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A systematic review of Quality of Life in adults on the autism spectrum

Abstract

Autism Spectrum Disorder (ASD) is associated with co-existing conditions that may adversely affect an individual's Quality of Life (QoL). No systematic review of QoL in adults on the autism spectrum has been conducted. Our objectives were: 1. review the evidence about QoL for adults on the autism spectrum; and 2. critically appraise current practise in assessing QoL in adults on the autism spectrum.

We searched bibliographic databases and other literature to identify studies using a direct measure of QoL in adults on the autism spectrum. Hand searching of reference lists, citation searching and personal communication with field experts was also undertaken.

827 studies were identified; 14 were included. All compared the QoL of autistic adults to neurotypical adults; QoL was lower in people on the autism spectrum. Only one QoL measure designed for use with the general autism spectrum population was identified.

QoL for adults on the autism spectrum is lower than that of typically developing adults, when measured with tools designed for the general population. There are no comprehensive ASD-specific QoL measurement tools validated for use with representative samples of adults on the autism spectrum. There is a pressing need to develop robust measures of QoL in autistic adults.

Background

Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder with an impact throughout the lifespan . Children and adults on the autism spectrum have an increased susceptibility to a range of comorbid psychiatric and neurological disorders such as anxiety disorders, depression and epilepsy (Stahlberg et al., 2004, Levisohn, 2007, Simonoff et al., 2008). Whilst approximately 1% of the UK child and adult population have ASD (Baird et al., 2006, Brugha et al., 2011), the majority of ASD research concerns children and adolescents, with little research evidence about the lives of adults on the autism spectrum (Mukaetova-Ladinska et al., 2012) and their quality of life (QoL) (van Heijst and Geurts, 2015).

The World Health Organisation (WHO) defines QoL as *"an individuals' perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns"* (Harper, 1998). The UK National Institute for Health and Care Excellence (NICE) recognised that ASD may have a significant effect on QoL, and stated that improving QoL should be one aim of any psychosocial intervention for people on the autism spectrum (NICE, 2013). Most current QoL measurement tools were designed for use with people from the general population. In recent years, QoL measures have been designed for people with a range of specific health and mental health conditions in order to capture aspects of life that are in some way 'different' from that of the general population due to their condition. In relation to autism, QoL measures often have a focus that makes them potentially inappropriate for use in adults on the autism spectrum – for example an emphasis on social integration and social relationships.

Several studies have investigated the QoL of individuals on the autism spectrum – for example changes in QoL across the lifespan (van Heijst and Geurts, 2015) and have identified factors that influence QoL (Chiang and Wineman, 2014). However, there is currently no systematic review evidence of the QoL of adults on the autism spectrum or the measures used in studies. Our systematic review aims were two-fold: first, to systematically identify and review the evidence for QoL in adults on the autism spectrum and second, to critically appraise the tools that have been used to assess QoL in adults on the autism spectrum.

Review Methods

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines for conducting systematic reviews were adhered to throughout the review process (Moher et al., 2009).

Review criteria

Randomised controlled trials (RCTs), observational, qualitative and mixed methods studies published in English from any country of origin were eligible for inclusion in the review. Eligible studies were required to include adult participants (aged ≥18 years) with a clinical diagnosis of ASD (confirmed by an appropriately qualified medical professional either for the purpose of the study or prior to recruitment) and use a direct measure of QoL (proxy, non-proxy, measures of QoL). Studies using indirect measures of QoL, such as employment status, educational level, accommodation and perceived loneliness among others, including measures of psychological well-being were excluded.

Search strategy

With specific guidance and expertise from an information scientist, and informed by scoping searches, a structured search strategy was developed using a combination of keywords and MeSH terms.

The following bibliographic databases were searched: MEDLINE (1966-October 2016), EMBASE (1980-October 2016), PsycINFO (1880-October 2016), Scopus (1960-October 2016) and Web of Science (1864-October 2016) and the Cochrane Library. Key search terms used were autis*, Asperger* and Quality of Life. Citation and hand searching of studies that fulfilled the eligibility criteria were also conducted. Experts in the field were consulted in May 2015 to identify any work in progress or grey literature.

Study Selection Process

Studies were selected through screening of titles and abstracts according to the inclusion criteria by one reviewer (MA). Studies identified by the primary selection process were independently assessed for inclusion using a study selection form. Disagreements were resolved by discussion (occurred

twice), or when agreement could not be reached, other co-authors were asked to adjudicate (occurred on three occasions).

Data extraction of included studies was undertaken using a data extraction form. When necessary, study authors were contacted with a request for additional information.

Assessment of Methodological Quality

Included studies were subjected to independent assessment of methodological quality by two reviewers (LA and DF). The Cochrane Collaboration tool for RCTs (Higgins and Green, 2009) was used to assess the risk of bias in six domains for the two experimental studies. A 5-item checklist (Flynn et al., 2012) adapted from the Newcastle-Ottawa Quality Assessment Scale for Case-Control Studies (Wells et al., 2014) was used to assess risk of bias in cross-sectional studies across five domains.

The 16-item Quality Assessment Tool for Studies with Diverse Designs (Sirriyeh et al., 2012) was used to assess the methodological quality of the remaining two pre-test post-test studies. The 14 items relevant for quantitative designs were used, which are scored from 0 to 3 (range of scores from 0 to 42), with total scores converted into percentages for reporting.

Data synthesis

Due to the expected heterogeneity of study designs and QoL measurement tools, and the small number of participants from individual studies, a narrative approach was used to synthesise the findings of the included studies. In synthesising the data from the methodological review tools emphasis was placed on the review criteria relevant to the objectives of this current review.

Results

Fourteen of 827 studies identified by the search strategy fulfilled the review criteria (Figure 1). Two were randomised studies (Garcia-Villamisar, 2010; Hesselmark et al., 2014). Ten studies were cross-sectional designs that assessed QoL in adults on the autism spectrum at a specific point in time (Jennes-Coussens et al., 2006, Saldaña et al., 2009, Kamp-Becker et al., 2010, Kamio et al., 2013,

Khanna et al., 2014, Lin, 2014, Gal et al., 2015, Katz et al., 2015, van Heijst and Geurts, 2015, Hong et al., 2016) and two were pre-test post-test intervention studies (Garcia-Villamisar, 2010, Hesselmark et al., 2014, Gal et al., 2015). Sample sizes ranged from 12 (Jennes-Coussens et al., 2006) to 291 adults (Khanna et al., 2014). The combined sample size from the 14 included studies was 959 adults. Collectively, these studies focussed on individuals aged from 18-55 years old with one study including participants up to 83 years (van Heijst and Geurts, 2015). As expected, there was a predominance of male participants.

Tools used to assess QoL in adults on the autism spectrum

Seven tools were used to assess QoL in adults on the autism spectrum. The psychometric properties of the tools and the populations in which the tools were used are described in the following section.

World Health Organisation Quality of Life – BREF (WHOQOL-BREF)

The WHOQOL-BREF tool is an abbreviated 26-item version of the original 100-item WHOQOL tool designed for use in subjective self-assessment of QoL across a range of population types. The tool assesses QoL across four domains: Physical Health, Psychological Health, Social Relationships and Environment. Reference values for different versions of the WHOQOL-BREF are available and QoL is scored out of a maximum total of 100. The WHOQOL-BREF has not been validated in research with autistic adults. Research using the tool with the general population indicated internal consistency coefficients (Cronbach's alpha) between 0.66 for the social domain to 0.84 for the physical domain. Test retest reliability ranges from 0.66 for the physical domain to 0.87 for the environmental domain. Data relating to construct validity and sensitivity to change are not available.

Quality of Life Questionnaire (QoLQ)

The QoLQ tool is a 40-item questionnaire designed for assessing QoL in individuals with Intellectual Disability (ID). It was designed to be administered in an interview format. The QoLQ assesses QoL across four domains: Satisfaction Competence/Productivity, Empowerment/Independence and Social Belonging/Community Integration. No normative reference values for QoL scores are available. A theoretical minimum total QoL score is 40 and a theoretical maximum score would be 120. Internal

consistency coefficient (Cronbach's alpha) has been reported for the total score as 0.9. Test retest reliability was 0.87. Data relating to construct validity and sensitivity to change are not available.

No guidance is available on categorisation of scores; for example as low, average or good. Findings from studies using the QoLQ cannot be easily compared to scores obtained using other QoL measurement tools because the domains of QoL assessed differ from other measures. The QoLQ has not been validated in research with autistic adults.

Quality of Life Inventory (QOLI)

The QOLI is a 32-item tool that measures QoL across four domains: Health, Relationships, Employment and Living Conditions. Internal consistency coefficient (Cronbach's alpha) for the domain total scores ranges from 0.77 to 0.89. Test retest reliability ranges from 0.80 to 0.91. Data relating to construct validity and sensitivity to change are not available. The QOLI has not been validated in research with autistic adults.

Comprehensive Quality of Life Questionnaire (ComQOL)

The ComQOL is a 35-item tool that assess QoL over seven domains: Material wellbeing, Health, Productivity, Intimacy, Safety, Place in Community and Emotional Wellbeing. No normative or control values for the ComQOL are available. Data relating to internal consistency, test retest reliability, construct validity and sensitivity to change are not available. The ComQoL has not been validated in research with autistic adults.

SF-36 (RAND-36)

The SF-36 a 36-item tool that assesses QoL over eight domains: Bodily pain, General health, Vitality, Social Functioning, Physical Functioning, Mental Health, Emotional well-being, Limitations to role caused by mental health problems, and Limitations to role caused by physical health problems. It has not been validated in research with autistic adults. Research with the general population indicates internal consistency coefficients (Cronbach's alpha) between 0.71 for the social domain to 0.92 for individual domains. Data relating to test retest reliability, construct validity and sensitivity to change are not available.

Medical Outcomes Study Short-Form Health Survey version 2 (SF-12 v.2)

The SF-12 v.2 is an abbreviated 12-item version of the SF-36 where items are grouped into Physical Component Summary and Mental Component Summary. The SF-12 v.2 has not been validated in research with autistic adults. Data relating to internal consistency, construct validity and sensitivity to change are not reported. Test retest reliability ranges from 0.89 for the physical component score to 0.76 for the mental component score.

Novel Quality of Life Measures (QOL1, QOL2)

The QOL1 is a proxy assessment of "autism-friendly environment" and includes assessment of the autism-specific knowledge held by the family, the provision of a structured education programme, the implementation of a personal treatment and care plan, the extent to which everyday life/employment is appropriate to the individual's ability and an overall rating of QoL. Each domain is scored on a scale of 1-5, with a score of 1 described as "very good", 2 as "good", 3 as "average", 4 as "poor" and 5 as "very poor". The QOL2 consists of a parent/carer rating of the wellbeing of an adult on the autism spectrum. This is scored on a scale of 1-5 and provides a marker of overall wellbeing in a residential setting. Cronbach's alpha for QOL1 is reported as 0.96 (Billstedt et al., 2011). Data relating to test retest reliability, construct validity and sensitivity to change are not available.

In summary, seven QoL measures were used across the 14 studies reviewed. A number of domains are assessed using the individual tools, the most common being physical health and mental health/wellbeing. Where they were reported, internal consistency and test-retest reliability were within acceptable levels, although it is important to note within the context of the current review that these metrics are derived from non-autistic samples, with no validation of these tools specifically for use with autistic adults (with the exception of QOL1 and QOL2). Only one study reported on the use of a QoL measure designed for use with adults on the autism spectrum (Billstedt et al., 2011). Billstedt et al. (2011) noted that their novel measure was brief and more work was required before it could be considered to be a valid QoL measure for autistic adults.

Results from studies using self-report QoL measures with autistic adults

The following section describes the results from eight identified papers reporting self-assessment of QoL by autistic participants.

Studies utilising the WHOQOL-BREF

Four studies included in the review utilised the WHOQOL-BREF (Jennes-Coussens et al., 2006, Kamp-Becker et al., 2010, Kamio et al., 2013, Lin, 2014). Jennes-Coussens et al. (2006) assessed the QoL of twelve 18-21 year old males with a diagnosis of Asperger Syndrome and compared this to a typically developing control group. Questionnaires were posted to participants. Results showed that participants with Asperger Syndrome rated their QoL lower than controls, (mean overall score of 3.7 (sd = 0.7) versus 4.1 (sd = 0.6) respectively). Of the four domains, only differences in the scores for Social Relationships reached statistical significance with participants with Asperger's syndrome reporting a mean score of 49.5 (sd = 23.0) compared to 78.4 (sd = 18.2) for typically developing controls.

In a substantially larger sample Kamio et al. (2013) reported on the QoL of 154 adults on the autism spectrum. Questionnaires were posted to participants. The authors only report on two domains of the tool, Psychological Health and Social Relationships. Autistic participants scored significantly lower than a typically developing reference group (2.71 vs. 3.19-3.25 respectively for Social Relationships; and 2.78 vs. 3.26-3.32 respectively for Psychological Health).

Kamp-Becker et al. (2010) assessed QoL of an all male sample of 26 adults on the autism spectrum compared to two age-specific reference samples (healthy individuals and individuals with schizophrenia-spectrum disorder) using the German version of the WHOQOL-BREF. QoL scores were significantly lower for autistic individuals than in the typically developing sample (mean 60.6 (sd = 26.1) compared to 75.5 (sd = 16.4) respectively). In three of four domains mean scores for autistic participants were significantly lower QoL than the reference population; Physical Health (70.1, sd = 19.1 vs. 87.2, sd = 13.6), Psychological Health (61.5, SD = 21.9 vs. 79.1, sd = 14.0) and Social Relationships (53.8, sd = 23.5 vs. 74.9, sd = 17.5). Autistic participants rated QoL in the Social Relationships domain lowest.

In Taiwan Lin (2014) conducted a cross-sectional study with 41 autistic adults (mean age 26.9 years) and compared to a control group of 41 adults without ASD selected from a community sample of 122 adults without ASD and matched to the autistic sample on age and gender. The autistic participants scored consistently lower than matched controls. Further descriptive comparisons were made without significance testing to a Taiwanese health population reference group of 9107 individuals for the Taiwanese version of the WHOQOL-BREF. Analysis showed autistic adults reported lower QoL in the Social relationships domain (mean 1.5 standard deviations below the reference group), and in the domains of Physical and Psychological Health (mean 1.0 standard deviation below the reference group).

Studies utilising the QOL-Q

Two included study used the QoL-Q, both of which provided data at more than one time point. Gal et al. (2015) sought to assess the impact of engaging in an army training and service programme on QoL in 25 autistic adults in Israel (Autistic adults in Israel are exempt from national military service, which is usually undertaken at age 18). The authors sought to examine the effect of enrolment in three month training programmes followed by a six month placement in a profession in the armed forces that "would match the 'special characteristics of people with ASD', including aerial photography interpretation. QoL was measured immediately before and after the three month aerial photography interpretation training programme and after six months of integration in an army unit. No significant difference in overall QoL or individual domains was found between the beginning of the training period and afterwards. However, a statistically significant self-reported increase in QoL was noted after six months of integration in the army unit across all domains.

Katz et al. (2015) investigated the influence of taking part in a nine month work placement on the self-reported QoL of 26 adults with 'High Functioning' ASD. Participants were either about to transfer from education to employment or were already seeking employment. QoL was measured every three months throughout their employment period. After nine months a non-significant increase from baseline in self-reported overall QoL was found, particularly in the self-competence domain.

Studies utilising QOL-I

Only one included study utilised the QOL-I. Hesselmark et al. (2014) assessed QoL in 60 autistic adults in an intervention study that evaluated the impact of Cognitive Behavioural Therapy (CBT) versus recreational activity on QoL and Self-Esteem. The CBT group received a programme aimed at introducing participants to concepts of acceptance and change; whereas those in the Recreational Activity intervention group received structured activity that promoted social interaction. The mean QOLI scores in the CBT and Recreational Activity groups were -0.11 and -0.28 at baseline, 0.64 and -0.01 post-intervention and 0.64 and 0.30 at follow-up respectively (negative scores indicate a response of 'dissatisfied'). The authors also quoted a mean QOLI score for non-clinical controls of 2.8 and note that the total participants' mean score was -0.19 in the autistic adults at baseline which *"indicated a general dissatisfaction with life (and) is similar to scores of untreated post-traumatic stress disorder (PTSD) patients (mean=0.3, SD 3.2) observed in other studies"*.

Studies utilising the SF-36

One study used the SF-36. In a cross-sectional study van Heijst and Geurts (2015) found QoL to be significantly lower in 24 autistic adults compared with controls (mean total score 66.7 (sd = 16.6) vs. 82.0 (sd = 13.2)). Age and IQ were not found to predict QoL but autism severity (as measured by the social responsiveness scale) was a significant predictor of QoL.

Studies utilising the SF-12 v.2

One study used a derivative of the SF36, the SF 12 V2. Khanna et al. (2014) conducted a cross-sectional study of QoL in 291 autistic adults registered with the Interactive Autism Network. The results indicated that the mental summary component of the SF-12 tool was lower for autistic adults compared to US norms. This statistically significant difference was present across both genders and all age groups. Scores for the physical summary component were only significantly lower in autistic adults aged 18-24 years compared to US norms. Severity of autism symptoms were assessed for each participant using the Adult Short Autism-Spectrum Quotient-10 Items (AQ-10). Physical Component Scores were higher in the high autism severity group (50.9) than the low autism severity group (46.9); Mental Component Scores were higher for the low autism severity group (42.6) than the high autism severity group (39.7).

Results from studies utilising proxy reports of QoL in autistic adults

Studies utilising the ComQOL

Saldaña et al. (2009) assessed the QoL of 25 autistic adults via parent/carer report. Informants completed an interview which included aspects of the ComQOL. The domains 'Material Wellbeing' and 'Intimacy with Family' were reported to be the domains with the highest QoL (83.6% and 81.4% respectively), followed by Place in Community (67.8%), Productivity (66.4%), Health (63.3%), Safety (61.3%) and Intimacy with Friends (55.8%). Assessment of the "Emotional Wellbeing" domain was excluded from the study as the authors concluded that this domain would be too complex for proxy assessment.

Studies utilising the QOL1 and QOL2

Billstedt et al. (2011) conducted a cross-sectional study of QoL with 108 parents/carers of autistic individuals using two novel parent/carer proxy report tools designed for the study, the QOL1 and QOL2. Findings indicated a mean score of average/good across participants utilising the QOL1 tool; using the QOL2 tool, parents and carers rated their family member's QoL as very good or good in 91/100 cases.

Results from studies using a combination of proxy and self-reported QoL

Studies utilising the WHOQOL-BREF

Hong et al. (2016) conducted a cross-sectional study investigating the relationship between autistic adult self-report, maternal proxy report and maternal report of QoL in a sample of 60 mother and adult-offspring dyads. The mean age of the participating autistic adults was 32.0 (SD=6.8). Correlation between autistic adult self-report and maternal proxy report was significantly higher than the correlation between autistic adult self-report and maternal self-report across 3 domains (Physical Health, Psychological Health and Social Relationships). For autistic adult self-report and maternal report mean scores were not significantly different for 3 of the domains (Physical Health, Psychological Health, and Environment) but autistic adults self-reported significantly higher QoL in the Social Relationships domain (m=71.2, sd=26.9) than on maternal report (m=60.2, sd=24.1). There

were no significant differences in mean domain scores between adult self-report and maternal proxy report.

Studies utilising the QOL-Q

Garcia-Villamisar (2010) evaluated the effect of a twelve month leisure programme intervention on the QoL of 37 adults on the autism spectrum. The intervention group were engaged in exercise groups, craft workshops, media-based activities and organised events and outings; with a wait-list control group of 34 autistic individuals. The authors assessed receptive and expressive language skills in autistic participants and administered the QoLQ to those deemed capable of self-reporting. For those who couldn't self-report, at least two individuals who knew the participant well completed the QoLQ on the individual's behalf (the number in this group is not reported). Compared with baseline scores, a statistically significant increase in QoL was reported in the intervention group after 12 months follow-up (improved from 50.6 to 63.6), but not in the control group (scores changed from 54.2 to 55.3).

Garcia-Villamisar et al. (2013) conducted a cross-sectional study to investigate the association between QoL, challenging behaviours and autistic traits in 70 autistic adults with intellectual disability (ID). As with Garcia Villamisar et al. (2010) participants' verbal skills were assessed and people considered able to do so completed the measure themselves (6 participants; therefore 64 had proxy report). The mean overall baseline QoL score was 72.5; however, the authors do not provide interpretation of the QoL scores.

Synthesis of findings across studies

The majority of included studies used self-assessment to assess QoL in participants. All studies where a comparison was made to a typically developing sample reported QoL to be lower in autistic adults. It is of note that 7 of the of 14 (50%) included studies that did not include reference values to enable QoL scores from their samples with typically developing individuals (Saldaña et al., 2009, Garcia-Villamisar, 2010, Billstedt et al., 2011, Garcia-Villamisar et al., 2013, Hesselmark et al., 2014, Gal et al., 2015, Katz et al., 2015). It is therefore difficult to draw any firm conclusions regarding the comparison of the QoL of autistic individuals participating in these studies to that of the general population.

In studies that reported individual domain scores, QoL tended to be reported to be pervasively lower for autistic adults and to be notably low in domains capturing social relationships and integration. For example, all four studies using the WHOQOL-BREF reported that QoL scores were lower for autistic adults overall and across all four domains, when compared to neurotypical comparison groups (Jennes-Coussens et al., 2006, Kamp-Becker et al., 2010, Kamio et al., 2013). Across all four studies, adults on the autism spectrum reported lowest QoL on the Social Relationships domain. Saldaña et al. (2009) found that scores were lowest in the "Intimacy with friends" domain of the ComQOL.

Evidence for the impact of autism severity on QoL has not been comprehensively examined. The two studies (Khanna et al., 2014, van Heijst and Geurts, 2015) that do examine the associations between autism severity and QoL report a significant correlation between greater severity of autistic symptoms and lower QoL. Furthermore, Saldaña et al. (2009) attribute an inability for family members to act as proxies in assessing the QoL for an autistic relative as increased symptom severity.

Methodological quality of included studies

The methods by which participants had been diagnosed with ASD were comprehensively reported in most of the included studies. Nine studies included evidence that the ASD diagnosis had been made by a Clinical Psychologist, Psychiatrist or multidisciplinary clinical team (Jennes-Coussens et al., 2006, Saldaña et al., 2009, Garcia-Villamisar, 2010, Kamp-Becker et al., 2010, Billstedt et al., 2011, Garcia-Villamisar et al., 2013, Hesselmark et al., 2014, Lin, 2014, van Heijst and Geurts, 2015). Further clarification about diagnosis was sought from the authors of two studies (Kamio et al., 2013, Khanna et al., 2014). Khanna et al. stated that they recruited through the Interactive Autism Network (IAN) database and that most of the individuals included on IAN had received a diagnosis by a clinician or their diagnosis had been clinically validated (personal communication, 29th July, 2015); however, some individuals had volunteered to be included without a clinician given ASD diagnosis but this applied to a minority of participants (Daniels et al., 2012). Kamio et al. (2013) did not offer sufficient information about the method of ASD diagnosis in their study for us to comment further.

The methodological quality assessment using structured checklists can be found in the web appendix.

Blinding of outcome assessment was inadequately addressed in the RCT by Hesselmark et al. (2014). The potential for other sources of bias was considered low risk for both experimental studies ((Garcia-Villamisar, 2010, Hesselmark et al., 2014).

All nine cross-sectional studies included an adequate description of the presenting condition, including a definition of the exposure and ascertainment of outcomes. However, six of these studies were regarded as unclear risk of bias from selection, and five from non-response in accordance with the quality checklists used.

The two pre-test post-test studies were of acceptable methodological quality. The total score percentages for Gal et al. (2015) and Katz et al. (2015) were 60% and 55% respectively.

Discussion

We identified fourteen studies that reported on QoL in autistic adults using either self or proxy report or a combination of the two. Narrative synthesis of data from these studies reveals a trend for QoL to be lower in autistic adults compared to typically developing adults. Thirteen studies used QoL measures that have previously been shown to be reliable and valid indicators of QoL in general population samples but importantly these measures have not been validated for use with autistic adults. One measure of QoL, composed of the QOL1 and QOL2 tools, had been validated for use with individuals with intellectual disability (Billstedt et al., 2011). The identification of widespread use of tools without established reliability and validity for measuring QoL in autistic adults is an important finding of this review because this practice may lead to misleadingly low or high scores.

A synthesis of the findings from across the included studies reveals that autistic adults are particularly likely to score lower than their typically-developing counterparts on QoL domains assessing social inclusion and interaction. Without a clear understanding of what specific aspects are important to assess in relation to QoL for autistic people it is difficult to make sense of these data. Two potential explanations are possible. It is possible autistic people's inherent difficulties regarding social interaction and communication are a risk factor for lower QoL reported in these domains. However,

we need to also be mindful that that autistic individuals may value some experiences and activities more/less than people without ASD (Tavernor et al., 2013). For example, time spent on repetitive behaviours, and on circumscribed or particular interests may be considered more desirable than some activities undertaken by typically developing adults; social and group activities may be valued less by autistic adults. Therefore the items included in these specific domains of assessment tools developed for and validated with the general population may lack validity when used with autistic individuals. Of course, these two explanations are not mutually exclusive. One firm conclusion that can be drawn from this review is that it is imperative we develop a greater understanding of what aspects of life matter most and affect the lives of autistic people and that we develop tools to measure these when assessing their QoL.

Seven of the fourteen included studies showed a bias towards recruiting adults on the autism spectrum without intellectual (learning) disability (ID) (Jennes-Coussens et al., 2006, Kamp-Becker et al., 2010, Kamio et al., 2013, Hesselmark et al., 2014, Khanna et al., 2014, Lin, 2014, van Heijst and Geurts, 2015), Thus the review does not provide a comprehensive picture of QoL for those 30-40% of autistic adults with co-occurring intellectual ability (Totsika et al., 2010) – and this review's interpretation of study results may or may not be applicable to this population.

Similarly, the findings highlight the conundrum of how best to disentangle the likely complex interactions between autism symptom severity and QoL. Two studies suggest QoL is lower when autism severity is greater (Khanna et al., 2014, van Heijst and Geurts, 2015). Saldaña et al. (2009) importantly remind us that a greater degree of autism severity may impact on people's capacity to self-report, and further may also impact on proxy raters accurately determining QoL for the person they are reporting for. Hong et al. present the first data directly comparing proxy and self-report of QoL data from autistic adults. Their results point to a correlation between maternal proxy report and self-report by autistic adults; however the majority of autistic adults who participated in the study were described as high functioning (Hong et al., 2016). The ability level of the autistic participants may have made it easier for proxy reporters to be make judgements about QoL for the autistic person they were reporting on. Furthermore, despite overall general agreement between self and proxy report, Hong and colleagues report a significant difference between adult self-report and maternal proxy report in the social relationship domain, where maternal ratings were lower. They conclude that although

mothers are generally accurate reporters of autistic adult offspring QoL there may be a tendency for proxy reporters to consider QoL in social relationships as poorer than autistic adults self-report. Interestingly, Hong et al report strongest agreement in the environmental domain and least agreement in the psychological health domain. This may suggest proxy reporters have more difficulty interpreting the internal or intrapersonal aspects of QoL of autistic adults – or may provide further evidence for lack of construct validity of particular neurotypically derived and validated domains of QoL for autistic adults.

Taken together the reviewed evidence highlights a clear take home message. To address the conceptual and methodological challenges, a measure of QoL either designed specifically for or validated with autistic adults is required to accurately assess their QoL. Improving measures already used with autistic adults rather than designing new tools from first principles is most likely to lead to the timely production of acceptable measures. Only one study identified in this review used a measure designed specifically for autistic people (Billstedt et al., 2011); interestingly this was the only study to report better QoL for autistic adults, compared with control populations. Billstedt et al. (2011) stated that the measure would benefit from further development to provide a more in-depth assessment across the multiple domains that constitute QoL.

Strengths and weaknesses of the current review

A key strength of this review is inclusion of only studies that reported on participants for whom a valid ASD diagnosis had been ascertained (although of course this could not be verified as part of this study). The review of the psychometric properties of the QoL measures was undertaken according to defined criteria. This further highlighted the shortcomings and methodological/design issues that should be addressed when investigating QoL in autistic adults. A potential weakness of this review is the exclusion of studies published in languages other than English, as these may contain relevant information. Similarly the exclusion of case studies, although limited in the reliability of their conclusions, may have excluded the results of studies using QoL measurement tools not identified in this review.

Implications for Future Research

The studies reviewed had several important methodological shortcomings, which should be addressed in future research. Bishop-Fitzpatrick et al. (2016) outline a need to address the conceptualisation of QoL among autistic people. They identify "normative" outcomes (employment, independent living, social engagement) and "objective QoL" (physical health, mental health, neighbourhood quality, family contact) as relevant concepts. The first step therefore is to reach consensus regarding what constitutes 'good' QoL for autistic individuals, across the lifespan. This cannot be achieved without active involvement of autistic people to define QoL - either as part of the research team, or as research consultants. Without the involvement of autistic adults in design, measures are unlikely to have content validity, or have acceptable wording and formatting (Parr, 2016). The participation of autistic adults and relatives' in discussion groups is required to ensure a tool takes account of the views of people with ID, or those who represent adults who are unable to participate themselves. It is possible that more than one QoL measurement tool will be required – for example for adults on the autism spectrum with and without ID, and potentially for younger and older adults. There are two possible approaches to optimal development of these tools; either the adaptation of an existing QoL measurement tool or the development of an entirely novel tool. The former would allow access to an existing framework and have the significant benefit of control data from a neurotypical population being available, while the latter allows greater freedom to fully incorporate the views and experiences of adults on the autism spectrum and those people who know them well. Either approach will require the investigation of the psychometric properties of any new or adapted tool and their acceptability to adults on the autism spectrum, including to people who might complete the measure on their behalf. At [TEXT REMOVED FOR BLIND REVIEW] and colleagues have started to consider how to develop aspects of the WHO-QoL BREF (with permission), with the aim of creating a reliable and valid way of measuring QoL in autistic adults (REFERENCE REMOVED FOR BLIND REVIEW).

Conclusions

Adults on the autism spectrum typically report lower QoL than typically developing adults when measured with tools designed for the general population. There is a pressing need to develop a reliable and valid measure of QoL in adults on the autism spectrum. Large studies of representative

populations of people with ASD are needed using measurement tools that are reliable and valid for use with autistic adults. Parallel collection of data about autism characteristics, and conditions that often accompany autism would allow investigation of how individual factors (such as mental health conditions) affect QoL. As we know from childhood autism research, planned multisite studies, data sharing and joint publication by groups working in collaboration would be desirable to ensure timely new knowledge is available to the autism community and those planning services.

Conflicts of interest

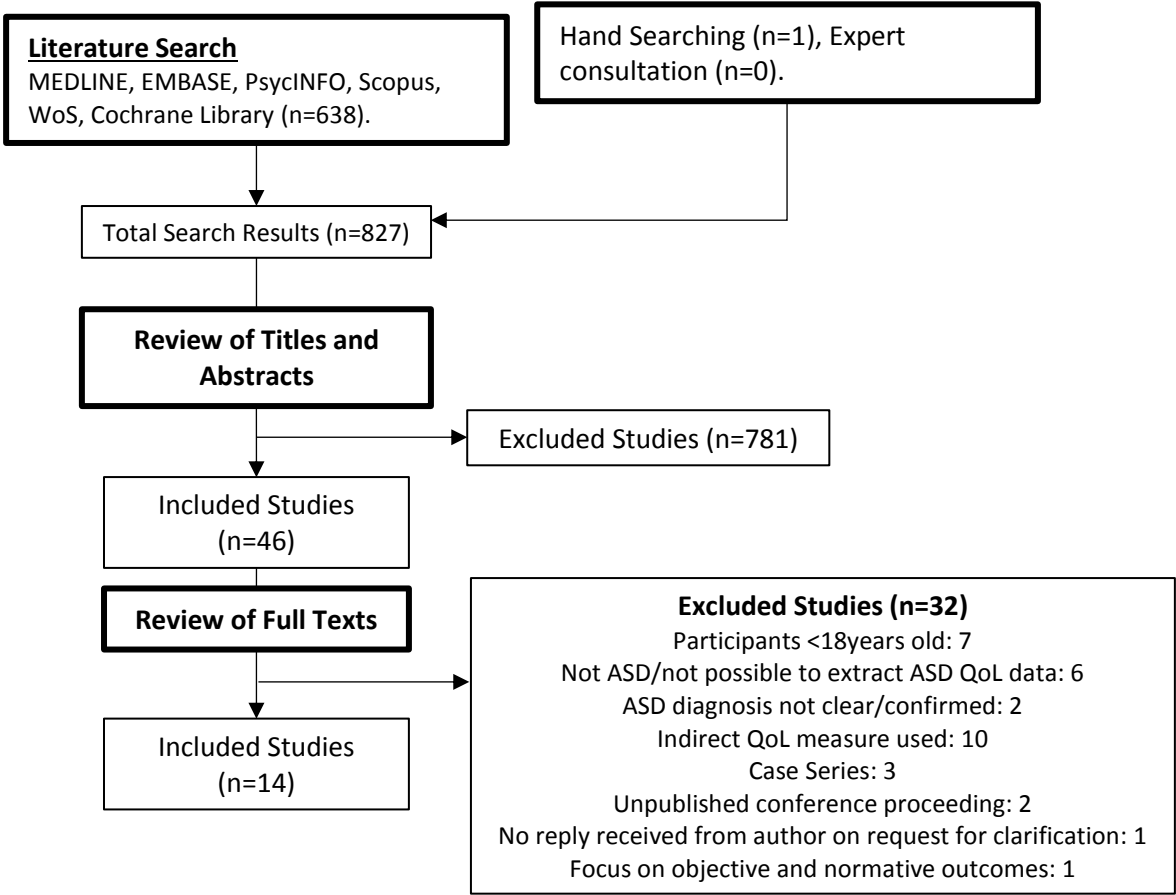None of the authors have any conflicts of interest.

Figure 1

Figure 1: PRISMA diagram showing the process used to identify studies to show Study Search and Selection

References

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). *The lancet,* 368**,** 210-215.

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**,** 7-20.

BISHOP-FITZPATRICK, L., HONG, J., SMITH, L. E., MAKUCH, R. A., GREENBERG, J. S. & MAILICK, M. R. 2016. Characterizing objective quality of life and normative outcomes in adults with autism spectrum disorder: An exploratory latent class analysis. *Journal of autism and developmental disorders,* 46**,** 2707-2719.

BRUGHA, T. S., MCMANUS, S., BANKART, J., SCOTT, F., PURDON, S., SMITH, J., BEBBINGTON, P., JENKINS, R. & MELTZER, H. 2011. Epidemiology of autism spectrum disorders in adults in the community in England. *Archives of general psychiatry,* 68**,** 459-465.

CHIANG, H.-M. & WINEMAN, I. 2014. Factors associated with quality of life in individuals with autism spectrum disorders: A review of literature. *Research in Autism Spectrum Disorders,* 8**,** 974-986.

DANIELS, A. M., ROSENBERG, R. E., ANDERSON, C., LAW, J. K., MARVIN, A. R. & LAW, P. A. 2012. Verification of parent-report of child autism spectrum disorder diagnosis to a web-based autism registry. *Journal of autism and developmental disorders,* 42**,** 257-265.

FLYNN, D., KNOEDLER, M. A., HESS, E. P., MURAD, M. H., ERWIN, P. J., MONTORI, V. M. & THOMSON, R. G. 2012. Engaging patients in health care decisions in the emergency department through shared decision-making: a systematic review. *Academic Emergency Medicine,* 19**,** 959-967.

GAL, E., SELANIKYO, E., EREZ, A. B.-H. & KATZ, N. 2015. Integration in the Vocational World: How Does It Affect Quality of Life and Subjective Well-Being of Young Adults with ASD. *International journal of environmental research and public health,* 12**,** 10820-10832.

GARCIA-VILLAMISAR 2010. Effects of a leisure programme on quality of life and stress of individuals with ASD. *Journal of Intellectual Disability Research,* 54**,** 611-619.

GARCIA-VILLAMISAR, DATTILO & MATSON 2013. Quality of life as a mediator between behavioral challenges and autistic traits for adults with intellectual disabilities. *Research in Autism Spectrum Disorders,* 7**,** 624-629.

HARPER, A. 1998. Development of the World Health Organisation WHOQOL-BREF quality of life assessment. *Psychological medicine,* 28**,** 551-558.

HESSELMARK, E., PLENTY, S. & BEJEROT, S. 2014. Group cognitive behavioural therapy and group recreational activity for adults with autism spectrum disorders: A preliminary randomized controlled trial. *Autism,* 18**,** 672-683.

HONG, J., BISHOP-FITZPATRICK, L., SMITH, L. E., GREENBERG, J. S. & MAILICK, M. R. 2016. Factors Associated with Subjective Quality of Life of Adults with Autism Spectrum Disorder: Self-Report Versus Maternal Reports. *Journal of autism and developmental disorders,* 46**,** 1368-1378.

JENNES-COUSSENS, M., MAGILL-EVANS, J. & KONING, C. 2006. The quality of life of young men with Asperger syndrome - A brief report. *Autism,* 10**,** 403-414.

KAMIO, Y., INADA, N. & KOYAMA, T. 2013. A nationwide survey on quality of life and associated factors of adults with high-functioning autism spectrum disorders. *Autism,* 17**,** 15-26.

KAMP-BECKER, I., SCHRODER, J., REMSCHMIDT, H. & BACHMANN, C. J. 2010. Health-related quality of life in adolescents and young adults with high functioning autism-spectrum disorder. *Psycho-social medicine,* 7.

KATZ, N., DEJAK, I. & GAL, E. 2015. Work performance evaluation and QoL of adults with High Functioning Autism Spectrum Disorders (HFASD). *Work,* 51**,** 887-892.

KHANNA, R., JARIWALA-PARIKH, K., WEST-STRUM, D. & MAHABALESHWARKAR, R. 2014. Health-related quality of life and its determinants among adults with autism. *Research in Autism Spectrum Disorders,* 8**,** 157-167.

LEVISOHN, P. M. 2007. The autism-epilepsy connection. *Epilepsia,* 48**,** 33-35.

LIN, L.-Y. 2014. Quality of life of Taiwanese adults with autism spectrum disorder. *PloS One,* 9.

MOHER, D., LIBERATI, A., TETZLAFF, J. & ALTMAN, D. G. 2009. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *Annals of internal medicine,* 151**,** 264-269.

MUKAETOVA-LADINSKA, E. B., PERRY, E., BARON, M. & POVEY, C. 2012. Ageing in people with autistic spectrum disorder. *International journal of geriatric psychiatry,* 27**,** 109-118.

NICE 2013. NICE Clinical Guideline 170.

PARR, J. R. 2016. How Can We Learn More About the Lives of Adults on the Autism Spectrum from across the age range, and Their Relatives? *In:* WRIGHT, S. (ed.) *Autism Spectrum Disorder in Mid and Later Life.* Jessica Kingsley Publishers.

SALDAÑA, D., ÁLVAREZ, R. M., LOBATÓN, 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**,** 303-316.

SIMONOFF, E., PICKLES, A., CHARMAN, T., CHANDLER, S., LOUCAS, T. & BAIRD, G. 2008. Psychiatric disorders in children with autism spectrum disorders: prevalence, comorbidity, and associated factors in a population-derived sample. *Journal of the American Academy of Child & Adolescent Psychiatry,* 47**,** 921-929.

SIRRIYEH, R., LAWTON, R., GARDNER, P. & ARMITAGE, G. 2012. Reviewing studies with diverse designs: the development and evaluation of a new tool. *Journal of evaluation in clinical practice,* 18**,** 746-752.

STAHLBERG, O., SODERSTROM, H., RASTAM, M. & GILLBERG, C. 2004. Bipolar disorder, schizophrenia, and other psychotic disorders in adults with childhood onset AD/HD and/or autism spectrum disorders. *Journal of neural transmission,* 111**,** 891-902.

TAVERNOR, L., BARRON, E., RODGERS, J. & MCCONACHIE, H. 2013. Finding out what matters: Validity of quality of life measurement in young people with ASD. *Child: Care, Health and Development,* 39**,** 592-601.

TOTSIKA, V., FELCE, D., KERR, M. & HASTINGS, R. P. 2010. Behavior problems, psychiatric symptoms, and quality of life for older adults with intellectual disability with and without autism. *Journal of Autism and Developmental Disorders,* 40**,** 1171-1178.

VAN HEIJST, B. F. C. & GEURTS, H. M. 2015. Quality of life in autism across the lifespan: A meta-analysis. *Autism,* 19**,** 158-167.

WELLS, G., SHEA, B., O'CONNELL, D., PETERSON, J., WELCH, V. & LOSOS, M. 2014. Newcastle-Ottawa Quality Assessment Scale, Cohort Studies.