follow a continuous distribution, with individuals with autism represented at the more severe end of this spectrum. Furthermore, subthreshold traits have also been identified in first-degree relatives of individuals with ASC, termed the broader autism phenotype (BAP). While there has been some research assessing the cognitive and behavioural symptoms associated with the autism spectrum using clinical samples, no studies to date have simultaneously evaluated the autism phenotype amongst individuals with a clinical diagnosis of ASC, first-degree relatives and community samples. This thesis uses structural equation modelling to evaluate the E-S theory of autism in order to understand the latent structure of the autism phenotype. Furthermore, it seeks to determine whether the latent structure of the autism phenotype is consistent amongst three groups stratified by genetic vulnerability: individuals with an autism diagnosis, parents of a child with ASC, and general population controls.

The first paper of the thesis assessed the relationship between self-report measures of autistic traits. Results highlighted two cognitive domains of empathy and systemising. These two dimensions were more strongly related in the ASC and parent group compared with controls, suggesting that the aetiological factors influencing behavioural traits may differ across controls, first-degree relatives and individuals with autism.

[1] Note that the terms autism spectrum conditions (ASC) and autism will be used interchangeably throughout this thesis

The second paper attempted to search for meaningful subgroups along these two dimensions, identifying three subgroups of individuals defined by their level of empathy, systemising and autistic traits. The first group contained individuals with high systemising and low empathy ability. The second group was defined by higher levels of empathy and a lower drive to systemise, while the third group consisted of individuals with comparable scores on both domains. The first group consisted predominantly of individuals with autism, providing support for the E-S theory. However, there was also evidence that some parents displayed the cognitive profile associated with autism, with approximately 20% falling into this subgroup, supporting the notion of the BAP.

In the third paper, the empathy factor was examined in more detail, highlighting four factors including cognitive empathy, emotional empathy, social skills and a performance-based measurement factor. Results indicated that the performance-based measures were not linked specifically to the cognitive empathy factor, and instead measured empathy more broadly.

The fourth paper of the thesis examined one of the less studied components of the non-social traits associated with autism, outlining the development of a measure assessing motivation and special interests. This paper assessed individuals' motivation to engage in their special interest, highlighting that individuals with autism display higher levels of intrinsic motivation. Results also indicated that individuals with ASC were motivated to engage in their special interest for the pursuit of knowledge. This fits with the notion that individuals with autism have an increased drive to systemise. Higher scores on this measure were also associated with the presence of more autistic traits, indicating that special interests form an important part of the autism phenotype.

Taken together, these studies provide support for the E-S theory of autism and that empathy and systemising can be measured quantitatively in groups with varying levels of genetic vulnerability to autism, ranging from clinical populations (high) to first-degree relatives (medium) and general population controls (low). However, results also suggest that even when these traits are measured quantitatively, meaningful homogeneous subgroups can be identified. The results of this thesis indicate that empathy is multifaceted and best measured using a combination of questionnaire and performance-based tasks. Support was also obtained for the existence of the BAP, particularly in fathers of children with an ASC diagnosis. The importance of special interests to individuals with autism was also highlighted, providing implications for clinical practice. Taken together, these studies exemplified the importance of utilising latent structural statistical modelling techniques to understand and refine the autism phenotype.

Chapter 1

General Introduction

This thesis uses statistical modelling to evaluate the Empathising - Systemising (E-S) theory of autism and the latent structure of the autism phenotype across individuals with autism, parents and general population controls. The first chapter of this thesis provides a general overview and background of the history of autism from its initial conceptualisation to current diagnostic criteria. Previous research assessing the biological and neurocognitive mechanisms associated with autism will then be presented, followed by an evaluation of previous research assessing the autism phenotype. Following this, the main theoretical perspectives that have been proposed to account for the autism phenotype including theory of mind, executive function, central coherence and the E-S theory of autism will be discussed. Finally, this chapter will outline the importance of using latent structural modelling techniques to evaluate the autism phenotype across individuals with varying genetic vulnerability to autism.

Autism spectrum conditions (ASC) are increasingly being diagnosed in young children, with a tenfold increase in the prevalence rate over the last two decades. Current estimates indicate an incidence of 1 in 68 in the US (Centers for Disease Control and Prevention, 2014), with estimates of at least 1 in 150 in the Australian population (Williams, MacDermott, Ridley, Glasson, & Wray, 2008). This increase in prevalence is likely due to the expansion of the diagnostic criteria for autism as well as an increase in the awareness and detection of these conditions (Elsabbagh et al., 2012; Fombonne, Quirke, & Hagen, 2011).

Autism was first described in the 1940s by Leo Kanner and Hans Asperger who delineated a small number of cases of children displaying a difficulty in social interaction as well as stereotypic movements and behaviours (Asperger, 1944; Kanner, 1943). Despite being identified in the 1940s, autism was first included in the third edition of the Diagnostic and Statistical Manual for Mental Disorders in 1980 (DSM-III; American

Psychiatric Association, 1980), labelled as Infantile Autism. Here it was described as involving impairment in social and communication development and an insistence on sameness occurring before the age of three (Rutter, 1978). A revised version of DSM-III was released seven years later, in which Infantile Autism was relabelled Autistic Disorder (American Psychiatric Association, 1987). The next full revision to the DSM emphasised a triad of impairment in social interaction, communication and repetitive behaviours and restricted interests (DSM-IV; American Psychiatric Association, 2000). This also saw the inclusion of a number of disorders including Autistic Disorder, Asperger's disorder, Childhood Integrative Disorder and Pervasive Developmental Disorder not otherwise specified.

Released in May 2013, the latest revision DSM-5 collapses previously defined diagnostic subtypes to encompass one set of diagnostic criteria for Autism Spectrum Disorder (American Psychiatric Association, 2013). Diagnostic criteria have also been revised, with the triad of impairment reduced to two components, combining the social interaction and communication symptoms and retaining the repetitive behaviour and restricted interests domain. The social and communication domain consists of three criteria assessing impairment in social reciprocity, non-verbal communication and understanding and maintaining relationships, all of which are essential for a diagnosis of autism (American Psychiatric Association, 2013). The non-social domain contains four criteria evaluating the presence of stereotyped or repetitive behaviour, insistence on sameness, restricted interests and sensory reactivity. Two criteria from the non-social domain need to be endorsed in order to receive a diagnosis (American Psychiatric Association, 2013). Since autism was first conceptualised by Kanner and Asperger through to the development of DSM-5, there has been an exponential growth in autism research (Lai, Lombardo, & Baron-Cohen, 2013). This has included research assessing the autism phenotype as well as its associated biological and neurodevelopmental underpinnings.

Genetics and autism spectrum conditions

Research suggests a strong biological component to autism (Frazier et al., 2014b), with a heritability estimate of approximately 80% (Ronald & Hoekstra, 2011). There is evidence to suggest that the recurrence rate for autism is 20% (Ozonoff et al., 2011), signifying an increased risk for ASC within families. However, no singular genetic explanation for autism has been identified. Rather, these complex conditions have been shown to result from simultaneous variations in a number of genes, as well as complex interactions between biological and environmental factors (Geschwind, 2011).

There is evidence to suggest that up to 1000 genes may be implicated in autism (Geschwind, 2011; Murdoch & State, 2013). Both rare gene mutations and copy number variations (CNVs) that exert larger effects (Levy et al., 2011; Sebat et al., 2007) are thought to play a role, as well as common genetic variation with relatively small effect sizes (Anney et al., 2012; Chakrabarti et al., 2009). Previous research has indicated that rare CNVs can occur de novo, that is, a new mutation can occur that was not inherited from either parent (Sanders et al., 2012). However, there is also evidence that these rare mutations can be passed down from parent to child (Levy et al., 2011).

Moreover, while some studies have identified potential common polymorphisms that are associated with autism, these result in effects that are not large enough to assume causality (Geschwind, 2011). It may be that combinations of these genetic variants can account for an increased risk of developing autism (Klei et al., 2012). Autism is therefore highly heterogeneous, represented as a spectrum of conditions that present differently in each individual. Due to this heterogeneity, researchers have suggested that there are many 'autisms' with different underlying causal explanations and neurodevelopmental pathways (Elsabbagh, 2012).

Neurodevelopment and autism

There has been a wealth of research assessing the underlying neurological processes associated with autism. For example, it has been suggested that impairment in social reciprocity and attention account for some of the social and communication difficulties associated with these conditions (Eapen, Črnčec, & Walter, 2013). There is also evidence that facial processing (Dawson, Webb, & McPartland, 2005), discrimination of facial emotions (Sucksmith, Allison, Baron-Cohen, Chakrabarti, & Hoekstra, 2013) and direction of gaze (Wallace, Coleman, Pascalis, & Bailey, 2006) is impaired in ASC. A recent theory proposes that many of the symptoms associated with autism can be explained by atypical neural connectivity (Brock, Brown, Boucher, & Rippon, 2002; Geschwind & Levitt, 2007; Just, Keller, Malave, Kana, & Varma, 2012). This is evidenced by lower levels of functional connectivity during tasks assessing language (Mizuno et al., 2011), social (Schipul, Williams, Keller, Minshew, & Just, 2012) and visuospatial (Damarla et al., 2010) processing. Further research has also highlighted an association between reduced (Jones et al., 2010) and increased (Shih et al., 2010) non-task related neural connectivity in autism. It has therefore been proposed that connectivity and communication between brain regions may have a significant impact on the processing deficits that are responsible for the traits associated with autism (Belmonte et al., 2004; Rippon, Brock, Brown, & Boucher, 2007).

However, there are a number of different suggestions as to the way in which connectivity in individuals with ASC differs from atypical development, none of which provide a comprehensive explanation for all symptoms associated with the autism phenotype (Lai et al., 2013). Understanding the autism phenotype provides a basis for a deeper knowledge of the underlying processes associated with autism. As Frith (2012) states, the study of cognition simplifies and guides the investigation of the neurobiology associated with ASC. Understanding the autism phenotype, or the cognitive and behavioural symptoms associated with ASC is therefore paramount.

Analysis of the phenotype

Research evaluating the autism phenotype suggests that the social and non-social traits associated with ASC are continuously distributed across the population and that autism represents the severe end of a continuous distribution or 'spectrum' (Constantino, 2011; Posserud, Lundervold, & Gillberg, 2006; Wing, 1988). This has lead to substantial research into the presence of subthreshold traits in the general population. This has been influenced by the development of quantitative measures like the Autism Spectrum Quotient (AQ; Baron-Cohen, Wheelwright, Skinner, Martin, & Clubley, 2001b). The AQ is designed to detect traits associated with the autism spectrum among adults with normal intelligence. It has been shown to be a good predictor of clinical diagnosis (Woodbury-Smith, Robinson, Wheelwright, & Baron-Cohen, 2005) as well as sensitive to variation in general population samples (Hoekstra, Bartels, Cath, & Boomsma, 2008). There is also evidence that the aetiological factors related to individual differences in the general population may (partly) overlap with the genetic and environmental factors affecting the extreme or clinical end of the autism spectrum (Robinson et al., 2012), further exemplifying the quantitative nature of autistic traits.

First-degree relatives have also been shown to display subthreshold traits, or the broader autism phenotype (BAP; Piven, Palmer, Jacobi, Childress, & Arndt, 1997b; Sucksmith, Roth, & Hoekstra, 2011), scoring in a similar pattern to individuals with autism on the AQ (Ruta, Mazzone, Mazzone, Wheelwright, & Baron-Cohen, 2012; Wheelwright, Auyeung, Allison, & Baron-Cohen, 2010). Parents have also been shown to have difficulty reading complex emotional states from viewing the eye region of the face (Baron-Cohen & Hammer, 1997; Losh & Piven, 2007; Sucksmith et al., 2013). This has been associated with low quality friendships and difficulty with pragmatic language use in parents (Losh & Piven, 2007). Such difficulties are similar to the social and communication difficulties implicated in autism (Baron-Cohen, Wheelwright, Hill, Raste, & Plumb, 2001a).

While there is less research assessing the non-social traits associated with autism and the BAP, some authors have shown that fathers display traits of rigidity and aloofness that map onto the characteristics of ASC (Losh, Childress, Lam, & Piven, 2008; Smith et al., 2009). There is also some evidence for increased levels of stereotyped behaviour (Bolton et al., 1994; Piven et al., 1997b) and restricted or intense interests (Narayan, Moyes, & Wolff, 1990; Wolff, Narayan, & Moyes, 1988) in parents of a child with autism. It has been argued that the incidence of common genetic variation may contribute to the existence of the BAP and the quantitative nature of autistic traits (Lai et al., 2013). Given these findings, it is important to attempt to understand and refine the autism phenotype incorporating groups containing individuals with a high (individuals with autism), medium (parents) and low (general population controls) genetic predisposition to autism.

It has been suggested that the social and non-social traits of the autism spectrum may be largely independent at genetic, cognitive and neural levels (Happé & Ronald, 2008). For example, it has been shown that within a community based sample, 10% of children display communication difficulties, social impairment or repetitive interests and behaviour independently, suggesting that these characteristics can occur in isolation (Ronald, 2006). Recent evidence also suggests a fractionation of these symptoms and that the social and non-social characteristics of these conditions may have distinct genetic causal explanations or developmental trajectories (Beuker et al., 2013; Georgiades et al., 2007; Happé & Ronald, 2008).

Following the release of DSM-5, there has been an increase in research assessing whether this new set of diagnostic criteria is able to account for the cognitive and behavioural phenotype associated with autism. A review of factor analytic studies assessing the symptoms of ASC highlighted consistent support for a social and communication domain that is distinct from repetitive behaviour and restricted interests (Shuster, Perry, Bebko, & Toplak, 2014). A number of recent studies also outline results consistent with the dyadic structure of the DSM-5. For example, Frazier and others (2014a) found support for the DSM-5 model within a large clinical and general population sample, finding three factors assessing social communication and two assessing repetitive behaviour and restricted interests. However, the substantial correlations between these factors indicated that partially overlapping mechanisms may be responsible for the underlying processes associated with autism (Frazier et al., 2014a). Guthrie and others (2013) also highlight a dyadic DSM-5 structure of social and communication and repetitive behaviour and restricted interests domains in a clinical sample of toddlers with autism. Further support was obtained for the DSM-5 model within two large samples from the United Kingdom and Finland including young people with both clinical and subthreshold symptoms of ASC (Mandy, Charman, Puura, & Skuse, 2014). While this suggests that the social and non-social aspects of ASC are separable, these studies showed moderate to large correlations, indicating that these two domains are not fully independent.

However, others argue that different structures can better account for the autism phenotype. For example, Duku and others (2013) show that a six factor structure provided a better fit than the two domains outlined in DSM-5 in a clinical sample of children and adolescents with autism. Norris and others (2012) also highlight no substantive differences between models containing one, two and three domains of functioning in an equivalent clinical sample. Given the varied results of this research as well as the inclusion of mostly clinical samples, it is important to attempt to understand and refine the autism phenotype within large samples containing a full range of genetic vulnerability; including individuals with and without clinical diagnoses.

In addition, it is important to evaluate theoretical perspectives that attempt to identify the underlying mechanisms and processes that are associated with the cognitive and behavioural profile of ASC. There have been a number of theoretical perspectives proposed that attempt to account for the symptoms associated with the autism phenotype.

Theoretical accounts of autism

Theory of mind

One of the first major theories that attempted to explain the cognitive and behavioural profiles implicated in autism posited that individuals with autism have difficulty with "Theory of Mind" (ToM), or the ability to attribute mental states to themselves and others (Baron-Cohen, Leslie, & Frith, 1985; Premack & Woodruff, 1978). The argument behind this hypothesis is that this ability to 'mentalise' is an innate cognitive mechanism underlying crucial aspects of the development of social skills; generally impaired in autism (Baron-Cohen et al., 1985). Evidence for this theory was originally provided by research using false belief tasks, requiring an inference regarding what another individual may be thinking (Baron-Cohen, 1989; Baron-Cohen et al., 1985). However, while initial results indicated that individuals with autism failed these tasks, a proportion were also able to make these inferences (Happé & Frith, 1996). This was discovered to be due to the relationship between verbal mental age and performance on false belief tasks (Happé, 1995). This finding led to the development of more advanced

ToM tasks involving language and facial emotion processing to assess the association between ToM and the symptomatology of autism.

Such advanced theory of mind tasks have highlighted differences between individuals with autism and typically developing peers. For example, it has been shown that autism is associated with difficulty in labelling simple emotions (Sucksmith et al., 2013), as well as with more complex emotion recognition tasks (Baron-Cohen, Jolliffe, Mortimore, & Robertson, 1997a; Baron-Cohen et al., 2001a). There is also evidence that individuals with ASC have trouble understanding nonliteral language, as well as the thoughts and feelings of characters in a narrative (Happé, 1994; Jolliffe & Baron-Cohen, 1999). Individuals with autism have also been shown to find it challenging to detect faux pas in social situations (Baron-Cohen, O'Riordan, Jones, Stone, & Plaisted, 1999). However, while difficulty understanding and reading emotion in others may contribute to the social and communication impairments associated with these conditions, there is considerable disagreement on whether a lack of ToM is central to the aetiology of ASC (Boucher, 2012; Carruthers, 1996; Tager-Flusberg, 2007). Indeed, this theory cannot account for the repetitive behaviours and restricted interests representative of autism.

Executive function

One theory that attempts to account for the non-social components of ASC argues that the symptom profile associated with autism can be explained by a deficit in executive function (Corbett, Constantine, Hendren, Rocke, & Ozonoff, 2009; Ozonoff, Pennington, & Rogers, 1991; Russell, 1997). Executive function involves a number of behaviours including planning, initiating, flexibility of thinking, inhibition and impulse control (Ozonoff et al., 1991). It has been argued that individuals with ASC have difficulty with cognitive flexibility, planning and inhibition (Hill, 2004). For example, children with autism have been shown to be impaired on tower tasks involving moving objects in a sequence to achieve a set goal (Ozonoff & McEvoy, 1994). Difficulties switching rules in a card sorting task have also been highlighted (Ozonoff & Jensen, 1999; Prior & Hoffmann, 1990). Others have argued that it is due to a lack of impulse control and inhibition that individuals with autism fail false belief tasks, rather than an impairment in ToM (Russell, Mauthner, Sharpe, & Tidswell, 1991).

However, impairment in executive function is also associated with a range of other neurodevelopmental disorders including Tourette's Syndrome and Attention Deficit Hyperactivity Disorder (Hill, 2004). It is therefore difficult to assert that this type of deficit is specifically linked to the autism profile. Furthermore, there is inconsistency between findings from different studies evaluating executive function in autism (Hill, 2004). This highlights that while executive dysfunction may account for some features of the autism phenotype, not all individuals with ASC display this impairment. Furthermore, while research has tended to focus on the impairments or deficits associated with autism, there is recognition that individuals with autism also exhibit specific areas of enhanced functioning and islets of ability (Baron-Cohen, 2002; Baron-Cohen & Bolton, 1993; Frith & Happé, 1994). The executive function hypothesis is a deficit theory that does not account for the abilities that have been associated with the autism spectrum.

Central coherence

The central coherence hypothesis suggests that both the deficits and assets associated with the autism spectrum may have the same origin, that is, in a different, rather than deficient mind (Frith, 1989). Central coherence is defined as the tendency to process information within a context to give it a higher meaning, often at the expense of memory for detail (Frith, 1989). This account argues that individuals with autism display weak central coherence; a processing bias in which individuals do the opposite, focusing on the local rather than global features of an object. Autism is therefore characterised as a cognitive style rather than a cognitive deficit (Happé, 1999).

Evidence for weak central coherence in autism is given through studies outlining exceptionally good performance by individuals with autism on tasks that benefit from the perception of local details rather than automatic global processing of the whole (Happé, Briskman, & Frith, 2001). For example, individuals with ASC show superior performance on the Wechsler block design (Shah & Frith, 1993) as well as display a preference for local over global coherence on sentence completion tasks (Booth & Happé, 2010). Although there is much evidence to support weak central coherence theory, there are also a number of studies that do not correspond with this account (Brian & Bryson, 1996; Pellicano, Maybery, Durkin, & Maley, 2006). For example, while there is some evidence that individuals with autism perform better than controls on the embedded figures test, where a figure is hidden within a global design (Jolliffe & Baron-Cohen, 1997), results from other studies do not support this finding (White & Saldaña, 2011).

While the earlier weak central coherence hypothesis argued that this bias was central to the aetiology of autism, this theory has evolved to suggest that weak central coherence may occur alongside deficits in social cognition (Happé & Frith, 2006; Rajendran & Mitchell, 2007). Moreover, recent research has also evaluated the impact of ToM, executive function and weak central coherence in unison, indicating that all three contribute significantly to behavioural indicators of autism (Best, Moffat, Power, Owens, & Johnstone, 2008). A recent review also suggests significant relationships between these three theoretical perspectives and argues that no single cognitive deficit (or style) theory is able to explain the autism phenotype (Brunsdon & Happé, 2014).

Empathising - systemising

The empathising-systemising (E-S) theory attempts to integrate both the social and communication difficulties as well as the repetitive behaviours and restricted interests to define a model that is able to account for both the impairment and abilities associated with ASC. This hypothesis argues that two cognitive processes, empathising and systemising, are able to explain the phenotype associated with autism. The term 'empathising' extends the idea of ToM to encompass a range of terms including "theory of mind", "mind reading" and "empathy" (Baron-Cohen, 2004; Dennett, 1987). In this conceptualisation, empathy involves two components; the ability to attribute mental states to oneself and others and to display an appropriate response to that emotion (Baron-Cohen, 2004, 2010b). The first component maps onto the previously defined concept of ToM, involving emotion recognition. This has also been termed cognitive empathy (Smith, 2006). The second component of empathising involves providing an affective response to another's emotional state; or emotional empathy (Smith, 2009). It has been argued that in most instances, cognitive and emotional empathy occur in unison and are difficult to separate (Baron-Cohen & Wheelwright, 2004). The E-S theory proposes that impairment in empathy is associated with the difficulties in communication and social interaction implicated in autism.

Systemising is conceptualised as the drive to understand and derive rules about various structures or systems (Baron-Cohen, 2002). A system is defined as anything that takes inputs and delivers outputs, and includes everything from technical systems through to natural, abstract, social, organisable and motoric systems that can be analysed or constructed (Baron-Cohen, 2004). Systemising allows an individual to predict the behaviour of a system and therefore to control it (Baron-Cohen, 2010b). The E-S theory argues that individuals with autism have an interest in systemising. For example, individuals with autism have special interests that are often in systemisable domains involving collecting, factual information, science, machines and technology (Caldwell-Harris & Jordan, 2014). Systemising can also account for the repetitive behaviour and resistance to change associated with the autism spectrum. Baron-Cohen (2009) argues that when an individual engages in systemising, one thing is varied at a time while everything else is held constant. Thus, repetitive behaviours and interests may be indicators of heightened systemising in individuals with autism.

According to the E-S theory, autism is best explained by impairment in empathy alongside intact or superior systemising (Baron-Cohen, 2004, 2010b; Baron-Cohen, Knickmeyer, & Belmonte, 2005; Baron-Cohen, Wheelwright, Lawson, Griffin, & Hill, 2002). In this way, the social and communication impairments seen in autism can be accounted for by empathy, with the islets of ability, repetitive behaviour and restricted interests explained by an interest in systemising (Baron-Cohen, 2004, 2010b). There is a large evidence base suggesting that individuals with ASC show impaired performance on both self-report and performance-based measures of empathy. For example, autism has been associated with lower levels of self-reported empathy (Baron-Cohen & Wheelwright, 2004), alongside difficulty recognising basic emotions (Sucksmith et al., 2013). There is also evidence that individuals with ASC have difficulty with more complex empathy tasks (Baron-Cohen et al., 2001a). However, while there is a large evidence base for impairment in cognitive empathy or emotion recognition in ASC, the relationship between emotional empathy and autism is less understood.

Further research has shown that individuals with autism display superior performance on tasks assessing systemising (Jolliffe & Baron-Cohen, 1997; Lawson, Baron-Cohen, & Wheelwright, 2004) as well as report increased levels of systemising behaviour compared with controls (Baron-Cohen, Richler, Bisarya, Gurunathan, & Wheelwright, 2003; Wheelwright et al., 2006). Individuals with ASC also display a preference for special interests involving mechanical systems, timetables, vehicles, factual information, animals, technology and numbers (Anthony et al., 2013; South, Ozonoff, & McMahon, 2005), all highly systemisable domains. While special interests have been associated with impairment in functioning (Turner-Brown, Lam, Holtzclaw, Dichter, & Bodfish, 2011), this component of the autism spectrum has also been shown to have a positive impact on individuals with ASC (Winter-Messiers, 2007). This highlights the potential of the E-S model to account for both the impairments and positive factors associated with the autism phenotype.

While there are a number of studies that highlight the plausibility of this theoretical explanation of ASC, there is a lack of research utilising statistical modelling in order to determine how the E-S account maps onto the autism phenotype. Furthermore, given the evidence for the quantitative nature of autistic traits and the existence of a BAP, specific analyses are needed in order to attempt to understand the latent structure of the autism phenotype across samples with a high (individuals with autism), medium (parents) and low (general population controls) genetic predisposition to autism.

Latent structure of the autism phenotype

Latent structure is defined as the fundamental nature of a construct, or the underlying structure that exists regardless of the way in which it is measured (Ruscio & Ruscio, 2002). A discrepancy between the latent structure of a construct and the observable way in which it is assessed has a substantial impact on the accuracy with which individuals can be defined (Ruscio & Ruscio, 2002). Given that psychological theories of autism attempt to identify the underlying processes and mechanisms associated with ASC, it is important to evaluate these theoretical perspectives using latent structural techniques.

Latent structural techniques have been utilised to determine whether psychopathology can be conceptualised as a continuum of severity or as discrete categories of disorder. The use of these statistical techniques provides a comprehensive and novel way of determining whether ASC denote distinct categories of disorder, or are represented at the extreme end of one or more dimensions. While there is growing support that autism represents the severe end of a continuum or spectrum (Constantino, 2011), it is important to statistically evaluate this notion. This can be achieved through an investigation of the latent structure of the autism phenotype. Given that a phenotype results from the interaction between genes and environment, there may be differences in latent structure for groups that contain varying levels of genetic risk for autism. Moreover, it is not known whether different autistic traits cluster together in a similar way across, controls, parents and individuals with ASC. This highlights the importance of evaluating latent structure across individuals with varying genetic vulnerability to autism.

To date there are no studies that assess the ability of the E-S theory to explain the underlying or latent structure of the autism phenotype or incorporate an evaluation of the latent structure of the phenotype amongst controls, parents and individuals with ASC. However, there has been some research that has attempted to understand the latent structure of symptoms of autism utilising numerous statistical methods and measures with varying results. These have consisted of research seeking to identify underlying dimensions, categories or a combination of both categories and continua.

Identifying latent dimensions

Factor analysis is a statistical method that is used to determine the underlying number of dimensions associated with a construct, as well as provide evidence for the strength of the relationship between different observed variables or symptoms (Muthén &

Muthén, 2012b). These underlying dimensions are referred to as continuous latent variables or factors. Factor analytical methods can be exploratory or confirmatory. Exploratory methods attempt to determine the factor structure when nothing is known about the relationship between the observed variables and the construct in question, whereas confirmatory methods test the feasibility of a theoretically determined structure (Muthén & Muthén, 2012b).

A number of authors have used factor analysis to determine the underlying dimensional structure of autism symptom profiles. These studies vary widely in terms of the measures used and factors identified, including models containing five (Frazier et al., 2014a) and six (Duku et al., 2013) interrelated dimensions. Others have emphasised an alternate three factor model comprising social communication, inflexible behaviour and language and repetitive sensory and motor behaviour (Georgiades et al., 2007), or that autism is represented by a single dimension (Constantino et al., 2004). While some research has found support for three underlying dimensions, in support of the DSM-IV triad of impairment (Lecavalier, Gadow, DeVincent, & Edwards, 2009; Sipes & Matson, 2014), there is reasonably consistent support that the autism phenotype consists of two dimensions that map onto the social and communication and repetitive behaviour and restricted interest domains outlined in DSM-5 (Shuster et al., 2014).

Identifying latent categories

A number of studies have also attempted to define distinct subgroups using categorical data analysis methods including taxometric analysis and latent class models. Taxometric analysis is designed to assess whether a construct is represented by a single dimension or a distinct categorical group or taxon. While some studies use taxometric analysis to provide evidence that autism can be conceptualised as a discrete category of disorder (Frazier et al., 2010), there are also studies that identify some constructs associated with autism that are categorical and other constructs that are not (Ingram, Takahashi, & Miles, 2008). However, when symptoms of ASC only were evaluated, results indicated a distinct ASC category (Ingram et al., 2008). There are also a number of papers that use latent class analysis to identify subgroups of individuals with ASC. Latent class analysis (LCA) is a statistical method used to identify classes or subgroups of individuals containing comparable levels of observable variables. Using LCA, two studies have identified four (Beuker et al., 2013) and six (Greaves-Lord et al., 2013) subgroups of children discriminated by their scores on the social and non-social traits of autism. Others have used latent class and taxometric methods to identify subgroups of individuals based on IQ and patterns of cognitive ability (Eagle, Romanczyk, & Lenzenweger, 2010; Munson et al., 2008). Categorical methods have also been used to assess the BAP, with a recent study identifying two distinct groups based on symptom severity, with significant familial clustering within these subgroups (Veatch, Veenstra-Vanderweele, Potter, Pericak-Vance, & Haines, 2014). Georgiades and others (2013b) have also used cluster analysis to provide evidence for the BAP in siblings.

Combining latent categories and dimensions

A further set of analytic techniques have been developed to assess the presence of a concurrently dimensional and categorical latent structure. Factor mixture models (FMM) incorporate both factor analytic and latent class methods (Lubke & Muthén, 2005; Muthén, 2008). The factors model quantitative dimensions, while the latent class variable allows for the classification of subgroups of individuals. There are three papers utilising mixture modelling to assess the latent structure of autism to date. Frazier and others (2012) examined the validity of the DSM-5 criteria amongst children aged two to eighteen years. The authors concluded that a model with two symptom dimensions (communication and social interaction /repetitive behaviours and restricted interests) and a categorical subgroup (ASC vs. non-ASC) provided the best fit to the data, providing support for the new diagnostic model (Frazier et al., 2012). Similarly, Georgiades and colleagues (2013a) obtained a two-factor three-class solution, indicating two dimensions based on DSM-5 domains as well as three classes of individuals displaying differing levels of both social and communication difficulties and repetitive behaviours. The authors also noted a subgroup of children who displayed a reduction in symptoms over time, with a follow up longitudinal analysis of the same sample indicating a two-factor two-class solution (Georgiades et al., 2014).

In summary, the majority of factor analytic studies of the autism phenotype suggest a multifactorial structure, consisting of at least one social and one non-social factor. The studies assessing the latent structure of autism to date have utilised a number of different methods, measures and diagnostic constructs. However, most of these studies have been conducted within clinical samples. Evaluating a multifactorial trait within clinical samples introduces a potential bias, given that in order to receive a diagnosis of autism, individuals need to display both the social and non-social traits associated with ASC (Happé & Ronald, 2008). This significantly limits the ability to identify whether the traits associated with ASC are represented by a single dimension of severity, or whether there are distinct social and non-social dimensions. However, the inclusion of population samples only is also problematic given the low base rate of autism within these groups. This thesis therefore employs an alternative research design, and examines the latent structure of the autism phenotype using a variety of latent structural techniques in samples covering the entire spectrum of vulnerability for autism, including not only a clinical group (high genetic predisposition), but also parents of a child with autism (medium genetic risk) and general population controls (low genetic vulnerability).

Aims of thesis

This thesis aims to evaluate the E-S theory of autism in order to understand the latent structure of the autism phenotype. Furthermore, it seeks to determine whether the latent structure of the autism phenotype is consistent across individuals with autism, parents of a child with autism and general population controls. It consists of four chapters containing papers in the format of manuscripts for publication. The manuscript contained in chapter two was published in the *Journal of Abnormal Psychology* in May 2013. The manuscript contained in the fourth chapter was published in *Molecular Autism* in August 2014. The manuscript in the third chapter is currently under review, while chapter five is being finalised for submission. Chapters two, three and four systematically evaluate the E-S theory of autism and the autism phenotype across groups stratified by genetic vulnerability. Chapter five provides an in depth assessment of motivations behind engaging in special interests in ASC and highlights the importance of understanding special interests and the role these interests play in the lives of individuals with autism. Collectively, these four chapters utilise latent structural statistical techniques in order to understand the latent structure of the autism phenotype.

Chapter two uses confirmatory factor analysis to determine whether empathy and systemising are best represented by a single continuum or as multiple dimensions. This chapter attempts to determine the latent structure of the autism phenotype based on the E-S theory. It also evaluates whether this structure differs across individuals with a high (individuals with autism), medium (parents of a child with autism) and low (general population controls) genetic vulnerability to autism.

Chapter three uses factor mixture modelling to determine whether the relationship between empathy and systemising is best conceptualised as dimensional, categorical or a combination of both categories and continua. This chapter builds on previous research highlighting that subgroups based on either severity of symptoms or clinical diagnosis can be identified across the DSM-5 dimensions of autism. This chapter will determine whether meaningful subgroups can be identified within a sample containing varying levels of genetic risk for autism based on their level of empathy, systemising and autistic traits.

Chapter four aims to assess the latent structure of empathy in more detail, evaluating the structure of cognitive and emotional empathy amongst individuals with autism, parents and controls. This chapter incorporates self-report questionnaire and performance-based task data in order to assess the relationship between these two methods of measurement and cognitive and emotional empathy.

Chapter five focuses on an aspect of non-social autistic traits that has so far received relatively little attention, namely special interests. The systemising account of autism provides an explanation for the development of particular special interests areas. However, at present little is known about the motivations to engage in special interests in autism. Moreover, there is conflicting research as to whether special interests are associated with negative or positive outcomes. This chapter outlines the development of a measure to assess motivation to engage in special interests in order to determine the relationship between motivation, special interests and autistic traits amongst individuals with autism, parents and controls.

Chapter 2

Empathising, systemising and autistic traits: Latent structure in individuals with autism, their parents and general population controls

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Abstract

Background: The search for genes involved in autism spectrum conditions (ASC) may have been hindered by the assumption that the different symptoms that define the condition can be attributed to the same causal mechanism. Instead the social and non-social aspects of ASC may have distinct causes at genetic, cognitive and neural levels. It has been posited that the core features of ASC can be explained by an impairment in empathy alongside intact or superior systemising; the drive to understand and derive rules about a system. First-degree relatives also show some mild manifestations that parallel the defining features of ASC, termed the broader autism phenotype.

Methods: Factor analyses were conducted to assess whether the latent structure of empathy, systemising and autistic traits differs across samples with a high (individuals on the spectrum), medium (first-degree relatives) or low (general population controls) genetic vulnerability to autism.

Results: Results highlighted a two-factor model, confirming an empathy and a systemising factor. The relationship between these two factors was significantly stronger in first-degree relatives and the autism group compared with controls.

Conclusions: The same model provided the best fit amongst the three groups, suggesting a similar latent structure irrespective of genetic vulnerability. However, results also suggest that whilst these traits are relatively independent in the general population, they are substantially correlated in individuals with ASC and their parents. This implies that there is substantially more overlap between systemising and empathy amongst individuals with an increased genetic liability to autism. This has potential implications for the genetic, environmental and cognitive explanations of autism spectrum conditions.

Introduction

Autism spectrum conditions (ASC) are characterised by impairment in the development of communication skills and reciprocal social interaction alongside the presence of unusually repetitive behaviours and restricted interests (DSM-IV-R; American Psychiatric Association, 2000). It is well established through family and twin studies that these conditions have a strong genetic component. A range of twin studies have indicated substantially higher concordance rates for clinical autism in monozygotic twins when compared with dizygotic twins. Altogether, these findings indicate strong genetic influences on ASC with a heritability estimate of around 80%, (Ronald & Hoekstra, 2011). A recent large scale family study suggests the recurrence rate for autism within families is close to 20% (Ozonoff et al., 2011). This, coupled with prevalence estimates of around 1% (Baron-Cohen et al., 2009; Brugha et al., 2011) suggests a markedly increased risk for autism within families, highlighting a strong influence of genetic effects. There is also a growing body of evidence from molecular genetic studies suggesting the involvement of multiple genetic variants and loci in the development of these conditions (Abrahams & Geschwind, 2008; Geschwind, 2011).

There is evidence to suggest that family members show mild manifestations that parallel the defining features of autism, a phenomenon termed the broader autism phenotype (BAP; see Sucksmith et al., 2011 for a review). With the development of quantitative psychometric instruments such as the Autism Spectrum Quotient (AQ; Baron-Cohen et al., 2001b), that assess autistic traits on a continuous scale, it is now possible to measure these subthreshold autistic traits with more precision. Use of such scales in family studies may provide insights into the genetic factors involved in ASC and the familial risk for developing autism.

A multitude of studies have reported mild impairments in relatives of individuals with autism, particularly in the social and communicative domains. For example, parents of individuals with ASC have been shown to display more social and communication difficulties, as measured by subscales of the AQ, as well as score lower on measures of pragmatics (Piven et al., 1997a; Ruser et al., 2007). Siblings also show difficulties in reciprocal social interaction (Nadig et al., 2007; Toth, Dawson, Meltzoff, Greenson, & Fein, 2007). Similarly, parents show some mild difficulties with social cognition, as measured by neuropsychological tests (Losh et al., 2009; Losh & Piven, 2007). Parents of children with an ASC also perform lower than a control group on a task assessing the ability to read complex emotional states from viewing the eye region of the face (Baron-Cohen & Hammer, 1997; Losh & Piven, 2007), providing evidence for the BAP at a cognitive level.

Whilst the evidence for some of the non-social aspects of ASC amongst relatives is more modest, a number of studies suggest an elevated rate of stereotyped behaviours and circumscribed hobbies in parents (Bolton et al., 1994; Briskman, Happé, & Frith, 2001; Piven et al., 1997b). Some studies suggest that first-degree relatives of individuals with ASC may also display the same 'cognitive style' that leads to superior performance on tasks where visual processing of local material is advantageous, including the embedded figures task (Baron-Cohen & Hammer, 1997; Bolte & Poustka, 2006; Happé et al., 2001) and the block design task (Scheeren & Stauder, 2008). However, findings related to tasks assessing local processing styles have been somewhat inconsistent, both in clinical groups (White & Saldaña, 2011) and in first-degree relatives (Sucksmith et al., 2011).

There has been much debate around whether the triad of features characteristic of autism (social impairments, communication impairments and repetitive behaviour/restricted interests) are influenced by the same genetic and environmental factors, or whether they are somewhat independent. Happé and Ronald (2008) suggest that the core features that define autism are largely 'fractionable'; that is, that they may have distinct causes at genetic, cognitive and neural levels. There are a number of family and twin studies that support this notion, showing that although the three sets of features are highly heritable individually, they are affected by largely independent genetic influences (Ronald et al., 2006a; Ronald, Happé, & Plomin, 2005; Ronald, Happé, Price, Baron-Cohen, & Plomin, 2006b). Moreover, 10% of children in a large general population study displayed only social impairment, only communication difficulties or only repetitive behaviours and restricted interests (Ronald, 2006), suggesting these characteristics can also occur in isolation.

Similarly, a review of factor analytic studies showed that, of the seven studies included, six found evidence for multiple factors underlying autistic features (Mandy & Skuse, 2008). Although the total number of factors identified varied across studies, all studies reported at least one social-communication factor and at least one distinct non-social factor (Mandy & Skuse, 2008). Taken together, these studies suggest that partially distinct causal explanations should be sought for the social and non-social aspects of ASC.

This hypothesis has so far mainly focused on features at a behavioural level. However, there are a number of theoretical explanations that attempt to account for the features in autism at the cognitive level. It has been suggested that these conditions are associated with difficulties in executive function (Corbett et al., 2009; Ozonoff et al., 1991; Russell, 1997), 'weak central coherence' (a processing bias in which individuals focus on the local rather than global features of an object) and 'Theory of Mind' (the ability to attribute mental states to oneself and others) (Baron-Cohen et al., 1985).

The term 'empathising' extends the idea of theory of mind and involves two components: the ability to attribute mental states to oneself and others, and the drive to respond with an appropriate emotion to that mental state (Baron-Cohen, 2004, 2010b). A different process, systemising, is conceptualised as the drive to understand and derive rules about a system (Baron-Cohen, 2002). Systemising allows an individual to predict the behaviour of a system and therefore to control it (Baron-Cohen, 2010b). A system is defined as anything that takes inputs and delivers outputs, and includes everything from technical systems (e.g., a machine) through to natural (e.g., the weather), abstract (e.g., mathematics), social (e.g., a company), collectible (e.g., a library), and motoric (e.g., a tennis top-spin) systems that the brain can analyse or construct (Baron-Cohen, 2004).

According to the Empathising-Systemising (E-S) theory, autism is best explained by an impairment in empathy alongside intact or superior systemising (Baron-Cohen, 2004, 2010b; Baron-Cohen et al., 2005; Baron-Cohen et al., 2002). In this way, the social and communication impairments seen in these conditions can be accounted for by empathy, and the islets of ability, repetitive behaviour and restricted interests or obsessions with systems can be accounted for by an interest in systemising (Baron-Cohen, 2004, 2010b). There is a large evidence base suggesting that individuals with ASC show impaired performance on measures of empathy and intact or elevated performance on tests of systemising ability (Baron-Cohen et al., 2003; Baron-Cohen & Wheelwright, 2004; Baron-Cohen, Wheelwright, & Jolliffe, 1997b; Baron-Cohen, Wheelwright, Stone, & Rutherford, 1999; Jolliffe & Baron-Cohen, 1997; Lai et al., 2011; Lawson et al., 2004).

As yet it remains unclear the extent to which empathising and systemising traits are related. Given that individuals with ASC tend to perform poorly on tasks of empathy and do well on tasks of systemising, an inverse correlation between the two traits would be expected in this group, provided that these traits are assessed on continuous scales that allow for sufficient variance within the clinical group. However, it is less clear whether this inverse association is linear across populations, or whether it would also apply to non-clinical samples. The current study aims to assess the association between empathy, systemising and social and non-social autistic traits across three distinct samples, stratified by their genetic risk for autism. This study reports on factor analyses employed in three distinct samples comprising individuals with a clinical ASC diagnosis (high genetic vulnerability), parents of a child with ASC (medium genetic risk), and a general population control group (low genetic vulnerability).

Methods

Participants

Individuals with a clinical ASC diagnosis and parents of a child with ASC were recruited via the participant database at the Autism Research Centre at the University of Cambridge (www.autismresearchcentre.com). In order to account for any potential response bias, the control sample was collected via a different portal at a general (non-clinical) volunteer psychology research webpage (www.cambridgepsychology.com). Participants were included in the study if they were 18 years and over and had completed all the measures. The individuals in the ASC group, and the children of the parent group were reported to all have received a formal ASC diagnosis from experienced clinicians in recognised clinics.

Individuals in the parent group did not report having an ASC diagnosis themselves. The control group was confined to individuals who did not report any past psychiatric history. The total sample consisted of 1034 individuals, comprising 363 individuals with ASC (males = 193, females = 170, mean age = 36 yrs, sd = 11), 439 parents of a child with ASC (males = 141, females = 298, mean age = 42 yrs, sd = 8) and 232 controls (males =

122, females = 110, mean age = 33 yrs, sd = 10). 80% of the control group had completed higher education, whilst 50% of the parent group and 54% of individuals with ASC had completed an undergraduate degree.

Measures

Individuals registered in either of the above websites were asked to fill out a range of well-validated questionnaires assessing empathy, systemising and quantitative autistic traits. Participants were able to complete the questionnaires in their preferred order. The measures used are designed as dimensional quantitative measures of empathy, systemising and autistic traits, in keeping with the paradigm that autism is best represented along a spectrum of symptoms. As these measures are not designed as clinical instruments, this allows for variance across the three sample groups.

Empathising and systemising

The Empathy Quotient (EQ; Baron-Cohen & Wheelwright, 2004) is a self-report measure of empathy. Items assess the ability to attribute mental states to oneself and others, and the drive to respond with an appropriate emotion to that mental state. An example of an item assessing recognising the mental state of another is ‘I am quick to spot when someone in a group is feeling awkward or uncomfortable’. An example of an item assessing the drive to respond emotionally to another’s mental state is ‘I tend to get emotionally involved with a friend's problems’. The EQ is comprised of 40 statements scored on a Likert scale including four response options; ‘strongly disagree’, ‘slightly disagree’ ‘slightly agree’ and strongly agree’. For approximately half the items, an ‘agree’ response is in line with high empathy abilities. On these items ‘strongly agree’ responses score two points, ‘slightly agree’ responses score one point and ‘strongly disagree’ and ‘slightly disagree’ both score zero. The other half of the items are reverse-scored as a ‘disagree’ response refers to better empathising in these items. Scores range from zero to 80 with a higher score reflecting increased empathising ability and follow a near normal distribution. Adults with high functioning autism or Asperger’s syndrome have been shown to score significantly lower on the EQ than age-matched controls (Baron-Cohen & Wheelwright, 2004).

The Systemising Quotient-Revised (SQ; Wheelwright et al., 2006) is a self-report measure of systemising consisting of 75 statements with four response options; ‘strongly agree’, ‘slightly agree’, ‘slightly disagree’ and ‘strongly disagree’. Scoring procedures are equivalent to those described for the EQ. Scores follow a continuous distribution ranging from zero to 150, with higher scores reflecting stronger systemising behaviour. Items include statements like ‘When I learn about a new category I like to go into detail to understand the small differences between different members of that category’ and ‘In maths, I am intrigued by the rules and patterns governing numbers’. Individuals with autism score higher on the SQ compared with age-matched controls (Baron-Cohen et al., 2003; Goldenfeld, Baron-Cohen, & Wheelwright, 2005; Goldenfeld, Baron-Cohen, Wheelwright, Ashwin, & Chakrabarti, 2007; Lai et al., 2011; Wheelwright et al., 2006).

Autistic traits

The Autism Spectrum Quotient (AQ; Baron-Cohen et al., 2001b) is a self-report quantitative measure of autistic traits. The AQ consists of 50 items assessing the core areas of difficulty in ASC including impaired social skills, communication difficulties, imagination and attention switching and a superior attention to detail. Participants were asked to rate themselves on a 4-point Likert scale with response categories ‘definitely disagree’, ‘slightly disagree’ ‘slightly agree’ and ‘definitely agree’. This study used the raw scoring method (as detailed in Hoekstra et al., 2008), with total scores following a normal distribution ranging from 50 to 200 and a score of 200 representing full endorsement of all autistic traits. Individuals with an ASC show significantly higher scores on the AQ compared with the general population (Baron-Cohen et al., 2001b).

Recent evidence suggests that the AQ can be split into two categories of items reflecting a broad social interaction factor comprising the social skills, attention switching, communication and imagination items and an attention to detail factor (Hoekstra et al., 2008). These two factors only correlate modestly and are therefore useful in making the distinction between social and non-social autistic traits (Hoekstra et al., 2008). The AQ was split into these two subscales for the current analysis.

Analytic strategy

There is a large evidence base suggesting that scores on the measures included in this study are affected by sex (Baron-Cohen et al., 2005; Baron-Cohen et al., 2011). The main focus of this paper was on the factor structure, not on sex differences in mean scores, which have been studied for our measures of interest in previous studies (Baron-Cohen et al., 2001b; Hoekstra et al., 2008; Sucksmith et al., 2013; Wheelwright et al., 2006). In order to account for the effects of sex as well as the potential confounding effect of age on the means, variables were standardised via regression analyses in SPSS for age and sex before analysis.

Following standardisation, a series of confirmatory factor (CFA) models were specified and estimated in MPlus version 6 (Muthén & Muthén, 2010) using the maximum likelihood estimator. A one-factor model encompassing all measures of empathy, systemising and autistic traits was fit for each of the three groups separately (Models 1 to 3). Following this, a two-factor model was fit across the three groups (Models 4 to 6). The EQ and the social interaction subscale of the AQ (AQ_soc) were predicted to load on one latent ‘empathy’ factor, whilst the scores on the SQ and the attention to detail factor (AQ_att) were expected to load on a ‘systemising’ factor. Scores on the social interaction subscale of the AQ (AQ_soc) were reverse scored to enable ease of interpretation. Empathising ability is therefore indicated by high scores on the EQ and high scores on the social interaction subscale of the AQ, whilst systemising ability is indicated by high scores on the SQ and on the attention to detail factor of the AQ.

In order to assess the full range of models available, and to test whether the hypothesised Empathising-Systemising model (tested in Models 4 to 6) really provided the best fit to the data, two further models were specified, the first including the SQ and the social subscale of the AQ (AQ_soc) on one factor, with the EQ and the attention to detail factor (AQ_att) loading on a second factor (Models 7 to 9). The second model included the EQ and SQ on the first factor and both sections of the AQ loading on a second factor (Models 10 to 12). Both models were fit across the three groups.

Models 1 to 12 were fit within the three individual groups to allow for a different factor structure relative to genetic liability. However, it is important to evaluate the equivalence of the parameters estimated in a CFA across groups (Brown, 2006). This can be achieved within one model using multigroup CFA. Multiple group models make it possible to pinpoint where any specific differences across groups may fall (Brown, 2006). Therefore, in order to assess whether theses traits function differently amongst the three groups, a further model was implemented, allowing all parameter estimates to vary (Model 13). A further model in which the factor loadings were constrained to be equal across groups was also implemented (Model 14).

To test whether the same factor structure was identified for males and females within each sample group a further three models were tested with varying restrictions across six groups split by sex and genetic vulnerability. Model 15 contains a multigroup CFA allowing all estimates to vary across the six groups. A second model was fit constraining the factor loadings to be equal (Model 16). A final model restricting the factor correlations to be equal for males and females as well as equal factor loadings across the six groups (Model 17) was also included in the analyses.

Model fit was evaluated using the following goodness of fit statistics; Akaike information criterion (AIC; Akaike, 1987), Bayesian information criterion (BIC; Schwarz, 1978), sample size adjusted BIC (SSABIC; Sclove, 1987), Root mean square error of approximation (RMSEA; Steiger & Lind, 1980), Comparative fit index (CFI; Bentler, 1987) and the Tucker-Lewis index (TLI; Tucker & Lewis, 1973). The AIC, BIC and SSABIC are parsimony-adjusted indices used to examine model fit, with lower values indicating a better fit to the data. It has been suggested that a RMSEA value <0.05 indicates a close model fit, with values up to 0.08 suggesting a reasonable error of approximation (Browne & Cudeck, 1993). Current recommendations state that a CFI and TLI value >=0.90 indicate acceptable fit with values >=0.95 indicative of very good fit to the data (Brown, 2006; Hu & Bentler, 1999). As well as taking into account the fit indices mentioned above, evaluation of model fit also took into account the strength and interpretability of the structural parameter estimates.

Results

Distribution of scores on the subscales of the AQ, the EQ and SQ, standardised for age and sex are given in Figure 1, showing adequate coverage of the possible range of responses. Model fit indices ascertained from the CFA models are given in Table 1. The one-factor models displayed poor fit amongst the three groups. In contrast, the two-factor model accounting for measures of systemising and empathy provided an excellent fit to the data within all three groups (see Models 4 to 6). RMSEA values of 0 occur due to a chi square value less than the number of degrees of freedom (Kenny, Kaniskan, &

McCoach, 2011). Similarly, CFI and TLI values are also affected by the chi square statistic as well as the degrees of freedom in the model (Brown, 2006). However, these values are indicative of almost perfect model fit (Savalei, 2010). The further two-factor models (Models 7 to 12) displayed poor fit amongst the three groups. Models 7 and 10 displayed a correlation greater than one between the two factors, indicating that there was no distinction between them. Similarly, fit statistics for models 8 and 9 fell under the required thresholds, suggesting that the empathising-systemising two-factor model (tested in Models 4 to 6) described the data best in all three groups.

Multiple group analyses were conducted in order to assess for specific group differences within the two-factor model where the EQ and the social interaction subscale of the AQ (AQ_soc) load on the latent 'empathy' factor, whilst the scores on the SQ and the attention to detail factor (AQ_att) load on a 'systemising' factor. Model 13 showed a good fit to the data, with a CFI and TLI above 0.97. Although the RMSEA is larger than the cut-off recommended for model fit, this value is affected by the number of free parameters in the model (Browne & Cudeck, 1993). As Model 13 includes more parameters, the RMSEA of this model is relatively high compared with models 4 to 6. Furthermore, with limited degrees of freedom the RMSEA value is of less concern given all other indices are strong and suggest a good fit (Brown, 2006). This is the case in Model 13 with CFI and TLI values falling above the specified threshold.

A model in which the factor loadings were constrained to be equal across the three groups (Model 14) resulted in a significantly poorer fit compared to the fit of Model 13 ($\Delta\chi^2 = 59.37$, $p < 0.001$). Therefore, Model 13, the two-factor model with equal form amongst the three groups, allowing the factor loadings to vary, provided the best fit to the data. Evaluation of Models 15 to 17 showed that the model constraining the factor correlations to be equal across males and females in each group (Model 17) provided the best fit to the data, indicating that the factor structure obtained in Model 13 does not differ when sex is taken into account.

Factor loadings, correlations and confidence intervals for the two-factor model taken from the Model 13 analysis are given in Figure 2. All factor loadings were salient and statistically significant ($p < 0.05$), reflecting that these measures are good indicators of their respective factors. Parents scored lower on the latent factor mean of empathy than the control group (Mean Difference = -0.31, $p < 0.01$). However, there was no significant difference between scores on the systemising factor between the parent and control groups. The ASC group showed lower scores on the latent factor empathy compared with controls (Mean difference = -2.68, $p < 0.01$) and the parent group (Mean difference = -2.37, $p < 0.01$) as well as superior latent mean scores on systemising compared with controls (Mean difference = 1.01, $p < 0.01$) and parents (Mean difference = 1.14, $p < 0.01$).

The correlation between empathy and systemising was significant across the three samples. The negative correlation between the two factors was significantly stronger in both the ASC group ($r = -0.61$) and the parent group ($r = -0.57$) compared with controls ($r = -0.22$). However, the correlations between the two factors for the ASC group and parent group did not differ (i.e., the confidence intervals for the correlations between empathy and systemising in the parent and ASC groups overlapped).

Discussion

The current study examined the structure of autistic characteristics across individuals with a low, medium and high genetic vulnerability for autism. Results indicated that a two-factor model provided the best fit across the three groups irrespective of sex. This model comprised an empathy factor including both the EQ and the social behavioural and cognitive traits measured by the AQ, and a systemising factor including the SQ and the 'attention to detail' traits measured by the AQ. The latent empathy factor and systemising factor were inversely correlated in all three groups. The factor correlations ranged from small to large, providing support for the notion that the social and non-social aspects of ASC may have distinct causes at a behavioural level (Happé & Ronald, 2008; Ronald, 2006; Ronald et al., 2006a; Ronald et al., 2006b).

Perhaps the most notable finding from the current study is the difference in the strength of the inverse relationship between empathising and systemising amongst controls, first-degree relatives and individuals on the spectrum. The association between empathy and systemising was substantially stronger in individuals with ASC and parents of a child with autism than in general population controls. Whilst a definitive explanation for these associations cannot be given without further research, there are a number of potential explanations why these constructs may be more strongly associated in individuals with autism and their first-degree relatives.

First, individuals on the spectrum are given a diagnosis of an ASC. This by definition includes symptoms from all three domains of social impairment, communication difficulties and repetitive behaviour/restricted interests (American Psychiatric Association, 2000). Since systemising and empathy are cognitive explanations of autism, individuals with ASC are likely to be both superior in systemising and weaker in empathy. It is therefore not surprising that these two factors are highly inversely related in this group, given that the presence of all three core symptoms of autism make high systemising and low empathising more likely. However, this account does not apply to the parent group as these parents do not have a diagnosis of ASC themselves and are therefore not directly selected to score high on systemising and lower on empathy.

An alternative explanation for our findings could be that empathy and systemising are highly correlated in individuals with ASC due to cognitive strategies used by this group. Due to their poor intuitive empathic abilities, individuals with autism may employ systemising strategies in empathy tasks. For example, when individuals with ASC engage in an activity requiring empathy, they may use systemising strategies to work out what particular emotion or mental state is relevant to the situation and how to respond appropriately. The use of such strategies may result in an association between empathy and systemising, and as such when attempting to measure empathy in this group we may actually be indirectly measuring the systemisation of empathy. If this strategy does not improve empathy ability, the correlation would remain strong and in the negative direction. However, if systemising is a helpful strategy and improves empathy ability, then it is likely to lower the negative correlation between empathising and systemising. Whilst the current study cannot identify whether such strategies are being used, our results call for further research into the types of strategies used by individuals with ASC in their approach to tasks of empathy.

Findings suggest that there is a relatively stronger overlap between empathy and systemising in individuals with a high and medium genetic risk for autism compared with individuals with a low genetic vulnerability. This overlap could be due to genetic and/or environmental influences. Although the design of the current study did not allow us to examine the nature of the association, we may consider possible genetic and environmental mechanisms that might underlie the different associations between empathy and systemising in groups of varying genetic vulnerability to autism.

One possible explanation for the high inverse association between empathy and systemising in the ASC and parent group compared with the modest association found in individuals with no relatives with autism could be genetic heterogeneity. The genetic risk for autism is thought to stem from a variety of different sources, including common genetic variants with relatively weak effects (Anney et al., 2010; Arking et al., 2008; Chakrabarti et al., 2009) and rare gene mutations and copy number variations (CNVs) with proportionally larger effects (Levy et al., 2011; Sebat et al., 2007). Although this is not within the scope of the current study, further investigation into whether common genetic variants may help to explain the variation in empathy and systemising traits in the general population is warranted. In contrast, rare CNVs and gene mutations with a relatively large effect may be more common in families affected by autism. Previous molecular genetic studies of ASC show that rare CNVs thought to have a role in autism aetiology can occur de novo (i.e., a new mutation that was not inherited from either parent) (Sanders et al., 2011), but can also be transmitted from parent to child (Levy et al., 2011). Further research would benefit from assessing whether gene variants with relatively large effects impact upon both systemising and empathy. Such heterogeneous genetic effects, although at present speculative, could possibly explain the strong relationship between empathising and systemising in individuals with ASC and parents, compared with the small association observed in control samples.

Alternatively, there may be heterogeneous environmental influences on empathy and systemising across the three different groups. As yet little is known about possible influences of environmental effects of autism, with peri and pre-natal complications one of the most consistently reported possible environmental risk factors (Kolevzon, Gross, & Reichenberg, 2007). Future research would benefit from direct assessment of the impacts of environmental factors on both systemising and empathy in samples with varying degrees of genetic vulnerability for autism.

Limitations

The current study had a number of limitations. The study was restricted in that the parent group contained a larger proportion of mothers (n = 298) than fathers (n = 141), whilst the sex ratio was approximately equal in the other two groups. The sex effects on the mean scores of the variables were accounted for by standardising for the effect of gender prior to conducting the factor analyses. Sex differences in latent structure were also explored, indicating that the factor structure obtained does not differ by sex. However, larger numbers in each group would serve to increase power for such types of comparisons. Secondly, the ASC sample group consisted of high functioning adults with an autism spectrum disorder. As is often the case in cognitive studies of autism (Hoekstra & Whatson, 2010), our study design using questionnaire self-ratings precluded the participation of individuals at the lower functioning end of the spectrum. It is less straightforward to test empathy and systemising in individuals on the spectrum who also have intellectual disability. Nevertheless, some characteristics of low functioning autism, such as the relative talent (compared to other abilities) in solving puzzles, a great interest in lawful systems and increased attention to small changes in the environment all hint towards a drive to systemise (Baron-Cohen et al., 2005), whilst delays and deficits in theory of mind development, even when compared with control children of similar mental age, suggest empathy impairments also apply to the lower functioning end of the autism spectrum (Abell, Happé, & Frith, 2000; Baron-Cohen, 1995). However, whether the factor structure between empathising and systemising found in high functioning individuals with autism in our study also generalises to individuals on the spectrum with intellectual disability remains unknown.

The measures used in this study were all questionnaire based and all concerned self-report. Future research should also incorporate cognitive performance measures and second person ratings of empathy and systemising. Of further interest would be to examine the extent to which other behavioural or psychiatric problems commonly found to be comorbid with autism (e.g., attention problems) may moderate the association between empathy and systemising.

As the study was conducted using an online volunteer register it was not possible to verify whether subjects met ASC diagnostic criteria. However, it has been reported that diagnoses in online volunteers are generally reliable (Lee et al., 2010). Furthermore, online data collection enabled the collection of data from a large number of respondents from a representative sample. The use of online research in this sample may also reduce selection bias due to the user friendly and non-invasive nature of the research and the difficulty that individuals on the spectrum or those parents taking care of a special needs child may have in attending a face-to-face laboratory setting. A possible drawback is that the sample may have been overrepresented by participants who feel comfortable using computers and are familiar with and interested in taking part in online research.

Conclusions

The current study assessed the latent structure of empathy, systemising and autistic traits across individuals with a low, medium and high genetic vulnerability to autism. Our results indicated that a two-factor model comprising a latent empathy and systemising factor provided the best fit across the three groups. The inverse relationship between both traits was substantial in individuals with high and medium genetic vulnerability, but only modest in individuals with low genetic risk for autism. We speculate that the varying strength in the association between empathising and systemising across groups may be explained by differences in cognitive style and by genetic and possibly environmental heterogeneity. However, further research is needed in order to establish the impact and causality of these associations.

Table 1 Fit indices and model comparisons for the alternative factor models of the autism phenotype

Model	Description	Fit indices							
		AIC	BIC	SSABIC	RMSEA	CFI	TLI	χ^2	r
One factor models									
1	1f control group (n = 232)	1982.193	2023.554	1985.520	0.464	0.664	0.008	101.87**	
2	1f parent group (n = 439)	4002.051	4051.065	4012.982	0.351	0.857	0.571	110.06**	
3	1f autism group (n = 363)	2959.359	3006.092	2968.021	0.338	0.845	0.536	84.73**	
Two factor models									
4	2f control group (n = 232)	1881.771	1923.132	1885.098	0.000	1.000	1.006	1.45	-0.22
5	2f parent group (n = 439)	3892.859	3941.873	3903.791	0.000	1.000	1.005	0.87	-0.57
6	2f autism group (n = 363)	2877.375	2928.002	2886.759	0.000	1.000	1.003	0.75	-0.62
7	2f control group (n = 232) (AQ_soc, SQ; AQ_att, EQ)	1982.767	2027.574	1986.371	0.655	0.665	-1.008	100.44**	>1.0
8	2f parent group (n = 439) (AQ_soc, SQ; AQ_att, EQ)	3997.030	4050.129	4008.873	0.482	0.865	0.189	103.04**	>1.0
9	2f autism group (n = 363) (AQ_soc, SQ; AQ_att, EQ)	2940.547	2991.174	2949.931	0.416	0.882	0.295	63.92**	>1.0
10	2f control group (n = 232) (AQ together and EQ and SQ together)	1976.182	2020.989	1979.786	0.633	0.687	-0.875	93.86**	>1.0
11	2f parent group (n = 439) (AQ together and EQ and SQ together)	3988.040	4041.138	3999.883	0.460	0.877	0.261	94.05**	>1.0
12	2f autism group (n = 363) (AQ together and EQ and SQ together)	2947.319	2997.946	2956.703	0.438	0.870	0.219	70.69**	>1.0
Multigroup models									$\Delta\chi^2$ (*df*)
13	2f multigroup all estimates allowed to vary (n=1034)	8672.204	8835.264	8730.452	0.085	0.986	0.972	31.26**	
14	2f multigroup equal factor loadings (n = 1034)	8717.568	8846.039	8763.460	0.116	0.953	0.947	90.63**	59.37(8) $p<0.01$
Multigroup models including sex									
15	2f multigroup factor all estimates allowed to vary (n = 1034)	8618.343	8934.580	8731.308	0.100	0.978	0.961	54.17**	
16	2f multigroup equal factor loadings (n = 1034)	8614.676	8881.500	8709.990	0.089	0.974	0.969	70.50**	16.33(10) $p>0.05$
17	2f multigroup factor correlations equal for males and females within each group and equal factor loadings (n = 1034)	8612.924	8864.925	8702.942	0.086	0.973	0.971	74.75**	4.25(3) $p>0.05$

**$p<0.01$

Note. AIC, Akaike information criteria; BIC, Bayesian information criteria; CFI, Comparative fit index; r, correlation coefficient; RMSEA, Root mean square error of approximation; SSABIC, Sample size adjusted BIC; TLI, Tucker-Lewis index; χ^2 = chi square statistic; $\Delta\chi^2$ (*df*) =chi square difference test.

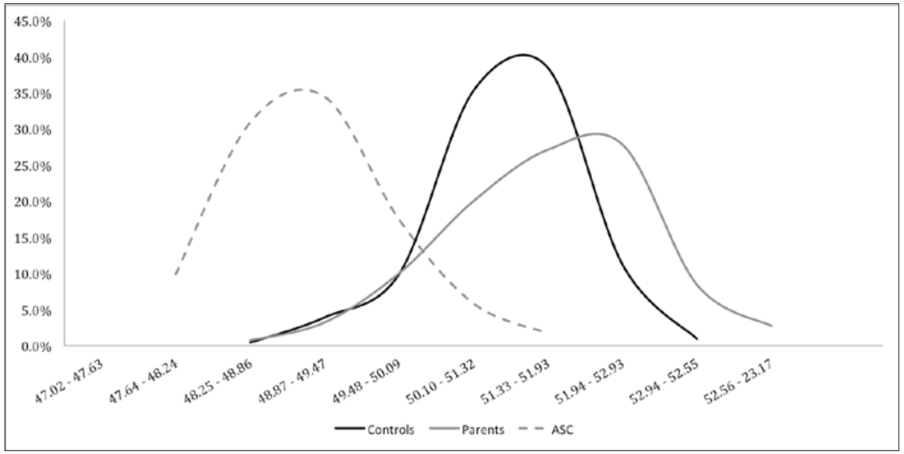

Figure 1a. Distribution of scores on the social factor of the AQ

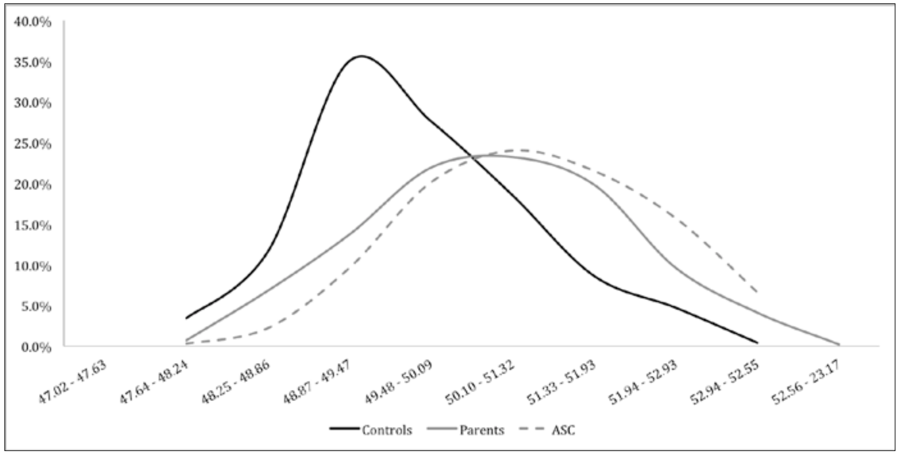

Figure 1b. Distribution of scores on the attention to detail factor of the AQ

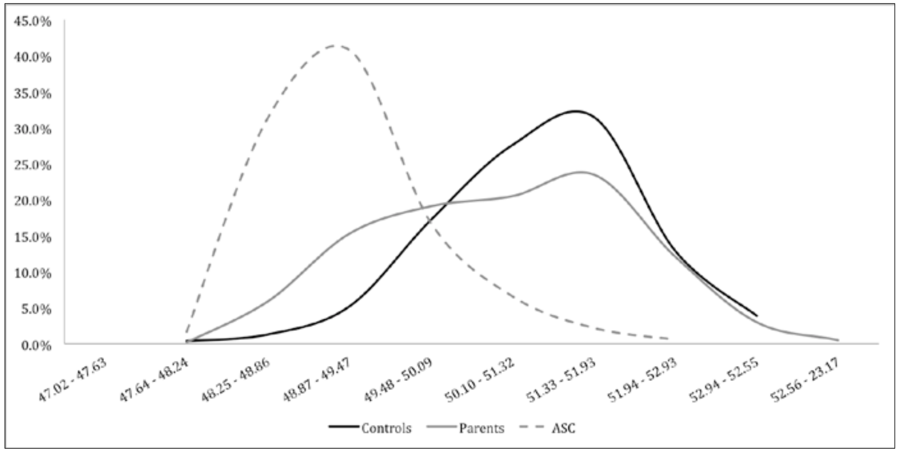

Figure 1c. Distribution of scores on the EQ

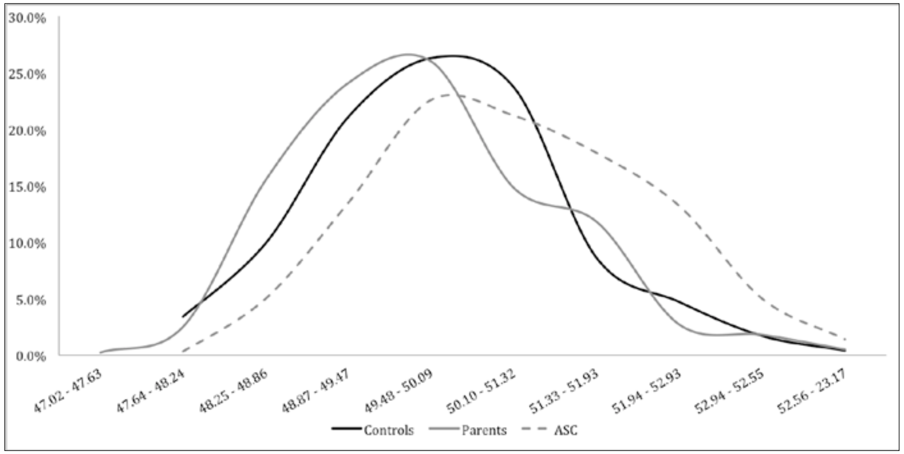

Figure 1d. Distribution of scores on the SQ

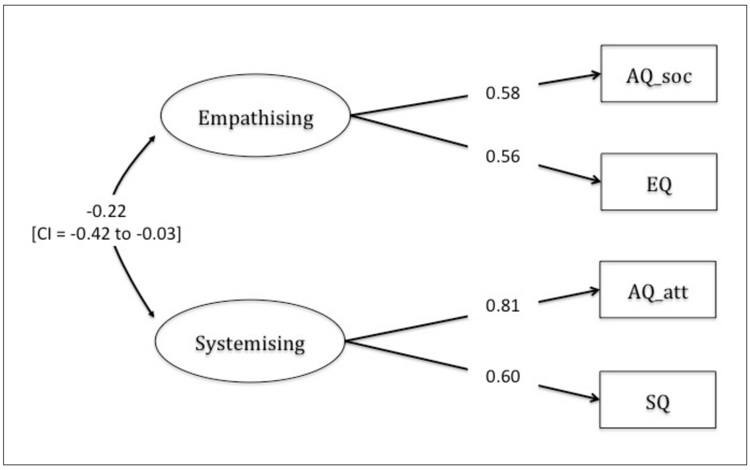

Figure 2a. Two factor multigroup model for the control group

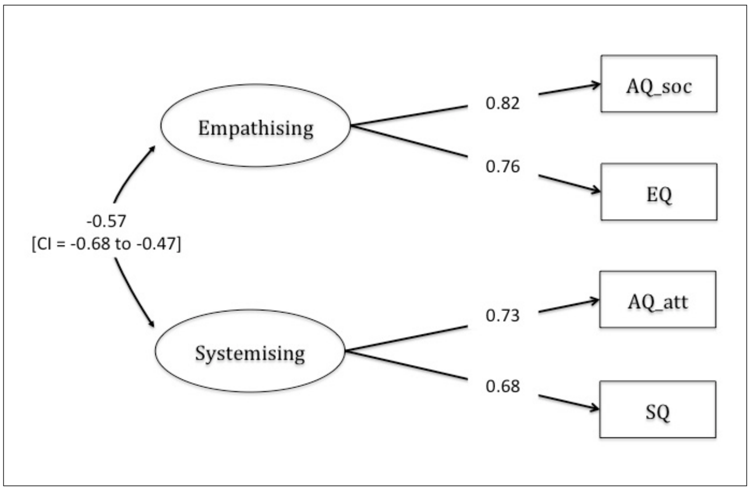

Figure 2b. Two factor multigroup model for the parent group

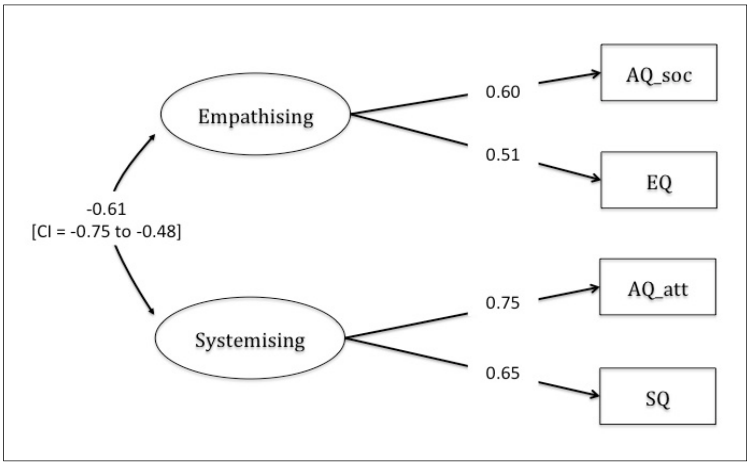

Figure 2c. Two factor multigroup model for the ASC group

Note. AQ_att, attention to detail factor of the Autism Spectrum Quotient; AQ_soc, social interaction factor of the Autism Spectrum Quotient; CI, 95% confidence interval; EQ, Empathy Quotient; SQ, Systemising Quotient

Chapter 3

Exploring the quantitative nature of empathy, systemising and autistic traits using factor mixture modelling

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Abstract

Background: Autism research has previously focused on either identifying a latent dimension or searching for subgroups. Research assessing the concurrently categorical and dimensional nature of autism is needed.

Aims: To investigate the latent structure of autism and identify meaningful subgroups in a sample spanning the full spectrum of genetic vulnerability.

Method: Factor mixture models were applied to data on empathy, systemising and autistic traits from individuals on the autism spectrum, parents and general population controls.

Results: A two-factor three-class model was identified, with two factors measuring empathy and systemising. Class one had high systemising and low empathy scores and primarily consisted of individuals with autism. Mainly comprising controls and parents, class three displayed high empathy scores and lower systemising scores, while class two showed balanced scores on both measures of systemising and empathy.

Conclusions: Autism is best understood as a dimensional construct, but meaningful subgroups can be identified based on empathy, systemising and autistic traits.

Introduction

A central debate in the development of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association, 2013) has been whether psychopathology is best conceptualised as a continuum of severity or as discrete categories of disorder, including the DSM-5 criteria for Autism Spectrum Disorder. Understanding the latent structure of autism, also referred to as Autism Spectrum Conditions (ASC), is important for guiding future conceptualisations of diagnostic criteria, as well as for informing the development of instruments assessing characteristics of ASC. A number of studies have assessed the latent structure of autism using either dimensional or discrete statistical techniques (Constantino et al., 2004; Frazier et al., 2010; Frazier et al., 2012; Ingram et al., 2008; Mandy, Charman, & Skuse, 2012). In contrast to these methods, factor mixture modelling allows for the presence of a concurrently dimensional and categorical latent structure (Lubke & Muthén, 2005). Three papers have previously applied mixture modelling to assess the latent structure of autism within children with a clinical ASC diagnosis and non-affected siblings (Frazier et al., 2012; Georgiades et al., 2014; Georgiades et al., 2013a). However, to date none of this work has focused on adult samples. Moreover, there is a large evidence base for the quantitative nature of autistic traits in the general population (Constantino, 2011), with undiagnosed first-degree relatives of individuals with autism displaying intermediate (or subthreshold) levels of autistic traits, also termed the broader autism phenotype (BAP; Piven et al., 1997b; Sucksmith et al., 2011). It is therefore important to assess the latent structure of autism across the full range of genetic vulnerability, from low risk general population samples to first-degree relatives (medium risk) and individuals with a clinical ASC diagnosis. The Empathising-Systemising (E-S) theory of autism argues that the persistent deficits in communication and social interaction in autism can be accounted for by an impairment in empathy, particularly cognitive empathy (also referred to as 'theory of mind'), whilst the repetitive behaviours and restricted interests can be explained by a strong drive to systemise (Baron-Cohen, 2009). Previous research in the same sample as reported here suggested that empathy and systemising are discrete constructs that can reliably be measured both in control samples, parents and individuals with ASC (Grove, Baillie, Allison, Baron- Cohen, & Hoekstra, 2013). The current study elaborates on these findings using mixture modelling methods. The study aims to assess the dimensional latent structure of empathy, systemising and autistic traits amongst individuals on the spectrum, first-degree relatives and the general population, whilst simultaneously examining whether meaningful subgroups can be identified.

Methods

Participants

Individuals with autism, parents of a child with autism and general population controls were recruited via two volunteer webpages at the University of Cambridge (www.autismresearchcentre.com; www.cambridgepsychology.com). The total sample consisted of 1034 individuals (controls = 232, parents = 439, ASC = 363). Controls were restricted to individuals with no previous psychiatric history and consisted of 110 females and 122 males (mean age = 33, sd = 10). The parent group contained 298 females and 141 males (mean age = 42, sd = 8). Parents were included in the study if they had a child with a formal ASC diagnosis, but reported no diagnosis of autism themselves. The ASC group comprised 170 females and 193 males (mean age = 36, sd = 11). These individuals had received a formal clinical diagnosis of autism. IQ was assessed via an online adapted version of the Ravens Progressive Matrices (Raven, Raven, & Court, 2003). The control group scored significantly higher on the Ravens than both the parent group ($p < 0.01$) and individuals with ASC ($p < 0.01$). There were no differences in IQ between parents and individuals with autism ($p = 0.112$).

Measures

Autistic traits

The Autism Spectrum Quotient (AQ; Baron-Cohen et al., 2001b) is designed to assess quantitative autistic traits including those related to communication, social skills, attention to detail, imagination and attention switching. 50 items are assessed with four response options; "definitely agree", "definitely disagree", "slightly agree" and "slightly disagree". A raw scoring method was used (Hoekstra et al., 2008), eliciting scores ranging from 50 to 200, with higher scores indicating more autistic traits. Previous research suggests the AQ can be split into two reliable subscales relating to social and non-social traits (Hoekstra et al., 2008). A broad social interaction factor (comprising 40 items assessing communication, social skills, imagination and attention switching) and an attention to detail factor (consisting of the remaining 10 items) were included in all analyses.

Systemising

The Systemising Quotient Revised (SQ; Wheelwright et al., 2006) is a measure designed to assess an individual's propensity to systemise; to construct and understand rule based systems for categorisation. This measure includes 75 items scored on a Likert response scale with four response options; "strongly agree", "strongly disagree", "slightly agree" and "slightly disagree". Strong responses score two points, with slightly agree/disagree responses receiving one point. Scores range from zero to 150, with higher scores indicative of a heightened drive to systemise.

Empathy

The Empathy Quotient (EQ; Baron-Cohen & Wheelwright, 2004) is a self-report measure of empathy. This 40-item measure includes equivalent response options and scoring methods to the SQ. Full endorsement of all items gives a score of 80, with higher scores indicative of a better capacity to empathise.

Apart from the AQ, SQ and EQ self-report questionnaires, which were included in the mixture analyses, data was also collected on two performance-based measures of empathy. Due to a large proportion of missing data in the sample of fathers, these measures were not included in the factor analyses. The 'Reading the Mind in the Eyes' test revised (Eyes; Baron-Cohen et al., 2001a) assesses how accurately an individual can read the emotion in another by viewing only the eye region of the face. 36 items are presented with four descriptions of mental states. Relatively subtle and complex mental states are used, for example, *joking, insisting, amused* and *relaxed*, making the task an advanced test of empathy. The total number of correct items is recorded, with higher scores reflecting better ability.

The Karolinska Directed Emotional Faces (KDEF; Lundqvist, Flykt, & Ohman, 1998) is a test of more basic emotion recognition, including happy, sad, angry, afraid, disappointed, surprised and neutral, and giving participants the opportunity to view the whole face. This measure consists of 140 items for which accuracy and response time information is recorded. Response times were weighted for accuracy (Sutherland & Crewther, 2010). In order for ease of interpretation, the KDEF was rescored so that higher values indicate higher ability rather than a slower response time.

Analytic strategy

Confirmatory factor analysis

Confirmatory factor analyses (CFA) were conducted to assess the dimensional structure of empathy, systemising and autistic traits using the EQ, SQ and the two subscales of the AQ. A one-factor model was implemented in order to assess whether these traits lie on a continuum of severity (Model 1). Next, a two-factor model representing the distinction between empathy and systemising was fit to the data (Model 2). The EQ and the social interaction factor of the AQ were predicted to load onto a factor representing empathy, with the SQ and the attention to detail factor of the AQ loading onto a second factor representing systemising.

Latent class analysis

Latent class analysis (LCA) is a technique designed to evaluate whether a number of observed variables (in this case empathy, systemising and autistic traits) can help to define an underlying categorical variable or class (Hagenaars & McCutcheon, 2002). A series of models ranging from one to five classes were implemented (Models 3 to 7).

Factor mixture models

Factor mixture models (FMM) combining both CFA and LCA models were also estimated. Measurement invariance is given when a measurement model relating observed variables to underlying latent variables does not vary across latent classes (Meredith, 1993). There is considerable debate over the invariance restrictions placed on classes in FMM and their varying effects on the results obtained (Lubke & Muthén, 2007). Lubke and Muthén (2005) show that correct class assignment is maximised by constraining factor loadings to be equal across classes while allowing thresholds to vary. This method was therefore applied to all models in the current study. Models were estimated up to and including the number of factors from the best fitting CFA model and the number of classes from the best fitting LCA model (Models 8 to 17).

Analyses were estimated using Mplus version 7 (Muthén & Muthén, 2012a) with the robust maximum likelihood ratio estimator. A number of fit indices were estimated including the Akaike information criterion (AIC; Akaike, 1987), Bayesian information criterion (BIC; Schwarz, 1978), sample size adjusted BIC (SSABIC; Sclove, 1987), entropy (Ramaswany, DeSarbo, Reibstein, & Robinson, 1993) and the Lo Mendel Rubin likelihood ratio test (LMR; Lo, Mendell, & Rubin, 2001). Lower AIC, BIC and SSABIC values are indicative of a better fit to the data. These fit statistics were used to evaluate the fit of the CFA models. In LCA and FMM, the BIC has been shown to be more reliable in obtaining the best fitting models than AIC and SSABIC (Gebregziabher, Shotwell, Charles, & Nicholas, 2012; Nylund, Asparouhov, & Muthén, 2007). Therefore the BIC was used to reach a final decision on the best fitting model in LCA and FFM analyses. In addition to these measures, the LMR test and entropy statistic are useful to determine the optimal number of latent classes. The LMR test is a goodness of fit test that indicates whether the addition of an extra class will provide an improvement to the fit of the model to the data (Lo et al., 2001). Similarly, the entropy statistic is a measure of the accuracy with which each individual can be categorised into a latent class, with higher entropy values indicating better categorisation (Ramaswany et al., 1993). In addition to AIC, BIC and SSABIC, the LMR and entropy statistic were also consulted when evaluating the LCA and FMM models.

As well as taking into account the fit indices mentioned above, in evaluating the model fit the conceptual appropriateness of the latent class profiles was also considered. Once the best fitting model was identified, further analyses assessing mean differences and demographic information for the classes identified was conducted using SPSS 21 (IBM Corp, 2012).

Results

As reported previously (Baron-Cohen et al., 2001a; Sucksmith et al., 2013; Wheelwright et al., 2006), there were a number of differences in the mean scores on the questionnaires for controls, parents and individuals with ASC (see Table 1). The ASC group scored significantly lower on the social interaction items of the AQ and higher on the attention to detail items compared with controls and parents ($p < 0.01$). Individuals with ASC also obtained significantly higher SQ scores and lower EQ scores compared with all other participants. Parents scored significantly lower on the social interaction factor of the AQ than controls ($p < 0.05$). Control females displayed higher scores on the EQ and on the AQ social interaction factor as well as lower scores on the SQ than control males. This pattern was also observed amongst parents. Interestingly, females with ASC displayed higher scores on the social interaction and attention to detail items of the AQ than males with autism. However, there were no sex differences within the ASC group on the other two measures.

Model fit indices for CFA, LCA and FMM analyses are given in Table 2. Results from the CFA indicated that a two-factor model (Model 2) consisting of empathy and systemising provided the best fit to the data, with lower AIC, BIC and SSABIC values than the one-factor model. There was a moderate negative correlation between the two factors, suggesting that better systemising is associated with lower empathy abilities. LCA models with up to five latent classes were then estimated. The LMR value for the five-class model did not reach significance, indicating that a model with one less class provided a better classification of individuals. The four-class LCA also had the smallest

AIC, BIC and SSABIC values of the remaining four models (Model 6) as well as an entropy statistic of 0.81. This model was therefore selected as providing the best fit to the data.

Mixture models consisting of one and two factors and up to five latent classes were then estimated. The smallest BIC values were identified in the two-factor three-class and two-factor four-class models. The three-class model had the smallest BIC value as well as the largest entropy value and provided the most parsimonious explanation for the data. This model, designating three classes with varying levels of empathy, systemising and autistic traits, was therefore selected as the best fitting model (Model 15). Model fit statistics were compared across Models 2 (CFA), 6 (LCA) and 15 (FMM) in order to establish whether empathy, systemising and autistic traits are best conceptualised as dimensional, categorical or a combination of both dimensional and categorical constructs. The mixture model (Model 15) provided the best fit across all three analyses and was therefore selected as providing the soundest explanation for the data.

Model 15 contained two moderately correlated factors ($r = -0.49$) representing empathy and systemising. This model identified three latent classes of individuals. Class one comprised 45% of the sample (n = 461, mean age = 38 yrs, sd = 11), class two approximately 30% (n = 310, mean age = 37, sd = 11) and class three the remaining 25% (n = 263, mean age = 40, sd = 9). Class three was significantly older than both class two ($p < 0.01$) and class one ($p < 0.05$). Class three also had a lower IQ score than class one ($p < 0.05$).

Mean differences on measures of empathy, systemising and autistic traits for each class are given in Figure 1. Scores on all measures were converted to the same scale via Z score transformation. These Z scores were then increased by two in order to remove any negative values before being plotted to ease interpretation. The first class identified (Class S) scored significantly higher on self-reported systemising (including the SQ and the attention to detail factor of the AQ) and lower on self-reported empathy (including the EQ and the social interaction factor of the AQ) than the other latent classes ($p < 0.001$). Class three (Class E) scored significantly higher on measures of empathy and lower on systemising than classes one and two ($p < 0.001$). Class two displayed scores on measures of systemising, empathy and autistic traits intermediate to that of the other two classes, showing a balance between empathy and systemising (Class B).

Subsequently, mean differences on performance-based measures of empathy were assessed. Class S scored significantly lower on the Eyes task than both Class B and Class E ($p < 0.01$). Class B also performed more poorly than Class E on this task (Mean difference = -0.26, $p < 0.01$). Class S scored significantly lower than both other classes on the KDEF task ($p < 0.001$), with no differences in mean scores between Class B and Class E.

The proportion of males, females, controls, parents and individuals with autism falling into each class is given in Figure 2. Class E primarily comprised females, while the gender division was similar in the other two classes. Individuals with ASC made up the majority of Class S (71%), along with 23% of parents and a very small proportion of controls (6%). Class B consisted of approximately 39% controls, 50% parents and 11% of individuals with ASC. Class E consisted predominantly of parents (67%) and controls (32%) with a very small proportion of those with ASC (1%). Within the parent group, 38% of fathers compared with 18% of mothers fell into Class S. Half of the sample of mothers fell into Class E compared with 20% of fathers.

Discussion

Structural equation modelling including CFA, LCA and FMM analyses in a large sample of individuals with ASC, parents and controls indicated that the characteristics of autism as measured in a sample spanning the full spectrum of genetic liability, are best described by a two-factor three-class mixture model. The quantitative nature of autistic traits is best captured by two moderately correlated latent factors representing systemising and empathy. In addition, three homogeneous latent classes of individuals could be identified by their mean scores on measures of empathy, systemising and autistic traits. Class one displayed superior performance on systemising, with significantly lower scores on both self-reported and performance-based tests of empathy (Class S). Class three demonstrated the opposite effect, showing increased scores on empathy tasks and lower performance on self-report measures of systemising (Class E). Class two appeared to be more balanced in terms of both empathy and systemising propensity (Class B).

The results provide support for the E-S theory, indicating that empathy and systemising are two separate constructs that together may partly provide a cognitive explanation of the characteristics of autism. The findings also lend indirect support to the current DSM-5 diagnostic criteria, including social and communication impairment (represented in the current study by difficulty with empathy) and repetitive behaviours and restricted interests (represented here by high systemising scores). EQ items map onto the social and communication domain assessing difficulties in social-emotional reciprocity, nonverbal communication and relationships. Examples include: "I can easily tell if someone else wants to enter a conversation", "I am quick to spot when someone in a group is feeling awkward or uncomfortable" and "Friendships and relationships are just too difficult, so I tend not to bother with them". The repetitive behaviour and restricted interests domain in DSM-5 contains four criteria relating to stereotyped and repetitive movement or use of objects, insistence on sameness, fixated interests and hyper or hypo reactivity to sensory stimuli. The first three of these DSM-5 criteria may be (partly) accounted for by a drive to systemise. By engaging in stereotyped and repetitive actions, insisting on sameness and focusing on circumscribed interests, the world becomes more predictable and therefore easier to negotiate. It is as yet less clear how sensory reactivity relates to empathy and systemising. A recent study reported an association between sensory sensitivity and autistic traits, with greater sensitivity associated with more traits on the autism spectrum (Tavassoli, Hoekstra, & Baron- Cohen, 2014). However, further research is needed in order to comprehensively understand the association between sensory reactivity, empathy and systemising.

The two-factor structure found in the current study is consistent with previous factor analytic studies directly assessing autistic characteristics via diagnostic instruments, suggesting that the autism phenotype follows a dyadic structure comprising social communicative difficulties and non-social autistic traits (Frazier et al., 2012; Grove et al., 2013; Mandy et al., 2014; Mandy et al., 2012). However, the two factors identified in the present study were moderately correlated (r = -0.49), indicating that empathy and systemising are not entirely independent.

The E-S theory posits that there are five different cognitive profiles that can be identified based on empathy and systemising. Type E (E > S) are individuals with stronger empathy than systemising ability; Type S (S > E) comprises individuals with systemising ability that is stronger than their empathy skills; Type B (E = S) includes individuals with similar empathy and systemising ability; Extreme Type E (E >> S) comprises individuals with above average empathy who have difficulty with systemising; and lastly, Extreme Type S (S >> E) includes individuals with above average systemising who have difficulty with empathy (Baron-Cohen, 2009). Individuals with autism are thought to be represented by the Extreme Type S cognitive profile, with varying combinations of the other cognitive profiles in the general population (Baron-Cohen, 2009). Using factor mixture modelling techniques, the current study identified classes that map very well onto the Type S, B and E profiles outlined in this theory.

Class S was characterised by low empathy and high systemising scores. It consisted predominantly of individuals with autism, with only a very small proportion of the control sample falling into this class. This finding follows the predictions of the E-S theory, with autism being characterised by an interest in systemising and difficulties with empathy. Approximately 20% of Class S comprised parents of a child with ASC, suggesting that these individuals display the BAP. Proportionally more fathers than mothers fell into Class S (38% versus 18% respectively). This is consistent with previous research suggesting that the BAP is more common in male relatives (Scheeren & Stauder, 2008). The presence of parents in Class S has clinical implications, as these parents may be especially well served by clinical advice and guidance provided in a systematic, factual manner. Parents who show difficulties with empathy themselves may also benefit from advice on how to manage and improve their own relationships with others.

A large proportion of parents, primarily mothers, were also represented in Class E, characterised by low systemising and high empathy scores. This finding highlights that not all parents show characteristics of the BAP. The high representation of mothers but not fathers in Class E further supports the notion of a gender difference in the BAP, with approximately half of mothers, but only 20% of fathers displaying an absence of any characteristics of the BAP.

A small proportion of individuals with ASC (approximately 10%) fell into Class B, represented by equivalent scores on empathy and systemising. It is surprising that a proportion of individuals with ASC would fall into this class. However, further analyses within this subgroup suggested a number of important differences. Although these individuals with an ASC diagnosis displayed similar scores on the EQ and SQ as the rest of Class B, they scored significantly lower on the social interaction items of the AQ

(Mean difference = 12.1, $p < 0.001$) and higher on the attention to detail items (Mean difference = 2.7, $p < 0.01$). They also scored lower on both performance-based measures of empathy (Eyes mean difference = 2.3, $p < 0.01$; KDEF mean difference = 82.5, $p < 0.001$), displaying equivalent scores to individuals in Class S. This indicates that while the self-report EQ and SQ scores suggest these individuals have equivalent empathy and systemising abilities, their scores on performance-based measures highlight a difficulty with empathy. Interestingly, the 10% of individuals with ASC who fell in class B were also significantly younger and had a lower IQ score than the other individuals in Class B. It could be that limited insight or understanding of their difficulties had an impact on scores on self-report measures, while their difficulties with empathy were picked up by the relatively poor performance on the KDEF and Eyes tasks. This highlights the importance of the use of performance-based measures in research and clinical practice as well as the potential impact of cognitive ability and age on self-report measures.

The current study identified homogeneous subgroups based on levels of empathy, systemising and autistic traits in a sample spanning the entire spectrum of vulnerability to autism, from general population controls to individuals with a clinical diagnosis. Most previous studies aiming to define phenotypic subgroups in autism included clinical samples only. These studies generally highlighted that distinct groups can be identified based on either the presence of an ASC diagnosis (Frazier et al., 2012) or severity of symptoms (Georgiades et al., 2014; Georgiades et al., 2013a). A recent study in a large sample of children with a DSM-IV diagnosis of pervasive developmental disorder identified six classes of individuals, including three groups displaying impairment on both social and non-social domains as well as three classes with impairment on only one of the two symptom dimensions (Greaves-Lord et al., 2013). One class comprised individuals displaying social and communication difficulties, but no restricted repetitive behaviours, suggesting that if these individuals had been diagnosed using current DSM-5 criteria rather than the DSM-IV, the new DSM-5 diagnosis of Social Communication Disorder (SCD; American Psychiatric Association, 2013) may have been appropriate. Our study, using a sample spanning from controls to individuals with autism, rather than a clinical sample only, did not identify a separate class characterised by low empathy and average systemising, a pattern that would perhaps be expected for individuals with SCD. Nevertheless it would be of interest to study empathy and systemising in individuals with SCD and explore any potential differences in these traits compared to individuals with autism.

Limitations

The findings of this study should be interpreted in the light of some limitations. While the gender ratio was balanced for the control and ASC groups, there was a larger proportion of mothers (n=298) than fathers (n=141) in the parent group. Future research would benefit from the inclusion of more fathers for comparison. Secondly, this study only included performance-based measures of empathy abilities; no performance-based measure of systemising was available to compare against the self-report questionnaire data. It should be stressed that as this study included self-rated measures, all individuals with ASC were high functioning. The results from this study can therefore not be generalised to individuals with ASC and intellectual disability. Given that the data were collected online, it was also not possible to verify diagnoses of autism. However, there is evidence to suggest that clinical diagnoses of ASC reported by online volunteers are generally reliable (Lee et al., 2010).

Conclusions

In conclusion, this study assessed the quantitative nature of empathy, systemising and autistic traits amongst individuals on the spectrum, first-degree relatives and general population controls. Results highlighted a two-factor three-class model in which two dimensions based on systemising and empathy were identified. This provides indirect support for the new diagnostic criteria outlined in DSM-5, which follow a dyadic rather than a triadic structure and include a dimensional rather than a categorical approach. Three homogenous classes were defined based on mean scores on empathy, systemising and autistic traits. Taken together, these results support the quantitative approach to autistic traits and confirm that even with the use of quantitative measures meaningful subgroups can be identified.

Table 1 Mean scores of controls, parents and individuals with autism on questionnaire data by sex

	AQ_soc	AQ_att	SQ	EQ
	Mean (sd)	Mean (sd)	Mean (sd)	Mean (sd)
Controls	117.2 (15.9)	25.3 (5.1)	63.7 (23.9)	43.1 (14.2)
Male	113.4 (15.3)	25.1 (5.0)	67.0 (23.0)	38.5 (12.9)
Female	121.5 (15.5)	25.4 (5.3)	60.0 (24.5)	48.1 (13.9)
Parents	113.6 (22.4)	24.3 (5.6)	57.8 (25.3)	42.4 (18.2)
Male	107.0 (23.2)	24.7 (5.4)	71.0 (26.9)	33.2 (17.1)
Female	116.7 (21.3)	24.1 (5.7)	51.6 (22.0)	46.8 (17.1)
ASC	75.2 (16.5)	29.9 (5.1)	77.4 (25.2)	18.4 (10.0)
Male	77.7 (17.1)	29.1 (4.9)	76.4 (25.3)	18.1 (10.6)
Female	72.3 (15.2)	30.8 (5.2)	78.5 (25.2)	18.7 (9.4)

Note. AQ_att, attention to detail factor of the Autism Spectrum Quotient; AQ_soc, social interaction factor of the Autism Spectrum Quotient; EQ, Empathy Quotient; SQ, Systemising Quotient Revised.

Table 2 Confirmatory factor analysis, latent class analysis and factor mixture models for empathy, systemising and autistic traits

Model	Analysis/Model description	Fit statistics				
		AIC	BIC	SSABIC	Entropy	LMR
Confirmatory factor analysis						
1	1f	33001.576	33060.870	33022.757		
2	**2f**	**32728.316**	**32787.610**	**32749.497**		
	Factor correlation (-0.65)					
Latent class analysis						
3	1c	35057.523	35097.053	35071.644		
4	2c	33498.692	33562.927	33521.638	0.872	0.0000
5	3c	33012.493	33101.434	33044.264	0.852	0.0002
6	**4c**	**32814.899**	**32928.546**	**32855.495**	**0.812**	**0.0172**
7	5c	32717.575	32855.928	32766.997	0.800	0.2492
Factor mixture models						
8	1f1c	33001.576	33060.870	33022.757		
9	1f2c	32753.226	32842.168	32784.997	0.626	0.0000
10	1f3c	32592.453	32711.041	32634.814	0.811	0.0563
11	1f4c	32505.882	32654.118	32558.834	0.801	0.0822
12	1f5c	32455.023	32632.905	32518.565	0.784	0.0003
13	2f1c	32728.316	32787.610	32749.497		
14	2f2c	32578.082	32676.906	32613.383	0.769	0.0035
15	**2f3c**	**32491.851**	**32625.264**	**32539.508**	**0.760**	**0.0232**
16	2f4c	32462.540	32625.599	32520.787	0.707	0.0249
17	2f5c	32443.404	32641.052	32514.007	0.726	0.0982

Note. AIC, Akaike information criteria; BIC, Bayesian information criteria; c, class; f, factor; LMR, Lo Mendel Rubin likelihood ratio test; SSABIC, sample size adjusted BIC. Boldface print indicates best model for the specific analysis.

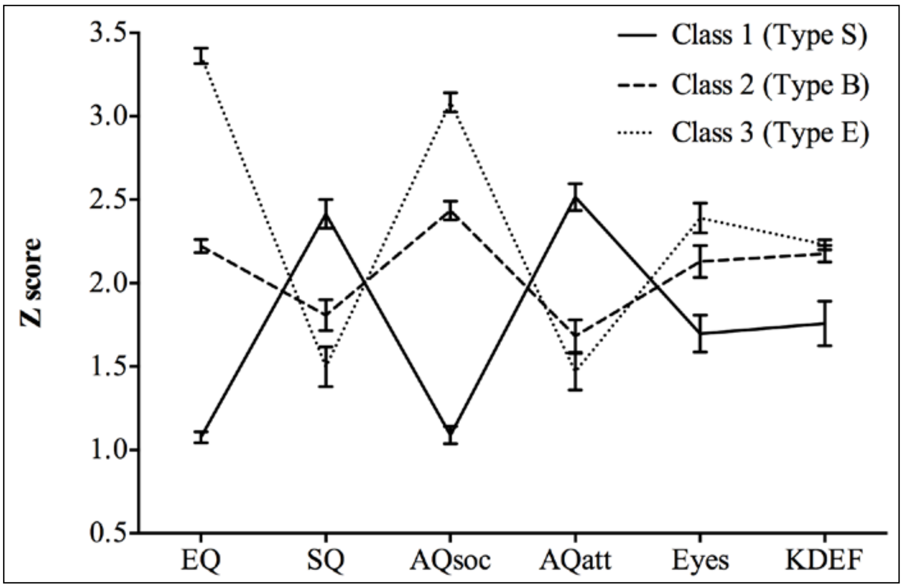

Figure 1. Mean scores on self-report and performance tasks across classes

Note. AQatt, attention to detail factor of the Autism Spectrum Quotient; AQsoc, social interaction factor of the Autism Spectrum Quotient; EQ, Empathy quotient; Eyes, Reading the Mind in the Eyes task; KDEF, Karolinska Directed Emotional Faces; SQ, Systemising Quotient Revised.

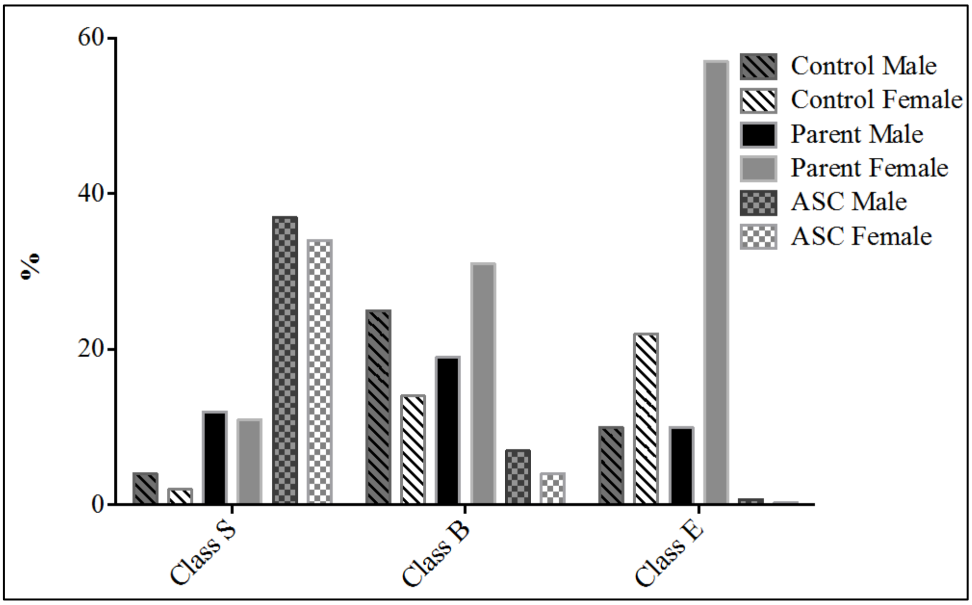

Figure 2. Sex and group membership in each class

Chapter 4

The latent structure of cognitive and emotional empathy in individuals with autism, first-degree relatives and typical individuals

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Abstract

Background: Empathy is a vital component for social understanding involving the ability to recognise emotion (cognitive empathy) and provide an appropriate affective response (emotional empathy). Autism spectrum conditions have been described as disorders of empathy. First-degree relatives may show some mild traits of the autism spectrum, the broader autism phenotype (BAP). Whether both cognitive and emotional empathy, rather than cognitive empathy alone, are impaired in autism and the BAP is still under debate. Moreover the association between various aspects of empathy is unclear. This study aims to examine the relationship between different components of empathy across individuals with varying levels of genetic vulnerability to autism.

Methods: Factor analyses utilising questionnaire and performance-based task data were implemented amongst individuals with autism, parents of a child with autism and controls. The relationship between performance-based tasks and behavioural measures of empathy was also explored.

Results: A four-factor model including cognitive empathy, emotional empathy, social skills and a performance-based factor fitted the data best irrespective of genetic vulnerability. Individuals with autism displayed impairment on all four factors, with parents showing intermediate difficulties. Performance-based measures of empathy were related in almost equal magnitude to cognitive and emotional empathy latent factors and the social skills factor.

Conclusions: This study suggests individuals with autism have difficulties with multiple facets of empathy, while parents show intermediate impairments, providing evidence for a quantitative BAP. Impaired scores on performance-based measures of empathy,

often thought to be pure measures of cognitive empathy, were also related to much wider empathy difficulties than impairments in cognitive empathy alone.

Introduction

Empathy has been defined as the drive to identify and respond appropriately to emotions and mental states in others (Baron-Cohen, 2002; Deutsch & Madle, 1975). It plays a vital role in human relationships and allows an individual to make sense of and predict the behaviour of another (Smith, 2006). Empathy involves both the ability to recognise and understand emotion in others (Smith, 2006) as well as an affective response to another's emotional state (Eisenberg & Strayer, 1987; Hoffman, 1987), respectively cognitive and emotional empathy (Baron-Cohen & Wheelwright, 2004; Eisenberg & Strayer, 1987).

Autism spectrum conditions (ASC) involve empathy deficits (Baron-Cohen & Wheelwright, 2004; Decety & Moriguchi, 2007; Gillberg, 1992; Wing, Gould, & Gillberg, 2011) and are characterised by communication and social difficulties as well as repetitive behaviours or restricted interests (American Psychiatric Association, 2013). Empathy dysfunction in autism has been demonstrated via research noting a theory of mind (ToM) impairment in children with ASC (Baron-Cohen et al., 1985); that is, that individuals with autism have difficulty reading the beliefs and intentions of others (Baron-Cohen et al., 1985; Perner, Frith, Leslie, & Leekam, 1989). ToM is often used interchangeably with cognitive empathy, perspective taking and 'mentalising' (Davis, 1996). However, as noted above, empathy has long been defined as a multifactorial construct including not only the representation of another's emotional state (i.e., ToM or cognitive empathy) but also an affective response (emotional empathy).

The Empathising-Systemising (E-S) theory (Baron-Cohen, 2009; Baron-Cohen, 2010a) expands the concept of ToM to include this affective component of empathy. The E-S theory argues that the social and communication difficulties seen in ASC can be accounted for by an empathy impairment (including both cognitive and emotional components), and the repetitive behaviours and restricted interests by an inclination for systemising (the drive to understand and derive rules about a system) (Baron-Cohen, 2010b). A recent factor analytic study by the authors (Grove et al., 2013) found support for the E-S model. This study, based on the same individuals as the research reported here, identified two factors representing empathy and systemising. These factors were found consistently across individuals with autism, first-degree relatives and general population controls (Grove et al., 2013). In concordance with the E-S theory, individuals with ASC showed elevated scores on the latent systemising factor and low scores on the empathy factor. This previous study included questionnaire measures of empathy and systemising only. However, other studies have indicated that individuals with autism also have difficulty with performance-based tasks involving the identification of emotions and perspective taking (Baron-Cohen et al., 1997b; Charman et al., 1997; Sucksmith et al., 2013). As these tasks involve the identification of emotion, they are generally conceptualised as performance-based tasks of cognitive empathy.

Although there is much evidence to suggest that individuals with autism display difficulties with ToM or cognitive empathy, there is more debate about the role of emotional empathy in autism. While mirror neuron theory (Williams, Whiten, Suddendorf, & Perrett, 2001) argues that individuals with ASC have weak emotional empathy, Dziobeck and others (2008) claim that emotional empathy is intact in autism. Other theorists have proposed that it is due to heightened emotional empathy that individuals with ASC find the social world more challenging, arguing that it is overwhelming rather than difficult to understand (Markram, Rinaldi, & Markram, 2007; Smith, 2006, 2009).

First-degree relatives of individuals with an ASC diagnosis may also show some mild traits of the autism spectrum (Piven et al., 1997b), also referred to as the broader autism phenotype (BAP; Bolton et al., 1994; Sucksmith et al., 2011). The finding of the BAP fits with the notion that autism is under polygenic influence, and that at least part of these genetic influences are inherited (rather than de novo genetic events) and can also be found in undiagnosed relatives displaying the BAP (Abrahams & Geschwind, 2008). The BAP has also been shown to apply to empathy, with parents and siblings of affected individuals scoring lower on performance-based tasks involving emotion recognition (Losh et al., 2009; Palermo, Pasqualetti, Barbati, Intelligente, & Rossini, 2006; Sucksmith et al., 2013; Wallace, Sebastian, Pellicano, Parr, & Bailey, 2010) and questionnaire measures assessing empathy (Sucksmith et al., 2013; Sucksmith et al., 2011). It is therefore important to examine cognitive and emotional empathy not only in clinical samples, but across the full range of genetic variability, including individuals on the autism spectrum, their relatives and general population controls.

A number of quantitative measures of empathy have been used in previous research, including the Interpersonal Reactivity Index (Davis, 1980) and the Empathy Scale (Hogan, 1969). However, one of the most widely used measures is the Empathy Quotient (EQ; Baron-Cohen & Wheelwright, 2004), a self-report measure of empathy assessing both cognitive and emotional components. The EQ has recently been studied in detail across three studies. Two studies highlight a three dimensional structure including cognitive empathy, emotional empathy and social skills (Lawrence, Shaw, Baker, Baron-Cohen, & David, 2004; Muncer & Ling, 2006), with the third highlighting a single dimension (Allison, 2011). The first two studies were based on student and general population samples, with the third including individuals with autism and first-degree relatives. Although individuals with ASC and family members were included in the third study, the factor structure and utility of the EQ was examined for the whole sample and not for each of the three groups (individuals on the spectrum, first-degree relatives and general population controls) separately.

Although the EQ and performance-based measures of cognitive empathy have been studied quite extensively by themselves in previous studies, the relationship between subscales of the EQ (a questionnaire-based measure), and performance-based measures of empathy have not been comprehensively assessed to date. The current study aims to evaluate the multifactorial nature of empathy utilising both behavioural and performance-based task data. It was assessed whether the latent structure of empathy differs across samples stratified by genetic risk (individuals with ASC, first-degree relatives and controls).

Methods

Participants

Individuals were recruited via two online databases from the Autism Research Centre (www.autismresearchcentre.com) and the Department of Psychology (www.cambridgepsychology.com) at the University of Cambridge. The total sample consisted of 1034 community-based participants including individuals with ASC (193 males, 170 females; mean age = 36, sd = 11), parents of a child with autism (141 males, 298 females; mean age = 42, sd = 8) and general population controls (122 males, 110 females; mean age = 33, sd = 10). Individuals who reported no previous psychiatric history were included in the control group. Individuals who had a formal ASC diagnosis were included in the autism group. The control group contained a significantly larger proportion of individuals with an undergraduate degree than the parent and ASC groups ($p < 0.001$).

Measures

Empathy

The Empathy Quotient (EQ; Baron-Cohen & Wheelwright, 2004) is a self-report measure assessing both cognitive (e.g., 'I can tune into how someone else feels rapidly and intuitively') and emotional empathy (e.g., 'Seeing people cry does not really upset me'). The EQ includes 40 statements with four response options; 'strongly disagree', 'slightly disagree' 'strongly agree' and slightly agree'. 'Strongly agree' responses are given two points, with 'slightly agree' responses receiving one point. Higher scores are indicative of increased levels of self-reported empathy. The EQ shows good test-retest reliability ($r = 0.97$, $p < 0.001$) (Baron-Cohen & Wheelwright, 2004).

Autistic traits

The Autism Spectrum Quotient (AQ; Baron-Cohen et al., 2001b) assesses quantitative autistic traits including communication, imagination, attention to detail, social skills and attention switching. 50 items are assessed on a 4-point Likert scale with response categories 'definitely disagree', 'slightly disagree' 'definitely agree' and slightly agree'. Hoekstra and others (2008) outline a raw scoring method, with total scores in the range of 50 to 200; higher scores indicating the presence of autistic traits. Previous research has highlighted that the AQ shows good test-retest reliability (Baron-Cohen et al., 2001b).

A previous factor analysis showed that the AQ can be reliably split into two factors assessing social and non-social autistic traits (Hoekstra et al., 2008). A broad social interaction factor was compiled using items assessing communication, social skills, imagination and attention switching (40 items). As the focus of the current study is on empathy, the further 10 items assessing attention to detail or non-social autistic traits were excluded from the current analysis.

Performance tasks

The 'Reading the Mind in the Eyes' test revised (Baron-Cohen et al., 2001a) is a performance task designed to assess how well an individual can read another's emotion based on viewing the eye area alone. This measure has been described as an advanced ToM task that assesses the ability to attribute mental states to oneself and others (i.e., cognitive empathy). Individuals are presented with a series of 36 photographs of the eye region of the face and asked to choose which of four words best describes the emotion depicted. The emotions used in the task are subtle and include, for example, a choice between *jealous, panicked, arrogant* and *hateful*. This test has been shown to detect meaningful individual differences, with individuals with AS or HFA scoring significantly lower than general population controls (Baron-Cohen et al., 2001a).

The Karolinska Directed Emotional Faces (KDEF; Lundqvist et al., 1998) is another task designed to assess the recognition of more basic emotions in others. In this modified version, participants were shown 140 photographs of faces expressing seven emotions (happy, sad, angry, afraid, disappointed, surprised and neutral). For each photograph, individuals were asked to select which of the seven emotions best described the emotion depicted. Results provide indications of accuracy and response time for each facial expression. Accuracy adjusted response time was calculated by dividing the mean response time for correct items by the proportion of items answered correctly. Weighted mean reaction times have been shown to provide a more sensitive measure, taking any potential speed-accuracy trade off into account (Sutherland & Crewther, 2010). The KDEF has good test-retest reliability and has been validated on emotional content, intensity and arousal (Goeleven, De Raedt, Leyman, & Verschuere, 2008). Individuals with autism have been shown to score lower than controls on this task (Sucksmith et al., 2013). To aid data interpretation, the KDEF was rescored so that lower values indicate higher accuracy adjusted response time and hence lower empathy ability.

Given that sex differences on the mean test scores were not the focus of this paper and have been reported elsewhere (Baron-Cohen et al., 2001a; Hoekstra et al., 2008; Sucksmith et al., 2013), any effects of sex and age on the mean test scores were regressed out prior to factor analysis. This enabled the comparison of the factor structure of empathy without the confound of sex differences on the mean. Furthermore, the standardisation of the items allowed for any differences in variance between the items of the EQ and the Eyes and KDEF tasks to be accounted for (as standardisation resulted in all variables having a mean of 0 and a variance of 1).

Analytic strategy

Previous research has shown that the EQ can be split into three factors: cognitive empathy, emotional empathy and social skills (Lawrence et al., 2004; Muncer & Ling, 2006). Although finding a comparable factor structure, these two papers showed differences in the number of items loading onto each latent factor. Allison and others (2011) have also explored the EQ in depth, highlighting a single dimension using Rasch analysis. The first stage of our analyses focused on determining the most appropriate factor structure for the EQ in the current sample.

Confirmatory factor analyses (CFA) were conducted in Mplus Version 7 using the maximum likelihood estimator (Muthén & Muthén, 2012a). Confirmatory models allow for a more direct test of previous models of empathy as well as greater control over model specification. The first set of analyses assessed the fit of a one-factor 26-item model (following the model identified by Allison et al., (2011)) across i) individuals with autism ii) parents and iii) general population controls (Models 1 to 3). Following this, three-factor models assessing cognitive empathy, emotional empathy and social skills were estimated based on Lawrence et al.'s (2004) 28-item model (Models 4 to 6) and Muncer et al.'s (2006) 15-item model (Models 7 to 9). The best fitting model identified in each of the three groups separately was then subjected to multiple group analysis to determine whether the same latent structure holds across individuals with autism, parents and controls (Models 10 to 11). In all subsequent analyses, the model that best described the EQ data across all three groups, a three-factor model including factors assessing cognitive empathy, emotional empathy and social skills (see results section) was utilised.

Following the analysis of the EQ alone, the study of the latent structure of empathy was extended by also including the AQ, Eyes and KDEF measures in the factor analysis. The AQ was not submitted to rigorous individual investigation as it has previously been studied extensively (Baron-Cohen et al., 2001b; Hoekstra et al., 2008; Horwitz, Sytema, Ketelaars, & Wiersma, 2005; Stewart & Austin, 2009; Wakabayashi, Baron-Cohen, Wheelwright, & Tojo, 2006; Wakabayashi, Tojo, Baron-Cohen, & Wheelwright, 2004; Woodbury-Smith et al., 2005).

First, a series of three-factor models (with latent factors Cognitive empathy, Emotional empathy and Social skills) were tested. The social interaction factor of the AQ was predicted to load on the Social skills factor due to the similarity between the content of the AQ items and the EQ items loading on this factor. The Eyes and the KDEF scores were expected to load onto the Cognitive empathy factor of the EQ as these two performance tests are thought to measure cognitive empathy (Models 12 to 14).

Secondly a series of four-factor models were estimated, in which the Eyes and KDEF scores loaded on to a separate fourth measurement factor representing performance-based assessment of empathy, rather than on the Cognitive empathy factor (Models 15 to 17). Multiple group models were used to determine whether the same structure was present amongst individuals with autism, parents and general population controls. The first multigroup CFA allowed all parameters to vary across the three groups (Model 18). A further model constraining the factor loadings to be equal across groups was also tested (Model 19).

In order to evaluate the possible impact of sex differences on the latent structure of empathy, three further models incorporating six groups based on genetic vulnerability (ASC, vs. parents vs. controls) and sex (males vs. females) were assessed using multigroup CFA (Models 20 to 22). As before, these models were run using test scores corrected for any mean sex (and age) differences, to ensure that these models focused on possible sex differences in *latent structure*, rather than sex differences in mean test scores. A number of fathers in the data set had missing data on the performance-based tasks (n = 104). In order to account for the effect of this missing data on the results, all six-group analyses were run both by imputing the data using the maximum likelihood estimator for these individuals as well as excluding these individuals for comparison.

Model fit was evaluated using the Bayesian information criterion (BIC; Schwarz, 1978), sample size adjusted BIC (SSABIC; Sclove, 1987), Akaike information criterion (AIC; Akaike, 1987), Tucker-Lewis index (TLI; Tucker & Lewis, 1973), Comparative fit index (CFI; Bentler, 1987) and the Root mean square error of approximation (RMSEA; Steiger & Lind, 1980). The BIC, SSABIC and AIC are used to assess model fit, with lower values reflective of a more parsimonious model. TLI and CFI compare the model under investigation with the null model, with CFI and TLI values >=0.95 indicating vey good fit and values >=0.90 representing adequate fit (Brown, 2006; Hu & Bentler, 1999). The RMSEA is a fit index that allows for modelling with large sample sizes. RMSEA values <0.08 indicate adequate fit, with values <0.05 suggesting excellent fit (Browne & Cudeck, 1993). Evaluation of model fit also included the interpretability of all other parameter estimates. Comparison of the nested models was based on chi-square difference tests. These have been shown to result in less type one error when the maximum likelihood estimator is implemented (French & Holmes Finch, 2006).

Results

Factor analyses of empathy as assessed by items of the Empathy Quotient

Model fit indices ascertained from the CFA models are given in Table 1. The model describing a one-factor solution of the EQ data, following Alison et al.'s (2011) model, displayed poor fit in all three groups (Models 1 to 3). Similarly, fit indices based on Lawrence et al.'s (2004) three-factor model of the EQ were below recommended thresholds (Models 4 to 6). The three-factor model of the EQ based on Muncer et al. (2006) provided the best fit to the data (Models 7 to 9). Multigroup CFA analyses indicated that this model displayed good fit across individuals with autism, parents and general population controls (Model 10). A model in which the factor loadings were constrained to be equal across the three groups (Model 11) resulted in a significantly poorer fit compared with Model 10 ($\Delta\chi^2 = 114.06$, $p < 0.001$). These findings suggest that the EQ assesses three constructs (Cognitive empathy, Emotional empathy and Social skills) in controls, parents and adults on the autism spectrum, but the association between each of these latent constructs is somewhat different across the three groups. For example, the factor correlations in the ASC group were higher than controls. This may account for why Model 11 did not provide a good fit to the data.

Factor analyses including both behavioural and performance-based measures of empathy

Next, the Eyes and KDEF tasks and the social interaction factor of the AQ were included in Muncer et al.'s (2006) three-factor model of empathy. Firstly, it was tested whether the performance-based tasks solely assess cognitive empathy, by including these two variables in the Cognitive empathy factor (Models 12 to 14). In these models, factor loadings of the Eyes and KDEF on the cognitive empathy factor were not statistically significant. This poor fit was also reflected in some of the fit indices, with CFI and TLI values below the recommended threshold in the ASC group. Secondly, a model in which the KDEF and Eyes data loaded onto a separate 'performance-based test factor' was implemented. This four-factor model (including factors Cognitive empathy, Emotional empathy, Social skills and a Performance-based factor) provided a good fit to the data in all individual groups (Models 15 to 17) as well as within the multigroup analysis (Model 18). Again, the model in which the factor loadings were constrained to be equal across the three groups (Model 19) resulted in a significantly poorer fit compared to the freely estimated model (Model 18) ($\Delta\chi^2 = 223.49$, $p < 0.001$).

Assessing sex differences in the factor structure of empathy

Lastly, the impact of sex on the factor structure of empathy was explored by running six-group analyses for the best fitting model identified, the four-factor model including factors Cognitive empathy, Emotional empathy, Social skills and a Performance-based test factor. As the effects on the mean test scores were regressed out prior to analysis, these models (Models 20 to 22) focus on sex differences in factor structure rather than in mean scores. In order to account for the effect of missing data on the performance-based tasks, all six-group analyses were also run both by imputing data for and excluding these individuals. There were no substantive changes in any of the analyses, indicating that this is not a confounding factor in the interpretation of the results. Model 20 with all estimates free to vary provided the best fit to the data for the six groups. However, comparison with the best fitting three-group multigroup model (Model 18) indicates that there is no significant difference between the latent structure of empathy when sex is taken into account. Therefore, Model 18, the four-factor model with equal form amongst the three groups allowing the factor loadings to vary provided the best fit to the data.

Empathy factor means and correlations in individuals with ASC, parents and controls

Parameter estimates for the four-factor model taken from the Model 18 analysis are given in Figure 1. All items loaded significantly onto their respective factors ($p < 0.05$). Mean differences between scores on the latent factors across the three groups are given in Table 2. Parents scored significantly lower than controls on Cognitive empathy, Emotional empathy and Social skills latent factors as well as on the performance-based tasks. Individuals with autism also scored significantly lower than controls across all four latent factor means. There was a significant difference between parents and individuals with ASC on Cognitive empathy, Emotional empathy and Social skills factors. However, these two groups scored similarly on the performance tasks (Mean difference = -0.30, $p > 0.05$). Please note that the current paper focused on latent factor means. Group differences on means of the different tasks under study have been reported elsewhere (Baron-Cohen & Wheelwright, 2004; Baron-Cohen et al., 2001a; Baron-Cohen et al., 2001b; Hoekstra et al., 2008; Sucksmith et al., 2013; Sucksmith et al., 2011; Wheelwright et al., 2006).

The correlation between the Performance-based test factor and the other empathy factors varied by sample group (see Table 3). In the control group the performance-based tasks were not significantly correlated with any other empathy factor. However, in both the parent and ASC group these tasks were significantly correlated with Cognitive empathy, Emotional empathy and Social skills. These correlations were of similar magnitude for all factors. To verify that these different correlation patterns between the performance-based test factor and the other empathy factors in the groups could not be explained by differences in score distributions on the performance-based tasks, the distributions of the KDEF and Eyes tasks were inspected. Both tasks showed very similar distributions in the control and parent groups. The differences in the correlation patterns are therefore unlikely to be due to differences in the test score distributions.

Discussion

Factor analyses in data from a large sample of individuals with ASC, parents and controls, using both questionnaire and performance-based measures of empathy, suggested a four-factor latent structure of empathy encompassing Cognitive empathy, Emotional empathy, Social skills and a Performance-based measurement factor. This structure was consistent across individuals deemed to have a high (individuals on the autism spectrum), medium (parents) or low (controls) genetic vulnerability for autism, indicating that the overall latent structure of empathy is consistent across both clinical and general population samples. However, there were some differences in the factor loadings and factor correlations across the three groups.

The latent structure identified in this study is consistent with previous research in that it identifies both a cognitive and emotional component of empathy (Davis, 1996; Dziobek et al., 2008; Eisenberg & Strayer, 1987; Smith, 2006). In addition, the analyses also identified a separate Social skills factor. Items measured by the Social skills factor of the EQ assess specific empathising skills within a social situation. For example, 'I find it hard to know what to do in a social situation' and 'I often find it difficult to judge if something is rude or polite'. Future research utilising other measures is needed to further assess the theoretical implications of this Social skills factor, which is shown to be separate from cognitive and emotional empathy.

It was expected that the performance-based emotion recognition tasks would be related to the Cognitive empathy factor. However, factor loadings of the Eyes and KDEF on the Cognitive empathy factor were low, with a model including a separate performance-based task component providing a better fit. Interestingly, the relationship between the Performance-based test factor and the other empathy factors was different across the three groups under study. In the control group, the performance tasks were not significantly correlated with any of the questionnaire-based empathy factors. Within parents and individuals with autism the performance measures were related in almost equal magnitude to all three components, rather than solely to cognitive empathy. The finding that these performance tasks do not directly and exclusively assess cognitive empathy is new. Previous research has operated on the assumption that these tasks are performance-based measures of cognitive empathy. The findings of the current study indicate that rather than being a direct measure of cognitive empathy, scores on performance-based tasks like the Eyes and the KDEF have a bearing on empathy more widely. Our results suggest that completion of either of these tasks requires engagement of more than just cognitive empathy abilities. Rather, impairment on these performance-based tasks is indicative of a broader impairment across all facets of empathy. This has important implications for future research involving the implementation of such tasks.

Individuals with autism showed greater impairment (as indexed by lower mean latent factor scores) across the Cognitive and Emotional empathy, Social skills and Performance-based empathy factors compared with controls. Similarly, the ASC group displayed greater impairment than parents across all factor means, with the exception of the Performance-based factor. This fits with the notion of autism as a disorder of empathy (Decety & Moriguchi, 2007; Gillberg, 1992; Wing et al., 2011). In contrast with some previous research (Dziobek et al., 2008; Markram et al., 2007; Smith, 2006, 2009), there was no evidence that individuals with autism exhibited intact or heightened emotional empathy.

Parents also showed mild impairment across all four factors compared to controls. However, with the exception of the Performance-based factor, impairment was not as strong as observed in the ASC group, placing their difficulties somewhere in between the clinical and the control group. This is consistent with previous accounts indicating that first-degree relatives show some difficulties on tasks of empathy (Sucksmith et al., 2013) compared with controls. Moreover, it fits with the notion that characteristics related to autism are distributed as quantitative traits rather than discrete entities (Baron-Cohen et al., 2001b; Constantino, 2011) and are likely to be influenced at least in part by common genetic variation (Abrahams & Geschwind, 2008).

Limitations

As mentioned previously, a number of fathers had missing data on the Eyes and KDEF tasks (n = 104). To assess whether these missing data had any effect on the results, all analyses were run both by imputing data for these individuals as well as excluding the missing cases. As there were no substantive changes within any of the models, it is highly unlikely that these missing data were a confounding factor.

The parent group also consisted of a larger proportion of mothers (n = 298) than fathers (n = 141). To ensure these differences would not bias the analyses, any sex effects on the means were regressed out prior to conducting the factor analyses. Moreover, the evaluation of sex differences in the latent factor structure indicated that it was similar across both sexes. Future studies including very large sample sizes would be of interest, as these could explore any possible sex differences in the latent factor structure in more detail than the current sample size permitted.

Lastly, the control group included in this study had completed a somewhat higher level of education than the parent and ASC groups. We can therefore not exclude the possibility that differences in educational level may explain some of the differences in factor structure of empathy observed between controls and the parent and ASC groups.

Conclusions

The current study assessed the latent structure of empathy across individuals with a low, medium and high genetic vulnerability to autism. Results highlighted that empathy shows evidence of multidimensionality, in which four factors can be distinguished irrespective of genetic vulnerability, including three components of empathy and a Performance-based factor. Unexpectedly, performance-based measures of empathy were related in almost equal magnitude to Cognitive empathy, Emotional empathy and Social skills, rather than solely to Cognitive empathy. This has implications for the nature of impairment indicated by performance on such tasks, suggesting that these effects are much wider than impairments in cognitive empathy alone. Individuals with autism displayed impairment on all four components of empathy, confirming the notion that autism is characterised by difficulties with multiple facets of empathy. Parents showed intermediate impairments of empathy, providing evidence for the BAP and highlighting the importance of assessing the characteristics of autism on quantitative scales.

Table 1 Fit indices and model comparisons

Model	Description	Fit indices							
		AIC	BIC	SSABIC	RMSEA	CFI	TLI	χ^2	$\Delta\chi^2$ (*df*)
One factor models EQ items (Allison et al., 2011)									
1	1f control group (n = 232)	13673.951	13942.797	13695.578	0.090	0.673	0.644	865.22**	
2	1f parent group (n = 439)	25650.600	25969.013	25721.480	0.083	0.833	0.818	1193.88**	
3	1f autism group (n = 363)	22451.458	22755.222	22507.762	0.087	0.748	0.726	1123.51**	
Three factor models EQ items (Lawrence et al., 2004)									
4	3f control group (n = 232)	14020.059	14319.925	14044.181	0.078	0.781	0.761	833.15**	
5	3f parent group (n = 439)	26186.672	26541.825	26265.731	0.071	0.883	0.873	1114.31**	
6	3f autism group (n = 363)	23768.627	24107.440	23831.427	0.080	0.802	0.785	1163.03**	
Three factor models EQ items (Muncer et al., 2006)									
7	3f control group (n = 232)	7731.634	7897.077	7744.943	0.048	0.942	0.930	132.71**	
8	3f parent group (n = 439)	14798.686	14994.632	14842.304	0.060	0.950	0.939	221.95**	
9	3f autism group (n = 363)	13040.850	13227.781	13075.499	0.055	0.932	0.918	184.27**	
Three factor multigroup models EQ items (Muncer et al., 2006)									
10	3f multigroup all estimates vary (n = 1034)	35574.089	36166.916	35785.782	0.056	0.938	0.931	589.85**	
11	3f multigroup equal factor loadings (n = 1034)	35628.149	36072.769	35786.918	0.060	0.921	0.921	703.91**	114.06 (30) *p*<0.01
Three factor model of cognitive empathy (EQ subscale, Eyes and KDEF), emotional empathy (EQ subscale) and social skills (EQ subscale and AQ_soc)									
12	3f control group (n = 232)	8331.507	8527.971	8347.312	0.044	0.945	0.936	1202.43**	
13	3f parent group (n = 439)	16721.291	16954.108	16773.218	0.055	0.947	0.938	3463.60**	
14	3f autism group (n = 363)	19949.121	20171.102	19990.266	0.067	0.894	0.877	2162.73**	
Four factor model of cognitive empathy (EQ subscale), emotional empathy (EQ subscale) social skills (EQ subscale and AQ_soc) and performance-based empathy (Eyes and KDEF)									
15	4f control group (n = 232)	8323.897	8530.701	8340.533	0.040	0.955	0.946	176.40**	
16	4f parent group (n = 439)	16712.141	16957.074	16766.664	0.055	0.949	0.939	298.78**	
17	4f autism group (n = 363)	15687.606	15921.270	15730.916	0.053	0.934	0.922	262.45**	

Four factor multigroup models of cognitive empathy (EQ subscale), emotional empathy (EQ subscale) social skills (EQ subscale and AQ_soc) and performance-based empathy (Eyes and KDEF)									
18	4f multigroup all estimates vary (n = 1034)	40743.301	41494.361	41011.591	0.052	0.938	0.932	808.00**	
19	4f multigroup equal factor loadings (n = 1034)	40894.787	41467.965	41099.535	0.061	0.909	0.907	1031.49**	223.49 (36) $p<0.01$
Four factor multigroup models specifying sex effects (6 groups)									
20	4f multigroup all estimates vary (n = 1034)	40757.204	42190.149	41269.074	0.058	0.923	0.916	1332.47**	
21	4f multigroup equal factor loadings and equal variance estimates (n = 1034)	40873.332	41861.570	41226.345	0.066	0.891	0.893	1628.59**	296.12 (90) $p<0.01$
22	4f multigroup equal factor loadings and free variance (n = 1034)	40745.441	41832.503	41133.757	0.059	0.914	0.914	1460.70**	128.23 (70) $p<0.01$

** $p<0.01$

Note. AIC, Akaike information criteria; AQ_soc, social interaction factor of the Autism Quotient; BIC, Bayesian information criteria; CFI, Comparative fit index; EQ; Empathy Quotient; Eyes, Reading the Mind in the Eyes task; KDEF, Karolinska Directed Emotional Faces; RMSEA, Root mean square error of approximation; SSABIC, sample size adjusted BIC; TLI, Tucker-Lewis index; χ^2 = chi square statistic; $\Delta\chi^2$ (*df*) =chi square difference test.

Table 2 Mean differences on factors scores in the multigroup confirmatory factor model

	Cognitive empathy	Emotional empathy	Social skills	Cognitive tests
	Mean (95% CI)	Mean (95% CI)	Mean (95% CI)	Mean (95% CI)
Control group (n = 232)	-0.04 (-0.16 to 0.08)	-0.06 (-0.16 to 0.04)	-0.03 (-0.15 to 0.10)	0.01 (-0.11 to 0.13)
Parent group (n = 439)	-0.36** (-0.51 to -0.22)	-0.27* (-0.44 to -0.10)	-0.38** (-0.52 to -0.24)	-0.29* (-0.55 to -0.04)
Autism group (n = 363)	-2.01** (-2.25 to -1.78)	-1.21** (-1.42 to -0.99)	-2.76** (-3.05 to -2.46)	-0.59** (-0.74 to -0.44)

* $p < 0.05$; ** $p < 0.01$ significantly different to controls

Table 3 Correlation between the cognitive test factor and the other components of empathy

	Cognitive empathy	Emotional empathy	Social skills
	r (95% CI)	*r* (95% CI)	*r* (95% CI)
Control group (n = 232)	0.15 (-0.10 to 0.39)	-0.04 (-0.23 to 0.17)	0.20 (-0.10 to 0.49)
Parent group (n = 439)	0.48** (0.27 to 0.70)	0.49** (0.25 to 0.74)	0.46** (0.25 to 0.68)
Autism group (n = 363)	0.32** (0.20 to 0.45)	0.32** (0.18 to 0.45)	0.32** (0.20 to 0.44)

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

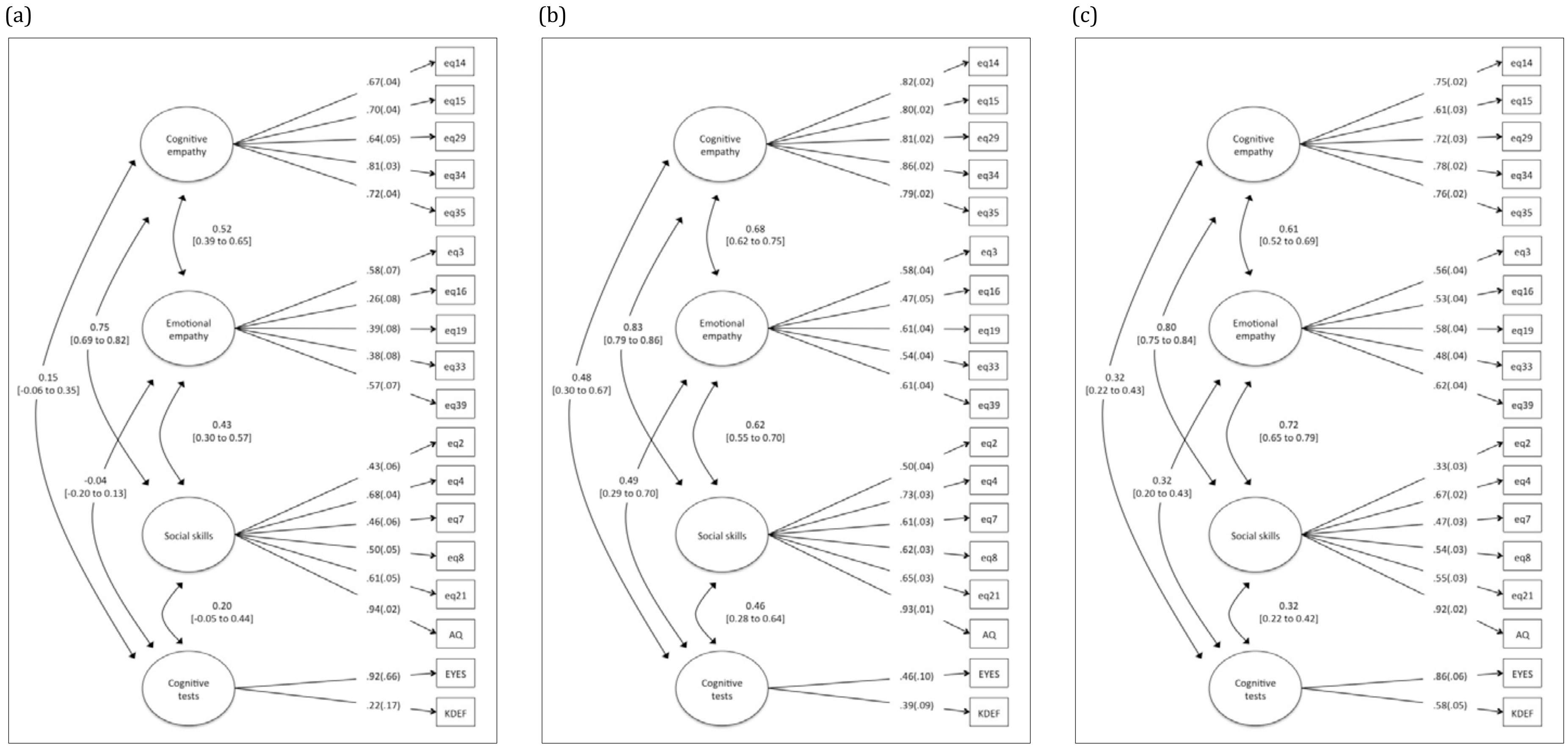

Figure 1. Four factor multigroup model for (a) controls, (b) parents and (c) ASC group

Chapter 5

The motivation behind special interests in individuals with autism and controls: Development and validation of the special interest motivation scale (SIMS)

Under review at *Autism Research*

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Abstract

Background: Clinical observations and anecdotal evidence suggests that individuals with autism are highly motivated to engage in special interests from infancy through to adulthood. Previous research assessing special interests has focused on children with autism spectrum conditions (ASC). It is therefore important to understand the significance of special interests for adults with ASC. This paper aims to systematically explore the motivations behind engagement in special interests, and whether these differ in individuals with autism, first-degree relatives and general population controls.

Methods: The Special Interest Motivation Scale (SIMS) was developed to assess motivation to engage in special interests. The internal structure of this scale was evaluated using factor analysis and mean scores on the SIMS factors were subsequently compared across individuals with autism, parents and general population controls.

Results: Factor analysis revealed a 20-item SIMS containing five factors assessing Personal life values and goals; Intrinsic interest and knowledge; Prestige; Engagement and "flow" and Achievement. Individuals with autism were more motivated by Intrinsic interest and knowledge and by Engagement and flow than controls.

Conclusions: The 20-item SIMS is a quick to administer measure that provides a reliable description of motivation to engage in special interests. This study indicates that individuals with ASC are highly motivated to engage in their special interest, and are more motivated than controls by intrinsic motivational factors, some of which are associated with positive affect. This has implications for research and clinical practice.

Introduction

Autism spectrum conditions (ASC) are characterised by impairment in social interaction and communication and repetitive behaviour and restricted interests (RRBI) (American Psychiatric Association, 2013). The social communication domain consists of three criteria including impairment in social and emotional reciprocity, difficulty understanding nonverbal communication and establishing and maintaining relationships. The RRBI domain consists of four criteria assessing stereotyped movement, insistence on sameness, sensory reactivity and restricted or intense interests (American Psychiatric Association, 2013). All three social-communication criteria and two of the four RRBI criteria need to be endorsed to receive a diagnosis of autism spectrum disorder. However, there is evidence that these impairments are largely independent, suggesting that the social and non-social characteristics of autism may have distinct causal explanations at a biological, behavioural and neural level (Happé & Ronald, 2008).

While the social symptom domain of autism has been studied extensively, the non-social traits relating to the autism spectrum are less well researched. There is some evidence to suggest that the non-social symptoms of ASC are heterogeneous, consisting of three distinct factors including repetitive motor behaviours, insistence on sameness and circumscribed interests (Lam, Bodfish, & Piven, 2008; Smith et al., 2009). Furthermore, it has been proposed that circumscribed or special interests are qualitatively different from repetitive behaviours (Jordan & Caldwell-Harris, 2012). Given that special interests appear to be a somewhat independent factor of the RRBI symptom domain, systematic assessment of special interests will provide vital information for understanding autism.

Special interests were first described in Kanner's seminal paper in the 1940s (Kanner, 1943), noting that special interests that dominated daily activities and caused high levels of preoccupation were present in an individual case study. Since this time, special interests have been recognised as being common across individuals with autism, with estimates of approximately 75-90% developing one or more special interests early in life (Klin, Danovitch, Merz, & Volkmar, 2007). Previous research indicates that individuals with autism have special interests that differ in subject matter compared with their typically developing peers (Caldwell-Harris & Jordan, 2014). Common interests involve mechanical systems, vehicles, dinosaurs, animals, factual information, timetables, technology and numbers (Anthony et al., 2013; South et al., 2005). This fits with Baron-Cohen's (2010a) notion that individuals with autism have a particular interest in systems and that individuals with autism seek to understand the underlying rules of these systems, thus engaging in 'systemising'.

Results from some previous studies suggest that special interests are associated with increased functional impairment in individuals with ASC (Turner-Brown et al., 2011). For example, special interests have been shown to be predictive of difficulties with social interaction and communication amongst a clinical group of children and adolescents with autism (Klin et al., 2007). Others have also argued that the persistence of special interests is problematic, and that they can be quite resistant to change (Mercier, Mottron, & Belleville, 2000). However, there is also research indicating that special interests can have a positive impact on individuals with ASC. Winter-Messiers (2007) reports that special interests are associated with self-confidence in individuals with autism. Mercier and others (2000) also argue that individuals with ASC and their families see their special interests as an area of great strength and skill. Moreover, there is evidence to suggest that special interests can increase socialisation and peer interaction when incorporated into treatment programs (Boyd, Conroy, Mancil, Nakao,

& Alter, 2007; Koegel, Vernon, Koegel, Koegel, & Paullin, 2012; Koegel, Kim, Koegel, & Schwartzman, 2013). This highlights the number of positive aspects associated with special interests for individuals with autism.

The majority of research into special interests in ASC has been conducted with children. Thus, the positive or negative aspects of special interests have mostly been measured via parent report. However, it may be that special interests are of significant importance to individuals with autism. Based on clinical observations and anecdotal evidence, it appears that individuals with ASC are highly motivated to engage in special interests through to adulthood. Characterising this motivation to engage in special interests provides an opportunity for understanding part of the non-social domain associated with the autism spectrum. There is also evidence to suggest that first-degree relatives display subthreshold levels of autistic traits, or the broader autism phenotype (BAP; Piven et al., 1997b; Sucksmith et al., 2011). It is therefore important to assess motivation and special interests across samples containing varying levels of genetic risk for autism. This paper aims to systematically explore the motivations behind engagement in special interests, and whether these motivations differ in individuals with autism, first-degree relatives (parents) and general population controls.

Development of the Special Interest Motivation Scale

Self-determination theory posits that behaviour can be intrinsically motivated, extrinsically motivated or amotivated (Deci & Ryan, 1985; Deci & Ryan, 2002). Intrinsic motivation describes motivation derived from the pleasure and satisfaction that occurs through engaging in an activity (Deci, 1975). This engagement is not dependent on external rewards or reinforcement but purely on an individual's enjoyment of the task (Deci & Ryan, 1985). It has been proposed that intrinsic motivation can be split into three specific goals or motives including 'to know', 'to accomplish' and 'to experience stimulation’ (Vallerand et al., 1992). The first motive ‘to know’ encompasses gaining satisfaction or pleasure from learning or understanding something new (Pelletier et al., 1995). ‘To accomplish’ describes engaging in an activity to derive a feeling of satisfaction or mastery (Pelletier et al., 1995). Finally, ‘to experience stimulation’ involves engaging in an activity in order to experience stimulating sensations or excitement (Pelletier et al., 1995).

In contrast to intrinsic motivation, which describes behaviours purely focusing on satisfaction or experience, extrinsic motivation describes engagement that is contingent on external factors or rewards (Ryan, Connell, & Grolnick, 1990). Three different aspects of extrinsic motivation have been described, including ‘external regulation’, ‘introjection’ and ‘identification’. External regulation refers to behaviour that is motivated by the expectation of external rewards or praise from others (Deci & Ryan, 1985).
‘Introjection’ applies to motivation that no longer requires the source of the external motivation to be present: an individual will engage in the behaviour having internalised the need for external recognition (Pelletier et al., 1995). For example, an individual may engage in a behaviour motivated by feelings of guilt or anxiety originally evoked by external factors. By contrast, identification relates to behaviour that is judged to be important and therefore performed by choice (Pelletier et al., 1995). However, the activity is still performed for extrinsic reasons, for example, in order to achieve an external goal. Finally, amotivation describes behaviour that is neither intrinsically nor extrinsically motivated. Individuals that are amotivated find it difficult to identify any reasons why they should continue to pursue an activity, and often give up (Pelletier et al., 1995). These three facets of motivation, and their subtypes, are all important in understanding motivation to engage in special interests.

The Special Interests Motivation Scale (SIMS) forms part of a comprehensive survey of special interests conducted by Roth and colleagues (Roth, Roelfsema, & Hoekstra, 2013). The SIMS was developed by Hoekstra and Roth based on the Sports Motivation Scale (Mallet, Kawabata, Newcombe, Otero-Forero, & Jackson, 2007; Pelletier et al., 1995), the Motivation at Work Scale (Gagné et al., 2010) and the Academic Motivation Scale (Fairchild, Horst, Finney, & Barron, 2005; Vallerand et al., 1992). The Sports Motivation Scale (Mallet et al., 2007; Pelletier et al., 1995) is a 24-item measure assessing seven factors related to intrinsic motivation, extrinsic motivation and amotivation. It consists of three factors relating to intrinsic motivation 'to know', 'to accomplish', 'to experience stimulation', three factors relating to extrinsic motivation 'external regulation', 'introjection' and 'identification' and one factor relating to amotivation.

The SIMS was developed to follow this same structure. Four items for each intrinsic and extrinsic motivation factor and two items assessing amotivation were included. Three items were taken from the Motivation at Work Scale (Gagné et al., 2010) and converted to reflect special interests. For example, 'I chose this job because it allows me to reach my life goals' was converted to 'I chose this special interest because it allows me to reach my life goals'. Twelve items were also directly converted from the Sports Motivation Scale (Mallet et al., 2007; Pelletier et al., 1995). A number of other items were also included, based on the definitions of the relevant motivational facets measured. Examples include, 'Because I enjoy broadening my knowledge about my special interest' assessing intrinsic motivation 'to know', 'Because when I do well at my special interest I feel important' assessing extrinsic motivation 'introjected' and 'I can't really give any good reason for doing my special interest' reflecting amotivation.

Altogether, the SIMS comprised 26 items, assessing a range of motivations to engage in special interests. A large sample of participants, spanning individuals with ASC, parents of a child with autism and general population controls were asked to complete a survey that included the SIMS. This paper aims to validate the SIMS as well as to assess the motivation behind special interests in individuals with autism, parents and controls.

Methods

Participants and measures

Participants included individuals with autism, parents of a child with autism and general population controls. Two modes of recruitment were utilised. Firstly, students of the Open University in the United Kingdom taking a range of first and second level modules spanning arts, health and science topics were approached via email to participate in the study. The Open University is a distance learning university and is therefore attractive to individuals who have carer responsibilities or have special needs themselves. Therefore, although most students fell into the control group, the sample also included some parents of children with ASC and some individuals with autism. Secondly, in order to increase numbers in the ASC and parent groups, registered research volunteers at the Autism Research Centre at Cambridge University received an invitation via email to take part in the study, including a link to the online survey. All participants completed the survey online, hosted at the Open University's Biomedical Online Research Network (www.open.ac.uk/born).

Only individuals with complete data on all items of the SIMS were included in the analyses (n = 610). The ASC group consisted of 158 individuals (Males = 86, Females = 72, Mean age = 41, sd = 13) who had received a formal diagnosis of autism made by a qualified clinician. Individuals who reported a self-diagnosis of autism were excluded from the sample. The parent group comprised 185 individuals (Males = 35, Females = 150, Mean age = 44, sd = 7) who reported having a child with a clinical diagnosis of autism but no diagnosis themselves. The control group consisted of 267 individuals (Males = 193, Females = 74, Mean age = 42, sd = 15). The control group was restricted to individuals who reported no previous psychiatric history. Controls who scored high on the Autism Spectrum Quotient (AQ; Baron-Cohen et al., 2001b) were excluded. The parent group was significantly older than the ASC group ($p < 0.001$). 64% of the ASC group had completed education above high school level, along with 69% and 52% of parents and controls respectively. There was a significant difference in education level between the parent and control groups ($p < 0.05$), most likely due to sampling methods.

Participants were administered an online version of the SIMS outlined above. The SIMS questionnaire was part of a larger set of questions, together comprising a comprehensive survey of special interests (Roth et al., 2013). Prior to completing the SIMS items, participants were asked to describe their 'most important' special interest, given that they may have more than one. Participants were asked to describe their special interest in 25 to 50 words, including an explanation of why they enjoy it. After this description and some other questions relating to their most important special interest, the participants were presented with the SIMS. The SIMS items were scored on a 7-point Likert scale assessing how well each statement describes why individuals engage in their special interest. The scale ranged from 'not at all' through to 'exactly', with 'moderately' as a midpoint. All items were summed, with higher scores reflecting increased motivation to engage in special interests.

The Autism Spectrum Quotient (AQ; Baron-Cohen et al., 2001b) was also administered in order to assess quantitative autistic traits including communication, imagination, social skills, attention to detail and attention switching. The AQ is a 50-item self-report measure rated on a 4-point scale with response options 'definitely agree', 'slightly agree', 'definitely disagree' and 'slightly disagree'. Hoekstra and others (2008) outline a raw scoring method that was implemented in the current study. Scores range from 50 to 200, with higher scores indicative of the presence of more autistic traits. Previous research has split the AQ into a broad social interaction factor and an attention to detail subscale (Hoekstra et al., 2008). These two subscales were used in all analyses.

Analytic strategy

Exploratory factor analyses (EFA) were conducted on the 26 items of the SIMS in order to evaluate the factor structure of the measure. Model fit indices including the Akaike information criterion (AIC) (Akaike, 1987), Bayesian information criterion (BIC) (Schwarz, 1978), Sample size adjusted BIC (SSABIC) (Sclove, 1987), Comparative fit index (CFI) (Bentler, 1987), Tucker-Lewis index (Tucker & Lewis, 1973) and the Root mean square error of approximation (RMSEA) (Steiger & Lind, 1980) were estimated. Smaller AIC, BIC and SSABIC values are indicative of better model fit. CFI and TLI values >=0.95 are indicative of very good fit to the data, with values >=0.90 indicating adequate fit (Brown, 2006; Hu & Bentler, 1999). RMSEA values <=0.08 are indicative of good fit, with values <=0.05 indicating excellent fit to the data (Browne & Cudeck, 1993).

The overall fit of the EFA models was evaluated by the statistics outlined above. In addition, the evaluation of how many items and factors to retain in the EFA was based on a range of different methods. Decisions regarding the removal of specific items followed the procedure outlined by Costello and Osborne (2005). This procedure recommends removing items containing cross loadings >=0.32 and factors measured by less than three items. In order to determine the number of factors to retain in each EFA model, a parallel analysis was conducted (Glorfeld, 1995; Horn, 1965). Parallel analysis generates 95% confidence intervals from random sets of data with the same sample size and number of variables as the original data. Eigenvalues from the EFA models that are larger than the values produced within the parallel analysis determined the number of factors retained (Glorfeld, 1995). Lastly, the interpretability of the various models was also taken into account when deciding which model provided the best description of the data.

Following scale development with EFA models, the factor structure of the SIMS was confirmed via confirmatory factor analysis (CFA), allowing for greater model specification and comparison of specific group differences. Furthermore, in order for mean comparisons on the SIMS to be made, invariance needed to be assessed across groups. Measurement invariance (Meredith, 1993) of a scale determines whether a measure assesses the same construct in a consistent way across different populations. Invariance was assessed by fitting a series of models that placed increasing levels of restrictions on the parameter estimates across each group to determine whether the SIMS is invariant for individuals with autism, parents and controls.

Analyses were estimated in Mplus version 7 (Muthén & Muthén, 2012a). Once the best fitting model was identified, further analyses were implemented in SPSS 21 (IBM Corp, 2012). Cronbach's alpha (1951) was calculated to estimate the reliability of the SIMS and any associated factors. Alpha scores between 0.6 and 0.7 are indicative of acceptable internal consistency, with scores ranging from 0.7 to 0.9 indicating good to excellent internal consistency (George & Mallery, 2003). Mean scores on the SIMS were also estimated for individuals with autism, parents and controls. The relationship between the SIMS and the social interaction and attention to detail factors of the AQ was also assessed.

Results

Exploratory factor analysis

EFA models were implemented in order to estimate the factor structure of the SIMS (see Table 1). An evaluation of the model fit indices highlighted that a model with six factors provided the best fit to the data (Model 1). Six eigenvalues in the EFA were also larger than those obtained in the parallel analysis, thus indicating a six-factor structure. However, three items in this six-factor model displayed significant cross loadings. The extrinsic motivation item "Because I would feel guilty or lazy if I didn't spend time doing my special interest" contained a loading >0.32 on three factors. Following the recommendations set by Costello and Osborne (2005), this item was dropped from the subsequent analysis. A second EFA model (Model 2) identified a six-factor structure in which two items contained cross loadings >0.32 on a number of factors. These two items "Because my special interest allows me to learn about many things that interest me" and "Because it is one of the best ways to develop myself" were therefore dropped from the third EFA model.

A further EFA was implemented including the 23 remaining items (Model 3). One item "Because people around me think it is important to engage in this activity" was the only item with substantial cross loadings on another factor. A subsequent EFA (Model 4) was therefore conducted with 22 items, revealing a six-factor model with salient cross loadings. However, factors containing less than three items are considered unstable and should be excluded from analysis (Costello & Osborne, 2005). Based on these recommendations, Factor 6, including the two items assessing amotivation was removed from the model. A final EFA model (Model 5) contained fit indices within the recommended thresholds. Results from the parallel analysis also indicated a 20-item five-factor structure provided the best fit. Moreover, this structure represented a model that is easy to interpret based on what is known from the literature on intrinsic and extrinsic motivation.

Confirmatory factor analysis

CFA models confirming the fit of the five-factor model identified in the exploratory analyses were then implemented. Results are given in Table 1. Firstly, a model assessing a five-factor structure in the total sample was estimated, indicating an adequate fit to the data (Model 6). Modification indices indicated the presence of correlated residual variances between similar items. Following the recommendations of Cole and others (2007), items that contained similar wording or meanings with large residual variances within each factor were allowed to correlate in all subsequent models. Three CFA models were estimated separately for controls (Model 7), parents (Model 8) and individuals with ASC (Model 9), all providing an adequate fit to the data.

Multiple group CFA models were then estimated in order to assess measurement invariance across the three groups (see Table 1). Firstly, a multiple group CFA was implemented, assessing the factor structure of the SIMS for all three groups concurrently, allowing all structural parameter estimates to be freely estimated for each group (Model 10). Further models with varying restrictions were then fit to the data, in which the factor loadings (Model 11), intercepts (Model 12), loadings and intercepts (Model 13) and loadings, intercepts and residual variances (Model 14) were restricted across individuals with autism, parents and controls. Chi square difference tests were computed to assess the relative fit of the more restrictive models. The factor loadings were determined to be invariant across the three groups (Model 11). However, initial examination of a model constraining the intercepts to be equal across groups was not supported by the data ($p < 0.05$). On further analysis, it was identified that one of the intercepts (item 19) did not show invariance for the ASC group. Several authors have argued that it is sufficient to obtain partial invariance when at least two indicators are shown to have equal factor loadings and intercepts across groups (Byrne, Shavelson, & Muthén, 1989; Steenkamp & Baumgartner, 1998). Given that the factor loadings of all 20 items and the intercepts of 19 items were shown to be invariant across individuals with autism, parents and controls, it was determined that it was acceptable to continue the measurement invariance analyses. The equality restraint on the intercept of item 19 in the ASC group was released in the subsequent models. There were no tenable differences between models 11 to 13, indicating that factor loadings and intercepts are invariant across the three groups (excluding item 19 for the ASC group). However, Model 14 containing equal residuals resulted in significantly poorer fit ($\Delta\chi^2 = 73.99$, $p < 0.001$).

Model 13, a five-factor model with invariant factor loadings and intercepts across individuals with autism, parents and general population controls therefore provided the best fit to the data. This indicates that the 20 items in the SIMS are interpreted in the same way by each of these three groups and allows for mean differences to be compared in subsequent analyses.

Factor structure of the SIMS

Exploratory and confirmatory methods suggested a 20-item five-factor structure for the SIMS. This is given in Figure 1. Note that items were relabelled to reflect the new 20-item scale of the SIMS (see Table 2). Factor one contained three items assessing motivation based on Personal life values and goals. Factor two consisted of three items assessing Intrinsic interest and knowledge, while factor three included four items assessing the feeling of Prestige associated with engaging in special interests. Factor four contained four items assessing Engagement and "flow", or the satisfaction experienced while completely absorbed in an activity (Csikszentmihalyi, 1990;

Csikszentmihalyi & Csikzsentmihalyi, 1988). Factor five included six items assessing motivation relating to experiencing a sense of Achievement. Cronbach's alpha estimates for each factor are presented in Table 3, indicating that all factors had good to excellent internal consistency.

Additional analyses

Mean comparisons for the SIMS are given in Table 3. Higher values are indicative of a higher level of motivation to engage in special interests. Individuals with autism scored significantly higher on Intrinsic interest and knowledge and Engagement and flow factors than the other two groups ($p < 0.01$). The control group also scored significantly higher than the parent group on Intrinsic interest and knowledge and Engagement and flow factors ($p < 0.05$). Individuals with ASC scored higher than parents but not controls on motivation due to Achievement ($p < 0.01$).

Correlations between the five factors of the SIMS are given in Table 4. Given that there were no significant differences between parents, controls and individuals with ASC, correlations are presented for the total sample. All factors were significantly correlated ($p < 0.01$). Correlations between the SIMS and the social interaction and attention to detail factors of the AQ are also given in Table 4. Similar to the correlations between the SIMS factors, all correlations between the AQ and SIMS subscales were comparable between the three groups (with overlapping confidence intervals) and are therefore presented together in Table 4, with one exception. The Prestige factor correlated significantly more strongly with the attention to detail subscale of the AQ in individuals with ASC ($r = -0.34$, $p<0.001$) compared to the other two groups. This appears to be driven by a subgroup of individuals in the ASC group with very high AQ scores (>120) who show very limited motivation due to prestige (as indicated by minimal scores on factor three). In all three groups there was a significant correlation between the Intrinsic interest and knowledge factor and the Engagement and flow factor and both subscales of the AQ, although all associations were modest (ranging between 0.11 and 0.19). There was also an association between the SIMS Achievement factor and the attention to detail factor of the AQ.

Discussion

This study presented the first systematic exploration of the motivation to engage in special interests, collected in a large sample of individuals with a clinical ASC diagnosis, parents of a child with autism, and general population controls. Results of an extensive factor analysis indicated that a 20-item version of the SIMS reliably assesses five dimensions of motivations to engage in special interests, including Personal life values and goals, Intrinsic interest and knowledge, Prestige, Engagement and "flow" and Achievement. This five-factor structure was invariant across controls, parents and ASC groups, indicating that the SIMS assesses the same construct within different populations. However, the equality restraint on the intercept of item 19 was released in the ASC group, indicating some imprecision within this factor and that this item could potentially be improved.

Factors one, two and four map onto three of the seven factors identified on the Sports Motivation Scale (Mallet et al., 2007; Pelletier et al., 1995). However, there were some differences in the factor structure of the SIMS and the original scales it was based upon. For example, factor five was found to consist of items designed to measure both extrinsic and intrinsic motivation. However, on closer evaluation, these items all appear to tap into motivation based on achievement.

Individuals with autism scored higher than controls and parents on factors assessing Intrinsic interest and knowledge and Engagement and flow, indicating that this group is more strongly motivated by intrinsic factors. The finding that individuals with ASC are particularly motivated to engage in their special interest due to sheer enjoyment and excitement (as measured by the Engagement and flow factor) is consistent with previous research highlighting that special interests are associated with feelings of enthusiasm, pride and happiness (Winter-Messiers, 2007). While there is evidence that individuals with autism have a negative self-image, engagement in special interests has been associated with a shift to a more positive sense of self (Winter-Messiers, 2007). This highlights the importance of special interests on positive outcomes for individuals with autism.

These results also broadly fit in with the systemising account of autism (Baron-Cohen, 2010a), which proposes that some of the characteristics of ASC can be explained by a strong drive to systemise; to understand, design and predict the underlying rules of systems. The finding that individuals with ASC are motivated to engage in special interests to further their knowledge and discover new aspects fits in with the notion that individuals with autism are motivated to fully understand the 'system' behind their special interest.

There were no differences between the groups on two extrinsic motivation factors assessing Personal values and goals and Prestige. Given the clinical symptoms that characterise autism, including impaired social communicative functioning, it could be expected that individuals with autism may be less motivated by extrinsic factors such as prestige than general population controls. Interestingly, this was not reflected in the results of the current study. There were no differences between the groups on both factors assessing achievement and prestige, suggesting that individuals on the autism spectrum, parents and controls are equally motivated by extrinsic factors. Furthermore, factor mean scores indicated that all three groups showed relatively higher intrinsic than extrinsic motivations. This indicates that extrinsic motivation does not appear to play a substantial role in engaging in special interests, at least not in engaging with one's most important special interest. However, the pursuit of interests that are perceived as less important may be motivated by different factors.

Results indicated that parents scored lower than both the ASC group and controls on Intrinsic interest and knowledge and Engagement and flow factors of the SIMS. Parents also scored lower than the ASC group on motivation due to Achievement. Previous research has suggested that special interests may form part of the BAP, and that relatives of individuals with autism may also display intense interests or preoccupations. For example, Smith and others (2009) highlighted that intense preoccupations in fathers were related to traits that have been associated with the BAP including rigidity and aloofness. However, in the current study parents displayed significantly lower levels of motivation to engage in their special interests due to engagement and knowledge than both the control and ASC group. In contrast to the study by Smith and others, the current study assessed the motivations behind engaging in special interests, rather than the intensity of the special interest itself. Moreover, parents of a child with autism are likely to be engaged for a significant amount of time in the care of a child with special needs, and these caring responsibilities may shape their motivations. This group may therefore not be representative of the wider BAP; for example, siblings of a child with autism, or second-degree relatives who do not have significant caring responsibilities. Further research is needed in order to fully understand the relationship between special interests and the BAP.

It has been suggested that individuals with autism engage in special interests in order to reduce anxiety or negative affect (Attwood, 2003; Spiker, Lin, Van Dyke, & Wood, 2012). Conversely, the results obtained in the current study indicate that individuals with autism are motivated to engage in special interests in order to obtain knowledge, experience engagement, flow and an overall sense of achievement. For example, item four "For the sense of sheer enjoyment I experience doing my special interest" assesses positive affect. There was no difference between the factor loading on this item between individuals with ASC and controls (0.57), indicating that enjoyment and special interests are measured in the same way across these two groups. Special interests therefore appear to be strongly related to positive affect and intrinsic engagement, rather than merely being an alleviation of negative emotion. This has implications for treatment practices and intervention strategies.

Previous research has highlighted the benefit of incorporating special interests into intervention programs for individuals with autism. For example, the inclusion of special interests in peer activities has been associated with increased socialisation, social engagement and peer interaction in adolescents (Koegel et al., 2013) and an increase in social behaviour in children with autism (Boyd et al., 2007; Koegel et al., 2012). Incorporating special interests has also been shown to increase pretend play and joint attention in children with ASC (Kryzak, Bauer, Jones, & Sturmey, 2013; Porter, 2012) and predict positive change in language, social communication, emotion regulation and motor skills (Winter-Messiers, 2007). Importantly, special interests are associated with a positive sense of self and an increase in self-confidence for individuals with autism and are vital for wellbeing (Winter-Messiers, 2007). The inclusion of special interests into case formulation and intervention programming therefore has the potential to significantly influence both behavioural and affective outcomes for individuals on the autism spectrum. As Winter-Messiers (2007) states, special interests capture the heart and mind of individuals with autism and provide a "lens through which they view the world" (p. 142). Future research evaluating the efficacy of incorporating special interests into intervention strategies is vital to improve quality of life and wellbeing for individuals with autism.

Results indicated an association between special interests and autistic traits. Motivation due to Intrinsic interest and knowledge and Engagement and flow was associated with higher levels of traits on both the social interaction and attention to detail subscales of the AQ. This suggests that the motivation to engage in special interests due to these factors is not only associated with non-social traits on the autism spectrum, but also with higher levels of social and communication difficulties. However, the associations between the AQ and the SIMS were small, indicating that while there is some association between motivation, special interests and the autism spectrum, there are substantial individual differences in the relationship between these constructs.

Limitations

The study included self-report measures of special interests and motivation and was therefore restricted to the inclusion of high functioning individuals with autism. This limits the generalisability of the results across the full autism spectrum, particularly in relation to individuals with associated intellectual disability. However, self-report measures are vital in assessing special interests in autism given the differences in how special interests are regarded by individuals and caregivers. The parent group was significantly older than the ASC group and had completed a higher level of education than controls. Future research would benefit from the inclusion of more specifically matched samples.

Conclusions

The SIMS is a 20-item scale providing a reliable assessment of five dimensions of motivations to engage in special interests, spanning personal life values and goals, intrinsic interest and knowledge, prestige, engagement and “flow” and achievement.

Individuals with ASC were more strongly motivated by intrinsic interests and knowledge as well as by a sense of engagement and flow than both controls and parents of a child with autism. This highlights that engagement in special interests is strongly related to positive affect, and not merely to a reduction of negative emotions. There was a significant relationship between autistic traits and motivation to engage in special interests, indicating that these interests are important in understanding the phenotype associated with ASC. This has significant implications for diagnosis, intervention and clinical practice. Moreover, the SIMS is a reliable measure that is quick to administer, and can thus be useful in future research or in clinical practice.

Table 1 Fit indices and model comparisons of exploratory and confirmatory factor analysis of the Special Interest Motivation Scale

Model	Description	Fit indices							
		AIC	BIC	SSABIC	RMSEA	CFI	TLI	χ^2	$\Delta\chi^2$ (*df*)
Exploratory Factor Analysis									
1	26 item SIMS								
	1f total sample (n=610)	59857.389	60201.639	59954.006	0.146	0.525	0.484	4173.101**	
	2f total sample (n=610)	58230.310	58684.896	58357.893	0.115	0.728	0.677	2496.022**	
	3f total sample (n=610)	57574.835	58135.345	57732.148	0.101	0.811	0.754	1792.548**	
	4f total sample (n=610)	57223.144	57885.163	57408.945	0.092	0.857	0.795	1394.856**	
	5f total sample (n=610)	56795.885	57555.000	57008.937	0.076	0.912	0.860	923.597**	
	6f total sample (n=610)	**56409.114**	**57260.912**	**56648.179**	**0.053**	**0.962**	**0.933**	**494.827****	
2	25 item SIMS								
	6f total sample (n=610)	54032.899	54849.389	54262.055	0.051	0.966	0.939	431.713**	
3	23 item SIMS								
	6f total sample (n=610)	49852.049	50597.924	50061.386	0.048	0.974	0.949	313.761**	
4	22 item SIMS								
	6f total sample (n=610)	47747.499	48458.066	47946.926	0.051	0.973	0.946	293.530**	
5	20 item SIMS								
	5f total sample (n=610)	43237.563	43811.313	43398.591	0.054	0.972	0.947	277.540**	
Confirmatory Factor Analysis									
6	5f total sample (n=610)	43489.975	43820.985	43582.876	0.072	0.923	0.906	639.953**	
7	5f controls (n = 267)	18759.443	19028.487	18790.693	0.072	0.923	0.905	372.391**	
8	5f parents (n = 185)	13239.568	13481.095	13243.547	0.078	0.916	0.897	330.704**	
9	5f ASC (n = 158)	11547.959	11777.654	11540.243	0.072	0.913	0.894	281.520**	

Measurement invariance analyses									
10	5f total sample (n = 610) free	43546.970	44539.999	43825.673	0.074	0.918	0.900	984.616**	
11	5f total sample (n = 610) factor loadings invariant only	43517.645	44334.135	43746.800	0.072	0.916	0.906	1035.290**	50.67 (40) $p > 0.05$
12	5f total sample (n = 610) intercepts invariant only	43521.314	44386.352	43764.095	0.072	0.918	0.905	1016.959**	32.34 (29) $p > 0.05$
13	**5f total sample (n = 610) factor loadings and intercepts invariant**	**43493.317**	**44181.817**	**43686.551**	**0.070**	**0.916**	**0.910**	**1068.963****	**83.63 (69) $p > 0.05$**
14	5f total sample (n = 610) factor loadings, intercepts and residual variances invariant	43491.494	44003.455	43635.180	0.070	0.910	0.910	1147.139**	162.52 (109) $p < 0.001$

** $p<0.01$

Note. AIC, Akaike information criteria; BIC, Bayesian information criteria; CFI, Comparative fit index; RMSEA, Root mean square error of approximation; SSABIC, Sample size adjusted BIC; TLI, Tucker-Lewis index; χ^2 = chi square statistic; $\Delta\chi^2$ (*df*) = chi square difference test. Bold text denotes best fitting model.

Table 2 Factor structure and items of the Special Interest Motivation Scale

Factor 1	**Personal life values and goals**
2	I chose this special interest because it allows me to reach my life goals.
13	Because it is a good way to learn lots of things that could be useful in other areas of my life.
17	Because my special interest fits my personal values.
Factor 2	**Intrinsic interest and knowledge**
1	Because it is satisfying to learn new things about my special interest.
8	Because I enjoy discovering new aspects about my special interest.
12	Because I enjoy broadening my knowledge about my special interest.
Factor 3	**Prestige**
3	Because it enables me to be well regarded by people I know.
7	For the prestige that comes with doing my special interest.
9	Because when I do well at my special interest I feel important.
18	To prove to others that I am good at my special interest.
Factor 4	**Engagement and "flow"**
4	For the sense of sheer enjoyment I experience doing my special interest.
10	Because I love being engaged in my special interest.
14	For the excitement I feel when I am really involved in my special interest.
19	Because I like the feeling of being totally immersed in my special interest.
Factor 5	**Achievement**
5	Because I love bettering myself at my special interest.
6	To prove to myself that I am capable of achieving something special.
11	For the sense of achievement I feel after accomplishing difficult aspects of my special interest.
15	Because I enjoy improving my special interest abilities.
16	Because I don't want to fail in pursuing my special interest.
20	Because it is satisfying to aim for excellence in my special interest.

Table 3 Mean scores on the Special Interest Motivation Scale and reliability estimates

	Values and Goals (F1)	Intrinsic (F2)	Prestige (F3)	Flow (F4)	Achievement (F5)
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)
Controls	11.4 (5.0)	15.4 (5.1)	10.9 (6.4)	22.3 (5.2)	26.6 (9.9)
Parents	11.9 (5.2)	14.4* (5.4)	10.9 (6.7)	21.3* (5.6)	25.1 (10.4)
ASC	12.1 (4.9)	16.8 ** (4.1)	11.4 (7.1)	23.6** (4.7)	27.9 (9.2)
Cronbach's alpha	0.68	0.91	0.87	0.78	0.87

* $p < 0.05$, ** $p < 0.01$ denotes significant group difference from controls

Table 4 Correlation between the five factors of the SIMS and the AQ

	Values and Goals (F1)	Intrinsic (F2)	Prestige (F3)	Flow (F4)	Achievement (F5)
F2	0.66** (0.61-0.70)				
F3	0.61** (0.56-0.66)	0.29** (0.22-0.36)			
F4	0.34** (0.27-0.41)	0.57** (0.51-0.62)	0.29** (0.22-0.36)		
F5	0.72** (0.68-0.76)	0.63** (0.58-0.68)	0.78** (0.75-0.81)	0.59** (0.54-0.64)	
AQ_soc	-0.01 (-0.09-0.07)	0.11* (0.03-0.19)	-0.02 (-0.10-0.06)	0.14* (0.06-0.22)	0.06 (-0.02-0.14)
AQ_att	0.08 (0.0-0.16)	0.19* (0.11-0.27)	0.09* (0.01-0.17)	0.17* (0.09-0.25)	0.12* (0.04-0.20)

* $p < 0.05$, ** $p < 0.01$; () = 95% confidence interval

Note. AQ_att = attention to detail factor of the Autism Spectrum Quotient; AQ_soc = social interaction factor of the Autism Spectrum Quotient.

Factor correlations did not differ between groups (with one exception, see text) and are therefore presented together.

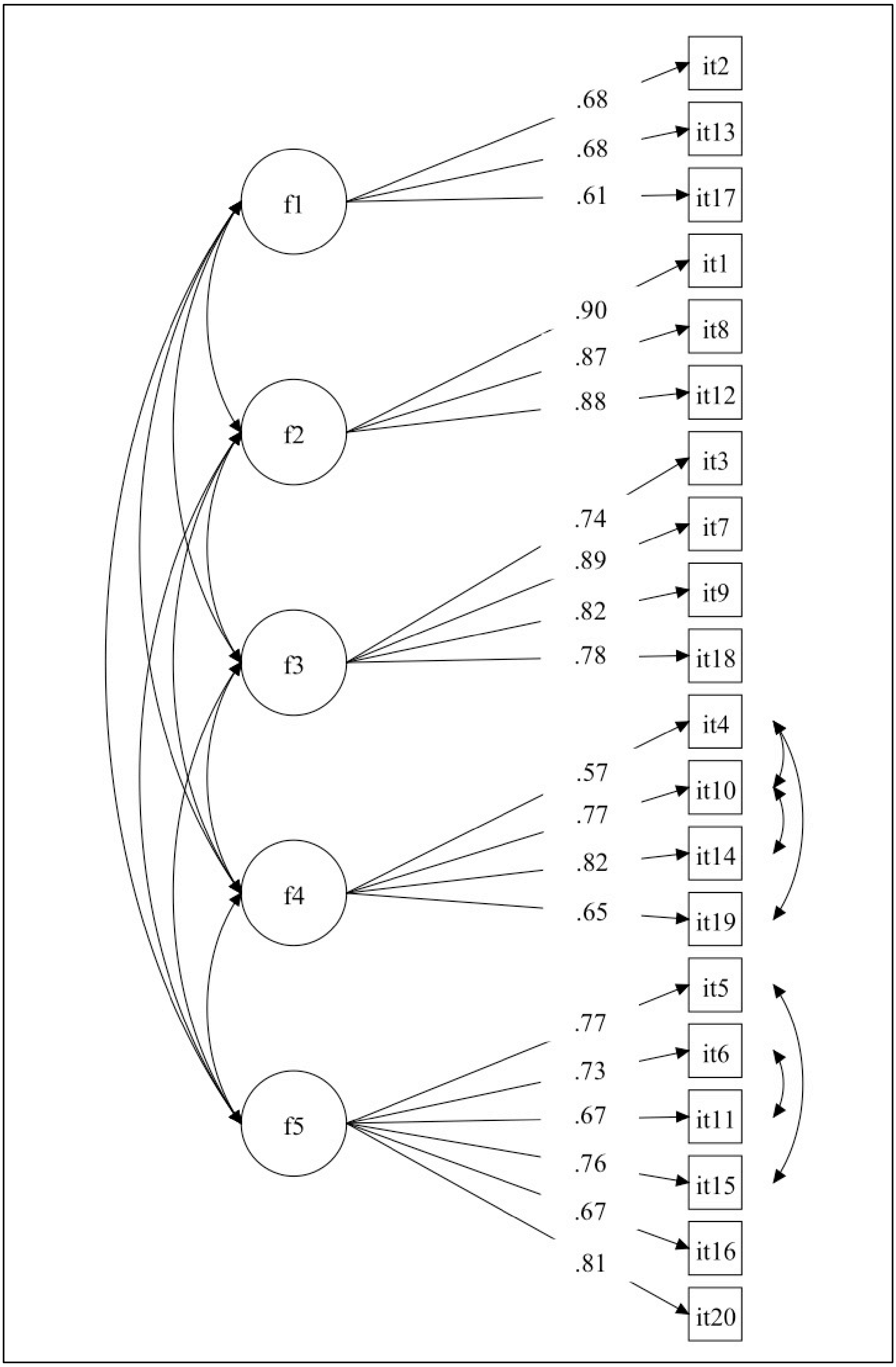

Figure 1. Factor structure of the Special Interest Motivation Scale

Chapter 6

General Discussion

Overview of findings

This thesis systematically evaluated the E-S theory of autism in order to further our understanding of the latent structure of the autism phenotype. Furthermore, it sought to determine whether the latent structure of the autism phenotype is consistent across individuals with a high (ASC), medium (parents) and low (general population controls) genetic predisposition to autism. It consisted of four chapters in the form of manuscripts prepared for publication.

The E-S theory of autism argues that the social and communication difficulties associated with ASC can be accounted for by difficulty with empathy, while the repetitive behaviours and narrow interests can be accounted for by a drive to systemise (Baron-Cohen, 2010a). Chapter two used confirmatory factor analysis to determine whether empathy and systemising traits are best conceptualised as lying on a single continuum or as multiple distinct dimensions. The latent structure was examined in three distinct groups, encompassing individuals with autism, parents and general population controls. Two correlated factors provided the best fit to the data, indicating that the social and non-social traits associated with ASC can be quantitatively measured by two dimensions representing empathy and systemising. This broad factor structure was found to be consistent for all three groups, providing support for the E-S theory of autism. Results are also consistent with previous research suggesting that the social and non-social traits of autism may be partly distinct at neural, genetic and cognitive levels (Happé & Ronald, 2008). However, the significant inverse correlation between the two factors (with strong systemising being associated with relatively poor empathy abilities) indicates that these dimensions are not completely separable. This inverse relationship was larger in the autism (r = -0.61) and parent group (r = -0.57) compared with controls (r = -0.22), indicating that the relationship between empathy and systemising increases with genetic risk. This suggests that the aetiological factors influencing behavioural traits, including genetic factors, may differ across controls, parents and individuals with autism. This highlights the quantitative nature of the autism spectrum as well as exemplifies the inherent heterogeneity of the autism phenotype.

Chapter three extended the factor analyses outlined in chapter two in order to determine whether homogenous subgroups could be identified based on varying levels of empathy, systemising and autistic traits. Results from the factor mixture modelling analyses indicated that a two-factor three-class structure provided the best fit to the data. Similar to the analyses presented in the second chapter, two dimensions reflecting empathy and systemising were identified within the total sample spanning the full range of genetic vulnerability to autism. In addition, through mixture modelling, this chapter also identified three homogenous subgroups across these dimensions containing differing levels of empathy and systemising. The first group contained individuals with high systemising and low empathy ability (Class S). The second group was defined by higher levels of empathy and a lower drive to systemise (Class E), while the third group displayed comparable scores on both domains (Class B). Most individuals with ASC displayed the Class S cognitive profile, providing support for the E-S theory of autism. A significant proportion of parents (20%), particularly fathers, also fell into this class, providing evidence for the BAP.

Chapter four evaluated the latent structure of empathy in more detail, using confirmatory factor methods on data obtained from individuals with a high, medium and low genetic risk for autism. This chapter included both self-reported empathy and performance-based emotion recognition tasks in order to evaluate the impact of measurement on the assessment of empathy. Results indicated a four-factor structure including cognitive empathy, emotional empathy, social skills and a performance-based measurement factor, highlighting the multifaceted nature of empathy. This structure was consistently identified across individuals with autism, parents and controls, indicating that the latent structure of empathy is maintained regardless of genetic vulnerability to autism. Unexpectedly, the emotion recognition tasks were not linked specifically to the cognitive empathy factor, and instead were related in equal proportion to the other three components of empathy. This suggests that the effects assessed by these performance-based tasks are much wider than impairment in cognitive empathy alone. This also indicates that performance-based tasks do not always map neatly onto self-report measures and provide an important additional assessment of an individual's empathy ability. Results from chapter four also provide further evidence for both the E-S theory and the BAP, highlighting that individuals with autism are impaired across all four components of empathy, with parents displaying intermediate difficulties across all empathy domains.

Chapter five systematically evaluated special interests, a component of the non-social domain of the autism phenotype that has so far received little attention. This chapter used exploratory and confirmatory methods to develop and evaluate a 20-item measure assessing motivation to engage in special interests amongst individuals with autism, parents and controls. This chapter sought to determine what motivates individuals to engage in their special interest, as well as to understand some of the more positive aspects associated with ASC. Chapter five demonstrated that individuals with autism are more motivated by intrinsic interest and knowledge and a sense of engagement and flow compared with controls. This indicates an association between positive affect and special interests in autism. These findings are also broadly in agreement with the systemising account of autism (Baron-Cohen, 2010). The strong motivation to further knowledge related to special interests highlighted in chapter five fits in with the notion that individuals with autism are driven by a desire to fully understand the underlying rules of the 'system' behind their special interest. There was also a relationship between autistic traits and motivation to engage in special interests, demonstrating that these interests form an important part of the autism phenotype.

This thesis presented an in depth evaluation of the E-S theory of autism, indicating that it provides a useful cognitive explanation of the autism phenotype. The results in this thesis also outlined evidence for the existence of the BAP as well as indirect support for DSM-5, with empathy and systemising mapping onto the two principal characteristics of autism; social and communication impairment and repetitive behaviours and restricted interests. Importantly, this thesis demonstrated that the latent structure of the autism phenotype is consistent across individuals with a high (ASC), medium (parents) and relatively low (controls) genetic vulnerability to autism, providing support for the quantitative nature of autistic traits. This thesis also exemplifies the important contribution latent structural modelling can make in advancing our understanding of the autism phenotype.

The latent structure of the autism phenotype

Fundamental to the understanding of autism, and indeed any psychological condition, is whether it is best understood as a discrete category of disorder or as representing the severe end of a continuum. This is important not only for diagnostic classification but also in determining the neurobiological mechanisms underpinning ASC. As Constantino (2011) states, while the difference between categorical and dimensional conceptualisations of disorder may appear trivial, this distinction has important implications for predicting developmental course, monitoring the effects of intervention and most importantly, for identifying the underlying processes associated with autism.

Substantial evidence has been reported for the quantitative nature of autistic traits, with a number of studies indicating that they are represented by one (Constantino et al., 2004) or more (Frazier, Youngstrom, Kubu, Sinclair, & Rezai, 2008; Georgiades et al., 2007; Kamp-Becker, Ghahreman, Smidt, & Remschmidt, 2009; Mandy, 2013; Snow, Lecavalier, & Houts, 2009) dimensions. This is reflected in DSM-5, which has moved from a classification system containing multiple discrete diagnostic categories to include a more dimensional conceptualisation of autism spectrum disorder (American Psychiatric Association, 2013). The results obtained in chapters two and three of this thesis lend support to the notion that the autism phenotype is best described by a set of quantitative traits, with clinical autism represented at one end of the distribution, intermediate traits in first-degree relatives, gradually blurring into variation observed in the general population. Chapter two outlined results indicating that the social and non-social traits associated with ASC can be quantitatively measured by two dimensions representing empathy and systemising. This latent structure was consistently identified across individuals with autism, parents and general population controls, indicating that both empathy and systemising can be measured quantitatively across individuals with varying degrees of genetic vulnerability to autism.

However, chapter two also identified a relatively stronger overlap between empathy and systemising in individuals with a high and medium risk for autism, indicating that these traits are not entirely separable. This finding potentially reflects the genetic heterogeneity associated with ASC. It may be that rare copy number variations and gene mutations that are more common in families at risk for autism have an impact on both empathy and systemising. There is evidence that these effects can occur de novo but can also be transmitted from parent to child (Levy et al., 2011). In addition to rare gene variants, common genetic variants that have minor effects are likely to contribute to the risk of developing autism (Gaugler et al., 2014). Altogether, these biological mechanisms may account for the increase in the relationship between empathy and systemising within the higher risk groups. The results obtained in the second chapter emphasise both the quantitative nature of the traits associated with the autism spectrum as well as the multidimensionality of the autism phenotype. This highlights the inherent heterogeneity of ASC.

There is substantial evidence for heterogeneity in autism (Charman et al., 2011; Geschwind, 2011) and that these conditions consist of many 'autisms' rather than a singular discrete disorder (Elsabbagh, 2012). In order to incorporate the quantitative nature of autistic traits as well as the heterogeneity associated with ASC, it may be beneficial to distribute individuals or subgroups along a number of quantitative dimensions based on individual variation or severity. There has been an attempt to measure this in DSM-5, which characterises autism on the basis of a social and non-social domain, with each individual assigned a severity indicator, reflecting three levels of support needs (American Psychiatric Association, 2013).

Previous research has demonstrated support for the distinction between the social and non-social traits associated with autism in both children (Frazier et al., 2012; Guthrie et al., 2013; Mandy et al., 2012) and adults (Frazier et al., 2014a; Frazier et al., 2008). There is also evidence for this DSM-5 model across cultures (Mandy et al., 2014). Support for the DSM-5 model was obtained in chapters two and three. These chapters identified that the autism phenotype is multidimensional, consisting of two moderately correlated factors assessing empathy and systemising. While the results of this thesis were unable to directly evaluate the distinction made between the specific social and non-social traits of the autism spectrum in DSM-5, it provided support that the latent structure of the autism phenotype can be consistently measured quantitatively across two dimensions of empathy and systemising amongst individuals with autism, as well as within first-degree relatives and general population samples.

In addition, through mixture modelling, the third chapter identified three homogenous subgroups across these dimensions containing differing levels of empathy and systemising. This corresponds with previous research utilising mixture modelling to evaluate the traits associated with autism. Three previous papers highlight two dimensions of social and non-social traits, with one distinguishing a diagnostic class (Frazier et al., 2012), a second identifying three classes defined by severity of symptoms (Georgiades et al., 2013a) as well as a follow up study noting a reduction to two severity classes over time (Georgiades et al., 2014). The use of factor mixture modelling techniques is relatively new to this area of research and represents an important paradigm shift in research evaluating the autism phenotype.

The investigation of the latent structure of the autism phenotype was continued in chapter four, providing evidence that empathy is a quantitative trait relevant to understanding the autism spectrum. Results designated four factors assessing cognitive empathy, emotional empathy, social skills and a performance-based measurement factor, highlighting the multidimensional nature of empathy. This is consistent with previous research demonstrating that cognitive and emotional empathy are represented by distinct dimensions. For example, assessment of the factor structure of the Empathy Quotient indicated the presence of distinct cognitive empathy, emotional empathy and social skills continuums (Berthoz, Wessa, Kedia, Wicker, & Grezes, 2008; Lawrence et al., 2004; Muncer & Ling, 2006).

Furthermore, Smith (2006) argues that cognitive and emotional empathy represent two separable but complementary systems. This is consistent with recent theory proposing a number of different underlying mechanisms and brain regions associated with varying aspects of social cognition. Kennedy and Adolphs (2012) propose four major social processing regions they label the amygdala, mentalising, empathy and mirror networks. The amygdala network is involved in emotion regulation while the mirror network contains neurons that are responsive to motor actions (Kennedy & Adolphs, 2012). The mentalising network is implicated in emotion recognition and maps onto the concept of cognitive empathy outlined in this thesis. The empathy network proposed is responsible for detecting and responding to the distress of another individual and thus maps more closely to the concept of emotional empathy.

There is some evidence to suggest the early differentiation of these two networks, signifying that each is associated with a different aspect of social functioning (Happé & Frith, 2014). It has been suggested that specific impairment in the mentalising network underpins the symptomatology associated with autism. The cognitive and emotional empathy dimensions identified in chapter four potentially represent distinct components of social cognition that may map onto different neural networks. However, while there was evidence for the differentiation of cognitive and emotional empathy in chapter four, individuals with autism were found to be equally impaired on both dimensions. Furthermore, it was shown that the performance-based tasks assessing emotion recognition were related in equal magnitude to both cognitive and emotional empathy factors. This is contrary to previous research that implicates the mentalising network in performance on the Eyes task (Holt et al., 2014). Happé and others (2014) suggest that perhaps the complexity of social interaction is such that disruption in one particular area results in flow on effects. This may be reflected in the results obtained in chapter four, which showed that the effects assessed by the performance-based tasks were much wider than impairment in cognitive empathy alone. This chapter highlights the multidimensional nature of empathy and that empathy forms a quantitative component of the autism phenotype.

Within chapter five, motivation to engage in special interests, a defining feature of ASC, was specifically examined. Results indicated that motivation to engage in special interests was best measured by five quantitative dimensions assessing i) personal life values and goals, ii) intrinsic interest and knowledge, iii) prestige, iv) engagement and flow and v) achievement. Compared with controls, individuals with autism displayed increased motivation related to intrinsic factors, while extrinsic motivations were similar in both groups. There was also a significant relationship between autistic traits and motivation to engage in special interests, indicating that special interests form an important part of the autism phenotype. Previous studies have suggested that the non-social characteristics of the autism spectrum are multidimensional, with distinct dimensions of stereotyped motor movements, insistence on sameness and special interests (Lam et al., 2008; Smith et al., 2009). The findings in chapter five indicate that these special interests are explained by a range of different motivations that can be reliably assessed on a quantitative scale.

Taken together, these studies reflect the quantitative nature of autistic traits, highlighting that the autism phenotype can be captured by two broad interrelated dimensions of empathy and systemising. Evidence was also obtained for the multidimensionality of empathy and motivation to engage in special interests. This has important implications for the conceptualisation of autism, and supports the approach taken in the DSM-5, which advocates assessing the severity of autistic symptoms using multiple dimensions. It is on the basis of this dimensional conceptualisation that we can now begin to search for homogenous subgroups that may give us some insight into the neurobiology associated with the autism spectrum. This will prove vital in deriving future conceptualisations of nosologic or diagnostic systems, identifying endophenotypes and understanding the aetiology of autism on a cognitive, neural and genetic level.

Broader autism phenotype

The quantitative nature of autistic traits is also reflected in the existence of the BAP, the finding that first-degree relatives of individuals with ASC display subthreshold levels of autistic traits (Piven et al., 1997b; Sucksmith et al., 2011). Support for the BAP was obtained in chapter two, with parents displaying a stronger overlap between empathy and systemising factors compared with controls. Further support for the BAP was also provided by the mixture modelling results in the third chapter, with approximately 20% of parents displaying the cognitive profile associated with autism. This subsample of parents displayed low levels of empathy and a high drive to systemise. This is consistent with previous research indicating that first-degree relatives have difficulty with empathy and social cognition (Sucksmith et al., 2013). While there is a wealth of research indicating that first-degree relatives display some of the social and communication difficulties associated with ASC, there is limited research evaluating the non-social traits of the autism spectrum in parents of a child with autism. However, there is some evidence that parents report higher levels of systemising ability (Bolte & Poustka, 2006), and have been shown to display special interests relating to systemisable domains (Briskman et al., 2001) as well as a preference for discussing their special interest area (Wolff et al., 1988). Parents have also been shown to display increased levels of stereotyped behaviours (Bolton et al., 1994; Piven et al., 1997b).

As reported in chapter three of this thesis, the subsample of parents that displayed the cognitive profile associated with autism consisted predominantly of fathers. This corresponds with previous research suggesting that the broader phenotype is more common in males (Scheeren & Stauder, 2008). Previous research has also shown that fathers score lower on the social domain of the AQ compared with fathers of typically developing children (Ruta et al., 2012). A large proportion of mothers fell into Class E, containing individuals with high levels of empathy and lower systemising propensity. This provides further evidence that not all parents display the BAP. In previous studies comparing parents, fathers were shown to more often display social deficits (Piven et al., 1997b) and to have significantly lower levels of social expressiveness than mothers (Dawson et al., 2007). Fathers have also been shown to have more difficulty on the Eyes task (Baron-Cohen & Hammer, 1997). These findings are consistent with the results presented in this thesis, suggesting that the cognitive profile associated with autism is especially common in fathers. Furthermore, this subsample of parents was identified through the use of statistical modelling techniques rather than on the basis of predefined criteria. This highlights the importance of implementing latent structural techniques in order to evaluate the quantitative nature of autistic traits and the broader phenotype.

Further evidence for the BAP was obtained in chapter four, with parents displaying better empathy ability than the ASC group, but scoring significantly lower across all facets of empathy than controls. This is consistent with previous research of the BAP, providing evidence for impaired abilities in first-degree relatives, particularly within the social communicative and social cognitive domain (Sucksmith et al., 2011). For example, Szatmari and others (2008) suggest that alexithymia, or the difficulty identifying and processing emotion, may form an important component of the BAP. It has also been demonstrated that parents score lower on advanced ToM tasks including the Eyes test (Gokcen, Bora, Erermis, Kesikci, & Aydin, 2009; Losh & Piven, 2007), a task also included in this chapter.

The results obtained in chapter two, three and four of this thesis provide support for the existence of the BAP, particularly in fathers. This provides important implications for future research assessing the biological and neurodevelopmental underpinnings of ASC and for future conceptualisations of diagnostic manuals and cognitive theories of autism.

Utility of the empathising - systemising theory of autism

The chapters outlined in this thesis provided evidence for the utility of the E-S theory to account for the cognitive and behavioural symptoms associated with the autism spectrum. Chapters two and three indicated that the autism phenotype could be conceptualised by two dimensions representing empathy and systemising. This is consistent with the E-S theory, which argues that the social and communication difficulties associated with autism can be accounted for by difficulty with empathy, while the repetitive behaviours and narrow interests can be explained by the drive to systemise (Baron-Cohen, 2010a). Previous research has attempted to evaluate the relationship between empathy and systemising. For example, Wheelwright and others (2006) reported a small but significant association between the Empathy Quotient and the Systemising Quotient that increased substantially in the ASC group. This is consistent with the results obtained in the second chapter, indicating a significant relationship between empathy and systemising dependent on genetic liability. It has been proposed that this inverse correlation may be representative of a trade-off between empathy and systemising that is further exacerbated in autism (Wheelwright et al., 2006). Previous research has explored this notion, evaluating whether high systemising ability is able to compensate for low empathy, conducting analyses to determine whether empathy and systemising “compete” at a neural level (Goldenfeld et al., 2005; Goldenfeld et al., 2007). The authors reported an effect within the control group, with relatively high systemising ability compensating for less well developed empathy in males, and high empathy ability for low systemising in females (Goldenfeld et al., 2007). However, elevated performance on systemising was not able to compensate for lower levels of empathy in the autism group (Goldenfeld et al., 2007).

The use of factor mixture modelling techniques in chapter three provided a novel way to establish support for the E-S theory of autism, with results identifying classes mapping onto the cognitive profiles outlined in the literature. The E-S theory proposes the existence of a number of cognitive profiles based on differing levels of both traits. The Type S profile reflects individuals who display stronger systemising and lower empathy skills. Type E is associated with greater empathy and decreased systemising skills, with Type B representing a more balanced cognitive profile (Baron-Cohen, 2009). The E-S theory also proposes two extreme cognitive profiles comprising individuals with above average empathy who have difficulty with systemising (Extreme Type E) and individuals who display above average systemising but have significant difficulty with empathy (Extreme Type S) (Baron-Cohen, 2009). Chapter three used mixture modelling to statistically determine whether homogenous subgroups can be identified within a sample including individuals with autism, parents and controls. Rather than specifying details about group membership prior to analysis, mixture models select subgroups of individuals on a statistical basis. Using these techniques, three groups of individuals were identified, mapping onto the Type E, S and B profiles outlined in the E-S theory. The vast majority of individuals with ASC included in the sample fell into the Type S profile, with controls and parents distributed across the other two groups. This demonstrates that the autism phenotype consists of low empathy and high systemising ability. These findings provide substantial support for the cognitive profiles outlined in the E-S theory of autism.

The E-S theory extended the idea of ToM, defining empathy as encompassing not only the ability to infer mental states but also to display an appropriate affective response. Thus, the E-S theory proposes that empathy consists of both cognitive and emotional components (Baron-Cohen & Wheelwright, 2004). This is reflected in the results obtained in chapter four, which provided evidence for distinct cognitive and emotional empathy factors. This chapter demonstrated that empathy is multidimensional and that individuals with autism and parents score lower than controls across all four identified factors. This is consistent with previous research outlining empathy difficulties in individuals with autism and first-degree relatives (Baron-Cohen & Wheelwright, 2004; Gokcen et al., 2009; Losh & Piven, 2007).

Chapter five focused predominantly on motivation and special interests, demonstrating that individuals with ASC were more motivated by intrinsic motivational factors to engage in special interests than controls. Previous research has highlighted a relationship between special interest areas and systemising (Caldwell-Harris & Jordan, 2014). This study in turn, suggests that motivation to engage in special interests is largely intrinsic, centring around a drive to further knowledge and be completely absorbed in the activity. This is broadly in agreement with the E-S theory, which proposes that individuals with autism are driven by a desire to fully predict and understand the underlying rules of the system behind their special interest (Baron-Cohen & Wheelwright, 1999).

Overall, the results of the thesis provide support for the E-S theory as a model for identifying the latent structure of the autism phenotype. Findings also demonstrate that this latent structure is consistently measured by two dimensions of empathy and systemising across individuals with autism, parents and general population controls. Results also highlight that three distinct cognitive profiles can be identified using factor mixture modelling techniques.

Clinical implications

The outcomes of this thesis have important implications for the understanding and conceptualisation of the autism phenotype as well as for the assessment, diagnosis and treatment of ASC. While the use of latent structural techniques formed the focus of this thesis, the use of self-report versus performance-based tasks also provided an indication of the impact of measurement on assessing empathy, systemising and autistic traits. Chapter four provided specific evidence for the impact of self-report versus performance-based tasks on the measurement of empathy. While it was expected based on previous research that the performance-based tasks assessing emotion recognition would be related to self-reported levels of cognitive empathy, in fact they were related in equal magnitude to all three components identified. This suggests that impaired scores on these performance-based tasks indicate a much broader difficulty with empathy than cognitive empathy alone. This finding has consequences for future research, as many previous studies have assumed that these performance-based tasks are pure indicators of cognitive empathy.

Chapter three also provided evidence for differences between assessment obtained via self-report or performance-based measures. While the majority of individuals with ASC were captured by the class displaying the cognitive profile associated with autism (Class S), a small proportion of the ASC group were allocated to Class B, characterised by comparable levels of empathy and systemising. While this group of individuals displayed similar scores on the self-report measures as the remainder of Class B, they performed at the same level on the performance-based tasks as Class S. This indicates that the performance-based tasks may provide a more sensitive measure of impaired empathy in the ASC group, particularly in those individuals with ASC who may have difficulty evaluating their own behaviour and skills. The individuals with ASC falling in Class B were significantly younger and scored lower on a proxy measure of intelligence than the group of ASC individuals falling in Class S. This illustrates the potential impact of age and cognitive ability on self-report measures. The results obtained in chapters two and three exemplify the importance of including both self-report and performance based measures in research and clinical practice.

The evidence outlined within chapter four indicated that parents displayed intermediate difficulties across all empathy factors, providing evidence for the BAP. Chapter three also demonstrated that a proportion of parents display the cognitive profile associated with ASC, involving high systemising and lower empathy ability. These findings have implications for clinical practice, as parents who have difficulty with empathy themselves may report differing treatment goals, and may have different skills in implementing intervention strategies for their child. Parents who display the cognitive profile associated with ASC may also benefit from the provision of clinical advice in a systematic, factual manner. Furthermore, parents who report difficulties with empathy may also benefit from advice on how to manage and improve their own relationships with others.

The results obtained in chapter five have important implications for intervention and outcomes for individuals with autism. There is increasing research into the importance of special interests for individuals with ASC, as well as the clinical utility of incorporating these interests into intervention programs. For example, a recent review revealed that interest based intervention programs resulted in a significant increase in prosocial behaviour amongst individuals with ASC (Dunst, Trivette, & Hamby, 2012). There is also evidence that the inclusion of special interests fosters peer interaction and social engagement (Boyd et al., 2007; Koegel et al., 2012; Koegel et al., 2013). Moreover, special interests have also been shown to be predictive of positive change in language, social communication, motor skills and emotion regulation following intervention (Winter-Messiers, 2007).

Importantly, special interests have been associated with feelings of pride, happiness and increased self-confidence in individuals with autism (Winter-Messiers, 2007). There is also evidence that special interests are acknowledged by families as an area of great strength and skill (Mercier et al., 2000). In a world where the social environment presents such a challenge for individuals with autism, it is of the utmost importance to capitalise on fostering a sense of pride and self-confidence in individuals with ASC. Incorporating special interests into case formulation and intervention programs has the potential to significantly influence outcomes for individuals with autism.

Limitations

There were a number of limitations within the data sets utilised in this thesis. Firstly, the parent group in the sample reported in chapters two, three and four contained a larger number of mothers than fathers, while the ratio of males and females was equivalent in both the control and ASC groups. Secondly, the fathers in this sample had missing data on the performance-based measures utilised in chapter four. While this was taken into account in the analyses, the inclusion of more fathers with full information on performance-based measures would benefit future research assessing the broader phenotype. This thesis was also limited by the fact that no performance-based measures of systemising were included in the data sets. The inclusion of performance-based measures of systemising in future studies would be beneficial.

Due to the online data collection methods utilised, there was no way to verify individual diagnoses of ASC. However, previous research has shown that diagnostic information reported online is generally accurate (Lee et al., 2010). Furthermore, the use of online data collection methods allowed for the large samples utilised in this thesis. Finally, the ASC group consisted of high functioning individuals only. While this is often the case for cognitive studies with autism (Hoekstra & Whatson, 2010) and is unavoidable when using self-report measures, this thesis precludes individuals who fall at the lower functioning end of the spectrum. Future research utilising this methodology to assess the autism phenotype across a range of ages and intellectual functioning would be beneficial.

Conclusions

This thesis utilised latent structural modelling in order to understand and refine the autism phenotype. The results obtained provided substantial independent support for the E-S theory and that it provides an effective framework for understanding the cognitive and behavioural symptoms associated with ASC. This thesis also highlighted the quantitative and multidimensional nature of autistic traits, thereby providing indirect support for the dimensional approach and dyadic grouping of autism symptoms included in the DSM-5. Furthermore, it provided evidence for the existence of a broader autism phenotype, particularly in fathers of a child with ASC. Through the examination of community samples as well as first-degree relatives and individuals with a clinical diagnosis of ASC, this thesis provides the first comprehensive evaluation of the latent structure of the autism phenotype simultaneously across the full genotypic spectrum. This thesis has significant implications for understanding the inherent heterogeneity associated with ASC as well as the quantitative and multidimensional nature of the autism phenotype. It provides significant value for future research aiming to elucidate the aetiology of autism on a cognitive, neural and biological level. It also significantly deepens our understanding of the autism phenotype, providing important implications for research and clinical practice.

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Appendix A

Special Interests Motivation Scale

Roth, I. & Hoekstra, R.A.

Special Interest Motivation Scale (SIMS)

Using the scale of 1 (Not at all) to 7 (Exactly) below, please indicate to what extent each of the following statements explains why you engage in your most important special interest. E.g. if a statement in no way explains why you engage with your special interest, click 'Not at all'; if a statement explains really well why you engage with your special interest, click 'Exactly'.

Not at all			Moderately			Exactly
1	**2**	**3**	**4**	**5**	**6**	**7**

1.	Because it is satisfying to learn new things about my special interest.	1	2	3	4	5	6	7
2.	I chose this special interest because it allows me to reach my life goals.	1	2	3	4	5	6	7
3.	Because it enables me to be well regarded by people I know.	1	2	3	4	5	6	7
4.	For the sense of sheer enjoyment I experience doing my special interest.	1	2	3	4	5	6	7
5.	Because I love bettering myself at my special interest.	1	2	3	4	5	6	7
6.	To prove to myself that I am capable of achieving something special.	1	2	3	4	5	6	7
7.	For the prestige that comes with doing my special interest.	1	2	3	4	5	6	7
8.	Because I enjoy discovering new aspects about my special interest.	1	2	3	4	5	6	7
9.	Because when I do well at my special interest I feel important.	1	2	3	4	5	6	7
10.	Because I love being engaged in my special interest.	1	2	3	4	5	6	7
11.	For the sense of achievement I feel after accomplishing difficult aspects of my special interest.	1	2	3	4	5	6	7
12.	Because I enjoy broadening my knowledge about my special interest.	1	2	3	4	5	6	7
13.	Because it is a good way to learn lots of things that could be useful in other areas of my life.	1	2	3	4	5	6	7
14.	For the excitement I feel when I am really involved in my special interest.	1	2	3	4	5	6	7
15.	Because I enjoy improving my special interest abilities.	1	2	3	4	5	6	7

16.	Because I don’t want to fail in pursuing my special interest.	1	2	3	4	5	6	7
17.	Because my special interest fits my personal values.	1	2	3	4	5	6	7
18.	To prove to others that I am good at my special interest.	1	2	3	4	5	6	7
19.	Because I like the feeling of being totally immersed in my special interest.	1	2	3	4	5	6	7
20.	Because it is satisfying to aim for excellence in my special interest.	1	2	3	4	5	6	7

Appendix B

Empathising, systemising and autistic traits: Latent structure in individuals with autism, their parents and general population controls

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Empathizing, Systemizing, and Autistic Traits: Latent Structure in Individuals With Autism, Their Parents, and General Population Controls

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The search for genes involved in autism spectrum conditions (ASC) may have been hindered by the assumption that the different symptoms that define the condition can be attributed to the same causal mechanism. Instead the social and nonsocial aspects of ASC may have distinct causes at genetic, cognitive, and neural levels. It has been posited that the core features of ASC can be explained by a deficit in empathizing alongside intact or superior systemizing; the drive to understand and derive rules about a system. First-degree relatives also show some mild manifestations that parallel the defining features of ASC, termed the broader autism phenotype. Factor analyses were conducted to assess whether the latent structure of empathizing, systemizing, and autistic traits differs across samples with a high (individuals on the spectrum), medium (first-degree relatives) or low (general population controls) genetic vulnerability to autism. Results highlighted a two-factor model, confirming an empathizing and a systemizing factor. The relationship between these two factors was significantly stronger in first-degree relatives and the autism group compared with controls. The same model provided the best fit among the three groups, suggesting a similar latent structure irrespective of genetic vulnerability. However, results also suggest that although these traits are relatively independent in the general population, they are substantially correlated in individuals with ASC and their parents. This implies that there is substantially more overlap between systemizing and empathizing among individuals with an increased genetic liability to autism. This has potential implications for the genetic, environmental, and cognitive explanations of autism spectrum conditions.

Keywords: autism, factor analysis, genetics, broader autism phenotype, family studies

Autism spectrum conditions (ASC) are characterized by impairment in the development of communication skills and reciprocal social interaction alongside the presence of unusually repetitive behaviors and narrow interests (*Diagnostic and Statistical Manual of Mental Disorders*, fourth edition, revised [*DSM–IV–TR*]; American Psychiatric Association, 2000). It is well established through family and twin studies that these conditions have a strong genetic component. A range of twin studies have indicated substantially higher concordance rates for clinical autism in monozygotic twins when compared with dizygotic twins. Altogether, these findings indicate strong genetic influences on ASC with a heritability estimate of around 80%, (Ronald & Hoekstra, 2011). A recent large scale family study suggests the recurrence rate for autism within families is close to 20% (Ozonoff et al., 2011). This, coupled with prevalence estimates of around 1% (Baron-Cohen et al., 2009; Brugha et al., 2011), suggests a markedly increased risk for autism within families, highlighting a strong influence of genetic effects. There is also a growing body of evidence from molecular genetic studies suggesting the involvement of multiple genetic variants and loci in the development of these conditions (Abrahams & Geschwind, 2008; Geschwind, 2011).

There is evidence to suggest that family members show mild manifestations that parallel the defining features of autism, a phenomenon termed the Broader Autism Phenotype (BAP; see Sucksmith, Roth, & Hoekstra, 2011 for a review). With the development of quantitative psychometric instruments such as the Autism Spectrum Quotient (AQ; Baron-Cohen, Wheelwright, Skinner, Martin, & Clubley, 2001), that assess autistic traits on a continuous scale, it is now possible to measure these subthreshold autistic traits with more precision. Use of such scales in family studies may provide insights into the genetic factors involved in ASC and the familial risk for developing autism.

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A multitude of studies have reported mild impairments in relatives of individuals with autism, particularly in the social and communicative domains. For example, parents of individuals with ASC have been shown to display more social and communication difficulties, as measured by subscales of the AQ, as well as score lower on measures of pragmatics (Piven, Palma, et al., 1997; Ruser et al., 2007). Siblings also show difficulties in reciprocal social interaction (Nadig et al., 2007; Toth, Dawson, Meltzoff, Greenson, & Fein, 2007). Similarly, parents show some mild difficulties with social cognition, as measured by neuropsychological tests (Losh et al., 2009; Losh & Piven, 2007). Parents of children with an ASC also perform lower than a control group on a task assessing the ability to read complex emotional states from viewing the eye region of the face (Baron-Cohen & Hammer, 1997; Losh & Piven, 2007), providing evidence for the BAP at a cognitive level.

Although the evidence for some of the nonsocial aspects of ASC among relatives is more modest, a number of studies suggest an elevated rate of stereotyped behaviors and circumscribed hobbies in parents (Bolton et al., 1994; Briskman, Happe, & Frith, 2001; Piven, Palmer, Jacobi, Childress, & Arndt, 1997). Some studies suggest that first degree relatives of individuals with ASC may also display the same "cognitive style" that leads to superior performance on tasks where visual processing of local material is advantageous, including the Embedded Figures Task (Baron-Cohen & Hammer, 1997; Bölte & Poustka, 2006; Happé, Briskman, & Frith, 2001) and the Block Design Task (Scheeren & Stauder, 2008). However, findings related to tasks assessing local processing styles have been somewhat inconsistent, both in clinical groups (White & Saldaña, 2011) and in first degree relatives (see Sucksmith et al., 2011 for a review).

There has been much debate around whether the triad of features characteristic of autism (social impairments, communication impairments and repetitive behavior/narrow interests) are influenced by the same genetic and environmental factors, or whether they are somewhat independent. Happé and Ronald (2008) suggest that the core features that define autism are largely "fractionable"; that is, that they may have distinct causes at genetic, cognitive, and neural levels. There are a number of family and twin studies that support this notion, showing that although the three sets of features are highly heritable individually, they are affected by largely independent genetic influences (Ronald, Happé, Bolton, et al., 2006; Ronald, Happé, & Plomin, 2005; Ronald, Happé, Price, Baron-Cohen, & Plomin, 2006). Moreover, 10% of children in a large general population study showed only social impairment, only communication difficulties, or only repetitive and restricted interests (Ronald, 2006), suggesting these characteristics can also occur in isolation.

Similarly, a review of factor analytic studies showed that, of the seven studies included, six found evidence for multiple factors underlying autistic features (Mandy & Skuse, 2008). Although the total number of factors identified varied across studies, all studies (see Constantino et al., 2004 for an exception) reported at least one social-communication factor and all but one also reported at least one distinct nonsocial factor (Mandy & Skuse, 2008). Taken together, these studies suggest that partially distinct causal explanations should be sought for the social and nonsocial aspects of ASC.

This hypothesis has so far mainly focused on features at a behavioral level. However, there are a number of theoretical explanations that attempt to account for the features in autism at the cognitive level. It has been suggested that these conditions are associated with difficulties in executive function (Corbett, Constantine, Hendren, Rocke, & Ozonoff, 2009; Ozonoff, Pennington, & Rogers, 1991; Russell, 1997), 'weak central coherence' (a processing bias in which individuals focus on the local rather than global features of an object), and "Theory of Mind" (the ability to attribute mental states to oneself and others) (Baron-Cohen, Leslie, & Frith, 1985).

The term "empathizing" extends the idea of Theory of Mind and involves two components: the ability to attribute mental states to oneself and others and the drive to respond with an appropriate emotion to that mental state (Baron-Cohen, 2004, 2010). A different process, systemizing, is conceptualized as the drive to understand and derive rules about a system (Baron-Cohen, 2002). Systemizing allows an individual to predict the behavior of a system and therefore to control it (Baron-Cohen, 2010). A system is defined as anything that takes inputs and delivers outputs, and includes everything from technical systems (e.g., a machine) through to natural (e.g., the weather), abstract (e.g., mathematics), social (e.g., a company), collectible (e.g., a library), and motoric (e.g., a tennis top-spin) systems that the brain can analyze or construct (Baron-Cohen, 2004).

According to the Empathizing–Systemizing (E-S) theory, autism is best explained by a deficit in empathy alongside intact or even superior systemizing (Baron-Cohen, 2004, 2010; Baron-Cohen, Knickmeyer, & Belmonte, 2005; Baron-Cohen, Wheelwright, Lawson, Griffin, & Hill, 2002). In this way, the social and communication impairments seen in these conditions can be accounted for by empathising, and the islets of ability, repetitive behavior, and restricted interests or obsessions with systems can be accounted for by an interest in systemizing (Baron-Cohen, 2004, 2010). There is a large evidence base suggesting that individuals with ASC show impaired performance on measures of empathizing and intact or elevated performance on tests of systemizing ability (Baron-Cohen, Richler, Bisarya, Gurunathan, & Wheelwright, 2003; Baron-Cohen & Wheelwright, 2004; Baron-Cohen, Wheelwright, & Jolliffe, 1997; Baron-Cohen, Wheelwright, Stone, & Rutherford, 1999; Jolliffe & Baron-Cohen, 1997; Lai et al., 2011; Lawson, Baron-Cohen, & Wheelwright, 2004).

As yet it remains unclear the extent to which empathizing and systemizing traits are related. Given that people with ASC tend to perform poorly on tasks of empathizing and do well on tasks of systemizing, an inverse correlation between the two traits would be expected in this group, provided that these traits are assessed on continuous scales that allow for sufficient variance within the clinical group. However, it is less clear whether this inverse association is linear across populations and would also apply to nonclinical samples. The current study aims to assess the association between empathizing, systemizing, and social and nonsocial autistic traits across three distinct samples, stratified by their genetic risk for autism. This study reports on factor analyses employed in three distinct samples comprising individuals with a clinical ASC diagnosis (high genetic vulnerability), parents of a child with ASC (medium genetic risk), and a general population control group (low genetic vulnerability).

Method

Participants

Individuals with a clinical ASC diagnosis and parents of a child with ASC were recruited via the participant database at the Autism Research Centre at the University of Cambridge (www.autismresearchcenter.com). To account for any potential response bias, the control sample was collected via a different portal at a general (nonclinical) volunteer psychology research webpage (www.cambridgepsychology.com). Participants were included in the study if they were 18 years and over and had completed all measures. The individuals in the ASC group and the children of the parent group were reported to all have received a formal ASC diagnosis from experienced clinicians in recognized clinics.

Individuals in the parent group did not report having an ASC diagnosis themselves. The control group was confined to individuals who did not report any past psychiatric history. The total sample consisted of 1034 individuals, comprising 363 individuals with ASC (males = 193, females = 170, mean age = 36 years, SD = 11), 439 parents of a child with ASC (males = 141, females = 298, mean age = 42 years, SD = 8), and 232 controls (males = 122, females = 110, mean age = 33 years, SD = 10). 79.9% of the control group had completed higher education, whereas 49.9% of the parent group and 54.0% of individuals with ASC had completed an undergraduate degree.

Measures

Individuals registered in either of the above websites were asked to fill out a range of well-validated questionnaires assessing empathizing, systemizing, and quantitative autistic traits. Participants were able to complete the questionnaires in their preferred order. The measures used are designed as dimensional quantitative measures of empathizing, systemizing, and autistic traits, in keeping with the paradigm that autism is best represented along a spectrum of symptoms. As these measures are not designed as clinical instruments this allows for variance across the three sample groups.

Empathizing and systemizing. The Empathy Quotient (EQ; Baron-Cohen & Wheelwright, 2004) is a self-report measure of empathizing. Items assess the ability to attribute mental states to oneself and others and the drive to respond with an appropriate emotion to that mental state. An example of an item assessing recognizing the mental state of another is 'I am quick to spot when someone in a group is feeling awkward or uncomfortable.' 'I tend to get emotionally involved with a friend's problems' is an example of an item assessing the drive to respond emotionally to another's mental state. The EQ is comprised of 40 statements scored on a Likert scale including four response options: *definitely disagree*, *slightly disagree*, *slightly agree*, and *definitely agree*. For approximately half the items an "agree" response is in line with high empathy abilities. On these items "definitely agree" responses score two points and "slightly agree" responses score one point, with "definitely disagree" and "slightly disagree" scoring zero. The other half of the items are reverse-scored, as a "disagree" response refers to better empathizing on these items. Scores range from zero to 80 and follow a near normal distribution, with a higher score reflecting increased empathizing ability. Adults with high functioning autism or Asperger's syndrome have been shown to score significantly lower on the EQ than age-matched controls (Baron-Cohen & Wheelwright, 2004).

The Systemizing Quotient-Revised (SQ-R; Wheelwright et al., 2006) is a self-report measure of systemizing consisting of 75 statements with four response options: *strongly agree*, *slightly agree*, *slightly disagree*, and *strongly disagree*. Scoring procedures are equivalent to those described for the EQ. Scores follow a continuous distribution ranging from zero to 150, with higher scores reflecting stronger systemizing behavior. Items include statements like 'When I learn about a new category I like to go into detail to understand the small differences between different members of that category,' and 'In math, I am intrigued by the rules and patterns governing numbers.' Individuals with autism score higher on the SQ-R compared with age-matched controls (Baron-Cohen et al., 2003; Goldenfeld, Baron-Cohen, & Wheelwright, 2005; Goldenfeld, Baron-Cohen, Wheelwright, Ashwin, & Chakrabarti, 2007; Lai et al., 2011; Wheelwright et al., 2006).

Autistic traits. The Autism Spectrum Quotient (AQ; Baron-Cohen et al., 2001) is a self-report quantitative measure of autistic traits. The AQ consists of 50 items assessing the core areas of difficulty in ASC including impaired social skills, communication difficulties, imagination and attention switching and a superior attention to detail. Participants were asked to rate themselves on a 4-point Likert scale with response categories *definitely disagree*, *slightly disagree*, *slightly agree*, and *definitely agree*. This study used the raw scoring method (as detailed in Hoekstra, Bartels, Cath, & Boomsma, 2008), with total scores following a normal distribution ranging from 50 to 200 and a score of 200 representing full endorsement of all autistic traits. Individuals with an ASC show significantly higher scores on the AQ compared with the general population (Baron-Cohen et al., 2001).

Recent evidence suggests that the AQ can be split into two categories of items, reflecting a broad social interaction factor comprising the social skills, attention switching, communication and imagination items and an attention to detail factor (Hoekstra et al., 2008). These two factors only correlate modestly and are therefore useful in making the distinction between social and nonsocial autistic traits (Hoekstra et al., 2008). The AQ was split into these two subscales for the current analysis.

Analytic Strategy

There is a large evidence base suggesting that scores on the measures included in this study are affected by sex (Baron-Cohen et al., 2005; Baron-Cohen et al., 2011). The main focus of this study was on the factor structure, not on sex differences in mean scores, which have been studied for our measures of interest in previous studies (see, e.g., Baron-Cohen et al., 2001; Hoekstra et al., 2008; Sucksmith, Allison, Baron-Cohen, Chakrabarti, & Hoekstra, 2013; Wheelwright et al., 2006). To account for the effects of sex as well as the potential confounding effect of age on the means, variables were standardized via regression analyses in SPSS for age and sex before analysis.

After standardization, a series of confirmatory factor (CFA) models were specified and estimated in MPlus version 6.11 (Muthén & Muthén, 2010a) using the maximum likelihood estimator (Muthén & Muthén, 2010b). A one-factor model encompassing all measures of empathizing, systemizing, and autistic traits was fit for each of the three groups separately (Models 1–3). After this a two-factor model was fit across the three groups (Models 4–6). The EQ and the social interaction subscale of the AQ (AQ_soc) were predicted to load on one latent "empathizing" factor, whereas the scores on the SQ-R and the attention to detail factor (AQ_att) were expected to load on a "systemizing" factor. Scores on the social interaction subscale of the AQ (AQ_soc) were reverse scored to enable ease of interpretation. Empathizing ability is therefore indicated by high scores on the EQ and high scores on the social interaction subscale of the AQ, whereas systemizing ability is indicated by high scores on the SQ and on the attention to detail factor of the AQ.

To assess the full range of models available, and to test whether our hypothesized Empathizing-Systemizing model (tested in Models 4–6) provided the best fit, two further models were specified, the first including the SQ-R and the social subscale of the AQ (AQ_soc) on one factor, with the EQ and the attention to detail factor (AQ_att) loading on a second factor (Models 7–9). The second model included the EQ and SQ-R on the first factor and both sections of the AQ loading on a second factor (Models 10–12). Both models were fit across the three groups.

Models 1 to 12 were fit within the three individual groups to allow for a different factor structure relative to genetic liability. However, it is important to evaluate the equivalence of the parameters estimated in a CFA across groups (Brown, 2006). This can be achieved within one model using multigroup CFA. Multiple group models make it possible to pinpoint where any specific differences across groups may fall (Brown, 2006). Therefore, to assess whether these traits function differently among the three groups, a further model was implemented, allowing all parameter estimates to vary (Model 13). A further model in which the factor loadings were constrained to be equal across groups was also implemented (Model 14).

To test whether the same factor structure was identified for males and females within each sample group, a further three models were tested, with varying restrictions, across six groups split by sex and genetic vulnerability. Model 15 contains a multigroup CFA allowing all estimates to vary across the six groups. A second model was fit constraining the factor loadings to be equal (Model 16). A final model restricted the factor correlations to be equal for males and females as well as equal factor loadings across the six groups (Model 17).

Model fit was evaluated using the following goodness of fit statistics; Akaike information criterion (AIC; Akaike, 1987), Bayesian information criterion (BIC; Schwarz, 1978), Sample size adjusted BIC (SSABIC; Sclove, 1987), Root mean square error of approximation (RMSEA; Steiger & Lind, 1980), Comparative fit index (CFI; Bentler, 1987), and Tucker-Lewis index (TLI; Tucker & Lewis, 1973). The AIC, BIC, and SSABIC are parsimony-adjusted indices used to examine model fit, with lower values indicating a better fit. It has been suggested that a RMSEA value <0.05 indicates a close model fit, with values up to 0.08 suggesting a reasonable error of approximation (Browne & Cudeck, 1993). Current recommendations state that a CFI and TLI value $>= 0.90$ indicate acceptable fit with values $>= 0.95$ indicative of very good fit to the data (Brown, 2006; Hu & Bentler, 1999).

As well as taking into account the fit indices mentioned above, evaluation of model fit also took into account the strength and interpretability of the structural parameter estimates.

Results

Distribution of scores on the subscales of the AQ, the EQ and SQ, standardized for age and sex, are given in Figure 1, showing adequate coverage of the possible range of responses. Model fit indices ascertained from the CFA models are given in Table 1. The one-factor models displayed poor fit among the three groups. In contrast, the two-factor model accounting for measures of systemizing and empathizing provided an excellent fit to the data within all three groups (see Models 4–6). RMSEA values of 0 occur as a result of a chi square value less than the number of degrees of freedom (Kenny, Kaniskan, & McCoach, 2011). Similarly, CFI and TLI values are also affected by the chi square statistic as well as the degrees of freedom in the model (Brown, 2006). However, these values are indicative of almost perfect model fit (Savalei, 2010). The further two-factor models (Models 7–12) displayed poor fit among the three groups. Models 7 and 10 displayed a correlation greater than 1 between the two factors, indicating that there is no distinction between them. Similarly, fit statistics for Models 8 and 9 fell under the required thresholds, suggesting that the empathizing-systemizing two-factor model (tested in Models 4–6) described the data best in all three groups.

Multiple group analyses were conducted to assess for specific group differences within the two-factor model where the EQ and the social interaction subscale of the AQ (AQ_soc) load on the latent "empathizing" factor, whereas the scores on the SQ-R and the attention to detail factor (AQ_att) load on a "systemizing" factor. Model 13 showed a good fit to the data, with a CFI and TLI above 0.97. Although the RMSEA is larger than the cut-off recommended for model fit, this value is affected by the number of free parameters in the model (Browne & Cudeck, 1993). Because Model 13 includes more parameters, the RMSEA of this model is relatively high compared with Models 4–6. Furthermore, with limited degrees of freedom the RMSEA value is of less concern given all other indices are strong and suggest a good fit (Brown, 2006). This is the case in Model 13 with CFI and TLI values falling above the specified threshold.

A model in which the factor loadings were constrained to be equal across the three groups (Model 14) resulted in a significantly poorer fit compared to the fit of Model 13 ($\chi^2 = 59.37$, $p < .001$). Therefore, Model 13, the two-factor model with equal form among the three groups, allowing the factor loadings to vary, provided the best fit to the data. Evaluation of Models 15 to 17 showed that the model constraining the factor correlations to be equal across males and females in each group (Model 17) provided the best fit to the data, indicating that the factor structure obtained in Model 13 does not differ when sex is taken into account.

Factor loadings, correlations, and confidence intervals for the two-factor model taken from the Model 13 analysis are given in Figure 2. All factor loadings were salient and statistically significant ($p < .05$), reflecting that these measures are good indicators of their respective factors. Parents scored lower on the latent factor mean of empathizing than the control group (Mean Difference $= -0.31$, $p < .01$). However, there was no significant difference between scores on the systemizing factor between the parent and control groups. The ASC group showed lower scores on the latent factor empathizing compared with controls (Mean difference $= -2.68$, $p < .01$) and the parent group (Mean difference $= -2.37$, $p < .01$) as well as superior latent mean scores on system-

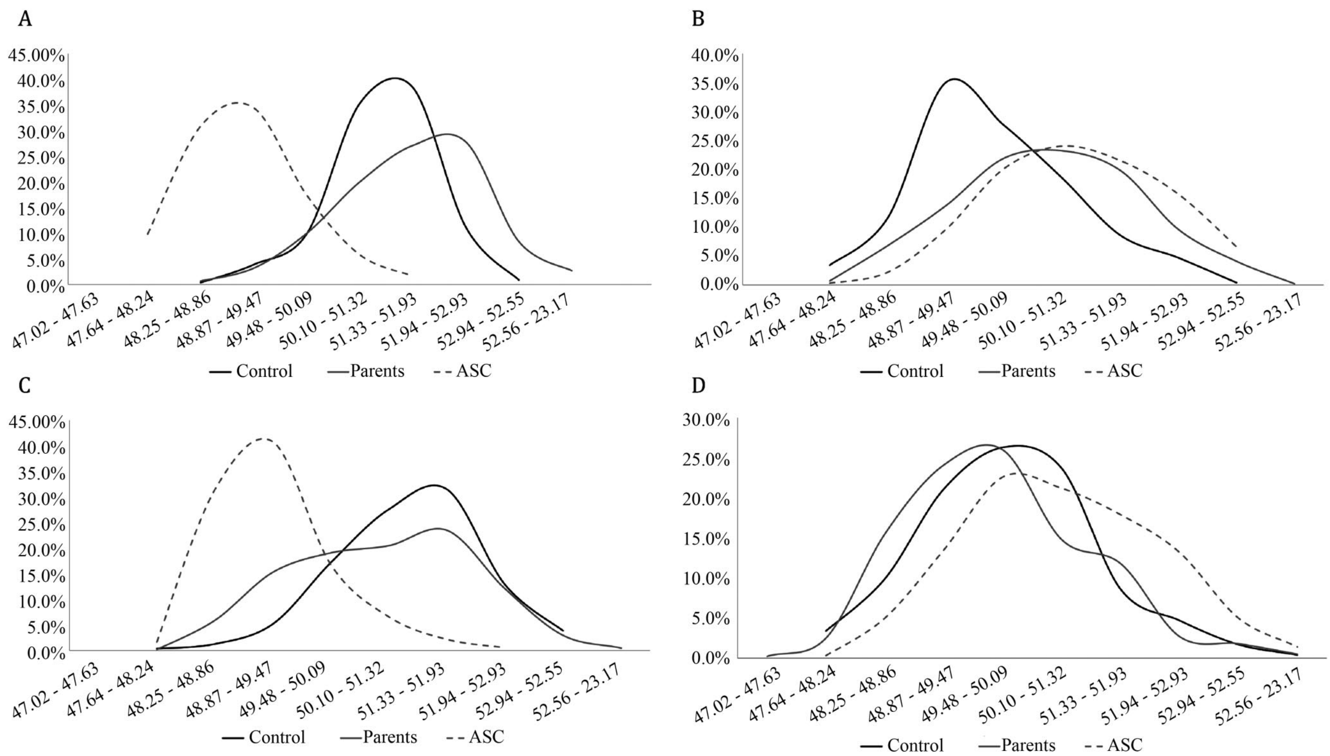

Figure 1. A, Distribution of scores on the social factor of the Autism Spectrum Quotient. B, Distribution of scores on the attention to detail factor of the Autism Spectrum Quotient. C, Distribution of scores on the Empathy Quotient. D, Distribution of scores on the Systemizing Quotient Revised.

izing compared with controls (Mean difference = 1.01, $p < .01$) and the parent group (Mean difference = 1.14, $p < .01$).

The correlation between empathizing and systemizing was significant among the three groups. The negative correlation between the two factors was significantly stronger in both the ASC group ($r = -0.61$) and the parent group ($r = -0.57$) compared with the control group ($r = -0.22$). However, the correlations between the two factors for the ASC group and parent group did not differ (i.e., the confidence intervals for the correlations between empathizing and systemizing in the parent and ASC groups overlapped).

Discussion

The current study examined the structure of autistic characteristics across individuals with a low, medium, and high genetic vulnerability for autism. Results indicated that the two-factor model provided the best fit across the three groups irrespective of sex. This model comprises an empathizing factor including both the EQ and the social behavioral and cognitive traits measured by the AQ, and a systemizing factor including the SQ-R and the "attention to detail" traits measured by the AQ. The latent empathizing factor and systemizing factor were inversely correlated in all three groups. The factor correlations ranged from small to large, providing support for the notion that the social and nonsocial aspects of ASC may have distinct causes at a behavioral level (Happé & Ronald, 2008; Ronald, 2006; Ronald, Happé, Bolton, et al., 2006; Ronald, Happé, Price, et al., 2006).

Perhaps the most notable finding from the current study is the difference in the strength of the inverse relationship between empathizing and systemizing among controls, first-degree relatives, and individuals on the spectrum. The association between empathizing and systemizing was substantially stronger in individuals with ASC and parents of a child with ASC than in general population controls. Although a definitive explanation for these associations cannot be given without further research, there are a number of potential explanations why these constructs may be more strongly associated in individuals with autism and their first-degree relatives.

First, individuals on the spectrum are given a diagnosis of an ASC. This by definition includes symptoms from all three domains of social impairment, communication difficulties, and repetitive behavior/narrow interests (American Psychiatric Association, 2000). Because systemizing and empathizing are cognitive explanations of autism, individuals with autism are likely to be both superior in systemizing and weaker in empathizing. It is therefore not surprising that these two factors are highly inversely related in this group, given that the presence of all three core symptoms of autism make high systemizing and low empathizing more likely. However, this account does not apply to the parent group as these parents do not have a diagnosis of ASC themselves and are therefore not directly selected to score high on systemizing and low on empathizing.

An alternative explanation for our findings could be that empathizing and systemizing are highly correlated in individuals with ASC due to cognitive strategies used by this group. Because of their poor intuitive empathic abilities, individuals with autism may

Table 1
Fit Indices and Model Comparisons for the Alternative Factor Models of the Autism Phenotype

Model	Description	Fit indices: AIC	BIC	SSABIC	RMSEA	CFI	TLI	χ^2	*r*	$\Delta\chi^2(df)$
One-factor models										
1	1f control group ($n = 232$)	1982.193	2023.554	1985.520	0.464	0.664	0.008	101.87**		
2	1f parent group ($n = 439$)	4002.051	4051.065	4012.982	0.351	0.857	0.571	110.06**		
3	1f autism group ($n = 363$)	2959.359	3006.092	2968.021	0.338	0.845	0.536	84.73**		
Two-factor models										
4	2f control group ($n = 232$)	1881.771	1923.132	1885.098	0.000	1.000	1.006	1.45	−0.22	
5	2f parent group ($n = 439$)	3892.859	3941.873	3903.791	0.000	1.000	1.005	0.87	−0.57	
6	2f autism group ($n = 363$)	2877.375	2928.002	2886.759	0.000	1.000	1.003	0.75	−0.62	
7	2f control group ($n = 232$) (AQ_soc, SQ; AQ_att, EQ)	1982.767	2027.574	1986.371	0.655	0.665	−1.008	100.44**	>1.0	
8	2f parent group ($n = 439$) (AQ_soc, SQ; AQ_att, EQ)	3997.030	4050.129	4008.873	0.482	0.865	0.189	103.04**	>1.0	
9	2f autism group ($n = 363$) (AQ_soc, SQ; AQ_att, EQ)	2940.547	2991.174	2949.931	0.416	0.882	0.295	63.92**	>1.0	
10	2f control group ($n = 232$) (AQ_soc, AQ_att; EQ, SQ)	1976.182	2020.989	1979.786	0.633	0.687	−0.875	93.86**	>1.0	
11	2f parent group ($n = 439$) (AQ_soc, AQ_att; EQ, SQ)	3988.040	4041.138	3999.883	0.460	0.877	0.261	94.05**	>1.0	
12	2f autism group ($n = 363$) (AQ_soc, AQ_att; EQ, SQ)	2947.319	2997.946	2956.703	0.438	0.870	0.219	70.69**	>1.0	
Multigroup models										
13	2f multigroup all estimates allowed to vary ($n = 1034$)	8672.204	8835.264	8730.452	0.085	0.986	0.972	31.26**		
14	2f multigroup equal factor loadings ($n = 1034$)	8717.568	8846.039	8763.460	0.116	0.953	0.947	90.63**		59.3 (8) $p < 0.001$
Multigroup models including sex										
15	2f multigroup factor all estimates allowed to vary ($n = 1034$)	8618.343	8934.580	8731.308	0.100	0.978	0.961	54.17**		
16	2f multigroup equal factor loadings ($n = 1034$)	8614.676	8881.500	8709.990	0.089	0.974	0.969	70.50**		16.33 (10) $p > 0.05$
17	2f multigroup factor correlations equal for males and females within each group and equal factor loadings ($n = 1034$)	8612.924	8864.925	8702.942	0.086	0.973	0.971	74.75**		4.25 (3) $p > 0.05$

Note. AIC, Akaike information criteria; AQ_att = Attention to detail factor of the Autism Spectrum Quotient; AQ_soc = Social interaction factor of the Autism Spectrum Quotient; BIC, Bayesian information criteria; CFI, Comparative fit index; EQ, Empathy Quotient; RMSEA, Root mean square error of approximation; SSABIC, Sample size adjusted BIC; SQ, Systemizing Quotient Revised; TLI, Tucker-Lewis index.
** $p < .001$.

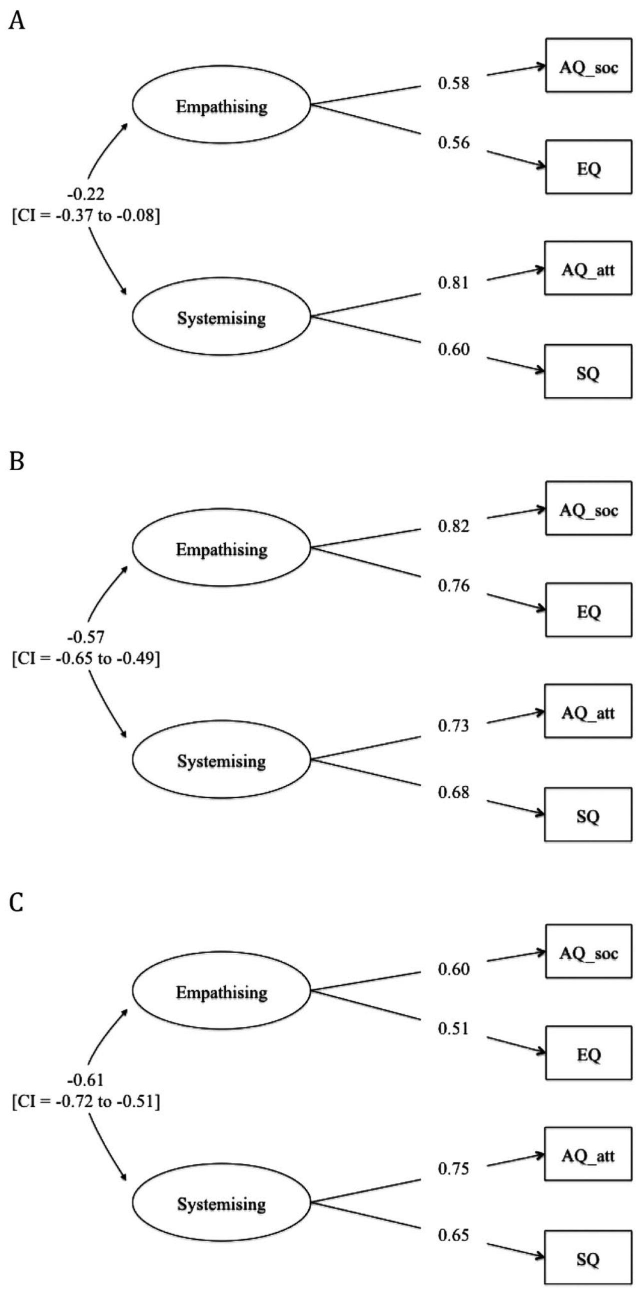

Figure 2. A, Two-factor multigroup model for the control group. B, Two-factor multigroup model for the parent group. C, Two-factor multigroup model for the ASC group. AQ_att = attention to detail factor of the Autism Quotient; AQ_soc = social interaction factor of the Autism Quotient; CI = 95% confidence interval; EQ = Empathy Quotient; SQ = Systemizing Quotient Revised.

use systemizing strategies in empathy tasks. For example, when an individual with ASC engages in an activity requiring empathizing, they may use systemizing strategies to work out what particular emotion or mental state is relevant to the situation and how to respond appropriately. The use of such strategies may result in an association between empathizing and systemizing, and as such when attempting to measure empathy in this group we may actually be indirectly measuring the systemization of empathy. If this strategy does not improve empathizing ability, the correlation would remain strong and in a negative direction. However, if systemizing is a helpful strategy and improves empathizing ability, then it is likely to lower the negative correlation between empathizing and systemizing. Although the current study cannot identify whether such strategies are being used, our results call for further research into the types of strategies used by individuals with ASC in their approach to tasks of empathy.

Our findings suggest that there is a relatively stronger overlap between empathizing and systemizing in individuals with a high and medium genetic risk for autism compared with individuals with a low genetic risk. This overlap could be attributable to genetic and/or environmental influences. Although the design of the current study did not allow us to examine the nature of the association, we may consider possible genetic and environmental mechanisms that may underlie the different associations between empathizing and systemizing in groups of varying genetic risk for autism.

One possible explanation for the high inverse association between empathizing and systemizing in the ASC and parent group compared with the modest association found in people with no relatives with ASC could be genetic heterogeneity. The genetic risk for autism is thought to stem from a variety of different sources, including common genetic variants with relatively weak effects (Anney et al., 2010; Arking et al., 2008; Chakrabarti et al., 2009) and rare gene mutations and copy number variations (CNVs) with proportionally larger effects (Levy et al., 2011; Sebat et al., 2007). Although this is not within the scope of the current study, further investigation into whether common genetic variants may help to explain the variation in empathizing and systemizing traits in the general population is warranted. In contrast, rare CNVs and gene mutations with a relatively large effect may be more common in families affected by autism. Previous molecular genetic studies of autism show that rare CNVs thought to have a role in autism etiology can occur de novo (i.e., a new mutation that was not inherited from either parent; Sanders et al., 2011), but can also be transmitted from parent to child (Levy et al., 2011). Further research would benefit from assessing whether gene variants with relatively large effects impact upon both systemizing and empathizing. Such heterogeneous genetic effects, although at present speculative, could possibly explain the strong relationship between empathizing and systemizing in individuals with ASC and parents, compared with the small association observed in control samples.

Alternatively, there may be heterogeneous environmental influences on empathizing and systemizing across the three different groups. As yet little is known about possible influences of environmental effects of autism, with peri and prenatal complications one of the most consistently reported possible environmental risk factors (Kolevzon, Gross, & Reichenberg, 2007). Future research would benefit from direct assessment of the impacts of environmental factors on both systemizing and empathizing in samples with varying degrees of genetic vulnerability for autism.

Limitations

The current study had a number of limitations. The study was restricted in that the parent group contained a larger proportion of mothers (n = 298) than fathers (n = 141), whereas the sex ratio was approximately equal in the other two groups. The sex effects on the mean scores of the variables were accounted for by standardizing for the effect of gender before conducting the factor analyses. Sex differences in latent structure were also explored, indicating that the factor structure obtained does not differ by sex. However, larger numbers in each group would serve to increase power for such types of comparisons. Second, the ASC sample group consisted of high functioning adults with an autism spectrum disorder. As is often the case in cognitive studies of autism (Hoekstra & Whatson, 2010), our study design using questionnaire self-ratings precluded the participation of individuals at the lower functioning end of the spectrum. It is less straightforward to test empathizing and systemizing in individuals on the spectrum who also have intellectual disability. Nevertheless, some characteristics of low functioning autism, such as the relative talent (compared to other abilities) in solving puzzles, a great interest in lawful systems, and increased attention to small changes in the environment all hint toward a drive to systemize (Baron-Cohen et al., 2005), whereas delays and deficits in Theory of Mind development, even when compared with control children of similar mental age, suggest empathy impairments also apply to the lower functioning end of the autism spectrum (Abell, Happé, & Frith, 2000; Baron-Cohen, 1995). However, whether the factor structure between empathizing and systemizing as found in high functioning individuals with autism in our study also generalizes to individuals on the spectrum with intellectual disability remains unknown.

The measures used in this study were all questionnaire based and all concerned self-report. Future research should also incorporate cognitive performance measures and second person ratings of empathizing and systemizing. Of further interest would be to examine the extent to which other behavioral or psychiatric problems commonly found to be comorbid with autism (e.g., attention problems) may moderate the association between empathizing and systemizing.

Because the study was conducted using an online volunteer register it was not possible to verify whether subjects met ASC diagnostic criteria. However, it has been reported that diagnoses in online volunteers are generally reliable (Lee et al., 2010). Furthermore, online data collection enabled the collection of data from a large number of respondents from a representative sample. The use of online research in this sample may also reduce selection bias attributable to the user friendly and noninvasive nature of the research and the difficulty that individuals on the spectrum or those parents taking care of a special needs child may have in attending a face-to-face laboratory setting. A possible drawback is that the sample may have been overrepresented by participants who feel comfortable using computers and are familiar with and interested in taking part in online research.

Conclusions

The current study assessed the latent structure of empathizing, systemizing, and autistic traits across individuals with a low, medium, and high genetic vulnerability to autism. Our results indicated that a two-factor model comprising a latent empathizing and systemizing factor provided the best fit across the three groups. The inverse relationship between both traits was substantial in people with high and medium genetic vulnerability, but only modest in individuals with low genetic risk for autism. We speculate that the varying strength in the association between empathizing and systemizing across groups may be explained by differences in cognitive style and by genetic and possibly environmental heterogeneity. However, further research is needed to establish the impact and causality of these associations.

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Appendix C

The latent structure of cognitive and emotional empathy in individuals with autism, first-degree relatives and typical individuals

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Molecular Autism

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The latent structure of cognitive and emotional empathy in individuals with autism, first-degree relatives and typical individuals

Rachel Grove[1*], Andrew Baillie[1], Carrie Allison[2], Simon Baron-Cohen[2,3] and Rosa A Hoekstra[2,4]

Abstract

Background: Empathy is a vital component for social understanding involving the ability to recognise emotion (cognitive empathy) and provide an appropriate affective response (emotional empathy). Autism spectrum conditions have been described as disorders of empathy. First-degree relatives may show some mild traits of the autism spectrum, the broader autism phenotype (BAP). Whether both cognitive and emotional empathy, rather than cognitive empathy alone, are impaired in autism and the BAP is still under debate. Moreover the association between various aspects of empathy is unclear. This study aims to examine the relationship between different components of empathy across individuals with varying levels of genetic vulnerability to autism.

Methods: Factor analyses utilising questionnaire and performance-based task data were implemented among individuals with autism, parents of a child with autism and controls. The relationship between performance-based tasks and behavioural measures of empathy was also explored.

Results: A four-factor model including cognitive empathy, emotional empathy, social skills and a performance-based factor fitted the data best irrespective of genetic vulnerability. Individuals with autism displayed impairment on all four factors, with parents showing intermediate difficulties. Performance-based measures of empathy were related in almost equal magnitude to cognitive and emotional empathy latent factors and the social skills factor.

Conclusions: This study suggests individuals with autism have difficulties with multiple facets of empathy, while parents show intermediate impairments, providing evidence for a quantitative BAP. Impaired scores on performance-based measures of empathy, often thought to be pure measures of cognitive empathy, were also related to much wider empathy difficulties than impairments in cognitive empathy alone.

Keywords: Empathy, autism, broader autism phenotype, factor analysis

Background

Empathy has been defined as the drive to identify and respond appropriately to emotions and mental states in others [1,2]. It plays a vital role in human relationships and allows an individual to make sense of and predict the behaviour of another [3]. Empathy involves both the ability to recognise and understand emotion in others [3] as well as an affective response to another's emotional state [4,5], respectively cognitive and emotional empathy [4,6].

Autism spectrum conditions (ASC) involve empathy deficits [6-9] and are characterised by communication and social difficulties as well as repetitive behaviours or restricted interests [10]. Empathy dysfunction in autism has been demonstrated via research noting a theory of mind (ToM) impairment in children with ASC [11]; that is, that individuals with autism have difficulty reading the beliefs and intentions of others [11,12]. ToM is often used interchangeably with cognitive empathy, perspective taking and 'mentalising' [13]. However, as noted above, empathy has long been defined as a multifactorial construct including not only the representation of an-

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other's emotional state (ToM or cognitive empathy) but also an affective response (emotional empathy).

The Empathising-Systemising (E-S) theory [14,15] expands the concept of ToM to include this affective component of empathy. The E-S theory argues that the social and communication difficulties seen in ASC can be accounted for by an empathy impairment (including both cognitive and emotional components) and the repetitive behaviours and narrow interests by an inclination for systemising (the drive to understand and derive rules about a system) [16]. A recent factor analytic study by the authors [17] found support for the E-S model. This study, based on the same individuals as the research reported here, identified two factors representing empathy and systemising. These factors were found consistently across individuals with autism, first-degree relatives and general population controls [17]. In concordance with the E-S theory, individuals with ASC showed elevated scores on the latent systemising factor and low scores on the empathy factor. This previous study included questionnaire measures of empathy and systemising only. However, other studies have indicated that individuals with autism also have difficulty with performance-based tasks involving the identification of emotions and perspective taking [18-20]. As these tasks involve the identification of emotion, they are generally conceptualised as performance-based tasks of cognitive empathy.

Although there is much evidence to suggest that individuals with autism display difficulties with ToM or cognitive empathy, there is more debate about the role of emotional empathy in autism. While mirror neuron theory [21] argues that individuals with ASC have weak emotional empathy, Dziobeck and others [22] claim that emotional empathy is intact in autism. Other theorists have proposed that it is due to heightened emotional empathy that individuals with ASC find the social world more challenging, arguing that it is overwhelming rather than difficult to understand [3,23,24].

First-degree relatives of individuals with an ASC diagnosis may also show some mild traits of the autism spectrum [25], also referred to as the broader autism phenotype (BAP) [26,27]. The finding of the BAP fits with the notion that autism is under polygenic influence, and that at least part of these genetic influences are inherited (rather than *de novo* genetic events) and can also be found in undiagnosed relatives displaying the broader phenotype [28]. The BAP has also been shown to apply to empathy, with parents and siblings of affected individuals scoring lower on performance-based tasks involving emotion recognition [20,29-31] and questionnaire measures assessing empathy [20,27]. It is therefore important to examine cognitive and emotional empathy not only in clinical samples, but across the full range of genetic variability, including individuals on the autism spectrum, their relatives and general population controls.

A number of quantitative measures of empathy have been used in previous research, including the Interpersonal Reactivity Index [32] and the Empathy Scale [33]. However, one of the most widely used measures is the Empathy Quotient (EQ) [6], a self-report measure of empathy assessing both cognitive and emotional components. The EQ has recently been studied in detail across three studies. Two studies highlight a three-dimensional structure including cognitive empathy, emotional empathy and social skills [34,35], with the third highlighting a single dimension [36]. The first two studies were based on student and general population samples, with the third including individuals with autism and first-degree relatives. Although individuals with ASC and family members were included in the third study, the factor structure and utility of the EQ was examined for the whole sample and not for each of the three groups (individuals on the spectrum, first-degree relatives and general population controls) separately.

Although the EQ and performance based measures of cognitive empathy have been studied quite extensively by themselves in previous studies, the relationship between subscales of the EQ (a questionnaire-based measure), and performance-based measures of empathy have not been comprehensively assessed to date. The current study aims to evaluate the multifactorial nature of empathy utilising both behavioural and performance-based task data. It was assessed whether the latent structure of empathy differs across samples stratified by genetic vulnerability (individuals with ASC, first degree relatives and controls).

Methods

Participants

Individuals were recruited via two online databases from the Autism Research Centre (www.autismresearchcentre.com) and the Department of Psychology (www.cambridgepsychology.com) at the University of Cambridge. The total sample consisted of 1,034 community-based participants including individuals with ASC (193 males, 170 females; mean age, 36 years; sd, 11), parents of a child with ASC (141 males, 298 females; mean age, 42 years; sd, 8) and general population controls (122 males, 110 females; mean age, 33 years; sd, 10). Individuals who reported no previous psychiatric history were included in the control group. Individuals who had a formal ASC diagnosis were included in the autism group. The control group contained a significantly larger proportion of individuals with an undergraduate degree than the parent and ASC groups (P <0.001). Ethics approval for data collection was given by the Cambridge Psychology Research Ethics Committee and all participants gave informed consent prior to taking part in the study.

Measures

Empathy

The EQ [6] is a self-report measure assessing both cognitive (for example, 'I can tune into how someone else feels rapidly and intuitively') and emotional empathy (for example, 'seeing people cry does not really upset me'). The EQ includes 40 statements with four response options; 'strongly disagree', 'slightly disagree', 'strongly agree', and 'slightly agree'. 'Strongly agree' responses are given 2 points, with 'slightly agree' responses receiving 1 point. Higher scores are indicative of increased levels of self-reported empathy. The EQ shows good test-retest reliability ($r = 0.97$, $P < 0.001$) [6].

Autistic traits

The Autism Spectrum Quotient (AQ) [37] assesses quantitative autistic traits including communication, imagination, attention to detail, social skills and attention switching. Fifty items are assessed on a 4-point Likert scale with response categories 'definitely disagree', 'slightly disagree', 'definitely agree' and 'slightly agree'. Hoekstra and others [38] outline a raw scoring method, with total scores in the range of 50 to 200; higher scores indicating the presence of autistic traits. Previous research has highlighted that the AQ shows good test-retest reliability [37].

A previous factor analysis showed that the AQ can be reliably split into two factors assessing social and non-social autistic traits [38]. A broad social interaction factor was compiled using items assessing communication, social skills, imagination and attention switching (40 items). As the focus of the current study is on empathy, the further 10 items assessing attention to detail or non-social autistic traits were excluded from the current analysis.

Performance tasks

The 'Reading the Mind in the Eyes' test revised [39] is a performance task designed to assess how well an individual can read another's emotion based on viewing the eye area alone. This measure has been described as an advanced 'theory of mind' task that assesses the ability to attribute mental states to oneself and others (i.e. cognitive empathy). Individuals are presented with a series of 36 photographs of the eye region of the face and asked to choose which of four words best describes the emotion depicted. The emotions used in the task are subtle and include, for example, a choice between jealous, panicked, arrogant and hateful. This test has been shown to detect meaningful individual differences, with individuals with AS or HFA scoring significantly lower than general population controls [39].

The Karolinska Directed Emotional Faces (KDEF) [40] is another task designed to assess the recognition of more basic emotions in others. In this modified version, participants were shown 140 photographs of faces expressing seven emotions (happy, sad, angry, afraid, disappointed, surprised and neutral). For each photograph, individuals were asked to select which of the seven emotions best described the emotion depicted. Results provide indications of accuracy and response time for each facial expression. Accuracy adjusted response time was calculated by dividing the mean response time for correct items by the proportion of items answered correctly. Weighted mean reaction times have been shown to be a more sensitive measure, taking any potential speed-accuracy trade off into account [41]. The KDEF has good test-retest reliability and has been validated on emotional content, intensity and arousal [42]. Individuals with autism have been shown to score lower than controls on this task [20]. To aid data interpretation, the KDEF was rescored so that lower values indicate higher accuracy adjusted response time and hence lower empathy ability.

Given that sex differences on the mean test scores were not the focus of this paper and have been reported elsewhere [20,38,39], any effects of sex and age on the mean test scores were regressed out prior to factor analysis. This enabled the comparison of the factor structure of empathy without the confound of sex differences on the mean. Furthermore, the standardisation of the items allowed for any differences in variance between the items of the EQ and the Eyes and KDEF tasks to be accounted for (as standardisation resulted in all variables having a mean of 0 and a variance of 1).

Analytic strategy

Previous research has shown that the EQ can be split into three factors: cognitive empathy, emotional empathy and social skills [34,35]. Although finding a comparable factor structure, these two papers showed differences in the number of items loading onto each latent factor. Allison et al. [36] have also explored the EQ in depth, highlighted a single dimension using Rasch analyses. The first stage of our analyses focused on determining the most appropriate factor structure for the EQ in the current sample.

Confirmatory factor analyses (CFA) were conducted in Mplus Version 7 using the maximum likelihood estimator [43]. Confirmatory models allow for a more direct test of previous models of empathy as well as greater control over model specification. The first set of analyses assessed the fit of a one-factor 26-item model (following the model identified by Allison et al. [36]) across: (1) individuals with autism; (2) parents; and (3) general population controls (Models 1 to 3). Following this, three-factor models assessing cognitive empathy, emotional empathy and social skills were estimated based on Lawrence et al.'s [34] 28-item model (Models 4 to 6) and Muncer et al.'s [35] 15-item model (Models 7 to 9). The best fitting model identified in each of the three groups separately was then subjected to multiple group analysis to determine whether the same latent structure holds across individuals with autism, parents and controls (Models 10 and 11). In all subsequent analyses, the model that best described the EQ data across all three groups, a three-factor model including factors assessing cognitive empathy, emotional empathy and social skills (see Results section) was utilised.

Following the analysis of the EQ alone, the study of the latent structure of empathy was extended by also including the AQ, Eyes and KDEF measures in the factor analysis. The AQ was not submitted to rigorous individual investigation as it has previously been studied extensively [37,38,44-48].

First, a series of three-factor models (with latent factors Cognitive empathy, Emotional empathy and Social skills) were tested. The social interaction factor of the AQ was predicted to load on the Social skills factor due to the similarity between the content of the AQ items and the EQ items loading on this factor. The Eyes and the KDEF scores were expected to load onto the Cognitive empathy factor of the EQ as these two performance tests are thought to measure cognitive empathy (Models 12 to 14). Second, a series of four-factor models were estimated, in which the Eyes and KDEF scores loaded on to a separate fourth measurement factor representing performance-based assessment of empathy, rather than on the Cognitive empathy factor (Models 15 to 17). Multiple group models were used to determine whether the same structure was present among individuals with autism, parents and general population controls. The first multigroup CFA allowed all parameters to vary across the three groups (Model 18). A further model constraining the factor loadings to be equal across groups was also tested (Model 19).

In order to evaluate the possible impact of sex differences on the latent structure of empathy, three further models incorporating six groups based on genetic vulnerability (ASC *vs.* parents *vs.* controls) and sex (males *vs.* females) were assessed using multigroup CFA (Models 20 to 22). As before, these models were run using test scores corrected for any mean sex (and age) differences, to ensure that these models focused on possible sex differences in latent structure, rather than sex differences in mean test scores. A number of fathers in the dataset had missing data on the performance-based tasks (n = 104). In order to account for the effect of this missing data on the results, all six-group analyses were run both by imputing the data for these individuals as well as excluding these individuals for comparison.

Model fit was evaluated using the Bayesian information criterion (BIC) [49], Sample size adjusted BIC (SSABIC) [50], Akaike information criterion (AIC) [51], Tucker-Lewis index (TLI) [52], Comparative fit index (CFI) [53] and the Root mean square error of approximation (RMSEA) [54]. The BIC, SSABIC and AIC are used to assess model fit, with lower values reflective of a more parsimonious model. TLI and CFI compare the model under investigation with the null model, with CFI and TLI values > = 0.95 indicating very good fit and values > = 0.90 representing adequate fit [55,56]. The RMSEA is a fit index that allows for modelling with large sample sizes. RMSEA values <0.08 indicate adequate fit, with values <0.05 suggesting excellent fit [57]. Evaluation of model fit also included the interpretability of all other parameter estimates. Comparison of the nested models was based on chi-square difference tests. These have been shown to result in less type one error when the maximum likelihood estimator is implemented [58].

Results

Factor analyses of empathy as assessed by items of the EQ

Model fit indices ascertained from the CFA models are given in Table 1. The model describing a one-factor solution of the EQ data, following Alison et al.'s [36] model, displayed poor fit in all three groups (Models 1 to 3 in Table 1). Similarly, fit indices based on Lawrence et al.'s [34] three-factor model of the EQ were below recommended thresholds (Models 4 to 6). The three-factor model of the EQ based on Muncer et al. [35] provided the best fit to the data (Models 7 to 9). Multigroup CFA analyses indicated that this model displayed good fit across individuals with autism, parents and general population controls (Model 10). A model in which the factor loadings were constrained to be equal across the three groups (Model 11) resulted in a significantly poorer fit compared with Model 10 ($\chi^2 = 114.1$, $P < 0.001$). These findings suggest that the EQ assesses three constructs (Cognitive empathy, Emotional empathy and Social skills) in controls, parents and adults on the autism spectrum, but the association between each of these latent constructs is somewhat different across the three groups. For example, the factor correlations in the ASC group were higher than controls. This may account for why Model 11 did not provide a good fit to the data.

Factor analyses including both behavioural and performance-based measures of empathy

Next, the Eyes and KDEF tasks and the social interaction factor of the AQ were included in Muncer et al.'s [35] three-factor model of empathy. First, it was tested whether the performance-based tasks solely assess cognitive empathy, by including these two variables in the Cognitive empathy factor (Models 12 to 14). In these models, factor loadings of the Eyes and KDEF on the cognitive empathy factor were not statistically significant. This poor fit was also reflected in some of the fit indices, with CFI and TLI values under the recommended threshold in the ASC group. Second, a model in which the KDEF and Eyes data

Table 1 Fit indices and model comparisons

Model	Description	Fit indices							
		AIC	BIC	SSABIC	RMSEA	CFI	TLI	χ^2	$\Delta\chi^2$ (*df*)
One-factor models EQ items (Allison et al., 2011 [36])									
1	1f control group (n = 232)	13,673.951	13,942.797	13,695.578	0.090	0.673	0.644	865.2[a]	
2	1f parent group (n = 439)	25,650.600	25,969.013	25,721.480	0.083	0.833	0.818	1,193.9[a]	
3	1f autism group (n = 363)	22,451.458	22,755.222	22,507.762	0.087	0.748	0.726	1,123.5[a]	
Three-factor models EQ items (Lawrence et al., 2004 [34])									
4	3f control group (n = 232)	14,020.059	14,319.925	14,044.181	0.078	0.781	0.761	833.2[a]	
5	3f parent group (n = 439)	26,186.672	26,541.825	26,265.731	0.071	0.883	0.873	1,114.3[a]	
6	3f autism group (n = 363)	23,768.627	24,107.440	23,831.427	0.080	0.802	0.785	1,163.0[a]	
Three-factor models EQ items (Muncer et al., 2006 [35])									
7	3f control group (n = 232)	7,731.634	7,897.077	7,744.943	0.048	0.942	0.930	132.7[a]	
8	3f parent group (n = 439)	14,798.686	14,994.632	14,842.304	0.060	0.950	0.939	222.0[a]	
9	3f autism group (n = 363)	13,040.850	13,227.781	13,075.499	0.055	0.932	0.918	184.3[a]	
Three-factor multigroup models EQ items (Muncer et al., 2006 [35])									
10	3f multigroup all estimates vary (n = 1,034)	35,574.089	36,166.916	35,785.782	0.056	0.938	0.931	589.9[a]	
11	3f multigroup equal factor loadings (n = 1,034)	35,628.149	36,072.769	35,786.918	0.060	0.921	0.921	703.9[a]	114.1 (30) $P < 0.001$
Three-factor model of cognitive empathy (EQ subscale, Eyes and KDEF), emotional empathy (EQ subscale) and social skills (EQ subscale and AQ_soc)									
12	3f control group (n = 232)	8,331.507	8,527.971	8,347.312	0.044	0.945	0.936	1,202.4[a]	
13	3f parent group (n = 439)	16,721.291	16,954.108	16,773.218	0.055	0.947	0.938	3,463.6[a]	
14	3f autism group (n = 363)	19,949.121	20,171.102	19,990.266	0.067	0.894	0.877	2,162.7[a]	
Four-factor model of cognitive empathy (EQ subscale), emotional empathy (EQ subscale) social skills (EQ subscale and AQ_soc) and performance-based empathy (Eyes and KDEF)									
15	4f control group (n = 232)	8,323.897	8,530.701	8,340.533	0.040	0.955	0.946	176.4[a]	
16	4f parent group (n = 439)	16,712.141	16,957.074	16,766.664	0.055	0.949	0.939	298.8[a]	
17	4f autism group (n = 363)	15,687.606	15,921.270	15,730.916	0.053	0.934	0.922	262.5[a]	
Four-factor multigroup models of cognitive empathy (EQ subscale), emotional empathy (EQ subscale) social skills (EQ subscale and AQ_soc) and performance-based empathy (Eyes and KDEF)									
18	4f multigroup all estimates vary (n = 1,034)	40,743.301	41,494.361	41,011.591	0.052	0.938	0.932	808.0[a]	
19	4f multigroup equal factor loadings (n = 1,034)	40,894.787	41,467.965	41,099.535	0.061	0.909	0.907	1,031.5[a]	223.5 (36) $P < 0.001$
Four factor multigroup models specifying sex effects (6 groups)									
20	4f multigroup all estimates vary (n = 1,034)	40,757.204	42,190.149	41,269.074	0.058	0.923	0.916	1,332.5[a]	
21	4f multigroup equal factor loadings and equal variance estimates (n = 1,034)	40,873.332	41,861.570	41,226.345	0.066	0.891	0.893	1,628.6[a]	296.1 (90) $P < 0.001$
22	4f multigroup equal factor loadings and free variance (n = 1,034)	40,745.441	41,832.503	41,133.757	0.059	0.914	0.914	1,460.7[a]	128.2 (70) $P < 0.001$

[a] $P < 0.001$.

AIC, Akaike information criteria; AQ_soc, social interaction factor of the Autism Quotient; BIC, Bayesian information criteria; CFI, Comparative fit index; Eyes, Items correct on Reading the Mind in the Eyes Task; KDEF, Weighted mean reaction time on the Karolinska Directed Emotional Faces task; RMSEA, Root mean square error of approximation; SSABIC, Sample size adjusted BIC; TLI, Tucker-Lewis index; χ^2, chi square statistic; $\Delta\chi^2$ (*df*), chi square difference test.

loaded onto a separate 'performance-based test factor' was implemented. This four-factor model (including factors Cognitive empathy, Emotional empathy, Social skills and a Performance-based test factor) provided a good fit to the data in all individual groups (Models 15 to 16) as well as within the multigroup analysis (Model 18). Again, the model in which the factor loadings were constrained to be equal across the three groups (Model 19) resulted in a significantly poorer fit compared to the freely estimated model (Model 18) ($\chi^2 = 223.5$, $P < 0.001$).

Assessing sex differences in the factor structure of empathy

Lastly, the impact of sex on the factor structure of empathy was explored by running six-group analyses for the best-fitting model identified, the four-factor model including factors Cognitive empathy, Emotional empathy, Social skills and a Performance-based test factor. As the effects on the mean test scores were regressed out prior to analysis, these models (Models 20 to 22) focus on sex differences in factor structure rather than in mean scores. In order to account for the effect of missing data on the performance-based tasks, all six-group analyses were also run both by imputing data for and excluding these individuals. There were no substantive changes in any of the analyses, indicating that this is not a confounding factor in the interpretation of the results. Model 20 with all estimates free to vary provided the best fit to the data for the six groups. However, comparison with the best fitting three-group multigroup model (Model 18) indicates that there is no significant difference between the latent structure of empathy when sex is taken into account. Therefore, Model 18, the four-factor model with equal form among the three groups, allowing the factor loadings to vary provided the best fit to the data.

Empathy factor means and correlations in individuals with ASC, parents and controls

Parameter estimates for the four-factor model taken from the Model 18 analysis are given in Figure 1. All items loaded significantly onto their respective factors ($P < 0.05$). Mean differences between scores on the latent factors across the three groups are given in Table 2. Parents scored significantly lower than controls on Cognitive empathy, Emotional empathy and Social skills latent factors as well as on the performance-based tasks. Individuals with autism also scored significantly lower than controls on all four latent factor means. There was a significant difference between parents and individuals with ASC on Cognitive empathy, Emotional empathy and Social skills factors. However, these two groups scored similarly on the performance tasks (Mean difference, -0.30, $P > 0.05$). Please note that the current paper focused on latent factor means. Group differences on means of the different tasks under study have been reported elsewhere [6,20,27,37-39,59].

The correlation between the performance-based test factor and the other empathy factors varied by sample group (see Table 3). In the control group, the performance-based tasks were not significantly correlated with any other empathy factor. However, in both the parent and ASC group these tasks were significantly correlated with Cognitive empathy, Emotional empathy and Social skills. These correlations were of similar magnitude for all factors. To verify that these different correlation patterns

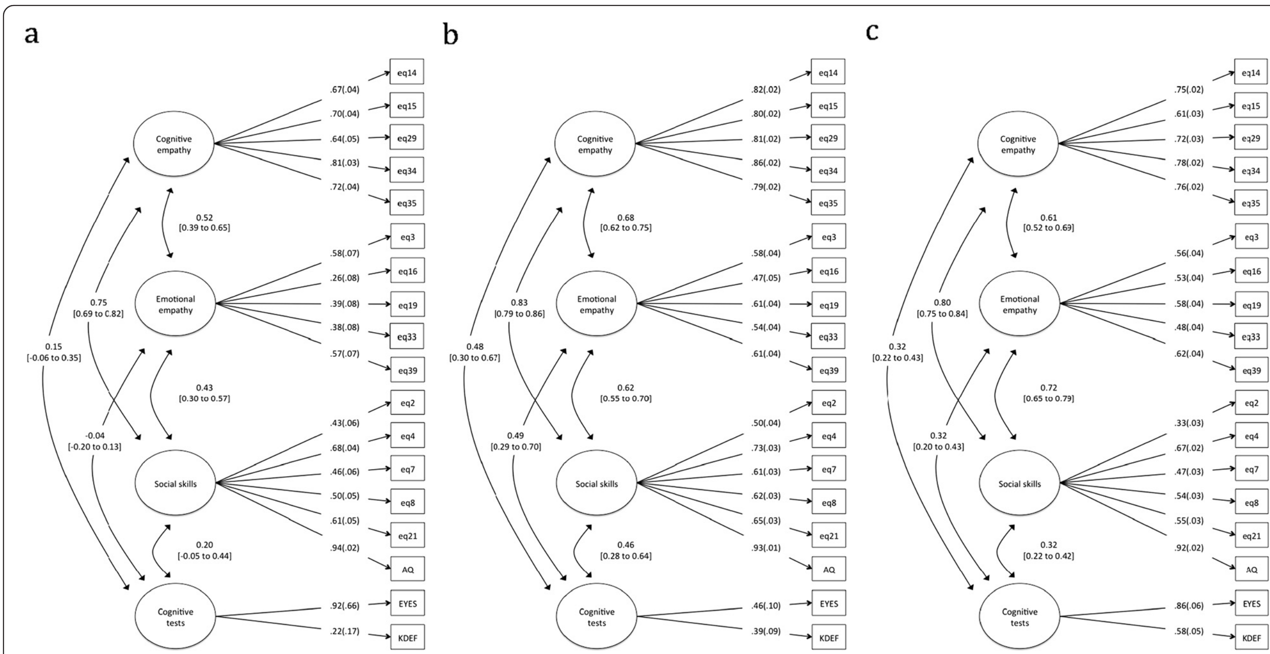

Figure 1 Four-factor multigroup model for (a) controls, (b) parents and (c) ASC.

Table 2 Mean differences on factors scores in the multigroup CFA model

	Cognitive empathy	Emotional empathy	Social skills	Cognitive tests
	Mean (95% CI)	Mean (95% CI)	Mean (95% CI)	Mean (95% CI)
Control group (n = 232)	-0.04 (-0.16 to 0.08)	-0.06 (-0.16 to 0.04)	-0.03 (-0.15 to 0.10)	0.01 (-0.11 to 0.13)
Parent group (n = 439)	-0.36[a] (-0.51 to -0.22)	-0.27[b] (-0.44 to -0.10)	-0.38[a] (-0.52 to -0.24)	-0.29[b] (-0.55 to -0.04)
Autism group (n = 363)	-2.01[a] (-2.25 to -1.78)	-1.21[a] (-1.42 to -0.99)	-2.76[a] (-3.05 to -2.46)	-0.59[a] (-0.74 to -0.44)

[a]$P < 0.01$ [b]$P < 0.05$ significantly different to controls.

between the performance-based test factor and the other empathy factors in the groups could not be explained by differences in score distributions on the performance-based tasks, the distributions of the KDEF and Eyes tasks were inspected. Both tasks showed very similar distributions in the control and parent groups. The differences in the correlation patterns are therefore unlikely to be due to differences in the test score distributions.

Discussion

Factor analyses in data from a large sample of individuals with ASC, parents and controls, using both questionnaire and performance-based measures of empathy, suggested a four-factor latent structure of empathy encompassing Cognitive empathy, Emotional empathy, Social skills and a Performance-based measurement factor. This structure was consistent across individuals deemed to have a high (individuals on the autism spectrum), medium (parents) or low (controls) genetic vulnerability for autism, indicating that the overall latent structure of empathy is consistent across both clinical and general population samples. However, there were some differences in the factor loadings and factor correlations across the three groups.

The latent structure identified in this study is consistent with previous research in that it identifies both a cognitive and emotional component of empathy [3,4,13,22]. In addition, the analyses also identified a separate Social skills factor. Items measured by the Social skills factor of the EQ assess specific empathising skills within a social situation. For example, 'I find it hard to know what to do in a social situation' and 'I often find it difficult to judge if something is rude or polite'. Future research utilising other measures is needed to further assess the theoretical implications of this Social skills factor, which is shown to be separate from cognitive and emotional empathy.

It was expected that the performance-based emotion recognition tasks would be related to the Cognitive empathy factor. However, factor loadings of the Eyes and KDEF on the Cognitive empathy factor were low, with a model including a separate performance-based task component providing a better fit. Interestingly, the relationship between the Performance-based test factor and the other empathy factors was different across the three groups under study. In the control group, the performance tasks were not significantly correlated with any of the questionnaire-based empathy factors. Within parents and individuals with autism, the performance measures were related in almost equal magnitude to all three components, rather than solely to cognitive empathy. The finding that these performance tasks do not directly and exclusively assess cognitive empathy is new. Previous research has operated on the assumption that these tasks are performance-based measures of cognitive empathy. The findings of the current study indicate that rather than being a direct measure of cognitive empathy, scores on performance-based tasks like the Eyes and the KDEF have a bearing on empathy more widely. Our results suggest that completion of either of these tasks requires engagement of more than just cognitive empathy abilities. Rather, impairment on these performance-based tasks is indicative of a broader impairment across all facets of empathy. This has important implications for future research involving the implementation of such tasks.

Individuals with autism showed greater impairment (as indexed by lower mean latent factor scores) across the Cognitive and Emotional empathy, Social skills and Performance-based empathy factors compared with controls. Similarly, the ASC group displayed greater impairment than parents across all factor means, with the exception of the Performance-based factor. This fits with the notion of autism as a disorder of empathy [7-9]. In

Table 3 Correlation between the cognitive test factor and the other components of empathy

	Cognitive empathy	Emotional empathy	Social skills
	r (95% CI)	*r* (95% CI)	*r* (95% CI)
Control group (n = 232)	0.15 (-0.10 to 0.39)	-0.04 (-0.23 to 0.17)	0.20 (-0.10 to 0.49)
Parent group (n = 439)	0.48[a] (0.27 to 0.70)	0.49[a] (0.25 to 0.74)	0.46[a] (0.25 to 0.68)
Autism group (n = 363)	0.32[a] (0.20 to 0.45)	0.32[a] (0.18 to 0.45)	0.32[a] (0.20 to 0.44)

[a]$P < 0.01$.

contrast with some previous research [3,22-24], there was no evidence that individuals with autism exhibited intact or heightened emotional empathy.

Parents also showed mild impairment across all four factors compared to controls. However, with the exception of the Performance-based factor, impairment was not as strong as observed in the ASC group, placing their difficulties somewhere in between the clinical and the control group. This is consistent with previous accounts indicating that first-degree relatives show some difficulties on tasks of empathy [20] compared with controls. Moreover, it fits with the notion that characteristics related to autism are distributed as quantitative traits rather than discrete entities [37,60] and are likely to be influenced at least in part by common genetic variation [28].

Limitations

As mentioned previously, a number of fathers had missing data on the Eyes and KDEF tasks (n = 104). To assess whether these missing data had any effect on the results, all analyses were run both by imputing data for these individuals as well as excluding the missing cases. As there were no substantive changes within any of the models, it is highly unlikely that these missing data were a confounding factor.

The parent group also consisted of a larger proportion of mothers (n = 298) than fathers (n = 141). To ensure these differences would not bias the analyses, any sex effects on the means were regressed out prior to conducting the factor analyses. Moreover, the evaluation of sex differences in the latent factor structure indicated that it was similar across both sexes. Future studies including very large sample sizes would be of interest, as these could explore any possible sex difference in the latent factor structure in more detail than the current sample size permitted.

Lastly, the control group included in this study had completed a somewhat higher level of education than the parent and ASC groups. We can therefore not exclude the possibility that differences in educational level may explain some of the differences in factor structure of empathy observed between controls and the parent and ASC groups.

Conclusions

The current study assessed the latent structure of empathy across individuals with a low, medium and high genetic vulnerability to autism. Results highlighted that empathy shows evidence of multidimensionality, in which four factors can be distinguished irrespective of genetic vulnerability, including three components of empathy and a performance-based factor. Unexpectedly, performance-based measures of empathy were related in almost equal magnitude to Cognitive empathy, Emotional empathy and Social skills, rather than solely to Cognitive empathy. This has implications for the nature of impairment indicated by performance on such tasks, suggesting that these effects are much wider than impairments in cognitive empathy alone. Individuals with autism displayed impairment on all four components of empathy, confirming the notion that autism is characterised by difficulties with multiple facets of empathy. Parents showed intermediate impairments of empathy, providing evidence for the BAP and highlighting the importance to assess characteristics of autism on quantitative scales.

Abbreviations

AIC: Akaike information criterion; AQ: Autism Spectrum Quotient; AQ_soc: Social interaction factor of the Autism Spectrum Quotient; ASC: Autism spectrum conditions; BAP: Broader autism phenotype; BIC: Bayesian information criterion; CFA: Confirmatory factor analysis; CFI: Comparative fit index; E-S theory: Empathising-Systemising Theory; EQ: Empathy Quotient; Eyes: Reading the Mind in the Eyes test revised; KDEF: Karolinska Directed Emotional Faces; RMSEA: Root mean square error of approximation; SSABIC: Sample size adjusted Bayesian information criterion; ToM: Theory of Mind; TLI: Tucker Lewis index.

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

RG, RH and AB designed the study. Data reported on in this study were collected by CA and SB-C. RG undertook all statistical analyses and drafted the manuscript. All other authors contributed to subsequent drafts and the final submitted manuscript. All authors read and approved the final version of the manuscript.

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Appendix D

Ethics Approval Letter

MACQUARIE UNIVERSITY

Ethics Secretariat <ethics.secretariat@mq.edu.au>

Ethics application ref: 5201100956 - External Approval Noted

Ethics Secretariat <ethics.secretariat@mq.edu.au> **Tue, Dec 13, 2011 at 10:22 AM**
To: A/Prof Andrew Baillie <andrew.baillie@mq.edu.au>
Cc: Ms Rachel Louise Grove <rachel.grove@students.mq.edu.au>

Dear A/Prof Baillie

Re: "The latent structure of the autism phenotype"

The above application was considered by the Executive of the Human Research Ethics Committee. In accordance with section 5.5 of the National Statement on Ethical Conduct in Human Research (2007) the Executive noted the final approval from the University of Cambridge Psychology Research Ethics Committee and your right to proceed under their authority.

This notification also applies to Ms Rachel Grove who will be conducting the research for the purposes of a combined Masters and PhD.

Please do not hesitate to contact the Ethics Secretariat at the address below, if you require a hard copy letter of the above notification.

Please retain a copy of this email as this is your official notification of external approval being noted.

Please do not hesitate to contact the Ethics Secretariat if you have any questions or concerns.

Yours sincerely

Dr Karolyn White
Director of Research Ethics
Chair, Human Research Ethics Committee