

Quality of Life Amongst Adults with a Childhood Diagnosis
of Autism and a Childhood IQ Outside the Intellectual
Disability Range

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Thesis declaration form

I confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

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Overview

This thesis considers the predictors and correlates of outcome amongst higher ability adults with Autism Spectrum Disorder (ASD). The literature review (Section one) critically appraises studies which have reported on the adult outcome of higher ability adults with ASD using the more traditionally used, objective adult social outcome measure. It examines which child and adult factors (specifically language level, intellectual ability and severity autism symptomatology) are associated with adult social outcome. The impact of methodological differences between and within studies on findings is addressed followed by discussion of the appropriateness of the adult social outcome measure, in isolation, as a way of determining how an individual with ASD is functioning in adulthood.

The empirical paper (Section two) considers an alternative measure of outcome, quality of life (QoL), with a sample of individuals who were diagnosed with autism in childhood and had a childhood IQ outside the intellectual disability range. It examines whether it is possible to determine which child and adult factors are associated with this more subjective measure of outcome and whether there is a difference between informant perceived and self-reported QoL scores.

The critical appraisal (Section three) evaluates the process of conducting a literature review and empirical study on these topics. It considers how decisions were guided by previous experiences, what methodological factors affected the process, and how these issues relate to conducting research with the wider ASD population.

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Part 1: Literature Review

Factors associated with Adult Social Outcomes Amongst Adults With Autism Spectrum
Disorders and an IQ Outside the Intellectual Disability Range

1.1 Abstract

Aim: To examine the factors associated with adult social outcome amongst higher ability adults with Autism Spectrum Disorders (ASD).

Method: Pubmed and Psycinfo databases were searched using the terms (i) ‘autis*’ or ‘Asperger’ and (ii) ‘outcome*’ or ‘follow-up’ in the title. ‘Adult’ was entered as a keyword to appear in any field. The search identified 1038 papers, of which 45 were selected based on their titles. Examination of the abstracts and full texts (where necessary) of these 45 papers resulted in 10 papers being selected for inclusion in the review. A further two papers were included from previous reviews.

Results: Three factors (language, IQ and autistic symptomatology) have been repeatedly demonstrated to be associated with adult social outcome in this population.

Conclusion: Child (and adult) language level, IQ and severity of autistic symptomatology are highly predictive of adult social outcome amongst higher ability adults with ASD. However, there remains a lack of consensus regarding which aspects of these three variables are most strongly associated with outcome. Furthermore, it is difficult to make predictions about outcome based on individual scores.

1.2 Introduction

1.2.1 Background to Autism Spectrum Disorders (ASD)

Leo Kanner (1943) originally used the term ‘autism’ to describe 11 children with “extreme autistic aloneness” (p. 242), “an anxious obsessive desire for the maintenance of sameness” (p. 245) and a “limitation in the variety of spontaneous activity” (p. 246). He later termed this syndrome “early infantile autism” (1956). Within a year of Kanner’s initial paper, Hans Asperger also published a paper describing a very similar group of individuals (1944). However, being published in German, his work remained largely unrecognised until Wing (1981) used the term ‘Asperger Syndrome’ (AS) to describe those individuals with autistic symptomatology who did not have language difficulties and were not socially aloof. AS was subsequently entered into the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV; American Psychiatric Association, 1994) and (ICD-10; World Health Organisation, 1993).

It is clear, even from these initial publications that a spectrum of severity existed. Both Kanner (1956) and Asperger (1944) described very similar disorders but there remained some clear differences between and within their accounts. In line with this, Wing and Gould (1979) proposed that autism was on a “continuum of severity” (p. 26). This is now a widely recognised concept, as demonstrated by the present day use of the term ASD within the new DSM-5 criteria (American Psychiatric Association, 2013a). At one end of this spectrum are individuals with autism and a low IQ and at the other end are individuals with high functioning autism ($\text{IQ} \geq 70$) or AS (see Section 1.2.2 for a discussion of these terms) and it is the latter ‘sub-group’ that will be the focus of the current review.

The variability in overall functioning in later life within this population was first queried by Kanner (1973). In observing the range of outcomes that his initial follow-up group displayed, he questioned what factors might be able to account for these differences. Thus, even since the earliest descriptive studies, the question of what predicts outcome in later life for individuals with ASD has been raised.

1.2.2 Defining ASD

The DSM-IV-TR criteria (American Psychiatric Association, 1994) previously defined ASD as a pervasive developmental disorder that is characterised by impairments in (i) social interaction, (ii) communication and (iii) restricted, repetitive and stereotyped patterns of behaviour. Additionally, these impairments must have occurred before three years of age.

These criteria have been modified in the DSM-5 (American Psychiatric Association, 2013). There are now two, not three, domains of impairment, which lie within a single diagnostic category, ASD; (i) social communication and interaction across contexts and (ii) restricted, repetitive patterns of behaviour, interests or activities. Additionally, symptoms must (i) be present in early development, (ii) lead to clinically significant levels of impairment in functioning and (iii) cannot be better explained by intellectual disability. ASD can also occur with or without intellectual and / or language impairment. This final criterion is particularly pertinent with regard to the distinction between AS and high-functioning autism.

High functioning autism and AS were originally distinguished from other ASDs by the presence of normal cognitive skills and from each other by an absence of early

language delays in AS (American Psychiatric Association, 2000). However, research now suggests that there is very little difference between the two groups (Howlin, 2003; Macintosh & Dissanayake, 2004; Witwer & Lecavalier, 2008). This is supported by the DSM-5 criteria, which states that a language delay is not an intrinsic part of the ASD construct, rather a factor affecting clinical symptoms (American Psychiatric Association, 2013). Consequently, AS has been removed from the DSM-5 criteria. In line with these new criteria, in this literature review, the term ASD refers to all individuals who fall within the autism spectrum. The term ‘higher ability ASD’ will be used throughout to refer specifically to individuals with a diagnosis of autism and an $IQ \geq 70$ (i.e. outside the intellectual disability range) or those who previously received a diagnosis of AS.

The term ‘higher ability’ is commonly used in the ASD literature to distinguish between those without a learning disability (i.e. $IQ \geq 70$) and the vast majority of the ASD population, who have significant learning difficulties. The cut-off of an $IQ \geq 70$ is in line with the DSM-V criteria for an intellectual disability (Association, 2013b) and current inclusion criteria for learning disability services in the NHS today. Additionally, the term ‘higher ability’ was used as it was considered to be a more appropriate way of describing this population than the more commonly used ‘high functioning autism’. This is because having an IQ outside the intellectual disability range does not necessarily mean that individuals are functioning highly in adulthood.

1.2.3 Background to the literature review

1.2.3.1 Early descriptive studies

The term ‘adult social outcome’ refers to an individual’s ability to cope in the adult world. Specifically, their achievements in employment, relationships and independent

living (Howlin et al., 2004; see Section 1.3.2 for details). Whilst the high lifetime dependency of individuals with ASD and a low IQ is widely recognised (Nordin et al., 1998), less is known about the outcomes for those individuals with normal cognitive ability.

In the earliest descriptive outcome studies that included higher ability individuals, Asperger and Kanner both reported on the potentially good outcomes for the sub-group of individuals with autism who were of higher ability. Asperger (1944) began to notice a link between IQ and outcome:

“one might expect...that social integration of autistic people is extremely difficult if not impossible...This bleak expectation, however, is born out only in a minority of cases and, in particular, almost exclusively in those people with considerable intellectual retardation in addition to autism...This is not so with intellectually intact autistic individuals” (p. 87, annotated translation of Asperger's initial paper, Frith, 1991).

Similarly, Kanner (1973) identified 11 out of 96 children who were “now in their twenties and thirties, mingling, working, and maintaining themselves in society” (p. 211). He proposed that some individuals can achieve more highly than others in adulthood and queried what affected this variability.

1.2.3.2 Adult social outcome in higher ability samples

Since these early descriptive studies, most outcome research has used a form of the adult social outcome measure (Howlin, Goode, Hutton, & Rutter, 2004), which was derived from the initial outcome ratings developed by Rutter et al. (1967). The current version (see Howlin, Moss, Savage, & Rutter, 2013; Howlin, Savage, Moss, Tempier, & Rutter,

2014), assesses employment, living status, relationships and friendships. Individuals are then placed into one of five outcome categories; ‘very good’, ‘good’, ‘fair’, ‘poor’ and ‘very poor’ depending on their scores on each of the domains assessed. An individual with a ‘very good’ outcome is likely to be achieving highly in all areas whereas someone with a ‘very poor’ outcome will have pervasive difficulties across all four categories.

The research to date has produced variable outcome results. The majority of studies have reported quite negative outcomes for their samples; Howlin et al. (2004) found that 74% of participants had a poor outcome. Some studies have reported relatively good outcomes for higher ability individuals with ASD; Farley et al. (2009) found that 48% of their sample had a ‘good’ or ‘very good’ outcome. However, even within studies that are reporting slightly more positive results, individuals with a ‘good’ or ‘very good’ outcome rarely rose above 50% (e.g. Farley et al., 2009; Rumsey, Rapoport, & Sceery, 1985; Szatmari, Bartolucci, Bremner, Bond, & Rich, 1989).

Comparisons between studies are hampered by various methodological issues including sample size, age, IQ, a lack of or inappropriate use of control groups, inconsistencies in assessment and diagnosis, and an overall discrepancy in the representativeness of samples. However, despite this, the overarching conclusion has been that the majority of higher ability individuals do not fare well in adulthood, with over half of participants having a ‘poor’ or ‘very poor’ outcome (Howlin & Moss, 2012).

These conclusions, regarding poor outcomes, make progression into mid- to late-adulthood a major concern for many families (Howlin, 2007); parents become increasingly concerned about what will happen when they are no longer able to support their children (Eaves & Ho, 2008; Howlin, 2004). A clearer understanding of what

factors are associated with outcome will improve our ability to inform families about the likely prognosis for their child with ASD and aid the development of effective intervention strategies.

1.3 Method

The current literature review examines which factors are associated with adult social outcome amongst higher ability adults with ASD. In this review, ‘predictors’ refers to early factors; typically childhood variables whereas ‘correlates’ refers to more current factors that are usually examined in adulthood at the same time that adult social outcome is assessed. The review intentionally has a narrow focus on social functioning in adulthood, as this is a prominent clinical issue (see Section 1.5.3 for details). Broadening the review further would have created an exceptionally large review, incorporating many types of outcome including autism symptomatology, cognitive and language ability and mental health.

1.3.1 Inclusion criteria

The following inclusion criteria were employed:

1. All studies were published in English and constituted complete articles from peer reviewed journals.
2. Participants received a diagnosis of autism or AS in childhood (in early papers other terms may have been used) by experts in the field / professionals who were trained in the diagnosis of ASD. Often, participants were diagnosed prior to the

development of diagnostic tools. Confirmation of diagnosis at a later date using standardised tools was preferable, but not essential¹.

3. Sample size greater than or equal to 10.
4. Measurement of outcome beyond 16 years for the majority of participants.
5. A rating of adult social outcome was conducted. Ideally, these ratings were based on the initial measure described by Rutter et al. (1967; 1967). When this was not the case, assessment of similar areas of functioning (for example, living and employment status, friendships and relationships) was sufficient, as were scores on the Vineland Adaptive Behavior Scale (VABS; Sparrow, Cicchetti, & Balla, 2005).
6. Inclusion of early and/or current factors that could be examined against adult social outcome scores.
7. Mean IQ of the sample was greater than or equal to 70 (see Section 1.3.3 for details).

1.3.2 *Search process*

The review is based on a search of articles published prior to 29th December 2014.

PubMed and PsycInfo databases were searched using the following terms in the title: (i) *autis** or *Asperger* and (ii) *outcome** or *follow-up*. The term ‘adult’ was also entered as a keyword to appear in any field. This narrow range of terms was generated based on the very specific inclusion criteria described in Section 1.3.1 and ensured that the maximum

¹ The requirement for a diagnosis of autism or AS was deliberate given the focus on higher ability ASD. Therefore, other specific terms such as Pervasive Developmental Disorder – Not Otherwise Specified (PDD-NOS) and/or more general terms including neurodevelopmental disorder were not included in the search. Studies involving participants with *only* these diagnoses were excluded.

possible number of suitable papers was obtained from the search. Figure 1.1 describes the paper selection process. The titles of the 1038 papers that were generated from the initial search, were screened for relevance. Following this, the abstracts, and where necessary full texts, were reviewed; resulting in 10 papers being selected. A further two papers were added from a recent narrative review (Howlin & Moss, 2012). Another review by Magiati, Tay and Howlin (2014), which addressed the wider topic of outcomes in general (not just social) and did not systematically examine predictors and correlates of outcomes, was also checked for relevant papers but no additional papers were selected. Most of the papers that were excluded at stage two focused on a different type of outcome, only included individuals with an intellectual disability or addressed an unrelated topic. At stage three, most of the exclusions were due to issues relating to the sample age or ability or because the study lacked variables that were available to compare against adult social outcome.

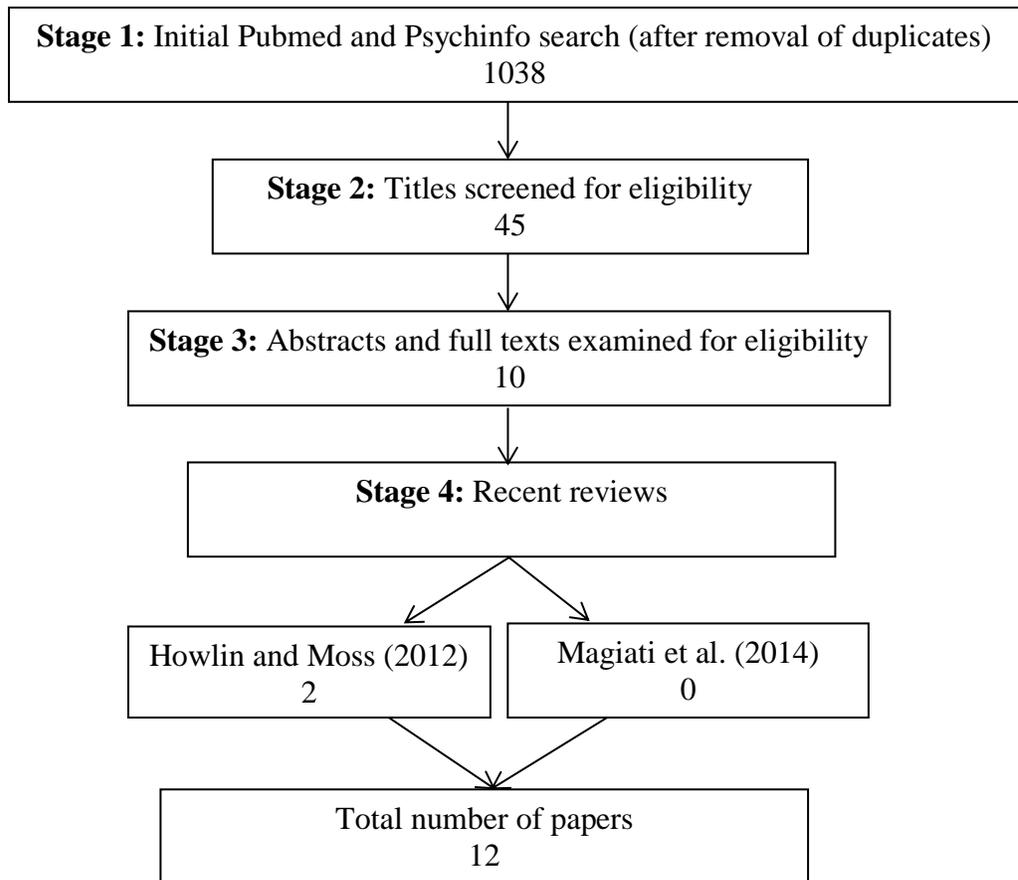


Figure 1.1 Literature search process

1.3.3 The ability level of samples

A primary inclusion criterion in the current review was that studies had to focus on participants who, at least in childhood, had an IQ outside the intellectual disability range (≥ 70). However, this proved complicated as many studies used mixed IQ samples and / or did not provide detailed IQ information. Consequently, an IQ decision-making hierarchy was developed to guide the systematic selection of papers based on IQ scores (Figure 1.2), a process which involved two stages. In stage one, if the sample had a reported mean IQ ≥ 70 then they were included but if the mean IQ was < 70 then they

were automatically excluded. If the mean IQ of the sample was not reported, then the second stage of the selection process was employed. At this stage, three different scenarios resulted in inclusion; (i) the majority of the sample had an $IQ \geq 70$, (ii) the lower limit of the IQ range was ≥ 70 or (iii) the mean developmental quotient was in the adult range.

1.3.4 Assessing the quality of the literature

When considering the quality of papers for use in systematic reviews, it is becoming increasingly common to use a quality assurance measure to rate each paper. These ratings can (i) aid the development of inclusion criteria, (ii) inform sensitivity analysis, (iii) weight studies for meta regressions and / or (iv) highlight studies or parts of studies with poor methodological quality (Stang, 2010).

These scales have been widely developed for reviews of Randomised Control Trials (RCTs). However, in recent years comparable scales for case control and/or cohort studies have begun to emerge (for example the Newcastle-Ottawa Scale, N-OS; Wells, 2004). This measure can be used as a checklist or a scale and uses a star rating system (range 0-9; 9 = highest quality), providing a semi-quantitative assessment of study quality. However, there is limited evidence for its reliability and validity and it may actually produce arbitrary results (Stang, 2010). Furthermore, Oremus, Oremus, Hall and McKinnon (2012) evaluated the Jadad (another quality assurance scale) and the N-OS and despite both having fair-to-excellent test re-test reliability, they both had poor-to-fair

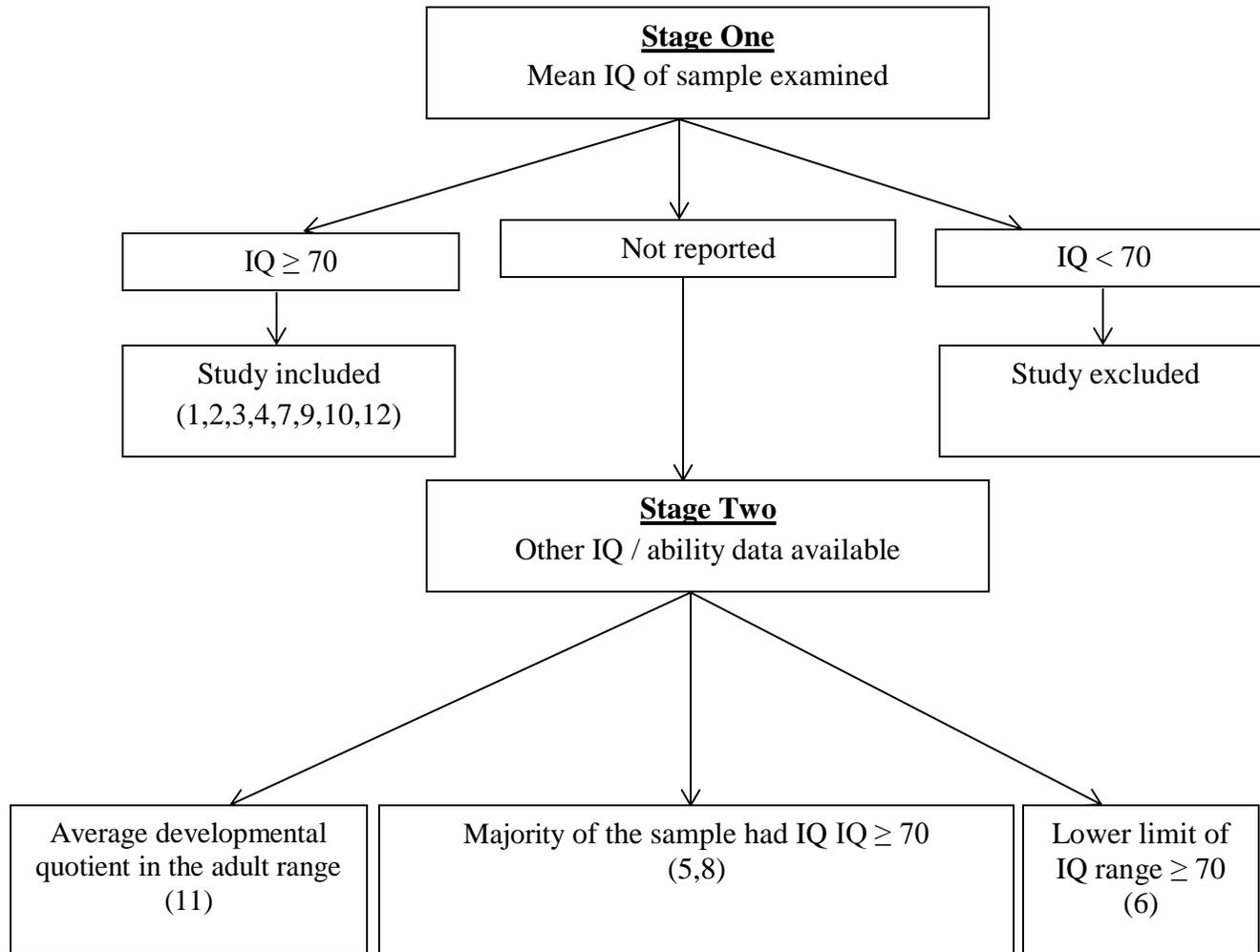


Figure 1.2 IQ hierarchy for inclusion in literature review

Nb: numbers refer to papers listed in Table 1 (page 23) that were included based on each level of the IQ hierarchy.

inter-rater reliability and their use by individuals who have not been trained properly was unclear. Lastly, Hartling et al. (2013) used the N-OS to assess 131 cohort studies that had been part of eight meta analyses. It was reported to be difficult to use and it was unclear whether it could identify biased results. Therefore, the research remains inconclusive regarding its suitability.

When deciding whether or not to use the N-OS for the current review, the above literature was considered along with the student's experience of piloting the measure with the papers that had been selected for the review. It became apparent that the inclusion criteria that had been devised were so specific that, in order to be included in the review, the studies were already of a high standard (almost all studies received an N-OS star rating of 7-9). The N-OS did not contribute anything further to the process as there was little distinction between each study based on the scores once they had been accepted into the review. Therefore, it was not used. Instead, the quality of the studies was discussed in Section 1.5.2 and any methodological factors that may have affected findings were considered. Quality assurance tools for cohort studies are still in their infancy and will require further refinement before they can be considered to be as useful as the more well-established tools that are available for RCTs. Until this is the case, it appears to be more appropriate to apply strict inclusion criteria when reviewing cohort studies to indirectly ensure that only high quality papers are selected.

1.4 Results

Table 1.1 describes the main characteristics of the 12 studies included in the review. This section will consider the factors that have been assessed with regard to their association with adult social outcome. The three main factors addressed are IQ, autistic

symptomatology and language level, followed by a discussion of any other factors that have been considered but received less attention.

1.4.1 IQ

The role that IQ plays in predicting outcome later in life amongst individuals with ASD has long been considered to be a significant one. This review examined whether this finding holds true amongst a sub-group of individuals with a childhood IQ outside the intellectual disability range or whether there comes a point above which there is no association with adult social outcome. Rutter et al. (1967) and Lotter (1974a; 1974b) were the first to examine this link empirically with a higher ability sample and both reported a significant association between IQ and social outcome. Individuals with a higher IQ obtained better outcome scores than those with a lower IQ (albeit still outside the intellectual disability range). Since these early studies, many research groups have continued to demonstrate the predictive value of IQ amongst higher ability ASD samples (for example, Farley et al., 2009; Howlin et al., 2004; Howlin et al., 2013; Larsen & Mouridsen, 1997).

The consensus view is that, even within higher ability samples, childhood IQ is broadly associated with outcome. However, there is still much debate regarding (i) the exact role that IQ plays, specifically which aspect of IQ is most strongly associated with outcome, (ii) whether there is an IQ cut-off point, above which IQ is no longer predictive of adult outcome and (iii) whether adult IQ is associated with adult social outcome. The following section will deal with each of these points in turn.

Table 1.1 Studies included in review

	Author	Sample Size <i>N</i>	Childhood Diagnosis	Mean childhood IQ (Range)	Mean age at follow-up (Range)	Adult Social Outcome % (<i>n</i>)	Factors examined in relation to outcome
1	Rutter et al. (1970; 1967; 1967)	63	Infantile autism	77.8 ¹ (ns)	16 years (ns)	Very poor – 48% (30) Poor – 13% (8) Fair – 25% (16) Good – 14% (9)	C: IQ, speech, autism symptomatology, schooling A: None
2	Lotter (1974a; 1974b)	29	Autism	71 (55-90)	ns (16-18 years)	Very poor – 48% (14) Poor – 14% (4) Fair – 24% (7) Good – 14% (4)	C: IQ, speech, VABS scores, autism symptomatology, developmental milestones, gender, epilepsy, years of schooling A: None
3	Rumsey et al. (1985) ²	14	Infantile autism	PIQ = 97.4 (55-129)	28 years (18-39 years)	Poor – 29% (4) Fair – 35% (5) Good – 35% (5) ³	C: None A: IQ
4	Szatmari et al. (1989) ²	16	Autism, Childhood schizophrenia and childhood psychosis	92.4 (68-110)	26 years (17-34 years)	Poor – 31% (5) Fair – 31% (5) Good – 38% (6)	C: None A: FSIQ, non-verbal problem solving, visuomotor, facial recognition, receptive language
5	Larsen and Mourisden (1997) ²	18	Autism and AS	ns (mixed)	36 years ¹ (ns)	Very Poor – 28% (5) Poor – 17% (3) Good – 28% (5) Very Good – 28% (5)	C: IQ A: None

14 = Average / near average IQ

	Author	Sample Size <i>N</i>	Childhood Diagnosis	Mean childhood IQ (Range)	Mean age at follow-up (Range)	Adult Social Outcome % (<i>n</i>)	Factors examined in relation to outcome
6	Howlin et al. (2000) ²	19	Autism	ns (70-117)	24 years (21-27 years)	Poor – 74% (14) Fair – 11% (2) Good – 16% (3)	C: language
7	Howlin et al. (2004) ²	67	Autistic Disorder	80.2 (51-137)	29 years (21-49 years)	Very Poor – 12% (8) Poor – 46% (31) Fair – 19% (13) Good – 11% (7) Very Good – 12% (8)	C: PIQ, Verbal ability A: PIQ, VIQ, language, autism symptomatology, reading and spelling
8	Cederlund et al. (2008) ²	140	Autism and AS	AS: 101.4 Autism: 14 ≥ 70	23 years (16-36 years)	Very Poor – 28% (39) Poor – 11% (16) Restricted but acceptable – 20% (28) Good – 27% (38) Very Good – 14% (9)	C: Age at diagnosis A: FSIQ, CIQ
9	Farley et al. (2009) ²	41	Autism	86.96 (69 - 137) Best IQ estimate	33 years (22-46 years)	Very Poor – 0% (0) Poor – 17% (7) Fair – 34% (14) Good – 24% (10) Very Good – 24% (10)	C: FSIQ, VIQ, PIQ, Age of single word and phrase speech A: VABS score, PIQ, caregiver support
10	Szatmari et al. (2009) ²	Autism: 36 AS: 21	Autism / AS	Autism: 84.5 AS: 101.33	Autism: 17.7 years AS: 17.6 years (17 – 19 years)	- ⁴	C: Autism symptomatology and Language A: ABS and VABS
11	Gillespie-Lynch et al. (2012) ²	20	Autism (one with PDD-NOS)	DQ: 54.7 (ns)	26.6 years (ns)	Poor – 50% (10) Fair – 20% (4) Good – 10% (2) Very Good – 20% (4)	C: Language, Response to joint attention, IQ, A: None

Author	Sample Size <i>N</i>	Childhood Diagnosis	Mean childhood IQ (Range)	Mean age at follow- up (Range)	Adult Social Outcome % (<i>n</i>)	Factors examined in relation to outcome
12 Howlin et al. (2013) ²	60	Autism	88.8 (70-133)	44.2 years (29 – 64 years)	Very Poor – 33% (20) Poor – 27% (16) Fair – 23% (14) Good – 10% (6) Very Good – 7% (4)	C: Autism symptomatology, language, IQ A: Autism symptomatology, IQ, mental health, age, gender, deprivation level, language

Nb. Summary ratings are based on authors' own classification where provided, otherwise: 'Good' = moderate to high levels of independence in job (or student) and / or living (may be at home with minimal supervision); some friends / acquaintances; 'Fair' = some degree of independence or job, may require moderate levels of support and supervision but does not need specialist residential accommodation; no close friends but may have some acquaintances; 'Poor' = requires specialist residential accommodation or hospital provision (or parental home with close supervision majority of the time); no friends / acquaintances.

Nb. The terminology that studies used to describe their own samples has been used in this table (for example, average / near average IQ)
ns = not specified, C = Measured in childhood, A = Measured in adulthood

¹ IQ and / or age based on a sub-sample because data not available for all participants (for example, died)

² Obtained from pubmed/psycinfo search

³ Outcome scores based on employment and living status only

⁴ VABS scores provided outcome data but exact scores not reported. Association between language impairment and VABS outcome examined

AS = Asperger Syndrome, VABS = Vineland Adaptive Behaviour Score, DQ = Developmental Quotient (mental age/chronological age), ABS = Applied Behavioral Scale, VIQ = Verbal IQ, PIQ = Performance IQ, FSIQ = Full Scale IQ

As research into outcomes has progressed, studies have attempted to delineate the role of childhood intelligence in relation to adult outcome by focusing on specific aspects of IQ. Howlin et al. (2004) reported a significant difference in independent living status and qualification level according to childhood PIQ bands but found that VIQ was not significantly associated with adult social outcome in a higher ability ASD sample. Additionally, childhood FSIQ was more reliable than both PIQ and VIQ. In contrast, Farley et al. (2009) found that VIQ explained 27% of the variance in adult outcome compared to PIQ, which only explained 14% of the variance.

In line with this, Howlin et al. (2013) commented on the limited predictive value of childhood PIQ. They reported that once other childhood factors had been taken into account, PIQ had the least predictive value in a regression which also included early autism symptoms and childhood language level. However, the regression model in this study only explained 44.1% of the variance in outcome and the predictive value of childhood VIQ was not examined. Therefore, it is unclear from this sample, how childhood PIQ and VIQ compare. Childhood PIQ and VIQ appear to have variable predictive value between studies once other factors have been taken into account.

Many studies have also considered whether there is a specific IQ point in higher ability samples, above which the association between childhood IQ and outcome diminishes because the IQ of the group is too homogenous. Howlin et al. (2004) commented on the relative ease of predicting outcome amongst lower ability samples compared to those of higher ability, suggesting that other, currently unknown factors must be at play in higher ability groups, as outcome was still variable within the sample. Supporting this, Farley et al. (2009) highlighted that within higher ability samples, childhood IQ alone cannot predict outcome. In their sample, the predictive value of VIQ

still left 73% of the variance in adult outcome unaccounted for. Subsequent studies have also found that childhood IQ has limited predictive value in a homogenous IQ sample, once other factors have been controlled for (Gillespie-Lynch et al., 2012; Howlin et al., 2013). The literature on childhood IQ in this population to date indicates that using childhood IQ alone to predict outcome amongst higher ability samples is complicated, with the exact nature and extent of the association unclear, and that the ability to make specific predictions based on individual IQ scores remains a challenge.

The association between *adult* IQ and outcome has also been addressed in the literature. Rumsey et al. (1985) reported that participant's outcomes were below what would have been expected given their current IQ but they did not report on overall adult social outcome, only specific outcomes on employment and living status. These early observations were later corroborated by Howlin et al. (2013); a regression analysis, including adult FSIQ, language level and total autism symptomatology scores, explained 70.5% of the variance in outcome, with IQ being the second strongest factor after symptomatology scores. Szatmari et al. (1989) also examined current FSIQ but focused on adaptive behaviour using the VABS. They reported a significant moderate correlation between FSIQ and VABS scores ($r = 0.6$), with individuals with higher IQs, particularly those >100 , having better outcomes.

Regarding specific aspects of adult IQ, Cederlund et al. (2008) reported that lower adult FSIQ *and* VIQ scores were associated with poorer outcomes in their AS sample. Additionally, Howlin et al. (2004) and Farley et al. (2009) both reported on the relative weakness of adult PIQ as a correlate of outcome. The former study found that adult VIQ was a better predictor of outcome than adult PIQ, and the latter reported that current PIQ scores had the weakest association with outcome compared to various other factors

considered. However, in both studies, the correlations were still significant and moderate-to-strong ($r = 0.66$ and $r = 0.55$ respectively).

Overall, the association between combined and specific aspects of childhood and adult IQ and outcome is evident but IQ (in childhood or adulthood) alone cannot reliably predict which adults with higher ability ASD are going to have the best outcomes. Other factors, such as language level and levels of autistic symptomatology, need to be considered in order to fully understand how to predict adult social outcome in this higher IQ population.

1.4.2 Autistic symptomatology

As with IQ, the association between autistic symptomatology and adult outcome has been repeatedly investigated in relation to higher ability adults with ASD. Some studies have reported on overall symptom severity whereas others have focussed more on specific aspects of ASD symptomatology.

Regarding the former, Rutter et al. (1967) found that less frequent and lower overall symptom scores were more common amongst individuals with ‘good’ adjustment at follow-up compared to those with ‘fair’ adjustment ($p < 0.01$). Supporting this, Lotter (1974a; 1974b) found that outcome was worse for individuals with autism compared to a control group. More recently, Howlin et al. (2013) found that Autism Diagnostic Interview (ADI; Le Couteur et al., 1989) total symptom scores at diagnostic confirmation were more predictive of adult outcome than childhood language level or PIQ. Similar results were also found for total adult symptomatology scores.

As with IQ, more recent studies have focused on specific domains of autism symptomatology. In a regression analysis, which included the three ADI domains,

childhood PIQ and language level, Howlin et al. (2013) found that childhood levels of Reciprocal Social Interaction (RSI), as assessed by the Autism Diagnostic Interview-Revised (ADI-R; Rutter, Le Couteur, & Lord, 2003), carried the greatest predictive value ($\beta = .049, p < .001$), in a model which explained 40.9% of the variance. *Adult* levels of RSI were also associated with adult outcome in this sample; in a multiple regression the RSI ADI-R domain explained most of the variance (76.1%; $\beta = 0.6, p < .001$), followed by IQ and then the Restricted and Repetitive Behaviours and Interests (RRBI) ADI-R domain ($\beta = 0.17, p = .02$).

Using a more fine-grained approach, Gillespie-Lynch et al. (2012) evaluated childhood scores on ‘response to joint attention’ (RJA) in relation to adult social outcome. They found an extremely high correlation with adult outcome ($r = -0.8$; greater impairments in RJA skills were associated with poorer adult outcomes). It is possible that this particularly strong correlation is a result of applying a more fine-tuned approach to examining the association between aspects of autism symptomatology and outcome. However, this extremely high correlation is uncommon in this area of research, particularly given the length of time between child and adult assessment. Additionally, the narrow focus on one aspect of symptomatology is unusual. Studies replicating this strong correlation are necessary before any firm conclusions can be drawn.

1.4.3 Language level

The presence of communicative, phrase speech has long been accepted as a key prognostic factor regarding outcome in adulthood for individuals with ASD, including the higher ability population. Despite an IQ outside the intellectual disability range, many individuals with higher ability ASD develop language late, if at all. Language

delay was previously a key feature of the disorder but it is now regarded as an associated difficulty (American Psychiatric Association, 2013).

Many researchers have attempted to clarify the age by which language acquisition is required in order to guarantee a better outcome in later life. Rutter et al. (1967) reported a moderately strong association between outcome and 'useful' speech by five years of age and observed a significant difference in the number of individuals who were lacking phrase speech by five years old between those with a 'good' and 'fair' outcome. They also found that after controlling for speech and IQ, many other factors were no longer significantly associated with outcome. Additionally, the combined association of speech and IQ with outcome was very high ($r = 0.89$).

Lotter (1974a) also found a strong, significant association between speech at 8-10 years and later outcomes ($r = 0.87$, $p < 0.001$). Again, when speech and IQ were combined, there was a slightly stronger correlation with outcome ($r = 0.89$) but, given the minimal increase in association, the authors concluded that it was the use of communicative speech that was the key prognostic factor. However, the association with language alone was similar to the one found in the control group (individuals with ASD symptoms who did not meet diagnostic criteria; $r = 0.88$).

The exceptionally high correlations reported by both of these early studies are unusual given that a basic measure of childhood language ability was compared with outcome scores many years later. The role played by other factors, which were not controlled for, must be considered. Additionally, this finding may be accounted for, at least in part, by the way in which language was measured. Lotter (1974a) split language ability into four categories, creating an arbitrary continuous variable. This would have increased the chance of an association between language and outcome due to the narrow

range of possible language scores. However, this is a common method of measuring language in this population (for example, Howlin et al., 2013), particularly if scores are based on the ADI/ADI-R ratings.

Since these early findings, many other studies have highlighted the importance of communicative / phrase speech by a certain age when predicting adult outcome (for example, Farley et al., 2009; Howlin et al., 2013). Additionally, the findings regarding the association between VIQ and adult outcome (Section 1.5.1) support the claim that language ability predicts outcome in adulthood.

Some research groups have gone one step further by attempting to delineate the exact type of language ability that predicts outcome. Howlin et al. (2000) reported that scores on the Peabody Picture Vocabulary Test in childhood contributed to 32% of the variability in adult outcome amongst higher ability adults with ASD and that current language skills were significantly associated with outcome. However, improvements in linguistic functioning over time and adult outcome scores did not appear to be associated. In contrast, although Gillespie-Lynch et al. (2012) also reported an association between early language and outcome ($r = -0.84$), they actually found a strong significant association between adult social outcome and change in language over time ($r = -0.89$); the greater the improvement in language, the better the outcome in later life. However, like Lotter (1974a), these associations are exceptionally high and warrant replication.

Szatmari et al. (2009) used a Structural Language Impairment (StrLi) in childhood (6-8 years) to distinguish between individuals with high functioning autism and those with AS. Those without a StrLi (i.e. individuals with AS) had better VABS

scores at follow-up than those with a StrLi (i.e. high functioning autism), suggesting that this specific language impairment impacted on individuals' outcomes in adulthood.

Despite the strong evidence supporting the predictive value of language abilities on adult social outcome outlined above, some studies have produced contrasting findings. Szatmari et al. (1989) reported that the association between VABS scores and receptive language was very small ($r = 0.14$) and Farley et al. (2009) did not find any association between early language skills and adult outcome. However, in the latter study all participants had communicative phrase speech by six years, limiting the variability of early language skills within the sample. Despite the lack of a statistically significant association, differences were observed between those with 'poor' and 'very good' adult outcome based on the age at which phrase speech was acquired, suggesting that childhood language ability holds some predictive value, even within a seemingly homogenous sample.

1.4.4 Other factors considered in relation to adult outcome

A number of studies have examined other possible factors that may be associated with outcome. These can be separated into individual and environmental factors.

Perhaps one of the more well-known individual factors to have been researched over the years is gender. Lotter (1974a) found that gender was significantly associated with outcome ($r = 0.42, p < 0.05$ versus $r = 0.32, p = ns$ in the control group) but the direction of this association was not specified. Furthermore, this association did not remain once speech and IQ had been controlled for. Conversely, Rutter et al. (1967), Howlin et al. (2004) and Howlin et al. (2013) all reported no significant gender differences between outcome groups. However, the lack of conclusive evidence

regarding the role of gender may be accounted for by limited statistical power (since the number of females in these samples was very small) and descriptive reports indicate that females may fare worse than males. For example, of the seven females in the Howlin et al. (2004) study ($N = 67$), five had a 'poor' or 'very poor' outcome and none had attended mainstream school or obtained any formal qualifications. Additionally, almost all were in day or residential centres and none had any friends or were living independently. There is currently no conclusive statistical evidence to suggest that gender plays a significant role in predicting outcome, but descriptive reports indicate that females may fare worse than males.

Some studies have examined the link between adult social outcome and mental health difficulties. Farley et al. (2009) found that individuals across all outcome categories reported mental health difficulties, indicating no association between the two, but they did not conduct any formal statistical analysis with the data. However, these reports were later corroborated by Howlin et al. (2013) who found no statistical association between adult social and mental health outcome. These results suggest that mental health difficulties in this population do not adversely affect social outcomes but, in the latter study in particular, a lack of variability in rates of mental health difficulties in the sample may have prevented any significant results from being identified.

Other individual factors that have been examined have included medical complications, specific areas of ability and age of diagnosis. Lotter (1974a) found an association between seizure history and outcome. However, as with other factors examined in this study, this association did not remain once speech and IQ had been accounted for. Szatmari et al. (1989) reported that outcome was associated with non-verbal problem solving, facial recognition and visuo-motor skills, but these factors have

never been investigated further. Finally, Cederlund et al. (2008) reported that individuals who were diagnosed with ASD at a younger age (5.5 – 9.5 years) were more likely to have a ‘good’ outcome than those who were older at diagnosis (16.0 – 24.5 years; 35% and 22% with a ‘good’ outcome respectively). However, no formal statistical analysis was conducted to confirm these findings.

There has been very limited examination of environmental factors within this population. Rutter et al. (1967) and Lotter (1974a) both examined the role of years of schooling on outcome. Rutter et al. (1967) found that the amount of schooling that a child received was associated with outcomes; 100% of children with a ‘good’ outcome had received at least 2 years of schooling compared with 20% of those with a ‘very poor’ outcome. However, it is possible that the length of schooling was a moderating factor between other individual factors and outcome. For example, IQ, language skills and/or levels of autistic symptomatology may have determined whether individuals were able to remain in school and, in turn, have better outcomes. In contrast, Lotter (1974a) reported a complicated association between years of schooling and employability in later life; commenting that despite having “adequate” intellectual abilities, not all individuals who attended school, were able to gain employment. It may be that many individuals, who were able to cope within the structured and predictable school environment, struggled when entering into employment where environments are less supported and predictable. Lastly, Farley et al. (2009) found that high levels of support from caregivers and local agencies were negatively correlated with social outcome scores.

This section has detailed other individual and environmental factors that have been considered by research groups in addition to the three main factors that have been repeatedly found to be associated with outcome in this population. Doing so,

demonstrates the ability of the field to remain open-minded and to continue to consider other possible factors that may be associated with outcome without being exclusively focussed on the key variables that have been repeatedly proven to be linked to adult outcome in this population. However, it also highlights a number of potentially important demographic factors that appear to have been overlooked over the years. For example, the role of socio-economic status (SES), ethnicity and other sample characteristics. Szatmari et al. (1989) noted that SES could be a prognostic factor affecting outcome in their discussion but did not examine its role with their sample. More recently, the student's PhD thesis (Moss, 2011) found no association between SES and adult social outcome in sample described by Howlin et al. (2013; 2014), but none of the other papers included in the current review have considered this with their own samples. Further examination of such factors would be an important area for future research, particularly given the large variability in demographics reported between samples. For example, diagnostic status, age and sample size. Section 1.5.2 considers this variability and potential the impact of these factors on the findings reported.

1.5 Discussion

1.5.1 Summary of results

This review has addressed the factors examined in relation to their association with social outcome in adult life amongst higher ability adults with ASD. Three key factors have been addressed in the literature: IQ, autistic symptomatology and language level. Using IQ alone to predict outcome within higher ability ASD samples is complicated. Whilst the predictive value of childhood IQ (particularly FSIQ) is evident, variability still remains within higher ability samples, suggesting that other factors are also likely to

be at play. Furthermore, whilst it is possible to make general predictions that those with a higher childhood IQ have a better chance of a positive adult outcome than those with a slightly lower IQ, making any more detailed predictions, based on specific IQ points, remains a challenge.

Total autism symptomatology scores in childhood are predictive of outcome in later life. In particular, skills in RSI (child and adult) have been found to be more predictive of outcome than the other domains. Some studies have attempted to delineate this further by focussing on specific autistic symptoms, for example RJA, but these findings require further examination before any firm conclusions can be drawn.

There is widespread evidence that early language abilities are predictive of outcome, even within higher ability samples. Many higher ability individuals with ASD often develop language late, if at all, and so this is a crucial factor in determining likely outcome in later life. This is supported by associations that have been identified between early VIQ scores and adult social outcome. Whilst some contradictory evidence exists about the role of language, in many cases it appears to be methodological factors that are contributing to the variability in findings reported.

Despite a focus on these three main factors a number of other individual and environmental factors have also been examined. These include gender, mental health, medical complications, years of schooling and caregiver support. To date, none of these have been consistently found to be associated with adult social outcome in this population. It seems that amongst higher ability individuals with ASD, IQ, autism symptomatology and language level (specifically in childhood) are the best indicators of social outcome in adult life. However, there is still no consensus regarding (i) which factor holds the highest predictive value, (ii) which aspect of each factor is most strongly

associated with outcome and (iii) whether the effect of variables are modified by group demographics. These questions have arisen due to great variability within and between studies. The following section will explore why this variability exists.

1.5.2 Critical review of papers

1.5.2.1 Diagnosis

Whilst one of the inclusion criteria for the current review was that individuals were diagnosed with autism in childhood, many studies did not use standardised criteria and/or did not clarify how diagnoses were made. This was particularly common amongst studies that recruited participants prior to the 1980's, when standardised diagnostic measures were first developed (For example, Lotter, 1974a; Rutter et al., 1967). Often, diagnoses were re-confirmed at follow-up using appropriate measures. For example, the individuals in the Howlin et al. (2013) sample were all diagnosed in childhood based on DSM-III criteria and their diagnoses were later confirmed using the retrospective ADI algorithm. However, not all studies took this approach. Furthermore, even individuals who did meet diagnostic criteria in childhood did not always meet criteria at follow-up. Rumsey et al. (1985) noted that only three out of their 14 participants met DSM-III autism diagnostic criteria at follow-up; the remaining 11 met criteria for 'autism, residual state'. As this term no longer exists, it is hard to know how to categorise these participants when comparing study findings or when applying results to individuals currently being diagnosed with higher ability ASD. Therefore, the criteria for the current review were not able to eliminate all variability in diagnosis between and within the samples.

1.5.2.2 Sample size

Only studies with a sample size ≥ 10 were included, but this is still a small N for reliable statistical analyses to be conducted and may have limited the generalizability of findings. For example, Szatmari et al. (1989) only reported on 16 people. Smaller sample sizes mean that one extra participant can significantly affect overall findings and, in turn, the conclusions drawn. It was also not always the case that the whole sample was of higher ability; Larsen and Mourisden (1997) reported on 18 participants, 14 of whom were of higher ability. Such studies, with higher ability sub-groups, can mean that the overall conclusions drawn by the authors do not necessarily reflect higher ability individuals and so care must be taken when examining the papers. Lastly, small samples that report null findings might lack sufficient power to detect significant results that exist. For example, Howlin et al. (2004) found no significant difference between males and females according to adult social outcome but only seven of their 67 participants were females. Whilst a higher proportion of males to females is expected in ASD samples, (male to female ratio = 3.3:1; Baird et al., 2006), such a small number of females would make it difficult to detect any significant findings. It is therefore important to examine results closely when reviewing papers with small samples and/or higher ability sub-groups.

1.5.2.3 Sample age

Many of the studies in this review included individuals whose adult social outcome was assessed in later adolescence / early adulthood (Cederlund et al., 2008; Lotter, 1974a; Rutter & Lockyer, 1967; Szatmari et al., 2009) whereas others focussed on mid-to-late adulthood (for example, Howlin et al., 2013). The age of a sample is crucial; younger

individuals are unlikely to be as independent as older individuals regarding living status and employment and this may impact on overall outcome scores and, in turn, factors found to be predictive of outcomes. Conversely, more recent, young samples could actually have better outcomes than older samples given that levels of specialist input have improved in recent years leading to improved overall outcomes (Eaves & Ho, 2008). Consequently, cohort effects combined with the range of ages both between and within samples is likely to be confusing our understanding of what impact age has on outcomes.

1.5.2.4 Range of IQs

This review had a specific focus on higher ability adults with ASD. However, examination of Table 1.1 demonstrated that even within this parameter, IQs can still be highly variable. Many papers lacked detailed IQ information, particularly the earlier studies (Rumsey et al., 1985). Moreover, even when studies seem to focus on higher ability samples, this was not always the case. Lotter (1974a) had a mean IQ of 71 but the range was 55 – 90 and Farley et al. (2009) had a mean IQ of 89 but a range of 50 – 140. The IQ decision-making hierarchy employed in this review (Figure 1.2), helped to simplify the selection process. However, the IQ variability that still existed between and within samples made it difficult to compare studies and/or to draw firm conclusions about the population.

More and more people are being recognised as having higher ability ASD and so it is increasingly important to focus on this group and to ascertain what the predictors and correlates of their outcomes are. Clear IQ data on samples and a focus on more homogenous IQ groups, even within larger mixed samples, will aid this. Furthermore,

literature reviews such as the current one need to be mindful of the variability in IQ that exists between and within study samples published to date and ensure that the inclusion criteria devised for the review take this into consideration.

1.5.2.5 Method of assessment and analysis

Comparisons between follow-up studies were also affected by differences in the outcome measures used. The majority of the studies reviewed used composite scores based on the initial ratings developed by Rutter et al. (1967), although usually with some modifications. An additional ‘restricted but acceptable’ category was used by Cederlund et al. (2008) and Howlin et al. (2004; 2000) added a ‘very good’ category *and* adjusted the measure to account for social relationships. This version was then used by Farley et al. (2009). Finally, Howlin et al. (2013) also included romantic relationships to reflect the age, and possible life stage, of the sample. In contrast, other studies devised their own outcome measures (Rumsey et al., 1985) or used VABS scores (Szatmari et al., 2009). The choice of outcome measure is important because studies need to use ‘like-for-like’ measures in order to be compared with one another.

Data collection methods also varied greatly across studies. Most data was collected using a combination of face-to-face informant and participant interviews and observations. However, some studies used less reliable methods; Larsen and Mourisden (1997) sourced their data from national registers, with no patient or informant contact at any point. This limits the reliability of findings based on non-standardised assessments.

1.5.2.6 Representativeness of samples

The representativeness of samples in this population is important as many samples were recruited from specialist clinics (For example, Rutter et al., 1967). Individuals who attend such clinics may have developed positive relationships with their clinicians and so have been more likely to continue to participate in research over the years. Moreover, they may have received more support than individuals who did not attend such clinics in childhood. Conversely, but perhaps most importantly, by virtue of attending a specialist clinic these participants may have experienced more difficulties, which could impact on the results. Either way, these samples cannot be considered truly representative of a wider ASD population.

The use of control groups is also important. Few ASD follow-up studies have used control groups and even amongst those that did, the appropriateness of the control group is questionable. Rutter et al. (1967) recruited their ‘non-infantile psychosis’ control group from the Maudsley Hospital Children’s Department. However, many of these participants had an intellectual disability and the majority had behavioural and / or mental health problems requiring in-patient care. Thus, comparisons with this control group cannot easily be generalised beyond the study. The lack of control groups in the majority of the studies is also noteworthy. This makes it difficult to determine how the results gleaned from an ASD sample compare to other neurodevelopmental disorders, for example, or the general population.

Population-based studies are considered to be more representative of the wider ASD population. However, even studies which appear to have recruited in this way can be biased. Farley et al. (2009) was a population-based study but the sample consisted almost entirely of members of the Church of Jesus Christ of Latter-Day Saints. This

community is known for its focus on family relationships and tight social cohesion, which may have contributed to the relatively better outcomes reported.

Cohort effects also play a role in the representativeness of ASD samples in follow-up studies such as these. Rising prevalence rates (Baird et al., 2006) mean that individuals diagnosed many years ago do not necessarily accurately reflect the ASD population today. Therefore, the generalizability of findings from studies conducted with samples that were diagnosed and recruited in the 50's, 60's and 70's to individuals diagnosed with higher ability ASD today may be limited.

1.5.3 Critical review of the research process

In addition to critiquing the papers themselves, it is important to evaluate the review process itself. This review could be criticised for not applying a quality assurance tool to evaluate the quality of the studies selected for inclusion in the review. Using such tools, particularly with reviews of RCT's, is becoming increasingly common. However, as explained in Section 1.3.4 using a tool was carefully considered for the current review but pilot work revealed that, by virtue of the inclusion criteria, all of the studies included in the review were already of a high standard that a quality assurance tool did not add anything useful to review process. Until quality assurance tools for non-RCT reviews are better established, it seems more appropriate to apply strict inclusion criteria and then to rigorously evaluate the quality of the studies on a case by case basis as was done in this review (Section 1.5.2).

A second limitation of the review process was that the literature search was not replicated by a second researcher to check that the search terms and inclusion criteria were robust. Ideally, this would have been done but given limited resources and time

this was not possible. However, to address this limitation care was taken to review other recent, more inclusive reviews (for example, Howlin and Moss, 2012; Magiati, Tay and Howlin, 2014), to ensure that no relevant papers had been overlooked.

This review has a number of strengths. Firstly, it examined adult social outcome in particular, rather than a range of outcomes. This narrow focus was intentional to ensure that the predictors and correlates of this widely used measure of outcome in ASD were considered in detail. The alternative, applying a broader stroke approach to examining all types of outcome, has proved useful in the past but does not allow for a fine grained evaluation of the factors affecting this type of outcome. Secondly, the very strict inclusion criteria, with respect to IQ, were novel. To the student's knowledge, this is the first systematic literature review in this field to focus exclusively on populations with an IQ outside of the intellectual disability range. Whilst many previous reviews have included such studies (for example, Magiati, Tay and Howlin, 2014), none have focussed exclusively on this group in adulthood. Tailoring the review in this way enabled a critical evaluation of the predictors and correlates of adult social outcome in this population which will, in turn, have clinical implications (see Section 1.5.4 for further discussion of this).

1.5.4 Conclusions, Clinical Implications and Future Research

This review has demonstrated that better child (and to a lesser extent adult) language and IQ scores and lower autistic symptomatology scores are associated with better social outcomes in later life, even amongst higher ability adults with ASD. A number of studies have begun to examine these factors in more detail and considered which specific scores / abilities are most predictive of outcome within these broader categories

but findings require further replication. There remain a number of questions to be addressed; (i) which of these three factors has the strongest association with outcome, (ii) which aspect of each factor is most strongly associated with outcome and (iii) whether the effects of variables are modified by group demographics. As yet, no other individual or environmental factors have been consistently identified as reliable factors associated with outcome. However, as discussed in Section 1.4.4, there has been a distinct lack of focus on the impact that other demographic factors (for example, socio-economic status and ethnicity) may have on adult social outcome. Future research would benefit from paying greater attention to these factors when considering outcomes in this population.

Individuals with higher ability ASD often suffer greatly with access to services, particularly in adulthood, because they do not meet criteria for learning disability services yet often have significant needs that generic services cannot address. Therefore, it is crucial to determine what predicts their outcome so that clinicians can know what skills to support them to develop in order to improve their achievements in adulthood. Further clarification of predictors and correlates of outcome will aid the development of appropriate interventions with a view to affecting outcomes in adulthood. For example, interventions that target the development of RSI skills in childhood could improve social outcomes in adulthood.

At present it is still difficult to make definite predictions about outcome based on individual childhood data for higher ability people with ASD. This is a challenge for clinicians who are currently diagnosing children in this population, because families want to know what to expect. By understanding more about predictors and correlates of outcome, clinicians will be able to advise families on (i) the likely impact of the

diagnosis throughout the lifespan, (ii) how best to support the person in childhood and adulthood in order to maximise their potential and (iii) how to prepare for the future based on the level of support that they are likely to require.

Regarding adult social outcome as a measure, it is important to consider its appropriateness as a way of determining whether or not an individual with high functioning ASD is achieving in adulthood. Achievements in independent living, employment, friendships and relationships are generally regarded as the hallmark of success in adulthood. However, these components map on to more traditional, societal expectations of what constitutes achievements in adulthood. This may be less appropriate for use, in isolation, with this unique population of higher ability adults with ASD than was previously thought. Such milestones are likely to be harder for these individuals to achieve and may not be what they themselves actually desire (for example, romantic relationships given the nature of their social and communication difficulties). Furthermore, this measure does not tap into more subjective aspects of outcome, such as perceived Quality of Life (QoL) so it cannot indicate whether or not the individual is satisfied with their current situation.

QoL is “the individual’s perception of their position in life in the context of culture and value systems in which they live, and in relation to their goals, expectations, standards and concerns” (The WHOQOL Group, 1995, p.1405). This is an important clinical issue because measuring QoL is thought to provide a more comprehensive, multidimensional measure of outcome due to its focus on more subjective variables, for example satisfaction and subjective wellbeing, than the adult social outcome measure, which focuses on more concrete achievements (Renty and Roeyers, 2006). The empirical paper in section two will consider whether perceived QoL is a more helpful

approach to determining outcome for individuals in mid-to-late adulthood with higher ability ASD than the more traditional adult social outcome measure and, in turn, predictors of outcome.

1.6 References

American Psychiatric Association. (1994). *Diagnostic and Statistic Manual of Mental Disorders (DSM-IV)* (Vol. 4th). Washington, DC: American Psychiatric Association.

American Psychiatric Association. (2000). *American Psychiatric Association: Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision* (Vol. 4th). Washington, DC: American Psychiatric Association.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Arlington, VA: American Psychiatric Publishing.

American Psychiatric Association. (2013b). Intellectual Disability Fact Sheet.

Asperger, H. (1944). 'Autistic psychopathy' in childhood. Translated and annotated by U. Frith (e.d) in *Autism and Asperger Syndrome* (1991). In U. Frith (Ed.), *Autism and Asperger Syndrome* (Vol. 1st, pp. 37-92). Cambridge: Cambridge University Press. (Reprinted from: NOT IN FILE).

Baird, G., Simonoff, E., Pickles, A., Chandler, S., Loucas, T., Meldrum, D., & Charman, T. (2006). Prevalence of disorders of the autism spectrum in a population cohort of children in South Thames: the Special Needs and Autism Project (SNAP). *Lancet*, 368(9531), 210-215.

Cederlund, M., Hagberg, B., Billstedt, E., Gillberg, I. C., & Gillberg, C. (2008). Asperger syndrome and autism: a comparative longitudinal follow-up study more than 5 years after original diagnosis. *Journal of Autism and Developmental Disorders*, 38(1), 72-85.

Eaves, L. C., & Ho, H. H. (2008). Young adult outcome of autism spectrum disorders. *Journal of Autism and Developmental Disorders*, 38(4), 739-747.

- Eisenberg, L., & Kanner, L. (1956). Childhood Schizophrenia; symposium, 1955. VI
Early infantile autism, 1943-55. *American Journal of Orthopsychiatry*, 26, 556-566.
- Farley, M. A., McMahon, W. M., Fombonne, E., Jenson, W. R., Miller, J., Gardner, M., . . . Coon, H. (2009). Twenty-year outcome for individuals with autism and average or near-average cognitive abilities. *Autism Research*, 2(2), 109-118.
- Gillespie-Lynch, K., Sepeta, L., Wang, Y., Marshall, S., Gomez, L., Sigman, M., & Hutman, T. (2012). Early childhood predictors of the social competence of adults with autism. *J Autism Dev Disord*, 42(2), 161-174. doi: 10.1007/s10803-011-1222-0
- Hartling, L., Milne, A., Hamm, M. P., Vandermeer, B., Ansari, M., Tsertsvadze, A., & Dryden, D. M. (2013). Testing the Newcastle Ottawa Scale showed low reliability between individual reviewers. *Journal of Clinical Epidemiology*, 66(9), 982-993. doi: 10.1016/j.jclinepi.2013.03.003
- Howlin, P. (2003). Outcome in high-functioning adults with autism with and without early language delays: implications for the differentiation between autism and Asperger syndrome. *Journal of Autism and Developmental Disorders*, 33(1), 3-13.
- Howlin, P. (2004). Outcome in autism *Autism and Asperger Syndrome: Preparing for Adulthood* (Vol. 2nd, pp. 19-45). London and New York: Routledge. (Reprinted from: NOT IN FILE).
- Howlin, P. (2007). The outcome in adult life for people with ASD. In F. R. Volkmar (Ed.), *Autism and Pervasive Developmental Disorders* (Vol. 2nd, pp. 269-306): Cambridge University Press. (Reprinted from: NOT IN FILE).

- Howlin, P., Goode, S., Hutton, J., & Rutter, M. (2004). Adult outcome for children with autism. *Journal of Child Psychology and Psychiatry*, 45(2), 212-229.
- Howlin, P., Mawhood, L., & Rutter, M. (2000). Autism and developmental receptive language disorder--a follow-up comparison in early adult life. II: Social, behavioural, and psychiatric outcomes. *Journal of Child Psychology and Psychiatry*, 41(5), 561-578.
- Howlin, P., & Moss, P. (2012). Adults with autism spectrum disorders. *Canadian Journal of Psychiatry*, 57(5), 275-283.
- Howlin, P., Moss, P., Savage, S., & Rutter, M. (2013). Social outcomes in mid- to later adulthood among individuals diagnosed with autism and average nonverbal IQ as children. *Journal of American Academy of Child and Adolescent Psychiatry*, 52(6), 572-581 e571. doi: 10.1016/j.jaac.2013.02.017
- Howlin, P., Savage, S., Moss, P., Tempier, A., & Rutter, M. (2014). Cognitive and language skills in adults with autism: a 40-year follow-up. *Journal of Child Psychology and Psychiatry*, 55(1), 49-58. doi: 10.1111/jcpp.12115
- Kanner, L. (1943). Autistic Disturbances of affective contact. *Nervous Child*, 2, 217-250.
- Kanner, L. (1973). How far can autistic children go in matters of social adaptation? *Childhood Psychosis: Initial studies and new insights* (Vol. 1st, pp. 189-213): V. H. Winston & Sons, Inc.
- Larsen, F. W., & Mouridsen, S. E. (1997). The outcome in children with childhood autism and Asperger syndrome originally diagnosed as psychotic. A 30-year follow-up study of subjects hospitalized as children. *European Child and Adolescent Psychiatry*, 6(4), 181-190.

- Le Couteur, A., Rutter, M., Lord, C., Rios, P., Robertson, S., Holdgrafer, M., & McLennan, J. (1989). Autism diagnostic interview: a standardized investigator-based instrument. *Journal of Autism and Developmental Disorders*, 19(3), 363-387.
- Lockyer, L., & Rutter, M. (1970). A five- to fifteen-year follow-up study of infantile psychosis. IV. Patterns of cognitive ability. *British Journal of Social and Clinical Psychology*, 9(2), 152-163.
- Lotter, V. (1974a). Factors related to outcome in autistic children. *Journal of Autism and Childhood Schizophrenia*, 4(3), 263-277.
- Lotter, V. (1974b). Social adjustment and placement of autistic children in Middlesex: a follow-up study. *Journal of Autism and Childhood Schizophrenia*, 4(1), 11-32.
- Macintosh, K. E., & Dissanayake, C. (2004). Annotation: The similarities and differences between autistic disorder and Asperger's disorder: a review of the empirical evidence. *Journal of Child Psychology and Psychiatry*, 45(3), 421-434.
- Moss, P. (2011). *Outcome in individuals with autism and their siblings: An investigation of the factors associated with functioning in adulthood*. (PhD), Kings College London, London.
- Oremus, M., Oremus, C., Hall, G. B., & McKinnon, M. C. (2012). Inter-rater and test-retest reliability of quality assessments by novice student raters using the Jadad and Newcastle-Ottawa Scales. *BMJ Open*, 2(4). doi: 10.1136/bmjopen-2012-001368
- Organisation, W. H. (1993). *The ICD-10 classification of mental and behavioural disorders: Diagnostic criteria for research* (Vol. 10th). Geneva: World Health Organisation.

- Rumsey, J. M., Rapoport, J. L., & Sceery, W. R. (1985). Autistic children as adults: psychiatric, social, and behavioral outcomes. *Journal of the American Academy of Child and Adolescent Psychiatry*, 24(4), 465-473.
- Rutter, M., Greenfeld, D., & Lockyer, L. (1967). A five to fifteen year follow-up study of infantile psychosis. II. Social and behavioural outcome. *British Journal of Psychiatry*, 113(504), 1183-1199.
- Rutter, M., Le Couteur, A., & Lord, C. (2003). *The Autism Diagnostic Interview-Revised*. Los Angeles: Western Psychological Services.
- Rutter, M., & Lockyer, L. (1967). A five to fifteen year follow-up study of infantile psychosis. I. Description of sample. *British Journal of Psychiatry*, 113(504), 1169-1182.
- Sparrow, S. S., Cicchetti, D. V., & Balla, D. A. (2005). *Vineland-II Vineland Adaptive Behaviour Scales*. Minnesota: AGS Publishing.
- Stang, A. (2010). Critical evaluation of the Newcastle-Ottawa scale for the assessment of the quality of nonrandomized studies in meta-analyses. *European Journal of Epidemiology*, 25(9), 603-605. doi: 10.1007/s10654-010-9491-z
- Szatmari, P., Bartolucci, G., Bremner, R., Bond, S., & Rich, S. (1989). A follow-up study of high-functioning autistic children. *Journal of Autism and Developmental Disorders*, 19(2), 213-225.
- Szatmari, P., Bryson, S., Duku, E., Vaccarella, L., Zwaigenbaum, L., Bennett, T., & Boyle, M. H. (2009). Similar developmental trajectories in autism and Asperger syndrome: from early childhood to adolescence. *Journal of Child Psychology and Psychiatry*, 50(12), 1459-1467. doi: 10.1111/j.1469-7610.2009.02123.x

- Wells, G., Shea, B., O'Connell, D., Peterson, J., Welch, V., Losos, M., & Tugwell, P. (2004). The Newcastle-Ottawa Scale (NOS) for assessing the quality of nonrandomised studies in meta-analyses.: Ottawa Health Research Institute.
- Wing, L. (1981). Asperger's syndrome: a clinical account. *Psychological Medicine*, *11*(1), 115-129.
- Wing, L., & Gould, J. (1979). Severe impairments of social interaction and associated abnormalities in children: epidemiology and classification. *Journal of Autism and Developmental Disorders*, *9*(1), 11-29.
- Witwer, A. N., & Lecavalier, L. (2008). Examining the validity of autism spectrum disorder subtypes. *Journal of Autism and Developmental Disorders*, *38*(9), 1611-1624.

Part 2: Empirical Paper

Adult and Child Correlates of Quality of Life Amongst Adults with Autism and a Childhood IQ Outside the Intellectual Disability Range

2.1 Abstract

Aim: To examine adult and child correlates of Quality of Life (QoL) amongst adults with autism, all of whom had a childhood IQ outside the intellectual disability range.

Method: Fifty-two individuals with a childhood diagnosis of autism and a childhood IQ ≥ 70 were administered the World Health Organisation Quality of Life – Brief (WHOQOL-BREF; informant and/or self-report). These scores were compared against UK norms and with a range of child and adult measures collected on the autism sample at previous time points.

Results: Participants' self-reported QoL was significantly better than a comparative psychiatric sample (physical, psychological and environmental QoL domains) and better than a healthy sample (environmental QoL domain). Childhood IQ and scores for Restricted and Repetitive Behaviours and Interests (RRBI) were significantly negatively associated with a number of self-reported QoL domains. Informant perceived psychological QoL was significantly poorer amongst individuals without language in childhood. Adult IQ and social outcome scores were negatively correlated with self-reported social QoL scores and age was negatively correlated with informant perceived physical QoL scores.

Conclusions: The QoL of adults with autism, who, as children had an IQ outside the intellectual disability range, is not as poor as has been previously suggested. The findings from this preliminary study suggest that it may be possible to anticipate who will have a poorer adult QoL than others. Adult social outcome and QoL are distinct constructs and both need to be considered when discussing outcomes in adult life.

2.2 Introduction

2.2.1 Defining Autism Spectrum Disorder (ASD)

Autism is a neurodevelopmental disorder that was first described by Leo Kanner (1943). In the DSM-IV (American Psychiatric Association, 1994) it was defined as a triad of impairments but the more recent DSM-5 (American Psychiatric Association 2013) stipulates just two domains of autistic impairment; (i) persistent deficits in social communication and interaction and (ii) restricted, repetitive patterns of behaviour, interests or activities. These impairments lie within a single diagnostic category, Autism Spectrum Disorder (ASD).

The DSM-5 has also addressed the distinction between high functioning autism and Asperger Syndrome (AS). The two disorders were originally distinguished from other ASDs by the presence of normal cognitive skills and from each other by an absence of early language delays in AS (American Psychiatric Association, 2000). However, a language delay is no longer essential for an ASD diagnosis (American Psychiatric Association, 2013a) and there is limited evidence of a difference between the two diagnostic groups (Howlin, 2003; Macintosh & Dissanayake, 2004; Witwer & Lecavalier, 2008). Therefore, neither category is included in the DSM-5 criteria.

For the purpose of this thesis, when discussing previously published studies, the term ASD will be used to describe individuals with a diagnosis of autism, AS/Asperger Disorder (AD), ASD or Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS). The term 'higher ability ASD' will be used to refer to individuals with a diagnosis of autism / ASD and an $IQ \geq 70$ or those previously diagnosed with AS/AD (Szatmari, 2000). When referring specifically to the current study sample, the term 'higher ability autism' will be used because as children, all participants had an IQ outside the intellectual disability range and were diagnosed

with autism at a time when the terms ASD and AS were not yet in use (see Section 2.3.1). Although the term ‘higher ability’ is not an official DSM-5 category, it helps to distinguish this sub-group from others on the spectrum who are intellectually impaired. The cut-off of an IQ ≥ 70 for this distinction is in line with the DSM-V criteria for an intellectual disability (Association, 2013b) and current inclusion criteria for learning disability services in the NHS today. Additionally, it was decided that the term ‘higher ability’ was more appropriate than the more commonly used ‘high functioning autism’ because having an IQ outside the intellectual disability range in childhood does not necessarily mean that individuals are functioning highly in adulthood.

2.2.2 *Why study outcomes in higher ability adults with ASD?*

Given the variability within the autism spectrum, it is important that studies focus on homogenous, well-defined groups. Recent American estimates suggest that almost 70% of all individuals with ASD have an IQ outside the intellectual disability range (≥ 70 ; 69%; CDC, 2014) making research focusing on this sub-group important. However, relatively little is known about this group in adulthood (Howlin and Moss, 2012).

Understanding the adult outcome for this population has important implications. Firstly, most individuals with ASD have continuing needs throughout the life span (Järbrink, McCrone, Fombonne, Zanden, & Knapp, 2007), although research on adults with an IQ outside the intellectual disability range (Howlin, 2004, 2005) is limited. Secondly, the estimated lifetime cost of supporting an adult with ASD without an intellectual disability is £0.92 million in the UK and \$1.4 million in the US (Buescher, Cidav, Knapp, & Mandell, 2014). Thirdly, ASD has a major

impact on society as a whole (Järbrink, Fombonne & Knapp, 2003; Järbrink & Knapp, 2001). Lastly, the transition into mid- to late-adulthood is an area of particular concern for many individuals (Howlin, 2007). People with ASD often rely heavily on familial support well into adulthood (Howlin, 2005; Howlin, Goode, Hutton, & Rutter, 2004) and older parents become increasingly concerned about the future for their children as their own age, or problems of ill health, inhibit their ability to continue to support them (Eaves & Ho, 2008; Howlin, 2004). A clearer understanding of the difficulties that adults with higher ability ASD experience could ensure that they and their families are appropriately supported and that clinicians have accurate guidance regarding the likely outcome for these individuals in order to help minimise the impact of ASD (National Audit Office, 2009).

2.2.3 Adult Social Outcome

When examining the outcome of higher ability adults with ASD, the most commonly used index is their social outcome; defined as the sum of their achievements in independent living, employment, friendships and relationships (Howlin & Moss, 2012). The most frequently used version of this measure (Howlin et al., 2004), was derived from the ratings developed by Rutter, Greenfield and Lockyer (1967).

For individuals with a childhood diagnosis of ASD and an IQ outside the intellectual disability range, outcomes vary widely across studies. Whilst some studies have reported slightly better outcomes for their samples (for example, Farley et al., 2009), most have reported that the majority of adults do not have a good social outcome (for example, Billstedt, Gillberg, & Gillberg, 2005; Howlin et al., 2004; Howlin, Mawhood, & Rutter, 2000; Lotter, 1974; Rutter et al., 1967). Furthermore, there has been limited research into mid- to late-adulthood. Howlin, Moss, Savage

and Rutter (2013) did focus on this age group and they, too, reported relatively poor outcomes for the majority of the sample. Variability in diagnostic ascertainment and in methods of data collection, however, compromise comparisons between studies and limit the overall conclusions that can be drawn.

The core components of adult social outcome map onto more traditional, societal expectations of what constitutes achievements in adulthood, for example, relationships. This has enhanced understanding of what aspects of society individuals with ASD struggle with and how best to support them. However, this measure in isolation may not be sufficient due to its narrow focus on objectively measurable concepts. Recently, there has been a move towards considering the fit between the individual and the environment in which they live (Henninger & Taylor, 2013).

This is particularly pertinent with regard to adults with higher ability ASD as they are often expected by society to achieve more than those with lower ability. However, whilst some do indeed want relationships, for example, this is not always the case. The Social Motivation Theory of ASD suggests that although many individuals with ASD lack social relationships, they also display a diminished preference for joint activities, score lower on friendship questionnaires and do not report greater levels of loneliness (Chevallier, Kohls, Troiani, Brodtkin, & Schultz, 2012), suggesting that they do not all desire relationships. Therefore, a broader outcome measure and / or other aspects of outcome need to be considered to ensure that the lived experience of the individual is addressed.

2.2.4 *Quality of Life (QoL)*

2.2.4.1 Why study QoL in ASD?

QoL is “the individual’s perception of their position in life in the context of culture and value systems in which they live, and in relation to their goals, expectations, standards and concerns” (The WHOQOL Group, 1995, p. 1405). Measuring QoL in ASD provides a comprehensive, multidimensional measure of outcome that accounts for more subjective variables, including satisfaction and subjective wellbeing (Renty & Roeyers, 2006). Perceived QoL amongst higher ability adults with ASD may differ, at least anecdotally, from their adult social outcome; individuals with ‘poor’ / ‘very poor’ social outcomes have often reported high levels of satisfaction with their lives (Moss, 2011). Supporting this, Billstedt, Gillberg and Gillberg (2011) concluded that QoL for individuals with ASD in a mixed IQ sample was better than expected given participants’ low levels of independence.

It has also been observed that parents tend to consider their son or daughter with ASD to be in greater need of support than they themselves believe to be the case (Engstrom, Ekstrom & Emilsson, 2003). This could, in turn, impact on parents’ perceptions of their sons’ or daughters’ QoL. By believing that they require more support, parents may also view their child’s QoL as poorer than the individual with ASD believes to be the case (Engstrom, Ekstrom, & Emilsson, 2003). However, these ideas have yet to be formally investigated.

2.2.4.2 Current research on QoL in ASD

There is a growing body of research on QoL in ASD (for example, Billstedt et al., 2011; Gerber, Baud, Giroud, & Galli, 2008; Persson, 2000; Saldaña et al., 2009). Most recently, van Heijst and Geurts (2015) published a meta-analysis on QoL in

ASD across the lifespan and found that individuals with ASD had a significantly poorer QoL than those without ASD.

Research focussing specifically on QoL in adults with higher ability ASD however, remains limited. Renty and Roeyers (2006) found that perceived informal support was associated with self-reported QoL while received informal and professional support were not, amongst high-functioning males with ASD. However, the study did not make comparisons with normative data, limiting the generalisability of findings. Kamio, Inada and Koyama (2012) did compare QoL scores against normative data and found that self-reported psychosocial QoL was significantly lower than that of the Japanese adult population. Additionally, diagnosis before four years and mother's support were positively associated with QoL. However, these findings must be considered within the context of the very different family culture in Japan, where societal factors contribute to the structure of support systems.

Barneveld et al. (2014) also found that QoL was significantly poorer for young adults with a childhood diagnosis of higher ability ASD than those with other childhood psychiatric disorders. Lastly, Lin (2014) found that the QoL of a higher ability ASD sample was poorer than that of a non-ASD sample and that within the ASD sample, the environmental and physical domains were significantly higher than the psychological and social domains. However, participants in all four studies were all in their early twenties, limiting the applicability of findings to mid-to-late adulthood.

Van Heijst and Geurts (2015) however, did focus on higher ability older adults with ASD; they had significantly lower QoL than a typically developing group but neither IQ nor age nor autism symptomatology were associated with QoL. However, IQ estimates were based on a reading test and the limited variability in IQ restricts conclusions about its potential predictive value.

To date, these are the only known studies to focus specifically on the QoL of higher ability adults with ASD. The variability between these studies regarding (i) the use of control groups, (ii) sample characteristics and (iii) the QoL measure used highlights the need for larger scale systematic QoL research with higher ability adults with ASD in mid-to-late adulthood. It is not yet clear what their QoL is, how it compares to the more traditionally used, society specific adult social outcome measure and what, if any, other factors are associated with QoL. It is widely accepted that lower IQ and language ability and higher levels of autism symptomatology are associated with poorer adult social outcome amongst individuals with ASD (Howlin & Moss, 2012). However, aside from the null results reported by van Heijst and Geurts (2015), these factors require further examination with regard to QoL.

2.2.4.3 Methods of assessing QoL in ASD

Assessing QoL in the ASD population is challenging due to the subjective and abstract nature of the concept and the known difficulties with self-report in this group. There are typically three approaches to measurement in this population; (i) proxy-report (parents/carers reporting what they think the individual with ASD believes their own QoL to be), (ii) informant report (parents'/carers' own views) and (iii) self-report. Proxy-reports are most commonly used amongst individuals with an IQ < 70 (Billstedt et al., 2011). However, discrepancies between self- and proxy-reports of QoL amongst the ASD population have been identified (Kamp-Becker et al., 2011). Furthermore, higher ability individuals with ASD also often struggle with introspection and expressive language (van Heijst & Geurts, 2015). Consequently, they too may lack the skills to effectively judge, and report on, their own QoL (Barneveld et al., 2014), which will, in turn, impact on parents'/carers' ability to accurately report on the self-perceptions of their children.

Using informant data does not require the parent / carer to try to predict the views of the individual with ASD but there are still challenges associated with this method. Sheldrick, Neger, Shipman and Perrin (2012) found lower correlations between informant- and self-reported QoL scores than between proxy- and self-report amongst adolescents with ASD and their parents. Additionally, informants may struggle to distinguish between their *own* perceptions and those of the individual with ASD. Lastly, inappropriate overuse of informant data risks ignoring the voice of the individual with ASD and so must be approached carefully.

As yet, there is no clear approach to measuring QoL in this population. Evidently, self-report is ideal but this is not always appropriate amongst individuals with developmental, cognitive or communication problems. Supplementing self-report data with informant reports could provide useful information but the association between informant and self-report QoL data amongst higher ability individuals remains unclear. Examination of this will help to further our understanding of QoL in this population. Even if discrepant from the self-report data, informant data can provide another perspective and enrich our understanding of the QoL of the person with ASD (van Heijst & Geurts, 2015).

2.2.5 *Research Questions*

The present study aimed to address the following questions with respect to higher ability adults with autism (i.e. those with a childhood IQ outside the intellectual disability range):

1. What is their self-report and informant perceived QoL?
2. Is there a difference between self-report and informant perceived QoL scores?

3. What childhood factors are associated with perceived QoL?¹
4. Is there an association between perceived QoL and adult social outcome scores?¹
5. What other adult factors are associated with perceived QoL?¹

2.3 Method

2.3.1 Clarification of terms

There were four stages to the longitudinal follow-up study that has spanned 40-50 years, of which the current study is in the fourth stage. These will be described in more detail throughout the method but are outlined here initially:

- Childhood Data:
 - Time point 1 (T1) – initial diagnosis and ability measurement
 - Diagnostic confirmation - using standardised measures
- Adult Follow-up study
- Current QoL study

2.3.2 QoL study participants

2.3.2.1 Inclusion criteria

All participants were recruited from a sample who recently took part in the adult follow-up study, conducted by the student for her PhD thesis (Howlin et al., 2013; Howlin, Savage, Moss, Tempier, & Rutter, 2014). All participants were diagnosed with autism as children (T1) based on ICD-9 and ICD-10 criteria and had a childhood Performance IQ (PIQ) outside the intellectual disability range (≥ 70).

Autism diagnoses were then confirmed at some point in childhood / early adulthood

¹ The use of informant and / or self-report data to answer research questions 3-5 depends on the results of research question 2.

(Fombonne, Bolton, Prior, Jordan, & Rutter, 1997; Howlin et al., 2004; Le Couteur et al., 1996) using the diagnostic algorithm of the Autism Diagnostic Interview (ADI; Le Couteur et al., 1989) retrospectively, based primarily on the 4-5 year period.

2.3.3.2 Group status

Whilst all participants had a PIQ outside the intellectual disability range as children, this was not always the case in adulthood. In the current QoL study, participants were divided into two groups; higher and lower adult IQ. Those who obtained a Full-Scale IQ (FSIQ) outside the intellectual disability range based on a Wechsler test at the adult follow-up study (Howlin et al., 2013; Howlin et al., 2014), and had a reading age above 5.1 years (based on the Wechsler Adult Attainment Test; WIAT; Wechsler, 2005), were placed in the 'higher adult IQ' group and the remainder in the 'lower adult IQ' group.

Informants (parents/carers) were invited to take part on behalf of all participants but in the 'higher adult IQ' group, the individual with autism was also invited to complete self-report data describing their QoL (see Section 2.3.6.3 for details). Individuals in the 'lower adult IQ' group were not asked to provide self-report data because the complexity of the QoL questionnaire was too great given their ability level. This decision was supported by a pilot study that was conducted during the adult follow-up study.

2.3.3 Power analysis

The power analysis is based on Research Question three (What childhood factors are associated with perceived QOL?). There have been no previous significant findings regarding the association between QoL and childhood factors. However, the recent

adult follow-up study (Howlin et al., 2013; Howlin et al., 2014) detected significant associations between adult social outcome and child and adult factors (range = 0.27 – 0.77), and two previous studies have reported significant associations between adult social outcome and various child factors (range = -0.34 - 0.83; Farley et al., 2009; Howlin et al., 2004). Based on this literature and the fact that a correlation ≥ 0.4 (i.e. a medium effect size; Pallant, 2007) could be considered sufficiently large to be of likely clinical significance, $r = 0.40$ was used as an estimate of expected effect size. A sample size of 47 was required for a Pearson correlation to have 80% power to detect a clinically and statistically significant association ($r = 0.40$, $\alpha = 0.05$) between two normally distributed continuous variables.

2.3.4 Recruitment process

There were three phases to recruitment for the current QoL study (Table 2.1). All families who were contacted ($N = 59$) gave their consent, during the adult follow-up, to be contacted again for future research.

Table 2.1 Recruitment Process

Phase	Process
1	Families received a newsletter (Appendix I; four similar ones were sent during the adult follow-up study) alerting them to the QoL study and its connection with the previous research.
2	Everyone was contacted by post, unless they requested no further contact.
3	Those who did not respond were contacted by telephone to discuss participation. ¹

¹ Of the 20 families contacted by telephone, 15 informants and one participant returned completed questionnaires. Of the remaining five, one agreed to participate but did not return the questionnaire and four had inactive numbers or had moved without leaving a forwarding address.

In most cases, personalised letters were sent to parents / carers². They were the first point of contact during the adult follow-up study as most individuals in this group rely heavily on support from parents / carers. All letters included an information sheet, two consent forms, the informant version of the World Health Organisation Quality of Life – Brief Version (WHOQOL-BREF; The Whoqol group, 1998; Appendices II & III) and a stamped addressed envelope. Informants were asked to return the completed questionnaire and consent form. In the ‘higher adult IQ’ group, autism specific information sheets, consent forms and the WHOQOL-BREF were also included (Appendices II & III).

2.3.5 Response rates

Figure 2.1 summarises the sample sizes at each stage of the longitudinal study. Of the 90 families who were seen in childhood, 60 participated in the adult follow-up and 52 of *those* families were seen again at the QoL study (positive response rates: 88% from adult follow-up to QoL study and 58% overall; T1 to the QoL study).

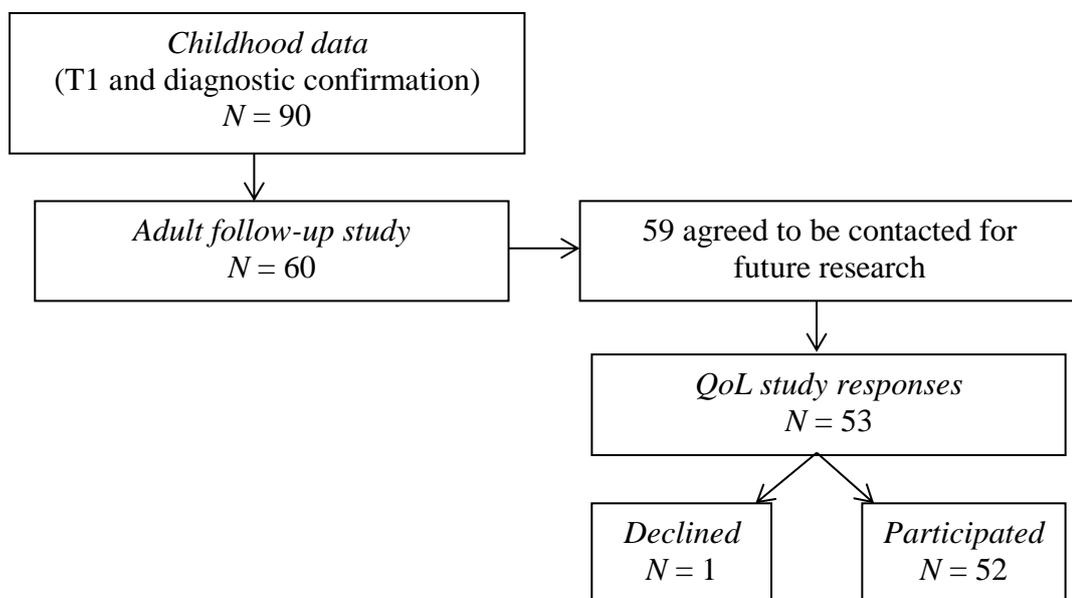


Figure 2.1. Sample sizes at the different time points

² No informant was available in two cases so letters were sent directly to the participant.

All data reported henceforth refers to the 52 individuals who participated in the current QoL study. Table 2.2 details the response rates according to the two QoL study sub-groups. In total, 50 informants and 22 higher ability individuals with autism returned completed WHOQOL-BREFs.

Table 2.2 Positive response rates according to group

	Lower adult IQ	Higher adult IQ ¹		Total ²
	<i>n</i> (%)	<i>n</i> (%)		<i>N</i> (%)
	Informant	Informant	Self-report	
Total contacted	30 (100%)	27 (93%)	29 (100%)	59 (100%)
Positive response	28 (93%)	22 (82%)	22 (76%)	52 (88%)
Negative response	0 (-)	1 (4%)	1 (3%)	1 (2%)
<i>Total response</i>	<i>28 (98%)</i>	<i>23 (85%)</i>	<i>23 (79%)</i>	<i>53 (90%)</i>

¹The 22 cases described in the higher IQ sub-group did not constitute matched pairs: two individuals with autism participated but did not have an informant and two informants in this sub-group participated but the individual with autism did not (i.e. there were 24 higher IQ families in total)

² ‘Total’ column refers to the total number of families who took part via informant and/or self-report

2.3.6 Measures

This sample completed a number of measures throughout the longitudinal follow-up study. However, only those measures used in the current QoL study are described below. See Howlin et al. (2013, 2014) for details of all other measures used.

2.3.6.1 Childhood data

When participants were seen at T1, their overall language level (based on the ADI categories of no language, few words or phrase speech) and PIQ scores were obtained. Some participants completed more than one IQ test so a scoring system

based on test quality and age, was used to select the score from the most appropriate test at the optimum age (Appendix IV). Autism diagnoses were re-confirmed at some stage between initial diagnosis and adult follow-up using the diagnostic ADI algorithm retrospectively based primarily on the 4-5 year period (Le Couteur et al., 1989), providing childhood autism symptomatology scores.

2.3.6.2 Adult follow-up study data

Cognitive ability, Language and Autism Symptomatology

Where possible, all participants completed an IQ test, ideally the Wechsler Adult Intelligence Scale-III (WAIS-III; Wechsler, Wycherley, & Benjamin, 1997). When this was not possible, due to limited verbal ability and/or attention span, an alternative test was completed (Figure 2.2). Expressive language level was rated using the five category system in the Autism Diagnostic Interview-Revised (ADI-R; Rutter, Le Couteur, & Lord, 2003; see Table 2.4 for categories). This differs from the three category system used at T1 because the ADI-R was not available then. The British Picture Vocabulary Scale (BPVS; Dunn, Dunn, Whetton, & Burley, 1997) measured receptive language level. Autism symptomatology was assessed using the *current* items from the ADI-R which was completed by parents / caregivers. This includes domain scores for Communication, Reciprocal Social Interaction (RSI), and Restrictive and Repetitive Behaviours and Interests (RRBI) and a total symptom severity score.

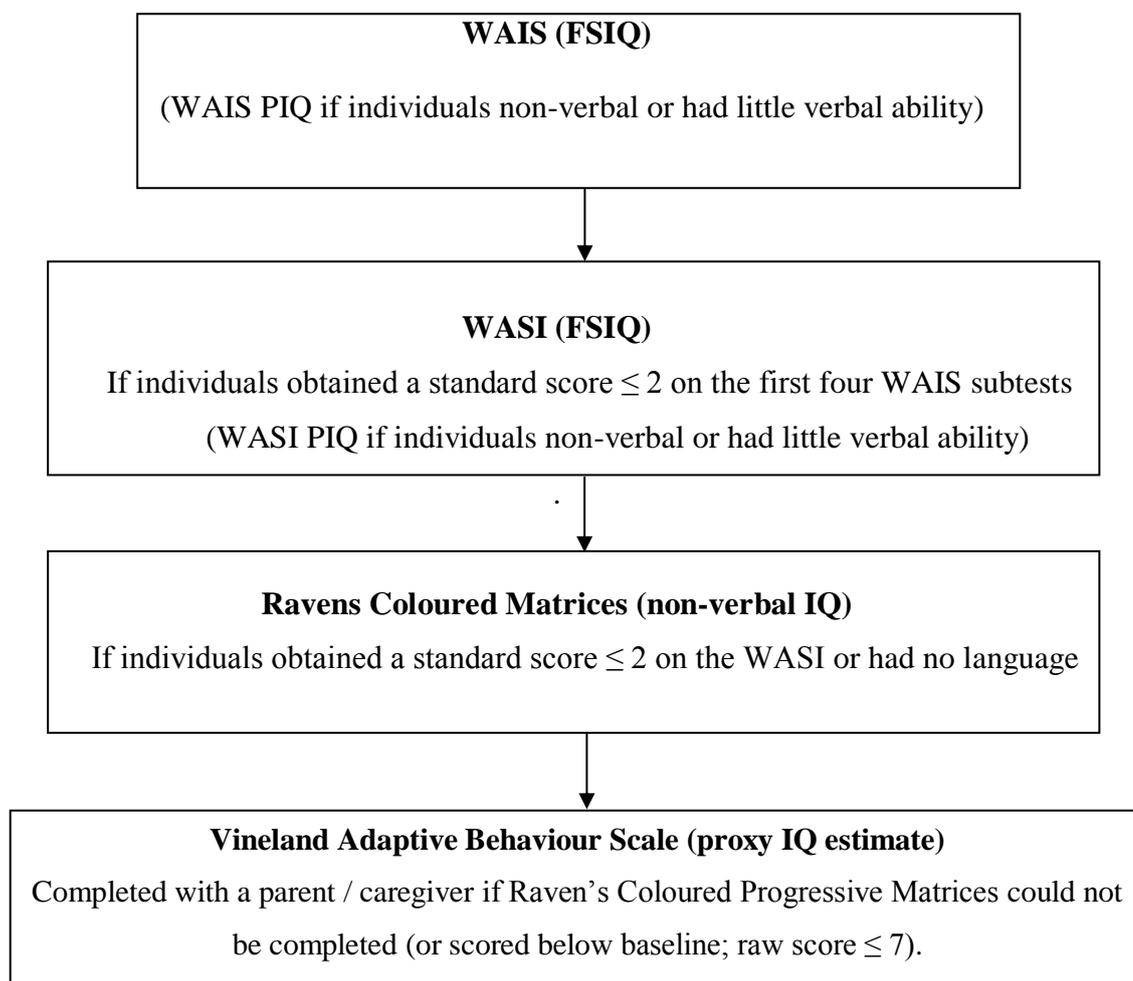


Figure 2.2 IQ test hierarchy³

Adult Social and Mental Health Outcome

Information on adult social and mental health outcome was assessed using the informant version of the Family History Schedule (FHS); a semi-structured interview that has been used as an indicator of outcome in many autism studies (Bolton et al., 1994; Pickles et al., 2000; Pinto et al., 2010; Sterling, Dawson, Estes, & Greenson, 2008). The adult social outcome ratings were based on data from the FHS and ADI-R, and was derived from the categories used by Rutter et al. (1967). The rating

³ Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999), Raven's Coloured Progressive Matrices (RCPM; Raven, 1986) and Vineland Adaptive Behavior Scale (VABS-II; Sparrow, Cicchetti, & Balla, 2005)

includes scores for employment, friendships, relationships and independent living (range = 0 – 3; 3 = lowest level of functioning). Scores on the four domains are then summed to create a composite outcome score for each individual (range = 0 – 12; 12 = poorest outcome), which is converted into a global ordinal adult outcome rating (very good, good, fair, poor and very poor; Appendix V).

A composite mental health score was based on FHS scores for five mental health difficulties (OCD, episodic depression, chronic depression and bipolar and anxiety disorder) according to whether the individual had ‘ever’ (i.e. since 16 years old) experienced the difficulty (range = 0 – 3; 0 = no difficulty, 3 = hospitalised because of the disorder). Scores for each disorder were then summed to create a composite mental health outcome score (range = 0 – 15; 15 = severe difficulties in all four areas). Figure 2.3 summarises the number of assessments completed at adult follow-up for all individuals with autism in the QoL study.

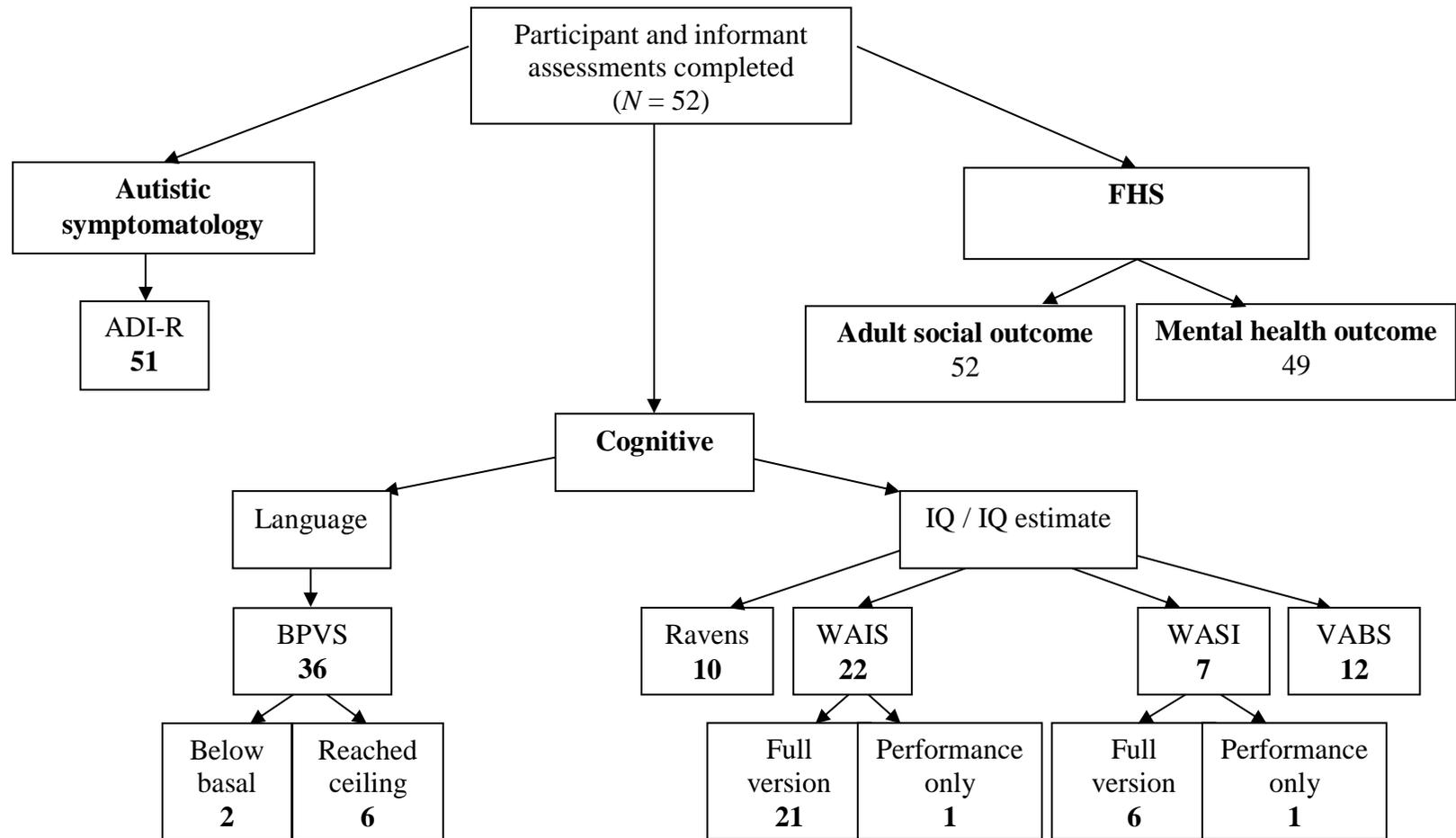


Figure 2.3 Assessments completed by the sample at adult follow-up
Not all participants and/or informants were able to complete all assessments.

2.3.6.3 Current QoL study

The WHOQOL-BREF (The WHOQOL Group, 1998, Appendix III) is a 26 item questionnaire based on the 100 item WHOQOL (The WHOQOL Group, 1995). Individuals rate items based on the past four weeks, using a five point scale (very poor, poor, neither good nor poor, good and very good), generating four domain scores; (i) physical health, (ii) psychological well-being, (iii) social relationships and (iv) quality of the environment. Scores are then transformed into a 0-100 scale in order to facilitate comparisons with UK normative data (Skevington and McCrate, 2012; Appendix VI). The measure also includes two separate general questions; (i) ‘how would you rate your quality of life?’ and (ii) ‘how satisfied are you with your health?’

The measure has been used with a range of populations and UK norms have been published (Skevington & McCrate, 2012). It has good-to-excellent psychometric properties in the general population including internal consistency, test re-test reliability and discriminant and concurrent validity (Skevington & McCrate, 2012). It has also been used with a higher ability adult ASD population (Kamio, Inada and Koyama, 2012). The standardised self-report version was sent to all participants in the higher ability sub-group and basic modifications were made to this version to make it suitable for informants of individuals with autism in both sub-groups. For example, “how would you rate *their* QoL?” (Appendix III).

2.3.7 *Situating the study within the context of the wider research project*

This study is not a standalone piece of research and cannot be viewed without consideration of the numerous research stages that came before it (see Section 2.3.1 for details), not least because data from these previous time points was used in the

analysis. Being part of a longitudinal follow-up study had implications for (i) measure selection and methods of data collection and (ii) determining who would be able to complete the self-report questionnaire. Regarding the former, given that the individuals were longstanding participants of a research project that has spanned 40-50 years, they had all taken part in many data collection phases over the years. The most recent of these was the adult follow-up study, which involved a lengthy face-to-face assessment process. Many participants reported that they found this process quite tiring. Therefore, care was taken during the current QoL study to ensure that participants were not overwhelmed by the volume of assessments again, hence the decision to use a single brief postal questionnaire to measure QoL.

Regarding the latter, in addition to the main criteria that were used to determine who could complete the self-report questionnaire (adult reading ability and FSIQ), clinical judgement was also used. Once the group was split into those who were of higher and lower adult ability, the groups were examined to ensure that no one was being asked to complete the self-report measure who would be unable to do so. These checks were conducted by the student and second supervisor (PH) who between them knew the entire sample well. Additionally, participants were clearly advised in the information sheet to seek support when completing the questionnaire if they needed to. Whilst the majority did seek support, none reported that they struggled with the questionnaire.

2.3.8 Ethics

The adult follow-up study had ethical approval from the NHS because all participants were initially recruited from a sample of individuals who were, as children, patients at the Maudsley hospital (project reference: 07/H0807/65).

However, this ethical approval had closed by the time of the current study. Upon advice from the UCL ethics committee, ethical approval for the current study was sought and received from UCL (project reference: 4111/001; Appendix VII), rather than the NHS, for the following reasons:

- All individuals were voluntary participants in the adult follow-up study.
- None were identified from current patient records.
- Only those who had agreed to be contacted for future research were approached.
- There were no capacity to consent concerns; (i) only individuals with autism with an adult FSIQ ≥ 70 and a reading age ≥ 5.1 years were asked to complete the self-report WHOQOL-BREF, and (ii) the informant questionnaire asked parents / carers for their *own* perception of the QoL of the individual with autism, not the views of the person with autism.

2.3.9 Data analysis

A mixed within and between subjects design was used. Where assumptions were met, parametric statistical analyses were conducted. Non-parametric tests were used when assumptions of normality were violated. ANOVA's compared the self-report data with normative data. Mann Whitney *U* / t-tests compared informant QoL scores between the lower and higher adult ability groups and correlations and t-tests compared informant and self-report QoL scores in the higher adult ability group.

Correlations were conducted between QoL scores and the various child and adult factors under consideration. Mann Whitney *U* or T-tests were used to assess the difference in QoL scores between those with and without language in childhood. No

analysis was conducted comparing males and females due to the very small numbers of females for each analysis.

The number of correlations conducted increased the risk of type I error. However, due to the sample size (particularly the self-report data; $n = 22$), reporting findings based on a bonferroni corrected p -value could have inflated the risk of type II error. Consequently, all findings with a p -value $< .05$ were treated as significant. However, p -values between $.01$ and $.05$ were reported separately and treated with caution.

2.4 Results

2.4.1 The sample

2.4.1.1 The current QoL study

In total, 52 individuals with a childhood diagnosis of higher ability autism took part (either themselves and/or their informant). Of these, 24 (21 male; gender ratio = 7 : 1) had an adult IQ ≥ 70 and 28 (22 male; gender ratio = 3.7 : 1) had an adult IQ < 70 . There were 50 informants (28 for the lower and 22 for the higher adult IQ group); 22 individuals (19 male; gender ratio = 6.3 : 1) in the higher adult IQ group also completed the self-report questionnaire⁴.

The mean age of the total sample was 49 years 4 months (SD = 9 years 7 months; range = 34 years 10 months – 69 years 6 months); the mean age of the lower adult IQ group was 47 years 10 months (SD = 9 years 5 months, range = 34 years 8 months – 69 years 6 months) and the mean age of the higher adult IQ group was 51 years 0 months (SD = 9 years 5 months, range = 36 years 9 months – 68 years 5

⁴ Two individuals with autism completed the self-report measure but their informant did not take part

months). Sections 2.4.1.2 and 2.4.1.3 provide descriptive data relevant to the QoL study from childhood and adult follow-up.

2.4.1.2 Childhood data

At T1, the mean age of the total sample was 6 years 4 months (SD = 2 year 1 months, Range = 2 years 9 months – 13 years 3 months). The mean age of the lower adult IQ group was 6 years 1 months (SD = 1 year 10 months, Range = 3 years 5 months – 10 years 0 months) and of the higher adult IQ group was 6 years 10 months (SD = 2 year 5 months, Range = 2 years 9 months – 13 years 3 months).

Table 2.3 describes the current participants' childhood language level, cognitive ability⁵ and autism symptomatology scores (obtained retrospectively at diagnostic confirmation). Scores for overall language level (using the ADI three category system) were based on assessments conducted at, or as close as possible, to the time that childhood IQ was established. Clinical reports were used when no formal language assessments were conducted.

⁵ See Appendix VIII for number of participants who completed each IQ test in childhood.

Table 2.3 Childhood data

		Lower adult IQ <i>n</i> = 28	Higher adult IQ <i>n</i> = 24	Group comparison ⁴	Total sample <i>N</i> = 52
<i>Language level</i> ¹					
<i>N</i> (%)					
Score	Category				
0	Good	8 (30%)	18 (75%)		26 (51%)
1	Few words	9 (33%)	2 (8%)	< .01	11 (22%)
2	None	10 (37%)	4 (17%)		14 (27%)
<i>Performance IQ</i>					
Overall Mean (SD, range)					
<i>N</i> (%) per test					
		86.10 (13.3, 70 - 118)	93.00 (15.2, 70 - 133)	.09	89.30 (14.4, 70 - 133)
<i>Autism Symptomatology on ADI algorithm (4-5 years)</i>					
Mean					
(SD, range)					
<i>N</i> (%) above cut off ²					
Reciprocal Social Interaction		20.75 (4.33, 12 – 28) 28 (100%)	17.04 (5.63, 5 – 26) 24 (100%)	< .05	19.04 (5.27, 5 – 28) 50 (96%)
Communication		13.75 (4.33, 6 – 22) 28 (100%)	14.00 (2.90, 8 – 20) 24 (100%)	.81	13.87 (3.71, 6 – 22) 52 (100%)
RRBI		7.25 (2.29, 2 – 11) 28 (100%)	7.63 (2.00, 4 – 11) 24 (100%)	.54	7.42 (2.15, 2 – 11) 51 (98%)
<i>Total</i> ³		41.75 (7.08, 26 – 56) 28 (100%)	38.17 (7.78, 25 – 51) 24 (100%)	.09	40.10 (7.42, 25 – 56) 52 (100%)

¹ One person in the lower IQ subgroup was missing a language score

² Cut-off scores: Reciprocal Social Interaction = 10, Communication = 8 (6 non-verbal), RRBI = 4.

³ Met cut-off on ≥ 3 diagnostic domains

⁴ *p*-values comparing scores between lower and higher ability sub-groups (Fisher's exact test used for categorical and t-tests for continuous variables). Mean scores used for ADI comparisons

RRBI = Restricted and Repetitive Behaviours and Interests

Maximum scores: Reciprocal Social Interaction = 28, Communication = 24, RRBI = 12, Total = 64

2.4.1.3 Adult Follow-up study

Table 2.4 summarises participants' language level, IQ and autistic symptomatology scores at adult follow-up. There was a significant difference between the two groups on all variables. Less than half of the lower adult IQ group was fully verbal and 85% had a receptive language level below eight years. Severity of autistic symptomatology was also higher ($\geq 79\%$ above cut-off in all three domains). The higher adult IQ group fared noticeably better (96% were fully verbal) but many still had language difficulties; 30% had a receptive language level below 13 years. Severity of autistic symptomatology was varied (<50% reached cut-off for RRBI's but 60% and 78% scored above cut-off for the RSI and Communication domains respectively).

Table 2.5 summarises adult social and mental health outcome scores. Most individuals were doing poorly in adulthood; no individual in the lower adult IQ group had a 'good' or 'very good' social outcome. Rates of mental health difficulties varied; some individuals had no problems but many reported a range of significant difficulties.

Table 2.4 Adult follow-up data

		Lower adult IQ <i>n</i> = 28	Higher adult IQ <i>n</i> = 24	Group comparison ¹	Total sample <i>N</i> = 52 ²
<i>Language level</i>					
<i>N</i> (%)					
Score	Category				
0	Fully verbal	13 (46%)	23 (96%)		36 (69%)
1	Spontaneous language without verbs/grammar	3 (11%)	1 (4%)	< .001	4 (8%)
2	Echoed and/or stereotyped speech, ≥ 5 words in last month and some phrases	3 (11%)	-		3 (6%)
3	> 5 words daily	5 (18%)	-		5 (10%)
4	Non-verbal	4 (14%)	-		4 (8%)
<i>British Picture Vocabulary Scale (BPVS)</i>					
<i>N</i> (%)					
Unable to complete test		15 (54%)	-		15 (29%)
0 – 7 years 11 months		11 (39%)	2 (8%)		13 (25%)
8 years 0 months–ceiling ³		2 (7%)	21 (88%)	< .001	23 (44%)
Declined test		-	1 (4%)		1 (2%)
<i>Single word reading age</i>					
Mean (SD, range) ⁴		9.30 (2.61, 6.0 – 14.0)	15.91 (3.61, 9.80 – 19.0)	< .001	14.15 (4.46, 6.0 – 19.0)
<i>IQ estimate</i>					
Mean (SD, range)		52.18 (30.35, 20 – 105)	91.57 (19.22, 70 – 139)	< .001	69.94 (32.40, 20 – 139)
<i>Autism Symptomatology</i> ²					
Mean score					
(SD, range)					
<i>N</i> (%) above cut-off ⁵					
RSI		16.71 (2.59, 11 – 20)	10.39 (4.43, 2 – 20)	< .001	13.86 (4.73, 2 – 20)
		28 (100%)	14 (60%)		42 (82%)

	Lower adult IQ <i>n</i> = 28	Higher adult IQ <i>n</i> = 24	Group comparison ¹	Total sample <i>N</i> = 52 ²
Communication	10.93 (4.87, 2 – 20) 22 (79%)	8.96 (2.44, 5 – 14) 18 (78%)	.07	10.04 (4.05, 2 – 20) 40 (78%)
RRBI	4.29 (2.16, 1 – 9) 22 (79%)	2.96 (2.01, 0 – 6) 11 (48%)	< .001	3.69 (2.18, 0 – 9) 33 (65%)
<i>Total</i>	<i>31.93</i> (5.43, 24 – 47) 28 (100%)	<i>22.39</i> (7.08, 7 – 33) 23 (96%)	< .001	<i>27.63</i> (7.81, 7 – 47) 51 (98%)

¹ Comparisons conducted between the lower and higher adult IQ sub-groups; Fisher's exact tests conducted with categorical and t-tests with continuous variables (Mann Whitney *U* with Restricted and Repetitive Behaviours and Interests (RRBI) domain). Language level, EOWPVT and BPVS categories compressed to allow for 2x2 comparison tables (i.e. language level above and below 8 years). Analysis based only on those who completed test.

² One higher adult IQ individual did not have an informant and did not consent to formal assessments.

³ Maximum age equivalent on the BPVS = 18 years 0 months.

⁴ Eight individuals in the lower adult ability and 22 in the higher adult ability sub-group completed the WIAT single word reading subtest.

⁵ Cut-off scores: Reciprocal Social Interaction (RSI) = 10, Communication = 8 (6 non-verbal), RRBI = 4
Maximum scores: RSI = 28, Communication = 24, RRBI = 12, Total = 64

Table 2.5 Adult social and mental health outcome scores

Outcome Category		Lower adult IQ <i>n</i> = 28	Higher adult IQ <i>n</i> = 24	Group Comparison ¹	Total sample <i>N</i> = 52
<i>Adult Social</i>					
	Mean	11.11	6.73	< .001	9.09
	(SD, range)	(1.41, 7 – 12)	(3.19, 0 – 12)		(3.24, 0 – 12)
Very good / good		0	5 (21%)		5 (10%)
Fair	<i>N</i> (%)	2 (7%)	13 (54%)	< .001	15 (29%)
Poor / Poor		26 (93%)	6 (25%)		32 (62%)
<i>Mental health</i> ¹					
	Mean	1.24	1.17	0.86	1.21
	(SD, range)	(1.57, 0 – 5)	(1.72, 0 – 5)		(1.62, 0 – 5)
Very good / good		15 (56%)	16 (73%)		31 (63%)
Fair	<i>N</i> (%)	1 (4%)	0	0.37	1 (2%)
Poor / Very poor		11 (41%)	6 (27%)		17 (34%)

¹ Comparisons conducted between the lower and higher adult IQ groups; Mann-Whitney *U* tests for continuous data, Fishers exact tests for categorical data (categories compressed down to three; good/very good, fair and poor/very poor).

² Mental health outcome scores missing for one participant in lower IQ and two in higher IQ group

2.4.2 WHOQOL-BREF

2.4.2.1 Overall Quality of Life (QoL)

Only individuals with autism in the higher adult IQ group were asked to complete the self-report version of the WHOQOL-BREF (see Section 2.3.3.2). However, this distinction, between higher and lower adult IQ, was not intended for the informant data (all parents / carers were asked to complete the questionnaire). Supporting this, there were no significant differences between the higher and lower adult IQ groups across all four informant perceived QoL domains (*p*-values > .05). Therefore, all subsequent analyses of informant based data combined these sub-groups.

Research Question 1: What is the self-report and informant perceived QoL for adults with autism and a childhood IQ outside the intellectual disability range?

Table 2.6 describes the informant and self-report WHOQOL-BREF scores (higher scores indicate better perceived quality of life).

Table 2.6 Summary scores on the WHOQOL-BREF

Quality of Life Domain	WHOQOL-BREF	
	<i>n</i> (%) ¹	
	Mean (SD, Range)	
	Informant	Self-report ²
Physical	47 (94%)	22 (100%)
	72.40 (13.65, 13 – 94)	81.09 (9.93, 63 – 100)
Psychological	45 (90%)	22 (100%)
	63.18 (13.23, 25 – 94)	72.14 (15.79, 31 – 100)
Social	45 (90%)	20 (95%)
	56.11 (14.71, 0 – 75)	69.53 (23.34, 6 – 100)
Environment	49 (98%)	22 (100%)
	74.35 (10.26, 50 – 100)	76.64 (11.02, 63 – 100)

N (%) = number and percentage of participants / informants who completed the measure.

2.4.2.2 Comparison with Normative data

Self-report data from the current study were compared with the healthy sample and a psychiatric disorders population (depression, chronic schizophrenia and mild dementia) from a UK population study investigating the WHOQOL-BREF (Skevington and McCrate., 2012). Self-report (not informant) data from the current autism sample was used as only self-report data was available for the comparison samples.

All ANOVA's were significant at the $p < .01$ level (Table 2.7). Tukey HSD post-hoc analyses revealed a significant difference between the psychiatric and other two samples in the physical and psychological domain (psychiatric < healthy, autism) and between the psychiatric and healthy sample in the social domain (psychiatric < healthy). In the environmental domain, there was a significant difference between all three samples (psychiatric < healthy < autism).

Table 2.7 ANOVAs comparing the autism sample self-report domain scores and samples from Skevington and McCrate (2012)

Quality of Life Domain	Quality of Life score			df (Between, Within)	<i>F</i>	<i>p</i> -value
	Mean (SD)					
	Autism (<i>n</i> = 20 – 22)	Healthy ¹ (<i>n</i> = 1324 – 1328) ²	Psychiatric ¹ (<i>n</i> = 77 – 80) ²			
Physical	81.09 (9.93)	76.49 (16.19)	54.57 (20.62)	2, 1423	67.27	< .001
Psychological	72.14 (15.79)	67.71 (15.56)	45.93 (25.99)	2, 1423	67.07	< .001
Social	69.53 (23.34)	70.52 (20.67)	61.91 (20.80)	2, 1421	6.37	< .01
Environment	76.64 (11.02)	68.20 (13.81)	61.0 (17.02)	2, 1423	14.13	< .001

¹Healthy sample = students, nurses, carers and dental practitioners, Psychiatric sample = depression, chronic schizophrenia and mild dementia

²Skevington and McCrate (2012) did not clarify the *n* for each domain for each sample, rather the range of *n* across all domains so mid-points were used for approximate *n* (i.e. healthy = 1326 and psychiatric = 78)

2.4.3 Informant versus self-report data

Research Question 2: Is there a difference between informant and self-reported QoL scores amongst adults with a childhood diagnosis of autism and adult IQ outside the intellectual disability range?

Only the informant perceived scores on the social domain ($p = .05$) and overall QoL (Q1; $p = .03$) were marginally significantly poorer than their respective self-report scores (Table 2.8). However, none of the self-report QoL domains were significantly associated with their respective informant perceived domains either (Table 2.9).

Given these findings, it is not appropriate to rely entirely on informant perceived QoL as a substitute for self-report data as they are evidently not strongly associated with one another. Therefore, both self-report and informant perceived QoL scores were used where available for all subsequent analyses.

Table 2.8 T-tests between informant and self-report data in the higher adult IQ group

Quality of Life		Mean	<i>n</i>	t-score	df	<i>p</i> -value
Domain	Data Source					
Physical	Informant	75.90	20	-1.92	19	.07
	Self-report	80.75				
Psychological	Informant	67.58	19	-1.38	18	.19
	Self-report	72.79				
Social	Informant	59.11	18	-2.08	17	.05*
	Self-report	71.28				
Environment	Informant	73.75	20	-.91	19	.37
	Self-report	76.75				
Q1	Informant	3.90	20	-2.44	19	.03*
	Self-report	4.35				
Q2	Informant	4.00	20	-1.55	19	.14
	Self-report	4.30				

* $p < .05$ ** $p < .01$

Question 1: How would you rate your / the person with autism's QoL?

Question 2: How satisfied are you with your / the person with autism's health?

Table 2.9 Correlations between informant and self-report data

Quality of Life domains	<i>n</i>	Correlation Coefficient	<i>p</i> -value
Physical	20	.35	.14
Psychological	19	.39	.10
Social	18	.01	.97
Environment	20	-.02	.95

2.4.4 Childhood factors associated with perceived QoL

Research Question 3: What childhood factors are associated with perceived QoL amongst adults with autism and a childhood IQ outside the intellectual disability range?

None of the informant domains were significantly correlated with any childhood factors (Table 2.10). At the $p < .01$ level, only ADI total score was significantly negatively correlated with self-reported overall satisfaction with health (Q2); higher levels of childhood autistic symptom severity were associated with lower levels of adult satisfaction with health. At the $p < .05$ level, three of the self-report QoL domains (physical, social and environmental) and self-report overall satisfaction with health (Q2) were significantly negatively correlated the diagnostic RRBI ADI domain; higher levels of childhood RRBI symptoms were associated with poorer adult QoL and satisfaction with physical health. Additionally, self-report overall QoL (Q1) was significantly negatively correlated with IQ; higher childhood IQ was associated with lower adult global QoL.

There were no significant differences between QoL scores and childhood language ability at the $p < .01$ level (Table 2.11). However, at the $p < .05$ level, there were significant differences in informant perceived psychological QoL; individuals

without language in childhood had poorer informant perceived adult psychological well-being than those with language in childhood.

Table 2.10 Correlations between adult QoL and childhood factors

Quality of Life		<i>n</i>	Childhood Variable Correlation Coefficient				
Data source	Domain		IQ	Diagnostic ADI algorithm			
				RSI	Communication	RRBI	Total
Informant	Physical	47	.02	.05	-.04	.11	.00
	Psychological	45	-.12	-.03	.14	.12	.01
	Social	45	-.04	.28	-.11	.23	.19
	Environment	49	-.05	.14	-.07	-.05	.03
	Q1	50	-.20	-.08	-.16	.02	-.16
	Q2	50	-.16	.00	-.09	-0.09	-0.10
Self-report	Physical	22	-.34	.14	-.02	-.44*	-.10
	Psychological	22	-.26	.21	-.07	-.50*	-.05
	Social	20	-.36	.36	.19	-.24	.22
	Environment	22	.04	-.03	-.12	-.51*	-.16
	Q1		-.44*	.02	.06	-.023	-0.1
	Q2		-0.24	-0.32	-0.36	-0.51*	-0.55**

* $p < .05$ (2-tailed)

** $p < .01$ (2-tailed)

ADI = Autism Diagnostic Interview, RSI = Reciprocal Social Interaction, RRBI = Restricted and Repetitive Behaviours and Interests

Question 1: How would you rate your / the person with autism's QoL?

Question 2: How satisfied are you with your / the person with autism's health?

Table 2.11 T-tests comparing QoL domain scores and childhood language level

Quality of Life		<i>n</i>		<i>t</i>	df	<i>p</i> -value
		Mean (SD)				
Data source	Domain	Language	No Language			
Informant	Physical	25	21	.31	44	.76
		72.80 (15.73)	71.52 (11.28)			
	Psychological	24	21	2.53	43	.02*
		67.58 (12.29)	58.14 (12.70)			
	Social	25	20	.23	43	.82
		56.56 (12.40)	55.55 (17.50)			
	Environment	25	23	-.54	46	.60
		73.56 (10.0)	75.17 (10.92)			
	Q1	25	24	.16	47	.87
		3.96 (0.93)	3.92 (0.93)			
Q2	25	24	.28	47	.78	
	4.0 (1.08)	3.92 (1.02)				
Self-report	Physical	17	5	-.13	20	.90
		80.94 (10.44)	81.60 (9.02)			
	Psychological	17	5	-1.45	20	.16
		70.71 (16.54)	82.60 (14.19)			
	Social	15	5	-1.54	18	.14
		68.0 (23.61)	85.0 (12.95)			
	Environment	17	5	-1.11	20	.28
		75.24 (10.33)	81.40 (13.20)			
	Q1	17	5	-.65	20	.52
		4.35 (0.79)	4.6 (0.55)			
Q2	17	5	-.32	20	.76	
	4.29 (0.69)	4.40 (0.55)				

* $p < .05$ (2-tailed)

** $p < .01$ (2-tailed)

Question 1: How would you rate your / the person with autism's QoL?

Question 2: How satisfied are you with your / the person with autism's health?

2.4.5 Adult factors associated with perceived QoL

Research Question 4: Is there an association between perceived QoL and adult social outcome scores amongst adults with autism and a childhood IQ outside the intellectual disability range?

None of the informant perceived QoL domains were associated with adult social outcome (Table 2.12). However, the self-reported social QoL domain was significantly positively associated with adult social outcome ($p < .01$); as adult social outcome improves, so does the self-reported quality of social relationships in adulthood.

Table 2.12 Correlations between adult social outcome and QoL domains

Quality of Life		<i>n</i>	Correlation coefficient
Data source	Domain		
Informant	Physical	47	-.23
	Psychological	45	-.25
	Social	45	-.04
	Environment	49	.09
Self-report	Physical	22	.19
	Psychological	22	.20
	Social	20	.57**
	Environment	22	.17

* $p < .05$ (2-tailed)

** $p < .01$ (2-tailed)

Research Question 5: What other adult factors are associated with QoL amongst adults with autism and a childhood IQ outside the intellectual disability range?

At the $p < .01$ level no adult variables were significantly associated with informant perceived or self-reported QoL (Table 2.13), although age and informant perceived physical QoL and IQ and self-reported social QoL were negatively correlated ($p < .05$). Thus, as age and adult IQ increased, informant perceived physical and self-reported social QoL declined.

Table 2.13 Correlations between QoL scores and adult factors

Quality of Life		<i>n</i>	Correlation Coefficient							
Data source	Domain		IQ ¹	Adult ADI-R algorithm ¹			Mental health outcome ²		Language ³	Age
				RSI	Communication	RRBI	Total			
Informant	Physical	47	.13	-.25	-.10	-.06	-.20	.03	-.02	-.34*
	Psychological	45	.25	-.25	.05	-.25	-.19	.07	-.20	-.09
	Social	45	-.04	-.04	-.20	-.16	-.16	-.02	.14	-.17
	Environment	49	-.10	.08	.10	-.10	.08	-.06	.05	.09
Self-report	Physical	22	-.17	.15	.17	-.22	.09	-.11	n/a	-.24
	Psychological	22	-.31	.07	.05	-.28	-.02	-.05	n/a	-.08
	Social	22	-.56*	.21	.13	.01	.17	.27	n/a	.03
	Environment	22	.04	.08	.06	-.05	.06	.20	n/a	-.09

* $p < .05$ (2-tailed)

** $p < .01$ (2-tailed)

ADI-R = Autism Diagnostic Interview – Revised, RSI = Reciprocal Social Interaction, RRBI = Restricted and Repetitive Behaviours and Interests

¹ One participant who completed the self-report WHOQOL-BREF was missing ADI and IQ scores at follow-up (i.e. ADI and IQ correlations with self-report data: $n = 21$ and between mental health outcome and self-report data: $n = 20$)

² Two participants who had informant data were missing a mental health outcome score

³ All participants who completed the self-report questionnaire had language at follow-up preventing any meaningful analysis

2.5 Discussion

In the current study, child and adult correlates of QoL were examined among 52 adults with autism and a childhood PIQ outside the intellectual disability range ($IQ \geq 70$). Informant- and self-report QoL scores were also compared, as were the current sample's QoL with normative data.

2.5.1 Summary and interpretation of results

2.5.1.1 Overall QoL

There was variability in the sample's QoL, some individuals had a good QoL (informant perceived and/or self-report) whereas others had a poor QoL. When compared with the normative data (Skevington and McCrate, 2012), the higher adult IQ sub-group self-reported a significantly better QoL than the psychiatric sample in three domains (physical, psychological and environmental) and the healthy sample on the environmental domain.

The better self-reported QoL in the higher adult IQ autism sample compared with the psychiatric sample contrasts with recent findings by Barneveld et al. (2014) who found that individuals with a childhood diagnosis of ASD had a poorer self-reported QoL than those with other childhood psychiatric disorders. However, the comparisons made in the current study were with a psychiatric sample that had mental health difficulties in adulthood not childhood (data obtained from Skevington and McCrate, 2012). Therefore, their *current* mental health difficulties are more likely to have impacted on their current QoL than childhood psychiatric diagnoses would have. These findings suggest that current mental health difficulties are more closely associated with current self-reported QoL than a childhood diagnosis of autism.

The significantly better self-reported QoL in the environmental domain for the higher adult IQ autism sample compared to the healthy sample is harder to understand. The result is actually in line with the pattern of findings reported by Lin (2014). Whilst Lin found that a higher ability ASD sample had a poorer QoL than a non-ASD sample in all four domains, they *did* report that the smallest difference between the two groups was in the environmental domain (the difference was only significant at the .05 rather than .01 and .001 level, as was the case with the other domains). Additionally, the environmental and physical domains were significantly higher than the other two domains in the ASD sample. Whilst the current findings do not replicate those reported by Lin (2014) exactly, the pattern of findings in the two studies does indicate that the self-reported environmental QoL of individuals with ASD is not in line with the pattern found for the other three domains and therefore warrants consideration.

It is important to consider why self-reported environmental QoL scores in the current study are higher in this sample than a healthy population. Self-reported QoL is shaped by an individual's personal frame of reference. It is possible that the individual with autism's personal frame of reference for an appropriate physical environment may differ from accepted standards (Barneveld et al., 2014). Thus, they may perceive their environment as better than others perceive it to be. Alternatively, despite not having a learning disability (i.e. $IQ \geq 70$), a diagnosis of autism means that an individual's physical environment is likely to be more carefully maintained by services and family than is the case for the general population. Consequently, their environmental QoL may well be of a high standard, by virtue of being so carefully monitored maintained, causing them to view it more positively. An interesting avenue for future research may be to examine, in more detail, how

individuals with higher ability autism perceive their environment and what constitutes a positive living environment for them.

Despite these possible explanations for the difference in environmental scores between the two groups, it is also important to consider how methodological factors may have affected the scores reported by the autism sample. Being a postal questionnaire, it was not possible to ascertain how well individuals understood each question that they answered. Individuals were advised to seek support where required when completing the questionnaire but it was not possible to monitor this support. Therefore, replication of the analysis using data from face-to-face assessments with individuals with higher ability ASD would be needed in order to confirm the accuracy of the finding reported.

2.5.1.2 Informant versus self-report data

Analysis comparing self-report and informant perceived QoL found that, despite the lack of a significant difference between the informant and self-report data (psychological, physical and environmental domains), the scores were not significantly associated with one another either. This lack of association between the two data sources corroborates previous evidence of differences between informant and self-report data in this population (Sheldrick et al., 2012). Whilst these analyses alone cannot definitively confirm that both the informant and self-report data are equally useful measures of outcome, they do demonstrate that the substitution of self-report data with informant data where the former is not available is inappropriate. However, more in-depth analysis of the utility of the two data sources is required in order to determine how they can be used to complement each other and enrich our understand of the QoL of this sample.

This lack of association also highlights the need to consider what the informant questionnaire is actually measuring. It was always intended to assess the views of the parent / carer but it is possible that these scores are representing more than just informant perceived QoL. Slightly lower informant scores (albeit not significantly lower) may be tapping into a discrepancy between parents' / carers' aspirations for the person with autism and the reality of their life in adulthood. Additionally, they may reflect a negative bias due to parents' / carers' own anxieties about the future for the person with autism (Moss, 2011). These anxieties may be exacerbated by the fact that their own increasing age is likely to affect their ability to support the adult with autism. Overall, findings support the decisions in the current study to (i) use self-report data wherever possible and (ii) not substitute the lack of self-report data in the lower adult IQ group with informant data.

2.5.1.3 Childhood factors associated with QoL

There was a significant difference between individuals with and without language in childhood and their informant-rated psychological domain scores (those without language had poorer informant-rated psychological QoL; $p < .05$). This is unsurprising given that it has been widely reported that limitations in language development in childhood are associated with poorer outcomes in general in ASD (Howlin and Moss, 2012). Additionally, individuals with ASD (particularly those with without / a delay in language) often struggle to report on their own experiences (especially the more abstract concepts involved in mental state; Stewart, Barnard, Pearson, Hasan, & O'Brien, 2006) and so informant's reports of poor psychological well-being may be due, in part, to these reporting difficulties.

Despite the fact that this finding is unsurprising for the reasons described above, the impact of methodological limitations regarding language measurement must also be considered. Language scores were based on a three category rating system (no language, few words, fully verbal), which was then compressed into two categories (language or no language) to facilitate analyses due to limited sample size. This basic measure of language may not fully reflect the range and complexity of language abilities in the sample and thus may be affecting the findings reported. Future studies, involving more detailed language measurement in childhood would be necessary to confirm whether or not the finding reported is accurate.

There was a significant negative association between self-reported overall QoL (Q1) and childhood IQ (as childhood IQ scores increased, overall QoL scores declined; $p < .05$). Interestingly, this contrasts findings by van Heijst and Geurts (2014) who reported no association between QoL and childhood IQ but they conducted regression analyses with a very small sample ($n = 24$), increasing the risk of type II error. Furthermore, IQ scores in this sample were based on an adult reading test rather than a standardised IQ assessment in childhood. Consequently, these methodological limitations call into question the reliability of the null finding reported.

Regarding the current study's finding, it is possible that individuals' increased awareness of their autism and its limitations (Eaves and Ho, 2008) means that they are more likely to report a poorer QoL because they desire more than they are able to achieve. However, there are also three key methodological issues that must be considered in relation to this unexpected finding. Firstly, individuals with a lower IQ may not have understood the questions as well, impacting on their ability to answer them accurately. Being a postal questionnaire it was not possible to ascertain how

well participants understood the questions or to explain the questions to those who struggled to understand them. Whilst individuals were encouraged to seek support where necessary, there was no way of monitoring this level of support. Secondly, the measurement of IQ in childhood was variable (see Section 2.5.1.5 for details) and, whilst an inevitable consequence of a longitudinal follow-up of individuals who were initially recruited from a clinical setting, it must be considered in relation to the current findings. Scores were all based on standardised measures but a range of measures were used to ascertain PIQ, not FSIQ, at different ages and this may have affected the reliability of the scores and thus the finding reported above. Lastly, this analysis was only conducted with individuals who had an *adult* FSIQ ≥ 70 . Ideally, this data would have been obtained for the entire sample in order to truly understand the impact of childhood IQ on adult QoL amongst individuals with a childhood PIQ outside the intellectual disability range but current ability and language levels prevented this. It is evident that methodological factors may have contributed to this unexpected finding between childhood IQ and adult QoL but it is unclear to what extent. Therefore, replication of this analysis once the issues described above have been addressed will be necessary before any firm conclusions can be drawn, particularly given that the finding was only significant at the .05 level.

Interestingly, the current results actually more closely reflect the widely reported findings that childhood IQ *is* a factor associated with adult social outcome in higher ability autism (Moss and Howlin, 2012), although the direction of the association differs. Higher IQ was associated with poorer perceived QoL in the current study, but a better social outcome in previous research. Whilst the exact reason for this requires further investigation, this difference in the direction of

association highlights the fact that adult social outcome and QoL are two distinct measurements of outcome.

Thirdly, total ADI scores in childhood were significantly negatively associated with self-reported overall physical health (Q2; $p < .01$) and the RRBI domain was significantly negatively correlated with the self-report psychological, physical and environmental domains ($p < .05$). That is, higher childhood symptomatology scores were associated with lower self-reported QoL in adulthood. This supports van Heijst and Geurts (2014), who found an association between the two factors and is in line with findings regarding the predictive value of early autistic symptomatology on adult social outcome (Howlin and Moss, 2012). Despite being in line with previous results, this is the first time that a possible association between QoL and childhood RRBI scores, in particular, has been identified in a higher ability autism sample in mid-to-late adulthood. However, given the limited significance of this ($p < .05$) and the other two findings reported above, further exploration of the links between childhood factors and later QoL is required, once all of methodological limitations described above have been taken into account.

2.5.1.4 Adult factors associated with QoL

There was a strong, significant positive association between the self-report social QoL domain and adult social outcome ($r = 0.57$, $p < .01$), which is unsurprising given that the social QoL domain and adult social outcome measure tap into similar concepts about friendships and relationships. However, there are still conceptual differences between the two measures. The former examines individuals' contentment with their social life whereas the latter focuses on more objective outcomes (for example, whether they have any friends). The lack of significant

findings between adult social outcome and all other QoL domains suggests that the traditional measures of social outcome in autism do not address other more subjective experiences of the individual that *are* covered by QoL.

Adult IQ and age were significantly negatively correlated with self-reported social QoL and informant perceived physical QoL scores respectively ($p < .05$). The former is unsurprising given that individuals with a higher IQ but a diagnosis of autism are likely to report a poorer social life. Their higher ability levels mean that they are often acutely aware that their autism is impacting on their ability to socialise and make friends (Eaves and Ho, 2008), hence the poorer reported QoL scores. The latter finding is also expected given that older individuals, in mid-to-late adulthood, are likely to have poorer physical health, by virtue of their age.

2.5.1.5 Changes in IQ scores from childhood to adulthood

The variability in IQ scores, from childhood to adulthood, is notable (Table 2.3 and 2.4). The difference in IQ scores between the lower and higher adult ability sub-groups is noticeably greater in adulthood compared to childhood. Whilst examining IQ change over time was not a primary aim of the study, this discrepancy requires consideration. It is most likely to be due to measurement differences at the two time points. In childhood, individuals completed numerous tests and so an IQ hierarchy was devised to use the best measure at the optimum age (Appendix IV), ideally a Wechsler test at 5-7 years. Such variability in measurement is an inevitable consequence of a longitudinal research project that was conducted with individuals who were initially recruited from a clinical setting 40-50 years ago when standardised assessments were less well established.

Additionally, PIQ rather than FSIQ was used to measure childhood IQ because this was available for all participants. Whilst some *did* obtain FSIQ scores in childhood, language impairments prevented others from doing so. However, it is possible that these language impairments may have actually been masking more widespread impairments for those individuals that were not reflected in their non-verbal IQ scores.

In adulthood, a different approach was taken. Best IQ scores were obtained based on a different IQ hierarchy (Figure 2.2). Where abilities permitted, this was a Wechsler FSIQ but when all other attempts failed, a proxy IQ based on the VABS was used. Findings are mixed regarding the comparability of VABS and IQ scores based on standardised intelligence tests. Some reports suggest that the VABS underestimates ability (Perry, Flanagan, Dunn, & Freeman, 2009; Sparrow et al., 2005). In contrast, Perry and Factor (1989) found that VABS scores compared well with IQ scores in children and adolescents with autism. Using VABS proxy IQ scores is not ideal but this method was only employed when all other attempts to use standardised measures had failed.

In the current sample, VABS proxy IQ scores accounted for 50% of the lower adult IQ sub-group but none of the higher adult IQ sub-group. Therefore, this is likely to have contributed for the decline observed in the former, given that VABS scores may underestimate IQ. However, what is not clear is whether this change reflects a true decline in ability or is a product of measurement changes over time. An alternative approach would have been to remove all participants with adult VABS IQ scores from the sample but this would have reduced the lower adult IQ group by 50% and the entire sample by 21%, and thus the statistical power of the study.

All of these factors need to be considered when examining the role that IQ plays on QoL in adulthood but they do not negate the use of the scores. The key focus of this study was on (i) understanding a group of individuals who, at least as children, had similar ability levels based on non-verbal IQ tests and (ii) on determining whether child and adult IQ scores are associated with adult QoL.

2.5.2 Strengths and limitations

2.5.2.1 Limitations

There are limitations to the study that must be addressed. Firstly, the representativeness of the sample, and thus the extent to which the findings can be generalised to individuals with autism more broadly, is a potential area of weakness. Participants were all diagnosed at a specialist clinic by international experts in the early stages of our understanding of ASD. Consequently, many such individuals may have had more severe autistic symptomatology than is typical of individuals diagnosed with higher ability ASD today. Population-based samples are preferable but few research groups have achieved this and even amongst those that have, the representativeness of samples remains questionable. For example, Farley et al. (2009) used a population-based sample but it focussed on members of the Church of Jesus Christ of Latter-Day Saints community. Nonetheless, the current sample provides a unique insight into the QoL of higher ability adults who were diagnosed in the UK with autism over the past 25 – 50 years.

Secondly, the assessment methods used at all stages of the research must be considered. A range of IQ and language tests were used at T1 and no standardised autism diagnostic measure was available at initial diagnosis. Attempts were made to address these issues and to make the process more systematic by using an IQ

hierarchy (Appendix IV), a three category language rating system and conducting diagnostic confirmation at a later stage using the standardised ADI but concerns regarding variability and reliability of scores still exist. Thus, these issues must be considered when examining the findings reported.

Additionally, the WHOQOL-BREF has not been formally assessed for its use with the ASD population, particularly as a postal questionnaire. Ideally, a pilot study would have been conducted where a certain number of individuals in the higher ability sub-group were administered the questionnaire face-to-face to ascertain its validity as a measure for this population. However, given limitations in time and resources, a pilot study was not feasible. Furthermore, the measure has been used with a higher ability ASD population before (Kamio, Inada and Koyama, 2012), its brevity made it suitable for a postal questionnaire and its psychometric properties and prior use with other populations (Skevington and McCrate, 2012) facilitated comparisons between studies. The lack of appropriately standardised measures available for research with the ASD population is a common problem in studies of this kind (Brugha, Doos, Tempier, Einfeld & Howlin, in press).

Given the known difficulties with self-report in this population it seemed appropriate to supplement the self-report data with informant reports. The WHOQOL-BREF was designed as a self-report measure and so its psychometric properties, as an informant measure, are unclear. However, it was considered more appropriate to adapt the WHOQOL-BREF for informants, facilitating analyses comparing the two groups, than to use a different measure entirely. The results from the current study demonstrate that using both informant and self-report can enhance our understanding of QoL amongst higher ability ASD samples.

Finally, although there was sufficient power for the analyses on informant data, this was not the case for the self-report data. Furthermore, the multiple correlations conducted means that results significant at $p < .05$ must be treated with caution due to the risk of type I error. A more stringent p -value could have been applied but this would have resulted in the loss of many interesting findings that would benefit from further exploration and increased the risk of type II error. Instead, all findings $p < .05$ were considered significant but those that were significant between .01 and .05 were treated as provisional and in need of replication.

2.5.2.2 Strengths

Many of the study's strengths pertain to the uniqueness of the sample. Firstly, the group was homogenous with respect to childhood PIQ. All participants had a childhood PIQ outside the intellectual disability range, contrasting with many other adult follow-up studies that include a range of IQs (see Section 2.2.3). Secondly, most individuals were in mid-to-late adulthood and there has been very little examination of QoL in this population at this age. Thirdly, the sample size ($N = 52$) is large compared to previous QoL studies which reported on smaller cohorts (for example, van Heijst and Geurts, 2014). Lastly, the response rate from adult follow-up to the current QoL study was high (overall = 90%, positive = 88%), especially for a postal study. This can be attributed, in part, to the strong links that the wider research team had made with families over the entire course of the research.

The range of child and adult factors available for comparison with the QoL data allowed exploration of a number of different research questions. Furthermore, standardised assessments were used where possible and the WHOQOL-BREF

facilitated comparison with other populations and will aid comparisons with future QoL research.

The direct comparison of informant perceived versus self-report QoL scores has not previously been examined in a sample such as the present one, and these findings will be important for future research with higher ability adults with ASD. Given that self and informant reports were not associated with one another, researchers and clinicians need to be aware that informant-based data provides supplementary information only, rather than a replacement for the views of individuals with ASD themselves.

2.5.3 Clinical Implications and Future Research

This study has far reaching clinical implications regarding the higher ability ASD population. By considering the subjective QoL of the individual, it enhances our understanding of overall outcome and helps us to predict who is likely to have a better adult QoL. It also broadens the scope of examining outcome in this sample, which will, in turn, facilitate the development of interventions throughout the lifespan to improve adult social outcome and also the subjective QoL of an individual.

By encouraging a focus on correlates of QoL, this study has highlighted a gap in the research. It is important to continue to systematically search for possible correlates of QoL in this population in mid-to-late adulthood. Of particular interest, is the finding that childhood RRBI scores are associated with self-reported adult QoL scores. As the RRBI domain covers a diverse range of symptoms, from stereotyped interests to sensory difficulties to challenges around flexibility of thought and

actions, it would be interesting to examine this further to ascertain which components of childhood RRBI symptomatology are most associated with adult QoL.

The representativeness of samples in follow-up studies is often compromised and so prospective longitudinal studies will be best placed to further our understanding in this field. Lastly, the WHOQOL-BREF has not been validated for use with an ASD sample but given its clear psychometric properties with other populations (Skevington and McCrate, 2012) and increasing use with ASD populations, it would be important to systematically evaluate this measure for use with this population.

2.5.4 Conclusion

This is the first known study to systematically assess adult and child correlates of QoL in a higher ability ASD population in mid-to-late adulthood who have been followed up since childhood. The lack of association between QoL and adult social outcome highlights the importance of research in this area, as they are clearly two distinct, but equally useful, measures of outcome.

2.6 References

- American Psychiatric Association. (1994). *Diagnostic and Statistical Manual of Mental Disorders (DSM-IV)* (Vol. 4th). Washington, DC: American Psychiatric Association.
- American Psychiatric Association. (2000). *American Psychiatric Association: Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision* (Vol. 4th). Washington, DC: American Psychiatric Association.
- American Psychiatric Association. (2013a). *Diagnostic and Statistical Manual of Mental Disorders* (5th ed.). Washington DC: Author.
- American Psychiatric Association. (2013b). Intellectual Disability Fact Sheet.
- Barneveld, P. S., Swaab, H., Fagel, S., van Engeland, H., & de Sonneville, L. M. (2014). Quality of life: a case-controlled long-term follow-up study, comparing young high-functioning adults with autism spectrum disorders with adults with other psychiatric disorders diagnosed in childhood. *Comprehensive Psychiatry*, *55*(2), 302-310. doi: 10.1016/j.comppsy.2013.08.001
- Billstedt, E., Gillberg, I. C., & Gillberg, C. (2005). Autism after adolescence: population-based 13- to 22-year follow-up study of 120 individuals with autism diagnosed in childhood. *Journal of Autism and Developmental Disorders*, *35*(3), 351-360.
- Billstedt, E., Gillberg, I. C., & Gillberg, C. (2011). Aspects of quality of life in adults diagnosed with autism in childhood: a population-based study. *Autism*, *15*(1), 7-20. doi: 10.1177/1362361309346066

- Bolton, P., Macdonald, H., Pickles, A., Rios, P., Goode, S., Crowson, M., . . . Rutter, M. (1994). A case-control family history study of autism. *Journal of Child Psychology and Psychiatry*, 35(5), 877-900.
- Brugha, T. S., Doos, L., Tempier, A., Enfield, E., & Howlin, P. (in press). Outcome measures in intervention trials for adults with autism spectrum disorders: a systematic review of assessments of core autism features and associated emotional and behavioural problems. *International Journal of Methods in Psychiatric Research*.
- Buescher, A. V., Cidav, Z., Knapp, M., & Mandell, D. S. (2014). Costs of autism spectrum disorders in the United Kingdom and the United States. *JAMA Pediatrics*, 168(8), 721-728. doi: 10.1001/jamapediatrics.2014.210
- CDC. (2014). Prevalence of autism spectrum disorder among children aged 8 years - autism and developmental disabilities monitoring network, 11 sites, United States, 2010. *MMWR Surveillance Summaries*, 63(2), 1-21.
- Chevallier, C., Kohls, G., Troiani, V., Brodtkin, E. S., & Schultz, R. T. (2012). The social motivation theory of autism. *Trends in Cognitive Science*, 16(4), 231-239. doi: 10.1016/j.tics.2012.02.007
- Dunn, L. M., Dunn, L. M., Whetton, C., & Burley, J. (1997). *The British Picture Vocabulary Scale* (Vol. 2nd). Windsor: NFER-NELSON.
- Eaves, L. C., & Ho, H. H. (2008). Young adult outcome of autism spectrum disorders. *Journal of Autism and Developmental Disorders*, 38(4), 739-747.
- Engstrom, I., Ekstrom, L., & Emilsson, B. (2003). Psychosocial functioning in a group of Swedish adults with Asperger syndrome or high-functioning autism. *Autism*, 7(1), 99-110.

- Farley, M. A., McMahon, W. M., Fombonne, E., Jenson, W. R., Miller, J., Gardner, M., . . . Coon, H. (2009). Twenty-year outcome for individuals with autism and average or near-average cognitive abilities. *Autism Research, 2*(2), 109-118.
- Fombonne, E., Bolton, P., Prior, J., Jordan, H., & Rutter, M. (1997). A family study of autism: cognitive patterns and levels in parents and siblings. *Journal of Child Psychology and Psychiatry, 38*(6), 667-683.
- Gerber, F., Baud, M. A., Giroud, M., & Galli, C. G. (2008). Quality of life of adults with pervasive developmental disorders and intellectual disabilities. *Journal of Autism and Developmental Disorders, 38*(9), 1654-1665.
- Group, T. W. (1995). World Health Organization quality of life assessment (WHOQOL): Position paper from the World Health Organization. *Social Science & Medicine, 41*(10), 1403-1409.
- Henninger, N. A., & Taylor, J. L. (2013). Outcomes in adults with autism spectrum disorders: a historical perspective. *Autism, 17*(1), 103-116. doi: 10.1177/1362361312441266
- Howlin, P. (2003). Outcome in high-functioning adults with autism with and without early language delays: implications for the differentiation between autism and Asperger syndrome. *Journal of Autism and Developmental Disorders, 33*(1), 3-13.
- Howlin, P. (2004). Outcome in autism *Autism and Asperger Syndrome: Preparing for Adulthood* (Vol. 2nd, pp. 19-45). London and New York: Routledge.
- Howlin, P. (2005). Outcomes in Autism Spectrum Disorders. In F. R. Volkmar, R. Paul, A. Klin & D. Cohen (Eds.), *Handbook of Autism and Pervasive Developmental Disorders* (Vol. third, pp. 201-220): John Wiley & Sons, Inc.

- Howlin, P. (2007). The outcome in adult life for people with ASD. In F. R. Volkmar (Ed.), *Autism and Pervasive Developmental Disorders* (Vol. 2nd, pp. 269-306): Cambridge University Press.
- Howlin, P., Goode, S., Hutton, J., & Rutter, M. (2004). Adult outcome for children with autism. *Journal of Child Psychology and Psychiatry*, *45*(2), 212-229.
- Howlin, P., Mawhood, L., & Rutter, M. (2000). Autism and developmental receptive language disorder--a follow-up comparison in early adult life. II: Social, behavioural, and psychiatric outcomes. *Journal of Child Psychology and Psychiatry*, *41*(5), 561-578.
- Howlin, P., & Moss, P. (2012). Adults with autism spectrum disorders. *Canadian Journal of Psychiatry*, *57*(5), 275-283.
- Howlin, P., Moss, P., Savage, S., & Rutter, M. (2013). Social outcomes in mid- to later adulthood among individuals diagnosed with autism and average nonverbal IQ as children. *Journal of American Academy of Child and Adolescent Psychiatry*, *52*(6), 572-581 e571. doi: 10.1016/j.jaac.2013.02.017
- Howlin, P., Savage, S., Moss, P., Tempier, A., & Rutter, M. (2014). Cognitive and language skills in adults with autism: a 40-year follow-up. *Journal of Child Psychology and Psychiatry*, *55*(1), 49-58. doi: 10.1111/jcpp.12115
- Jarbrink, K., Fombonne, E., & Knapp, M. R. (2003). Measuring the parental, service and cost impacts of children with autistic spectrum disorder: a pilot study. *Journal of Autism and Developmental Disorders*, *33*(4), 395-402.
- Jarbrink, K., & Knapp, M. (2001). The economic impact of autism in Britain. *Autism*, *5*(1), 7-22.

- Jarbrink, K., McCrone, P., Fombonne, E., Zanden, H., & Knapp, M. (2007). Cost-impact of young adults with high-functioning autistic spectrum disorder. *Research in Developmental Disabilities, 28*(1), 94-104.
- Kamio, Y., Inada, N., & Koyama, T. (2012). A Nationwide Survey on Quality of Life and Associated Factors of Adults With High-Functioning Autism Spectrum Disorders. *Autism*.
- Kamp-Becker, I., Schroder, J., Muehlan, H., Remschmidt, H., Becker, K., & Bachmann, C. J. (2011). Health-related quality of life in children and adolescents with autism spectrum disorder. *Z Kinder Jugendpsychiatr Psychother, 39*(2), 123-131. doi: 10.1024/1422-4917/a000098
- Kanner, L. (1943). Autistic Disturbances of affective contact. *Nervous Child, 2*, 217-250.
- Le Couteur, A., Bailey, A., Goode, S., Pickles, A., Robertson, S., Gottesman, I., & Rutter, M. (1996). A broader phenotype of autism: the clinical spectrum in twins. *Journal of Child Psychology and Psychiatry, 37*(7), 785-801.
- Le Couteur, A., Rutter, M., Lord, C., Rios, P., Robertson, S., Holdgrafer, M., & McLennan, J. (1989). Autism diagnostic interview: a standardized investigator-based instrument. *Journal of Autism and Developmental Disorders, 19*(3), 363-387.
- Lin, L. Y. (2014). Quality of life of Taiwanese adults with autism spectrum disorder. *PLoS One, 9*(10), e109567. doi: 10.1371/journal.pone.0109567
- Lotter, V. (1974). Social adjustment and placement of autistic children in Middlesex: a follow-up study. *Journal of Autism and Childhood Schizophrenia, 4*(1), 11-32.

- Macintosh, K. E., & Dissanayake, C. (2004). Annotation: The similarities and differences between autistic disorder and Asperger's disorder: a review of the empirical evidence. *Journal of Child Psychology and Psychiatry*, 45(3), 421-434.
- Moss, P. (2011). *Outcome in individuals with autism and their siblings: An investigation of the factors associated with functioning in adulthood*. (PhD), Kings College London, London.
- National Audit Office. (2009). Supporting people with autism through adulthood (pp. 1-59). London: The Stationary Office.
- Pallant, J. (2007). *SPSS Survival Manual* (Vol. 3rd). Maidenhead, Berkshire: Open University Press.
- Perry, A., & Factor, D. C. (1989). Psychometric validity and clinical usefulness of the Vineland Adaptive Behavior Scales and the AAMD Adaptive Behavior Scale for an autistic sample. *Journal of Autism and Developmental Disorders*, 19(1), 41-55.
- Perry, A., Flanagan, H. E., Dunn, G. J., & Freeman, N. L. (2009). Brief report: the Vineland Adaptive Behavior Scales in young children with autism spectrum disorders at different cognitive levels. *Journal of Autism and Developmental Disorders*, 39(7), 1066-1078.
- Persson, B. (2000). Brief report: A longitudinal study of quality of life and independence among adult men with autism. *Journal of Autism and Developmental Disorders*, 30(1), 61-66.
- Pickles, A., Starr, E., Kazak, S., Bolton, P., Papanikolaou, K., Bailey, A., . . . Rutter, M. (2000). Variable expression of the autism broader phenotype: findings

from extended pedigrees. *Journal of Child Psychology and Psychiatry*, 41(4), 491-502.

Pinto, D., Pagnamenta, A. T., Klei, L., Anney, R., Merico, D., Regan, R., . . .

Betancur, C. (2010). Functional impact of global rare copy number variation in autism spectrum disorders. *Nature*, 466(7304), 368-372.

Raven, J. C. (1986). *Coloured Progressive Matrices*. Essex: E T Heron (Print) Ltd.

Renty, J. O., & Roeyers, H. (2006). Quality of life in high-functioning adults with autism spectrum disorder: The predictive value of disability and support characteristics. *Autism*, 10(5), 511-524.

Rutter, M., Greenfield, D., & Lockyer, L. (1967). A five to fifteen year follow-up study of infantile psychosis. II. Social and behavioural outcome. *British Journal of Psychiatry*, 113(504), 1183-1199.

Rutter, M., Le Couteur, A., & Lord, C. (2003). *The Autism Diagnostic Interview-Revised*. Los Angeles: Western Psychological Services.

Saldaña, D., Alvarez, R. M., Lobaton, S., Lopez, A. M., Moreno, M., & Rojano, M. (2009). Objective and subjective quality of life in adults with autism spectrum disorders in southern Spain. *Autism*, 13(3), 303-316. doi: 10.1177/1362361309103792

Sheldrick, R. C., Neger, E. N., Shipman, D., & Perrin, E. C. (2012). Quality of life of adolescents with autism spectrum disorders: concordance among adolescents' self-reports, parents' reports, and parents' proxy reports. *Quality of Life Research*, 21(1), 53-57. doi: 10.1007/s11136-011-9916-5

Skevington, S. M., & McCrate, F. M. (2012). Expecting a good quality of life in health: assessing people with diverse diseases and conditions using the

WHOQOL-BREF. *Health Expectations*, 15(1), 49-62. doi: 10.1111/j.1369-7625.2010.00650.x

Sparrow, S. S., Cicchetti, D. V., & Balla, D. A. (2005). *Vineland-II Vineland Adaptive Behaviour Scales*. Minnesota: AGS Publishing.

Sterling, L., Dawson, G., Estes, A., & Greenson, J. (2008). Characteristics associated with presence of depressive symptoms in adults with autism spectrum disorder. *Journal of Autism and Developmental Disorders*, 38(6), 1011-1018.

Stewart, M. E., Barnard, L., Pearson, J., Hasan, R., & O'Brien, G. (2006). Presentation of depression in autism and Asperger syndrome: a review. *Autism*, 10(1), 103-116.

Szatmari, P. (2000). The classification of autism, Asperger's syndrome, and pervasive developmental disorder. *Canadian Journal of Psychiatry*, 45(8), 731-738.

van Heijst, B. F., & Geurts, H. M. (2015). Quality of life in autism across the lifespan: A meta-analysis. *Autism*, 19(2), 158-167. doi: 10.1177/1362361313517053

Wechsler, D. (1999). *The Wechsler Abbreviated Scale for Intelligence*. San Antonio, TX: The Psychological Corporation.

Wechsler, D. (2005). *Wechsler Individual Attainment Test 2nd Edition (WIAT II)*. London: The Psychological Corporation.

World Health Organisation. (1998). Development of the World Health Organization WHOQOL-BREF quality of life assessment. The WHOQOL Group. *Psychological Medicine*, 28(3), 551-558.

Wechsler, D., Wycherley, R. J., & Benjamin, L. (1997). *Wechsler Adult Intelligence Scale: Administration Manual* (Vol. 3rd). UK: The Psychological Corporation.

Witwer, A. N., & Lecavalier, L. (2008). Examining the validity of autism spectrum disorder subtypes. *Journal of Autism and Developmental Disorders*, 38(9), 1611-1624.

Part 3: Critical Appraisal

3.1 Introduction

This appraisal considers some of the issues that arose during the process of the research study. The first section will consider how my previous experiences of working with the sample influenced the aims of the current study and decisions that were made about methodology and analysis. The second section will address the wider issue of assessment with an Autism Spectrum Disorder (ASD) sample and how this relates to the current study.

3.2 How previous experiences influenced this study

3.2.1 Inspiration for the research

The current study aims were inspired by the data collection process for my PhD thesis (Moss, 2011), which formed the adult follow-up phase of the longitudinal research study. Whilst visiting people to conduct a range of assessments, I was struck by two discrepancies. Firstly, based on the informant rated adult social outcome measure used, a lot of people were scoring very poorly but when meeting with the individual with autism, they seemed fairly content with their life and their situation. In many cases they indicated no desire for a wider range of relationships, a more complex job and/or to live independently if this was not currently the case. There was one individual in particular who caught my attention. He was living with his mother and step-father, working in a packing factory and had never had a romantic relationship. When asked about friendships, he proudly stated that his step-father was his best friend. It struck me that this gentleman would be one of the highest scoring participants on the social outcome measure (a higher score indicates a poorer social outcome) yet he appeared to be very content with his life and current situation. It was cases such as these that led to me to question whether adult social outcome was

sufficient on its own to determine how people with higher ability ASD were coping in adulthood. Whilst its use and importance are indisputable, I found myself wondering whether this alone gave the full picture of how people were functioning in adult life and, critically, whether they were satisfied with their current situation.

The second discrepancy that I observed was the apparent difference, at least anecdotally, between parents / carers views of the person with autism's life and the views of the individual themselves. There was no significant difference in adult social outcome scores between informant and self-report data when statistical analysis was conducted (Moss, 2011). However, this is unsurprising given the objective nature of the social outcome measure, where the status of the individual is clear (for example, where they are living). Consequently, I wondered whether the difference that I was observing could not be identified through measuring objective outcomes but was instead a discrepancy in subjectively measurable outcomes, such as Quality of Life (QoL), between informants and the individual with autism. Consequently, I sought to investigate how informant perceived versus self-reports of QoL differed within this sample.

3.2.2 Advantages of knowing the sample well

Working with this sample during the adult follow-up study two years prior to the current QoL study meant that my experiences were able to inform many decisions regarding the methodology. I was aware of the complexities (as well as advantages) that came with working with this unique group of individuals who had been followed up for 40-50 years, some of whom were the first individuals in the country to be diagnosed with autism.

The sample was well known to the wider research team, specifically my external supervisor (PH), and I knew many of the participants and their families personally from the adult follow-up data collection phase. Consequently, families had built strong relationships with various members of the team, particularly those who had been working with the families in a research (and at times clinical) capacity for many years. This was a great strength of the study and it enabled me to anticipate relatively high response rates. Indeed, 90% of the 59 families contacted for the current QoL study responded, with a positive response rate from adult follow-up to the current study of 88% and 58% from childhood to the current study. These extremely high response rates to a postal questionnaire were definitely achieved, in part, due to the strong relationships that had been built with families over the years.

Having worked with the sample recently during the adult follow-up study, I was also in the fortunate position of being able to make an informed judgement regarding what assessments people were likely to be able and / or willing to complete. The adult follow-up study had involved lengthy face to face interviews, in depth cognitive, language and mental health assessments (where ability levels permitted). Whilst individuals were happy to participate again, a number of them reported that they found the volume of assessments included in the adult follow-up study quite challenging and tiring. Consequently, I was keen to ensure that they did not feel overwhelmed with another large set of assessments so close to the previous study. Doing so would have been unethical and inevitably limited the number of people willing to take part. I was therefore careful to ensure that the measure that I selected was not too lengthy or complicated. Additionally, given that participants were located all over the UK (and in some cases abroad), it was not possible to visit

people in their homes again and so participation had to be short and simple enough to be completed by post.

Knowing the sample so well also enabled me to take decisions, based on clinical judgement, regarding which individuals would be able to complete the self-report QoL questionnaire. There were two criteria that were initially used to divide the group into those who should and should not be sent the self-report questionnaire before clinical judgement was applied. At the recent adult follow-up study, individuals had to have obtained a minimum reading ability level of 5.1 years based on the single word reading subtest of the Wechsler Individual Achievement Test (WIAT-II; Wechsler, 2005) *and* an full-scale IQ (FSIQ) ≥ 70 . Regarding the former, despite this low cut-off, the average reading ability of the sample was actually much higher (15.9 years, range = 9.08 – 19.0 years). Regarding the latter, adult FSIQ scores were based on Wechsler tests which require a certain level of verbal comprehension skills in order to complete the verbal and non-verbal tests to a sufficient level and obtain a score outside of the intellectual disability range.

Once the group had been split according to these criteria, my second supervisor and I each examined the groups in detail. Using my recent assessment experiences with the participants (from the adult follow-up study) and my supervisor's longstanding experience of conducting assessments with these individuals, we were able to confirm that no-one in the 'higher adult ability' subgroup was being sent the questionnaire who we thought would struggle to complete it, even with support. Our checks confirmed that everyone who had been allocated to this group would be able, based on our judgement, to complete the test. Furthermore, the information sheet clearly advised individuals in this group to seek support when completing the questionnaire if they required it and to indicate on the questionnaire

whether or not they had done so. The majority of the group did seek support but none of them reported any difficulties completing the questionnaire. Using a combination of inclusion criteria, the guidelines in the information sheet and the research team's knowledge of the sample, ensured that only individuals who would be able to complete the self-report questionnaire were asked to do so. Consequently, a balance was struck between trying to get as much self-report data as possible and being mindful of the current ability level of the sample.

3.2.3 Challenges of working with a longitudinal sample

The current study aim, of examining the factors associated with QoL amongst higher ability adults with autism, was a focussed one. However, because the sample was part of a 40-50 year follow-up research process the current study was quite complex. This was a particular challenge in the method section of the empirical paper. It was important to be concise but also to clearly explain the complex background to and different phases of the study. This was vital to ensure that the uniqueness of the sample and the research team's in-depth knowledge of the participants and their ability level were conveyed. In order to achieve this, a separate section was added to the methodology (Section 2.3.1). This section situated the current study within the wider research project and thus demonstrated that, whilst being a separate phase of the follow-up process, it could not be considered without awareness of the longitudinal nature of the project.

With the longitudinal nature of the study, came the vast amount of data that I had at my disposal to work with. This meant that the variables available with which I could run comparative analyses with the QoL scores were numerous. It was important to keep the research questions focussed and to achieve a balance between

being inclusive without aimlessly searching for significant correlations with multiple variables. Addressing too many topics would have further limited the statistical power of the analyses and thus the risk of type I error. Additionally, without a clear rationale for the measures included it would have been confusing as a piece of research. When conducting longitudinal follow-up research such as this, it is important to select variables that are (i) guided by previous research and (ii) based on the best quality measures available.

A potential criticism of this study was that the self-report questionnaires were sent by post and no formal method of assessing the validity of the responses was applied. However, there are a number of reasons why these decisions were taken. Firstly, clinical judgement was used to check that no-one was being sent the self-report questionnaire whom myself or my second supervisor (PH) thought would struggle to complete it. During the recent adult follow-up study, all participants were administered a number of self-report questionnaires during the face-to-face assessment session. Consequently, I was able to draw on these experiences to determine whether the higher adult ability sub-group would be able to complete the questionnaire without a researcher present. Secondly, it was not possible to visit everyone again to support them to complete the questionnaire. Ideally, assessors blind to the study hypotheses would have sat with participants to review their comprehension and responses in vivo. However, not only were time and resources too limited for this but participants reported finding the previous assessment phase tiring. Therefore, care was taken to ensure that minimal effort was required from them for the current study, hence a postal questionnaire. Lastly, no participant indicated in the space provided at the end of the questionnaire that they had experienced any difficulties completing the measure. Whilst ideally all measures

with this population would be completed face-to-face or methods would be taken to check the validity of the responses, a number of steps were taken to overcome the fact that this could not be done on this occasion.

3.2.4 What I would have done differently

With endless amounts of time, finances and participants, it is possible to be ambitious with the research process but this is not always appropriate. Ideally, all assessments would have been standardised for use with the ASD population and would have been conducted face-to-face. This would have allowed me to be confident that individuals, particularly the participants with autism, fully understood the questions being asked. Furthermore, meeting face-to-face would have allowed me to include more assessments to enhance our understanding of QoL in this sample. However, the reality was that this sample had already taken part in three large studies over the years, requiring numerous hours of assessment. Indeed, many had reported finding the recent adult follow-up study quite strenuous. Furthermore, they lived all over the UK and abroad and parents and/or carers were increasing in age and for some their health was also beginning to fail. This inevitably limited the amount of assessments that people could / were willing to participate in.

In hindsight, a pilot study involving a few face-to-face assessments with individuals in the higher adult ability sub-group to examine the validity of the self-report data would have been useful. However, time and financial resources were limited and so I concluded that the research team's personal experiences of conducting assessments with the individuals in the sample were sufficient. This oversight may have been due to my longstanding involvement with the wider research project which may have, at times, hindered by ability to step back from it

and make balanced judgements about what methodological approaches would guarantee the most reliable and valid data. The research process is a constant balancing act between finances, time, availability of suitable measures and the needs of the participant versus those of the researcher. I hope that I was sensitive to all of these factors and that this allowed me to arrive at a study that provided a useful insight into the questions being asked, whilst remembering that the needs of the participant are paramount.

3.3 Assessment of people with ASD

The issue of using suitable measures is a challenge faced not only in this study but by many researchers working with ASD populations (Brugha, in press), particularly when examining more abstract topics such as QoL. Few measures are designed specifically for this population and standardised measures that are used with the general population are not always suitable (Brugha, in press). Instead, measures have to be selected that have been developed for the general population but have been used with people with ASD before, albeit with a few adjustments where necessary.

The issue of selecting appropriate measures for use with the ASD population also pertains to deciding whether to use informant or self-report data, something that had to be considered carefully with the current sample. Measurement of QoL is very subjective and service user involvement is essential to understand QoL and meet the needs of the individual with ASD. This is particularly relevant in the current NHS climate where Patient Reported Outcome Measures (PROMs) are being routinely used in services to measure outcomes and Skevington and McCrate (2012) are recommending that the WHOQOL-BREF is also considered for this purpose. Given that individuals with ASD with an IQ ≥ 70 do not meet criteria for learning disability

services and so are often expected to access generic mental health services, for example, it is important to establish whether the WHOQOL-BREF is appropriate for use as a self-report measure of QoL with this population.

It is well documented that individuals with ASD *do* struggle with self-report measures, particularly when addressing more abstract concepts such as QoL (see Section 2.2.4.3 of the empirical paper for details) and so the use of self-report data has to be considered carefully. However, recent reports which indicate that adolescents with ASD are able to validly and reliably report on their own QoL (Shipman, Sheldrick, & Perrin, 2011). Therefore, focussing entirely on informant data in this sample would have been inappropriate.

In the current study, having an IQ outside the intellectual disability range in childhood meant that all participants were labelled as having higher ability autism for research purposes. However, this did not necessarily mean that as adults they were all able to independently complete a postal questionnaire about their QoL. In hindsight, conducting a pilot study with a number of individuals and administering the WHOQOL-BREF face-to-face to assess the validity of participant's responses would have lead greater confidence to the validity of the self-report dataset as a whole. However, given limited time and resources this was not possible. To overcome this, when deciding who would be asked to complete the self-report WHOQOL-BREF the inclusion criteria were combined with my clinical judgement and that of my second supervisor (PH), who both knew the sample well, (see Section 3.2.2 for details). This resulted in a much smaller sample for the self-report data was much smaller ($n = 22$) than the informant data ($n = 50$), limiting the statistical power of the results obtained from the former. Given this, and the concerns described above

about the validity of the self-reported responses, the self-report data was not used in isolation but was considered alongside the larger informant dataset.

Overall, the decisions that were taken regarding the self-report data (administration and analysis) were determined by three factors; ensuring that the voice of the individual was accurately heard and reported, ethical issues around not asking any individuals to complete the self-report questionnaire who might struggle with it and sample size. My decision to use both forms of data was based on (i) statistical analysis which demonstrated that the informant and self-report data were not similar enough to warrant any substitutions in data, (ii) recent findings regarding the reliability of self-report QoL in younger ASD populations, (iii) the need to hear the voice of the individual with ASD and (iv) the way in which including informant responses increased statistical power and enhanced our understanding of the perceived QoL of the individual with ASD.

3.4 Data analysis

Due to the large number of analyses being conducted, particularly once informant *and* self-report data were included, the issue of declining statistical power and thus the risk of type I errors became a consideration. It was therefore important to decide whether to include findings that were significant at $p < .05$, albeit with caution attached to any conclusions drawn, or to discount them entirely. The decision was taken to apply the former approach as applying more stringent p -value based on a bonferroni correction would have meant the loss of many interesting findings from the study. Instead, it was considered more appropriate to include such findings but acknowledge that the limited statistical power means that the conclusions drawn

from findings with p values between .01 and .05 are extremely tentative at present and that further investigation is required.

3.5 Conclusion

The overarching theme that has struck me throughout this process has been that of the fine balance that has to be achieved at every stage of the research process. This applied during the literature review process; between the use of strict inclusion criteria versus a quality assurance measure, at the assessment stage; when deciding how to measure QoL and at the data analysis stage; when selecting how many variables to examine, which data source to use (informant and/or self-report) and whether to apply more stringent criteria regarding p -values. Overall, these decisions come down to an even wider issue of the balance that needs to be achieved between the needs of the participant versus those of the researcher. I believe that any keen researcher will have the ambition and drive to collect as much data as possible, with the maximum possible sample size and to conduct as many analyses as is feasible with that data. However, this always has to be weighed up against various methodological challenges. First and foremost are the needs of the participant and ensuring that the process is beneficial to them both as participants in the study and as part of a wider higher ability ASD population.

3.6 References

- Brugha, T. S., Doos, L., Tempier, A., Enfield, E., & Howlin, P. (in press). Outcome measures in intervention trials for adults with autism spectrum disorders: a systematic review of assessments of core autism features and associated emotional and behavioural problems. *International Journal of Methods in Psychiatric Research*.
- Moss, P. (2011). *Outcome in individuals with autism and their siblings: An investigation of the factors associated with functioning in adulthood*. (PhD), Kings College London, London.
- Shipman, D. L., Sheldrick, R. C., & Perrin, E. C. (2011). Quality of Life in Adolescents With Autism Spectrum Disorders: Reliability and Validity of Self-Reports. *Journal of Developmental & Behavioral Pediatrics*, 32(2), 85-89. doi: 10.1097/DBP.0b013e318203e558
- Skevington, S. M., & McCrate, F. M. (2012). Expecting a good quality of life in health: assessing people with diverse diseases and conditions using the WHOQOL-BREF. *Health Expectations*, 15(1), 49-62. doi: 10.1111/j.1369-7625.2010.00650.x
- Wechsler, D. (2005). *Wechsler Individual Attainment Test 2nd Edition (WIAT II)*. London: The Psychological Corporation.

Appendix I

Newsletter

January 2013

Impact of Autism in Adult Life Study

Department of Psychology, Institute of Psychiatry & Department of Clinical, Educational and Health Psychology, University College London,

Contact:

Tel:

/

Email:

Dear Families,

Firstly we would like to wish you all a very



We last contacted you in February 2012, to tell you that the main study was complete and to summarise the initial findings from the adult follow-up study in which so many of you very kindly took part. We hope that you found the results interesting.

What happens with the results now?

We are now in the process of presenting these findings and publishing them in the relevant journals. We hope that the results of the study will provide adult services with the information that they need to improve the support that they can offer to older individuals with autism and their families. This research would not have been possible without your input so a huge

THANK
YOU!

to everyone who helped to make the study a success!

What are we doing next?

We are now starting a new study, looking into the quality of life of adults with autism. During our visits, many of you commented on this issue. Although some people were considered to have a good quality of life, there were many whose quality of life was felt to be poor, and could be greatly improved. We believe that this is an extremely important but poorly understood area within the field of autism research and it is clearly a crucial issue for many adults with autism and their families.

How will we be assessing quality of life?

We appreciate all of the time you have already given up in helping with our research and so all that this next stage will involve will be the completion of a short postal questionnaire on the quality of life of the adult with autism. We will be contacting you in the next few weeks with further details and hope that you will not mind us contacting you again. If, however, you do not want to be contacted any further for this research, please let us know as soon as possible using the contact details above. If you have any other questions, please do not hesitate to contact us.

In the meantime, we hope that all will go well for you in the year ahead and we look forward to being in touch with you soon.

With best wishes,

and

Appendix II

Information sheets and consent forms

Information sheet 1 – for individuals with autism

Information sheet 2 – for parents / carers of individuals with autism and an IQ \geq 70

Information sheet 3 – for parents / carers of individuals with autism and an IQ $<$ 70

Consent form 1 – for individuals with autism

Consent form 2 – for parents / carers of individuals with autism

Information Sheet for Individuals with Autism in Research Studies (1)

Title of Project: **Quality of life in higher ability adults with autism**

This study has been approved by the UCL Research Ethics Committee

Project ID Number: 4111/001

Contact name: [REDACTED] / [REDACTED]

Work address: [REDACTED]
[REDACTED]

Contact details: [REDACTED]
[REDACTED]

We would like to invite individuals with autism to participate in this research project.

You and your family recently kindly took part in a follow-up study of individuals with autism in adulthood for Professors [REDACTED] and [REDACTED]. You may remember being visited by either [REDACTED] or [REDACTED] who spoke with you about your experiences of autism in adulthood and carried out some tasks with you. We would like to invite you to take part in a new stage of this study about the quality of life for adults with autism. Before you decide to take part you need to understand why the research is being done, and what it will involve for you and your family. Please take time to read the following information carefully and ask others to help you if there is anything that you do not understand. Please contact [REDACTED] if you have any questions or worries about the study or any difficulties completing the questionnaire.

Aims of the research and possible benefits.

We hope that this research will help us to understand people's experiences of autism in adulthood, particularly their quality of life. The results of the study will not benefit you directly but knowing about the quality of life of adults with autism will help us to understand the needs of this group of individuals. It will also help us to identify younger adults who might be at risk of difficulties in adulthood so that we can support them. We also believe that the findings will help us to improve services for adults with autism.

Who are we contacting for this study?

We are contacting all families of individuals with autism who took part in the most recent follow-up study and agreed to be contacted again for future research.

What will happen if you agree to take part?

If you agree to take part, we will ask you to complete the enclosed questionnaire about your quality of life. The questionnaire should take no more than 10-15 minutes to complete but please ask a family member / carer / friend to help you if you need support. We would be

grateful if you could then return this questionnaire, and the completed consent form enclosed, to us in the stamped addressed envelope provided. We will also be contacting your parents / carers to ask them to complete a questionnaire about their views on your quality of life. The information that you and your parents / carers provide will be linked to the information that you provided during the previous study so that we can learn more about the factors affecting the quality of life of adults with autism.

Please feel free to call me about this if there is anything that you would like to talk about or anything that you are unsure about on [REDACTED] / [REDACTED] (or via email [REDACTED]).

Are there any risks?

There are no expected risks associated with this study. However, if you do experience any difficulties after completing the questionnaire, or any other issues arise, please contact us via email ([REDACTED] / [REDACTED]) or phone ([REDACTED] / [REDACTED]).

Will my taking part in the project be kept confidential?

All information collected from the questionnaires will be confidential and kept anonymous and will only be available to those working on the project. Nothing that you tell us will be repeated to any other members of your family who may be taking part. None of the information in the final report will contain any personal information about you. However, in the unlikely event that you report that you or someone else is / has been at risk of harm, the researcher (Dr [REDACTED]) will speak to Dr [REDACTED] and / or Professor [REDACTED] who will then take advice on how to deal with this information and how to help you.

What will happen after I participate?

As with the previous study, we will be sending out a regular newsletter, to keep you updated on the study. The final newsletter will summarise the results of the study.

What will happen if I no longer want my data to be used for the study?

If you decide that you no longer want to take part, you can stop at any time. If this is your decision, any information you have provided will still be very useful to us but it can also be withdrawn and destroyed if you wish, and without you giving any reason. However, it is important that you tell us about this decision before the results are included in the final report (April 2014). If you decide not to take part in the study, or you want to stop at any time, this will not affect any help or treatment that you may be currently receiving.

Who do I contact if there is a problem?

If you are unhappy about any part of the study, you should speak to the researcher (Dr [REDACTED]) directly ([REDACTED] or tel: [REDACTED]) or contact Dr William Mandy on ([REDACTED] or tel: [REDACTED]) and we will do our best to answer your questions. If you are still unhappy, you can get more advice from the University College London complaints procedure and we will let you know how to go about doing this. If something does go wrong or you are harmed during the research and this is due to something that anyone in the research team had done then you may have grounds for a legal action against University College London, but you may have to pay for this. The normal University College London complaints procedure will still be available to you.

Information Sheet for Parents / Carers of Individuals with Autism in Research Studies (2)

Title of Project: **Quality of life in higher ability adults with autism**

This study has been approved by the UCL Research Ethics Committee

Project ID Number: 4111/001

Name: [REDACTED] / [REDACTED]

Work Address: [REDACTED]
[REDACTED]

Contact Details: [REDACTED] / [REDACTED]
[REDACTED]

We would like to invite parents / carers of adults with autism to participate in this research project.

You and your son / daughter / the person that you care for recently kindly participated in a follow-up study of individuals with autism in adulthood. We would like to invite you to take part in this new stage of the study investigating the quality of life for adults with autism. Before you decide whether or not to take part, you need to understand why the research is being done, and what it will involve for you and your family. Please take time to read the following information carefully and talk to others if you wish. Please contact Dr [REDACTED] via email ([REDACTED]) or phone ([REDACTED] / [REDACTED]) if you have any questions or concerns about the study.

Aims of the research and possible benefits.

We hope that this research will help us to understand people's experiences of autism in adulthood, in particular what their quality of life is and what factors might affect this. The results of the study will not benefit you or the individual with autism that you know directly but knowing about the quality of life of this population will be vital for understanding the needs of older people with autism and their families in general. It will also enable us to identify younger individuals who might be at risk of difficulties in later adulthood in order to support them. Finally, we believe that the findings will also help us to improve services for older people with autism.

Who are we contacting for this study?

We are contacting all families / carers of individuals with autism who participated in the most recent follow-up study and agreed to be contacted again for future research

What will happen if you agree to take part?

If you agree to participate, we will ask you to complete the enclosed questionnaire about your perception of the quality of life of the individual with autism. The questionnaire should take no more than 10-15 minutes to complete. We would be grateful if you could then return this to us, along with a completed consent form, in the envelope provided. The information that you provide will then be linked to the information that you (or another family member / carer) provided during the previous follow-up study in order to

learn more about any factors that contribute to the quality of life of an individual with autism in adulthood.

Are there any risks?

There are no anticipated risks associated with the study. However, if you experience any distress as a result of the study, or any other issues arise, please contact us via email ([REDACTED]) or phone ([REDACTED] / [REDACTED]).

Will my taking part in the project be kept confidential?

All information collected from the questionnaires will be kept on a confidential database that is only accessible to those working on the project. Nothing that you tell us will be made available to any other members of your family / team who may be taking part. All personal details will be kept separately from the information collected and participants will be identifiable only by means of a code throughout the study to ensure anonymity. If published, information will be presented without reference to any identifying information. However, in the unlikely event that you report that you believe that yourself or the individual with autism is / has been at risk of harm, this information will be disclosed by the research worker (Dr [REDACTED]) to Dr [REDACTED] and / or Professor [REDACTED], who will then take legal advice on how to deal with this information. All data will be collected and stored in accordance with the Data Protection Act 1998.

What will happen after I participate?

As with the previous study, we will be sending out a regular newsletter, to keep you updated on the progress of the study. The final newsletter at the end of the study will provide a summary of the results.

What will happen if I no longer want my data to be used for the study?

If you decide that you no longer wish to be involved in the research you are free to withdraw at any time. If you decide to do this, any information you have provided will still be very useful to us but this can also be withdrawn and destroyed if you wish, and without you giving any reason. A decision to withdraw at any time, or decision not to take part, will not affect the standard of care / education that you or the individual with autism receives. You may withdraw your data from the project at any time up until it is transcribed for use in the final report (*April 2014*).

Who do I contact if there is a problem?

If you have concerns about any aspect of the study, you should speak to the researcher (Dr [REDACTED]) directly ([REDACTED]) or tel: [REDACTED] / [REDACTED]) or Dr [REDACTED] on ([REDACTED]) or tel: [REDACTED]) and we will do our best to answer your questions. If you remain unhappy and wish to complain formally you can do this through University College London complaints procedure. Details of this procedure can be obtained from University College London. In the event that something does go wrong or you are harmed during the research and this is due to negligence by anyone in the research team then you may have grounds for a legal action for compensation against University College London, but you may have to pay your legal costs. The normal University College complaints mechanism will still be available to you.

Information Sheet for Parents/Carers of Individuals with Autism in Research Studies

(3)

Title of Project: **Quality of life in higher ability adults with autism**

This study has been approved by the UCL Research Ethics Committee

Project ID Number: 4111/001

Name: Dr [REDACTED] / Dr [REDACTED]

Work Address: [REDACTED]
[REDACTED]

Contact Details: [REDACTED] / [REDACTED]
[REDACTED]

We would like to invite parents / carers of adult with autism to participate in this research project

You and your son / daughter / the person that you care for recently kindly participated in a follow-up study of individuals with autism in adulthood. We would like to invite you to take part in this new stage of this research study investigating the quality of life for adults with autism. Before you decide to participate you need to understand why the research is being done, and what it will involve for you and your family. Please take time to read the following information carefully and talk to others if you wish. We encourage you to discuss your decision about whether or not to take part with your son or daughter with autism. However, some parents / carers may decide that, given their son or daughters ability level, this is not appropriate. Please contact Dr [REDACTED] via email ([REDACTED]) or phone ([REDACTED] / [REDACTED]) if you have any questions or concerns about this or any other aspect of the study

Aims of the research and possible benefits

We hope that this research will help us to understand people's experiences of autism for individuals in adulthood, in particular what their quality of life is and what factors might affect this. The results of the study will not benefit you or the individual with autism that you know directly but knowing about this will be vital in understanding the needs of older people with autism and their families. It will also enable us to identify younger individuals who might be at risk of difficulties in later adulthood in order to support them. Finally, we believe that the findings will help us to improve services for older people with autism.

Who are we contacting for this study?

We are contacting all families / carers of individuals with autism who participated in the most recent follow-up study and agreed to be contacted again for future research.

What will happen if you agree to take part?

If you agree to participate, we will ask you to complete the enclosed questionnaire about your perception of the quality of life of the individual with autism. The questionnaire should take no more than 10-15 minutes to complete. We would be grateful if you could then return

this to us, along with a completed consent form, in the envelope provided. The information that you provide will then be linked to the information that you (or another family member / carer) provided during the previous follow-up study in order to learn more about any factors that contribute to the quality of life of an individual with autism in adulthood

Are there any risks?

There are no anticipated risks associated with this study. However, if you do experience any distress as a result of this study, or any other issues arise, please contact us via email ([REDACTED]) / phone ([REDACTED] / [REDACTED]).

Will my taking part in the project be kept confidential?

All information collected from the questionnaires will be kept on a confidential database that is only accessible to those working on the project. Nothing that you tell us will be made available to any other members of your family / team who may be taking part. All personal details will be kept separately from the information collected and participants will be identifiable only by means of a code throughout the study to ensure anonymity. If published, information will be presented without reference to any identifying information. However, in the unlikely event that you report that you believe that yourself or the individual with autism is / has been at risk of harm, this information will be disclosed by the research worker ([REDACTED]) to Dr [REDACTED] and / or [REDACTED], who will then take legal advice on how to deal with this information. All data will be collected and stored in accordance with the Data Protection Act 1998.

What will happen after I participate?

As with the previous study, we will be sending out a regular newsletter, to keep you updated on the progress of the study. The final newsletter at the end of the study will provide a summary of the results.

What will happen if I no longer want my data to be used for the study?

If you decide that you no longer wish to be involved in the research you are free to withdraw at any time. If you decide to do this, any information you have provided will still be very useful to us but this can also be withdrawn and destroyed if you wish, and without you giving any reason. A decision to withdraw at any time, or decision not to take part, will not affect the standard of care / education that you or the individual with autism receives. You may withdraw your data from the project at any time up until it is transcribed for use in the final report (April 2014).

Who do I contact if there is a problem?

If you have a concern about any aspect of the study, you should speak to the researcher directly ([REDACTED] or tel: [REDACTED] / [REDACTED]) or Dr [REDACTED] on ([REDACTED] or tel: [REDACTED]) and we will do our best to answer your questions. If you remain unhappy and wish to complain formally you can do this through University College London complaints procedure. Details of this procedure can be obtained from University College London. In the event that something does go wrong or you are harmed during the research and this is due to negligence of anyone in the research team then you may have grounds for a legal action for compensation against University College London, but you may have to pay your legal costs. The normal University College complaints mechanism will still be available to you.

Informed Consent Form for Individuals with Autism (1)

Please complete this form after you have read the Information Sheet and/or listened to an explanation about the research.

Title of Project: Quality of life of higher ability adults with autism

This study has been approved by the UCL Research Ethics Committee

Project ID Number: 4111/001

Thank you for your interest in taking part in this study. Before you agree to take part, please make sure you understand the study by reading the information sheet. Please ask someone to help you with this if you are unsure about anything.

If you have any questions please contact [REDACTED] or [REDACTED] via email ([REDACTED] / [REDACTED]) or phone ([REDACTED] / [REDACTED]).

You have been given two copies of this Consent Form so that you can keep one for yourself.

Participant's Statement

- I have read the Information sheet about this study, and understand what the study is about.
- I understand that I can stop taking part at any time and ask for my information to be removed from the study without giving any reason before the final report is written (April 2014).
- I have asked all the questions that I want to and have had them answered in a way that I understand.
- I agree to my personal information being used for the purpose of this study.
- I understand that the information that I provide will be kept private and that my personal information will not be given to anyone else.
- I agree to take part in this study

Name (print):

Signed:

Date:

Informed Consent Form for Parents / Carers of Individuals with Autism (2)

Please complete this form after you have read the Information Sheet and/or listened to an explanation about the research.

Title of Project: Quality of life of higher ability adults with autism

This study has been approved by the UCL Research Ethics Committee
Project ID Number: 4111/001

Thank you for your interest in taking part in this research. Before you agree to take part, please make sure you understand what is involved by reading the information sheet provided. If you have any questions arising from the Information Sheet, please contact the researcher via email ([REDACTED] / [REDACTED]) or phone ([REDACTED] / [REDACTED]) before you to decide whether to join in. You have been given two copies of this Consent Form so that you can keep one and refer to it at any time.

Participant's Statement

I(full name)

- have read the notes written above and the Information Sheet, and understand what the study involves.
- understand that if I decide at any time that I no longer wish to take part in this project, I can notify the researchers involved and withdraw immediately without giving any reason. Furthermore, I understand that I will be able to withdraw my data at any time before the final report is written (April 2014).
- consent to the processing of my personal information for the purposes of this research study.
- understand that such information will be treated as strictly confidential and handled in accordance with the provisions of the Data Protection Act 1998.
- agree that the research project named above has been explained to me in a way that I understand
- I agree to take part in this study.

Name (print):

Signed:

Date:

Appendix III

WHOQOL-BREF (Self-report and informant versions)

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Appendix IV
IQ test hierarchy

Table 1. IQ test hierarchy

		Score ¹
Test completed	Merrill Palmer with less than 5 subtests	1
	Full Merrill Palmer, Ravens or Leiter	2
	Part Wechsler test	3
	Full Wechsler test	4
Age test completed	≥ 11 years	1
	8-10 years	2
	3-4 years	3
	5-7 years	4

¹ 1 = lowest quality test / least reliable age, 4 = highest quality test / optimum age

Appendix V
Adult Social Outcome Measure

Table 2. FHS sub-domains of functioning in adulthood

Sub-domains	Level	Score
Independence	Living independently	0
	Semi-sheltered accommodation (or at home) but with high degree of autonomy	1
	Living with parents, some limited autonomy/In residential accommodation with some limited autonomy	2
	In specialist autistic or other residential accommodation with little or no autonomy / in hospital care or at home because nowhere else would accept the individual	3
Friendships	1/+ close reciprocal relationships, in own age group	0
	1/+ close reciprocal relationships but limited in terms of restricted interests or less than normal reciprocity	1
	Seeking of contact but only in group situation/school/work	2
	No peer relationships that involves selectivity	3
Highest Level of Employment ¹	Professional or highly skilled and non-manual skilled jobs	0
	Manual skilled	1
	Partly skilled or unskilled and untrained	2
	Chronically unemployed, homemaker for over one year, sheltered employment, full-time education or never worked	3
Current Employment ¹	Employed or self-employed	0
	Out of work up to 5 years	1
	Out of work 5 years+ Never had a job	2 3
Relationships	Has maintained reciprocal relationships	0
	Reciprocal relationships but shorter than normal for peer group	1
	No enduring relationships / very brief with reduced sharing	2
	No reciprocal relationships longer than one month / never had a relationship	3

¹ Overall employment score was based on an average of (i) the highest level of employment and (ii) current level of employment, to reflect an individuals' overall employment status in adulthood.

Table 4. Overall ratings of adult social outcome

Description	Total Scores	Overall outcome rating
Achieving a high level of independence, having some friends; maintained reciprocal relationships. Employed at some level, now or in the past.	0-2	Very Good
Requiring some degree of support in daily living; some friends/acquaintances; has experienced relationships but typically shorter than normal. Possibly employed at some level, now or in the past.	3-5	Good
Some degree of independence but requires support and supervision does not need specialist residential provision; no close friends but some acquaintances; reciprocal relationships with reduced sharing. Possibly employed at some level, now or in the past.	6-8	Fair
Specialist residential provision / high level of support; no friends outside of residence; no enduring relationships. Possibly employed at some level, now or in the past.	9-11	Poor
High-level hospital care or specialist autistic accommodation, no friends; no autonomy; never had a job; no relationships.	12	Very Poor

Appendix VI

WHOQOL-BREF scores transformation process

Table 4. Method for converting raw scores to transformed scores

Domain 1			Domain 2			Domain 3			Domain 4		
Raw	Transformed										
	4-20	0-100		4-20	0-100		4-20	0-100		4-20	0-100
7	4	0	6	4	0	3	4	0	8	4	0
8	5	6	7	5	6	4	5	6	9	5	6
9	5	6	8	5	6	5	7	19	10	5	6
10	6	13	9	6	13	6	8	25	11	6	13
11	6	13	10	7	19	7	9	34	12	6	13
12	7	19	11	7	19	8	11	44	13	7	19
13	7	19	12	8	25	9	12	50	14	7	19
14	8	25	13	9	31	10	13	56	15	8	25
15	9	31	14	9	31	11	15	69	16	8	25
16	9	31	15	10	38	12	16	75	17	9	31
17	10	38	16	11	44	13	17	81	18	9	31
18	10	38	17	11	44	14	19	94	19	10	38
19	11	44	18	12	50	15	20	100	20	10	38
20	11	44	19	13	56				21	11	44
21	12	50	20	13	56				22	11	44
22	13	56	21	14	63				23	12	50
23	13	56	22	15	69				24	12	50
24	14	63	23	15	69				25	13	56
25	14	63	24	16	75				26	13	56
26	15	69	25	17	81				27	14	63
27	15	69	26	17	81				28	14	63
28	16	75	27	18	88				29	15	69
29	17	81	28	19	94				30	15	69
30	17	81	29	19	94				31	16	75
31	18	88	30	20	100				32	16	75
32	18	88							33	17	81
33	19	94							34	17	81
34	19	94							35	18	88
35	20	100							36	18	88
									37	19	94
									38	19	94
									39	20	100
									40	20	100

Appendix VII
Ethics approval document

[REDACTED]

15 October 2012

Dear Mr [REDACTED]

Notification of Ethical Approval

Project ID: 4111/001: Quality of life in higher ability adults with autism

I am pleased to confirm that further to receipt of your amended Information Sheets your study has been approved by the UCL Research Ethics Committee for the duration of the project i.e. until September 2014.

In relation to committee members insistence that individuals with autism should be informed that their parents/carers would be asked to complete an informant QOL questionnaire to ascertain their own perception (not the view) of the QOL of the individual with autism, your response has been reviewed and your approach (outlined below) approved:

- Those with higher-ability autism will be informed;
- Parents of individuals in the lower ability autism sub groups will be encouraged to discuss their own participation in this study with the person with autism if they think that this is appropriate, based on the ability level of that individual. In this way, they can determine whether or not it would be appropriate to discuss the study with the person with autism directly.

Approval is also subject to the following conditions:

1. You must seek Chair's approval for proposed amendments to the research for which this approval has been given. Ethical approval is specific to this project and must not be treated as applicable to research of a similar nature. Each research project is reviewed separately and if there are significant changes to the research protocol you should seek confirmation of continued ethical approval by completing the 'Amendment Approval Request Form'.

The form identified above can be accessed by logging on to the ethics website homepage: <http://www.grad.ucl.ac.uk/ethics/> and clicking on the button marked 'Key Responsibilities of the Researcher Following Approval'.

2. It is your responsibility to report to the Committee any unanticipated problems or adverse events involving risks to participants or others. Both non-serious and serious adverse events must be reported.

Reporting Non-Serious Adverse Events

For non-serious adverse events you will need to inform Helen Dougal, Ethics Committee Administrator (ethics@ucl.ac.uk), within ten days of an adverse incident occurring and provide a full written report that should include any amendments to the participant information sheet and study protocol. The Chair or Vice-Chair of the Ethics Committee will confirm that the incident is non-serious and report to the Committee at the next meeting. The final view of the Committee will be communicated to you.

Reporting Serious Adverse Events

The Ethics Committee should be notified of all serious adverse events via the Ethics Committee Administrator immediately the incident occurs. Where the adverse incident is unexpected and serious, the Chair or Vice-Chair will decide whether the study should be terminated pending the opinion of an independent expert. The adverse event will be considered at the next Committee meeting and a decision will be made on the need to change the information leaflet and/or study protocol.

On completion of the research you must submit a brief report (a maximum of two sides of A4) of your findings/concluding comments to the Committee, which includes in particular issues relating to the ethical implications of the research.

With best wishes for the research.

Yours sincerely

Professor [REDACTED]
Chair of the UCL Research Ethics Committee

Cc: [REDACTED]

Appendix VIII
IQ tests completed

Table 5. IQ tests completed

	Lower IQ sub-group	Higher IQ sub-group	Total sample
	Total N = 28	Total N = 24	Total N = 52
Merrill Palmer	20 (71%)	8 (33%)	28 (54%)
WISC	5 (18%)	14 (58%)	19 (37%)
WPPSI	2 (7%)	1 (4%)	3 (6%)
Leiter International Performance Scale	1 (4%)	1 (4%)	2 (4%)