



UvA-DARE (Digital Academic Repository)

Quality of life in autism across the lifespan: A meta-analysis

van Heijst, B.F.C.; Geurts, H.M.

DOI

[10.1177/1362361313517053](https://doi.org/10.1177/1362361313517053)

Publication date

2015

Document Version

Final published version

Published in

Autism

License

Article 25fa Dutch Copyright Act

[Link to publication](#)

Citation for published version (APA):

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

General rights

It is not permitted to download or to forward/distribute the text or part of it without the consent of the author(s) and/or copyright holder(s), other than for strictly personal, individual use, unless the work is under an open content license (like Creative Commons).

Disclaimer/Complaints regulations

If you believe that digital publication of certain material infringes any of your rights or (privacy) interests, please let the Library know, stating your reasons. In case of a legitimate complaint, the Library will make the material inaccessible and/or remove it from the website. Please Ask the Library: <https://uba.uva.nl/en/contact>, or a letter to: Library of the University of Amsterdam, Secretariat, Singel 425, 1012 WP Amsterdam, The Netherlands. You will be contacted as soon as possible.

Quality of life in autism across the lifespan: A meta-analysis

Autism
2015, Vol. 19(2) 158–167
© The Author(s) 2014
Reprints and permissions:
sagepub.co.uk/journalsPermissions.nav
DOI: 10.1177/1362361313517053
aut.sagepub.com



Barbara FC van Heijst¹ and Hilde M Geurts^{1,2,3}

Abstract

Autism is a lifelong neurodevelopmental disorder, with a known impact on quality of life. Yet the developmental trajectory of quality of life is not well understood. First, the effect of age on quality of life was studied with a meta-analysis. Our meta-analysis included 10 studies (published between 2004 and 2012) with a combined sample size of 486 people with autism and 17,776 controls. Second, as there were no studies on quality of life of the elderly with autism, we conducted an empirical study on quality of life of the elderly (age range 53–83) with autism ($N = 24$) and without autism ($N = 24$). The meta-analysis showed that quality of life is lower for people with autism compared to people without autism, and that the mean effect is large (Cohen's $d = -0.96$). Age did not have an effect on quality of life. The study concerning the elderly with autism showed that the difference in quality of life is similar in the elderly. Age, IQ and symptom severity did not predict quality of life in this sample. Across the lifespan, people with autism experience a much lower quality of life compared to people without autism. Hence, the quality of life seemed to be independent of someone's age.

Keywords

age, autism, meta-analysis, quality of life, review

Introduction

Autism¹ is a lifelong neurodevelopmental disorder, yet the developmental trajectory of people with autism is not well understood. A minority of adults with autism, although continuing to be affected by their autism, can find work, live independently and develop meaningful relationships with others. However, the majority have an overall poor outcome (Billstedt et al., 2005). They remain dependent on their parents or others, are either unemployed or underemployed and lead fairly isolated lives (Howlin et al., 2004, 2013). Moreover, a large number of adults with autism remain without appropriate services and effective interventions (Barnard et al., 2001; Gerhardt and Lainer, 2011). Consequently, many parents of children with autism do not know what to expect for the future of their children. They worry about what will happen to their children when they are not around to care for them anymore, and they fear that the adult services are not as good as those for children (Eaves and Ho, 2008). These factors combined highlight a growing need to describe the developmental trajectory of people with autism, so that the appropriate steps in provision of care can be taken.

The majority of research on outcomes in autism has focused on one or a few domains (e.g. work, friendships) or only on objective measures (e.g. employment status, but

not employment satisfaction). Quality of life (QoL) is a more comprehensive, multidimensional concept that includes subjective well-being, and is well suited to assess people with autism (Burgess and Gutstein, 2007). The World Health Organization (WHO, 1995) defines QoL as the individual's perception of his or her position in life in the context of the culture and value system, and in relation to one's goals, expectations, standards and concerns. It incorporates the individual's physical health, psychological state, level of independence, social relationships, personal beliefs and his or her relationship to salient features of the environment in a complex way. Therefore, the first goal of this study is to investigate the QoL of people with autism.

¹d'Arc, Brain & Cognition, Department of Psychology, University of Amsterdam, The Netherlands

²Dr Leo Kannerhuis, The Netherlands

³Cognitive Science Center Amsterdam, University of Amsterdam, The Netherlands

Corresponding author:

Hilde M Geurts, d'Arc, Brain & Cognition, Department of Psychology, University of Amsterdam, Weesperplein 4, 1018 XA Amsterdam, The Netherlands.

Email: H.M.Geurts@uva.nl

Previous research on unidimensional outcome measures suggests that one of the predictors contributing to better adult outcomes are less severe autism symptoms (Eaves and Ho, 2008; Kuhlthau et al., 2010). Autism symptoms seem to show modest improvement with age (Esbensen et al., 2009; Happé and Charlton, 2012). Age also has an effect on other aspects of autism, as older people have shown fewer sensory abnormalities (Kern et al., 2006) and less maladaptive behaviour (Shattuck et al., 2007) than younger people with autism. However, not all individuals improve. Some people reach a plateau in their development, and others decline (McGovern and Sigman, 2005). In addition, while some symptoms show modest improvements, this seldom leads to functioning in the normal range, which reinforces the notion that autism is a lifelong condition (Seltzer et al., 2004). Therefore, the second goal of this study is to assess the effect of age on QoL.

To our knowledge, there is currently no quantitative review of QoL in autism, but there are two qualitative reviews of QoL in autism by the same authors: Kamp-Becker et al. (2010) – 7 studies, and Kamp-Becker et al. (2011) – 9 studies, of which 5 studies overlap with those of the 2010 paper. The authors did not draw a general conclusion across studies, as between studies there was a wide variety in patient population, design, treatments and outcome measures. However, most studies show that QoL is relatively low in people with autism. Moreover, the effect of age was not studied, despite reports that in the general population QoL decreases with age (StatLine, 2010). Therefore, a direct comparison between the QoL of people with autism and people without autism is needed in order to control for the effects of aging on QoL found in the general population.

In this study, we investigate how the QoL of people with autism can be described over the entire lifespan. In Study 1, we will examine QoL quantitatively by performing a meta-analysis on QoL studies. We will describe the magnitude of the difference between QoL of people (i.e. children, adolescents and adults) with and without autism. As elderly people with autism are likely to be underrepresented in the existing studies (Mukaetova-Ladinska et al., 2012), we conducted Study 2, which will focus on QoL among the elderly with autism. In both studies, we explore the effect of age as well as other factors that influence QoL.

We hypothesize that independent of age, QoL will be lower for people with autism than for people without autism. Although there will be individual differences in people's life trajectory (Seltzer et al., 2004), we hypothesize that age plays a role in QoL, because autism symptoms often reduce with age. As people might learn to compensate, accept impairments and follow successful treatment (García-Villamisar et al., 2002), QoL could improve with age. However, a contrasting hypothesis would be that QoL will decrease with age. Age brings about increased responsibilities and decreased parental support, which may cause impairments to be perceived more intensively in adulthood

than in childhood. There may even be an accumulative impact of autism impairments on finances, health and social life, as was found for attention deficit hyperactivity disorder (ADHD) impairments (Bernardi et al., 2012; Brod et al., 2012). For these reasons, we will explore the direction of the hypothesized age effect.

Methods study I: quantitative review

Literature search

The databases PubMed, PsycINFO and Web of Knowledge were searched for studies (including theses and dissertations) that focus on QoL in autism (up to January 2013). Search terms relating to autism (e.g. *autism*, *autism spectrum disorder (ASD)*, *Asperger*, *pervasive developmental disorder-not otherwise specified (PDD-NOS)*) were combined with QoL and measures of QoL (e.g. *quality of life*, *health-related QoL (HRQOL)*, *World Health Organization Quality of Life-Brief (WHOQOL)*, *PedsQL*). We did not search unpublished work by authors, but cross-references of the obtained studies were checked for studies that might have been missed in the electronic search.

Inclusion criteria

The inclusion criteria were that a study: (a) concerned participants that were diagnosed with autism by clinical consensus (usually verified with standardized instruments); (b) included a self-report or proxy-report measure of QoL; (c) used a standardized, valid and reliable measure of QoL; and (d) addressed a comparison between groups with and without autism. Studies concerning people with autism and a co-morbid disorder were included, as this would increase generalizability.

Obtained studies

The literature search generated 3231 hits; based on titles and abstracts, 42 studies were selected. After full text screening, 28 out of the 42 studies did not meet inclusion criteria. Reasons for excluding studies were that the autism diagnosis was not validated ($N = 3$), the studies used a non-standardized QoL measure ($N = 5$), no control group was included ($N = 17$) or a different type of QoL (family QoL, communication QoL) was assessed ($N = 3$). Of the 14 studies that met inclusion criteria, 9 studies did not include all of the necessary information; authors were contacted and 5 provided the requested information. This resulted in 10 studies available for the meta-analysis, with 486 participants with autism and 17,776 participants without autism (Table 1). The large number of control participants is mainly due to the use of normative comparison groups. All the information in Table 1 was recorded by the first author and verified by the second author.

Table 1. Main characteristics of the studies included in the meta-analysis.

Author	Cohen's <i>d</i>	Variance	Autism <i>N</i>	Control <i>N</i>	Age ^a	Control ^b	Informant	Questionnaire	Diagnosis ^c	Co-morbidity ^d
Bastiaansen et al. (2004)	-0.43	0.04	28	215	9.7 (2.4)	Clinical ^e	Self	PedsQL	CC, SI	NM
Bastiaansen et al. (2004)	-0.38	0.04	28	215	9.7 (2.4)	Clinical ^e	Proxy	PedsQL	CC, SI	NM
Bennet et al. (2005)	-0.24	0.17	9	22	40.7 (9.3)	Learning Disability ^f	Proxy	LEC	CC, Q	NM
Jennes-Coussens et al. (2006)	-0.62	0.18	12	13	20.3 (1.3)	TD	Self	WHOQOL-BREF	CC, Q	NM
Limbers et al. (2009)	-2.26	0.05	22	427	9.3 (2.2)	TD	Proxy	PedsQL	CC, Q	NM
Kamp-Becker et al. (2010)	-0.81	0.05	26	124	21.6 (3.4)	TD	Self	WHOQOL-BREF	CC, SI	M
Kamp-Becker et al. (2011) ^g	-0.20	0.03	40	9947	12.7 (2.6)	TD	Self	ILK	CC, SI	M
Kamp-Becker et al. (2011) ^g	-1.14	0.02	42	1721	12.7 (2.6)	TD	Proxy	ILK	CC, SI	M
Shipman et al. (2011) ^g	-0.65	0.03	39	1170	14.8 (1.9)	TD	Self	PedsQL	CC	NM
Shipman et al. (2011) ^g	-1.69	0.03	39	1384	14.8 (1.9)	TD	Proxy	PedsQL	CC	NM
Tavernor et al. (2012) ^g	-1.81	0.10	10	1033	10.9 (1.4)	TD	Self	PedsQL	CC	NM
Tavernor et al. (2012) ^g	-2.58	0.10	11	427	10.9 (1.4)	TD	Proxy	PedsQL	CC	NM
Kamio et al. (2012) ^g	-0.97	0.01	154	828	27.6 (6.5)	TD	Self	WHOQOL-BREF ^h	CC	M
Cottenceau et al. (2012) ^g	0.34	0.04	26	250	15.0 (2.5)	TD	Self	VSP-A ⁱ	CC	M

ADHD: attention deficit hyperactivity disorder; CC: diagnosis via clinical consensus; ILK: Inventory for the Assessment of Quality of Life in Children and Adolescents; LEC: Life Experiences Checklist; M: mentioned; NM: not mentioned (no full assessment); PedsQL[™]: Pediatric Quality of Life Inventory; Q: questionnaire and rating scales; SI: structured instrument such as structured interviews and observation schedules; TD: typically developing; VSP-A: Experience and Perceived Health for Adolescents; WHOQOL-BREF: World Health Organization Quality of Life abbreviated version.

^aThe age of the autism group.

^bWhen possible a TD control group was chosen as a comparison group.

^cThe autism assessment procedure as described by the authors.

^dHow co-morbidity was addressed.

^eBastiaansen et al. (2004) compared to multiple clinical groups (people with ADHD, anxiety disorders, mood disorders and other disorders). Scores were combined into one clinical group by using the formula for combining groups from the Cochrane handbook (Higgins and Deeks, 2011). The group 'No diagnosis' was neither included as a clinical control group (because no Diagnostic Manual of Statistic Disorders—Fourth Edition (DSM-IV) diagnosis could be made) nor TD control group (because there were problems for which they were referred to mental health services).

^fBennet et al. (2005) had two comparison groups: (1) high scorers on an autism questionnaire, but no autism diagnosis and (2) low scorers on an autism questionnaire and no autism diagnosis. Group 2 was chosen as a comparison group since that group did not have any autism symptomatology.

^gInformation to calculate effect size obtained through personal communication with the author.

^hKamio et al. (2012) measured two subscales from the WHOQOL-BREF: psychological health and social relationships. To calculate the effect size psychological health was chosen, because this subscale consists of more items, and has a high test reliability, internal consistency, construct validity and a strong correlation with, and high factor loading on, overall QoL (Skevington et al., 2004; Wang et al., 2006).

ⁱCottenceau et al. (2012) calculated two global scores from the VSP-A: Index 2 without 'relationships with medical staff' and 'affective and sexual life' dimensions and Index 4 without 'relationships with medical staff' but with the 'affective and sexual life' dimension. To calculate an effect size, Index 4 was chosen since this included more dimensions.

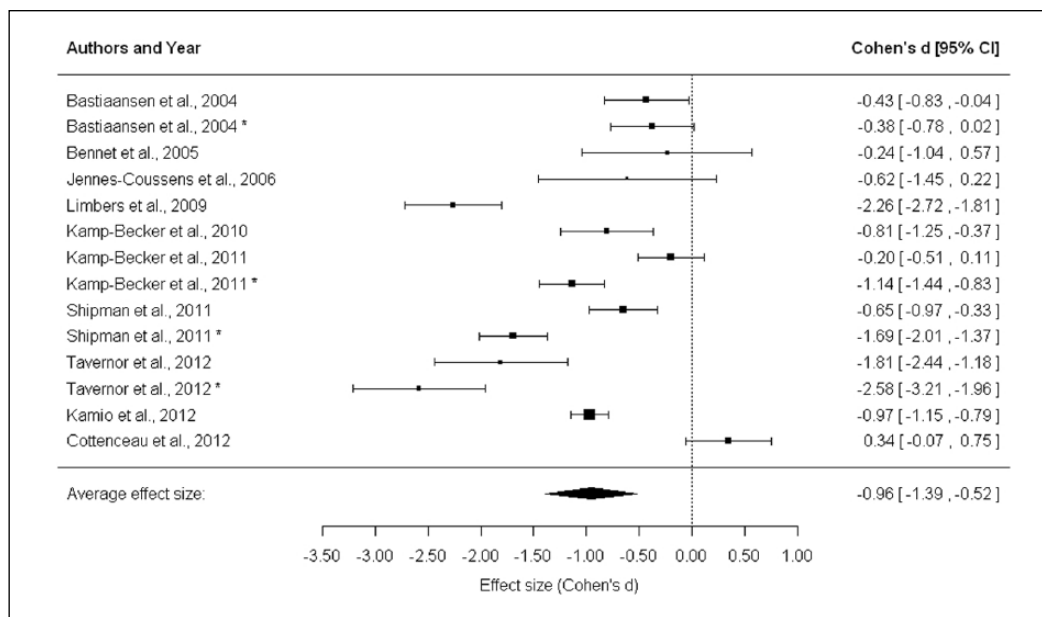


Figure 1. Forest plot of the standardized mean difference (Cohen's d) and 95% confidence interval of QoL. Negative effect sizes imply that the QoL is lower in people with autism as compared to controls while positive effect sizes suggest the opposite pattern. *Effect sizes were calculated separately for self-report and proxy-report. Thus, 14 effect sizes were calculated from 10 studies.

Independent variables

For each study, the age of the autism participants was coded as a continuous variable. The following categorical variables were also coded: the respondent to the QoL questionnaire (self-report or proxy-report), the comparison group (typically developing (TD) or clinical control group) and the QoL questionnaire utilized (WHOQOL-BREF, Pediatric Quality of Life Inventory (PedsQL) or other). The study characteristics served as the independent variables in order to determine whether these characteristics moderated the effect size.

Dependent variable

The QoL questionnaires generated continuous outcomes, for which a standardized mean difference (Cohen's d) was calculated. This effect size is widely used, is easily interpretable and can be calculated from t -test statistics (Thalheimer and Cook, 2002; Turner and Bernard, 2006).

Data analyses

The data were analysed using the Metafor package for R (Viechtbauer, 2010). A random effects meta-analysis was performed to provide an average effect for the population of studies, as the included studies were assumed to be a random selection of the entire study population. We conducted a mixed effects meta-analysis to determine the moderating effects of the study characteristics. Effect sizes were regressed on the study characteristics in a restricted maximum likelihood meta-regression (Viechtbauer, 2010).

We tested the heterogeneity with conventional Q tests and with the I^2 statistic. The I^2 statistic estimates how much of the total variability in the effect size estimates (composed of heterogeneity and sampling variability) can be attributed to heterogeneity among the true effects. We checked for publication bias with funnel plots and the fail-safe analysis (Rosenthal, 1979).

Results study I

Overall result

Based on our qualitative analysis, there were many differences in QoL between people with and without autism. The most affected domain of QoL seemed to be social functioning (Bastiaansen et al., 2004; Jennes-Coussens et al., 2006; Kamio et al., 2012; Kamp-Becker et al., 2010). Other relevant reported findings were that the autism group had fewer friends and more special education (Bastiaansen et al., 2004); lower physical health (Jennes-Coussens et al., 2006); lower QoL relating to relationships with friends, leisure, affective and sexual relationships; and placed less importance on activities with peers and more importance on activities with parents (Cottenceau et al., 2012).

Our quantitative analysis showed that the mean effect size is large, $d = -0.96$; 95% confidence interval = $-1.39, -0.52$; range = -2.58 to 0.34 (Figure 1). People with autism had a lower QoL than people without autism, $z = -4.29$, $p < 0.001$. There was significant heterogeneity between the effect sizes, $\tau^2 = 0.63$, $Q(13) = 168.80$, $p <$

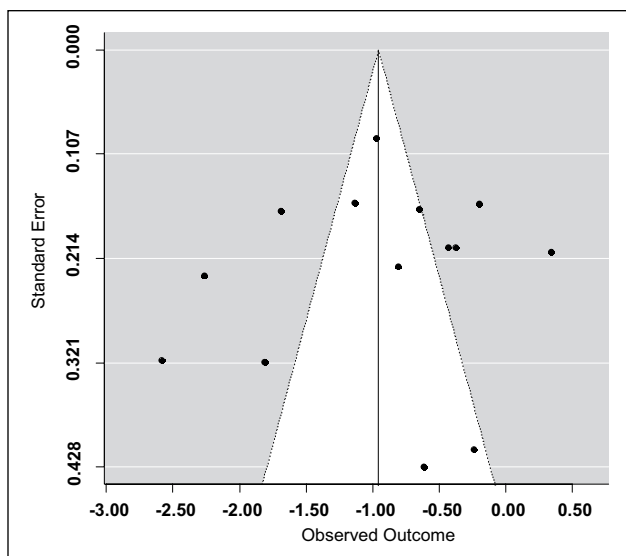


Figure 2. Funnel plot of effect sizes (Cohen's d) against the standard errors. The dots represent the studies included in the analysis.

0.001, $I^2 = 94.6\%$. Leave-one-out sensitivity analyses indicated that the effect remained significant regardless of which study was dropped (Cohen's d ranged from -1.06 to -0.84 , all $p < 0.001$). A QQ-plot confirms that there is a normal distribution of effect sizes.

Visual inspection and a regression test for funnel plot asymmetry indicated no asymmetry in the funnel plot ($z = -0.40$, $p = 0.696$; Figure 2). As the number of effect sizes is 14, the fail-safe N should be greater than $14 \times 5 + 10 = 80$, that is, the fail-safe $N = 1544$. Thus, there is no indication of publication bias.

Moderator analysis

At least part of the heterogeneity in the model may be due to the influence of moderators. First, age was added into the model but this did not have a moderating effect, $p = 0.265$. Second, we added control group, questionnaire and respondent to the model to explore whether these would moderate the effect size. The omnibus test was significant, $Q_M(5) = 28.46$, $p < 0.001$, and the three moderators were significant (control group, $p = 0.002$; questionnaire, $p = 0.005$; respondent, $p = 0.004$). The estimated amount of residual heterogeneity was equal to $\tau^2 = 0.19$, suggesting that $(0.63 - 0.19)/0.63 = 69.8\%$ of the total heterogeneity could be accounted for by the included moderators. There was still a significant amount of residual heterogeneity, $Q_E(8) = 37.44$, $p < 0.001$, indicating that study characteristics not considered in the model also moderate the effect size.

Further inspection of the moderators with univariate analyses revealed that there was no significant difference in effect size between studies that compared people with autism to a TD or clinical group ($p = 0.153$). There was no

difference in effect size between studies that used the WHOQOL-BREF questionnaire and those that used the PedsQL, $p = 0.446$. Studies concerning a self-report ($d = -0.63$, $p = 0.018$) and studies concerning a proxy-report ($d = -1.39$, $p < 0.001$) both showed a difference in QoL between people with and without autism. The difference between these two estimates approached significance, $z = -1.87$, $p = 0.062$, indicating that a larger difference in effect size with a proxy informant as compared to self-reports made by people with autism. As five of the six studies that included a proxy informant had young participants (aged 9.3–14.8 years old), we tested for an interaction between age and respondent, but this was not significant. Hence, the moderator analyses suggest that when taken together, control group, questionnaire and respondent moderate the effect size, but only the respondent is a marginal moderator when other moderators are not considered.

Discussion study I

The meta-analysis shows a large difference in QoL between people with and without autism. Age does not have a moderating effect, which might be due to the mean age (range 9.3–40.7) of the participants; the elderly with autism were underrepresented in the meta-analysis.

Studies that assess QoL with different types of questionnaires do not produce different results, which indicates a high comparability between the various QoL measures. Studies that compared to either a TD or a clinical control group do not produce different results; however, only three studies included a clinical control group.

The informant was found to be the only factor that may influence QoL. Parents rate the QoL of their children with autism lower than the children themselves do (Bastiaansen et al., 2004; Kamp-Becker et al., 2011). There may be several reasons for this discrepancy. First, parents of children with health concerns generally report a lower QoL than children themselves (Upton et al., 2008), and there is a greater agreement for observable (e.g. physical) functioning than for non-observable (e.g. emotional or social QoL) functioning (Eiser and Morse, 2001). Since social impairments form a substantial part of the autism symptomatology, this may explain the discrepancy between self- and proxy-report. Second, the discrepancy may be due to the difficulties that people with autism have when reporting on their own QoL. They may have a deficit in self-representation that affects the identification of emotional states in others as well as in themselves (Johnson et al., 2009; Lombardo et al., 2007). In addition, they may lack the emotional vocabulary, which could affect their ability to respond to items about their mood and feelings (Tavernor et al., 2012). However, this is debatable as it has been reported that self-reports of QoL from children with autism are valid and reliable (Shipman et al., 2011), and that

Table 2. Group means and standard deviations for age, gender, estimated IQ and Social Responsiveness Scale Adult version score.

Variable	Groups		Statistics
	Autism (<i>N</i> = 24)	Control (<i>N</i> = 24)	
Age	63.7 (7.4) Range 53–83	63.5 (8.0) Range 51–84	$F(1,46) = 0.01, p = 0.911, \eta^2 = 0.00$
Gender	19M, 5F	18M, 6F	$\chi^2(1, N = 48) = .12, p = .731$
IQ	109.5 (10.3)	109.6 (7.8)	$F(1,45) = 0.001, p = 0.982, \eta^2 = 0.00$
SRS-A	94.9 (26.1)	25.7 (11.2)	$F(1,46) = 143.11, p < 0.001, \eta^2 = 0.76$

SRS-A: Social Responsiveness Scale Adult version.

children with autism have sufficient ability to rate their emotions appropriately (Hobson et al., 2006). Third, children may perceive their limited social and emotional functioning as a less severe problem, as compared to their parents. This is illustrated by a quote of a parent: ‘I answered the question producing negative scores, but upon reflection I think my child actually prefers being alone’ (Tavernor et al., 2012). Therefore, the discrepancy between parent- and self-report seems to be derived from a perceptual difference concerning children’s activities and appraisals, and not from a lack of parental knowledge. When parents are asked to respond as they think their child would, the correlation with self-report increases (Sheldrick et al., 2012). In conclusion, disagreement between parent’s and children’s reports of QoL is, in itself, unlikely to indicate that either is wrong or right, but rather is a consequence of each individual’s beliefs about the child’s health and well-being. Parent-reports, as an addition to self-reports, can provide another perspective, and thus, produce a greater understanding of QoL in autism.

Study 2: new data on QoL in the elderly with autism

One shortcoming observed in the meta-analysis was the lack of studies that assessed QoL in elderly with autism. Therefore, in Study 2 we conducted an empirical study in order to focus on QoL in the elderly with autism. Moreover, there was a large amount of unexplained heterogeneity in the meta-analysis even after including several potentially important moderators (control group, questionnaire and informant). Unfortunately, information regarding the participants’ symptom severity (Eaves and Ho, 2008; Kuhlthau et al., 2010), IQ (Howlin et al., 2004; McGovern and Sigman, 2005) and early language development (Billstedt et al., 2005; Szatmari et al., 2003) were too sparse to include in the meta-analysis, while these participant characteristics are relevant predictors of adult outcome. Hence, in Study 2 we assessed symptom severity and intelligence in relation to QoL. Moreover, two more moderators of QoL were explored. First, we know that QoL is affected in early stages of cognitive decline (Bárrios et al., 2013); therefore, we investigated whether regular cognitive mistakes influence QoL in the elderly with

autism. Second, we explored whether psychological problems related to psychopathology also influence QoL of the elderly with autism because psychological health is an important part of QoL (WHO, 1995) and co-morbid psychopathology is common among people with autism (Geurts and Jansen, 2012).

Methods Study 2

Participants

The autism group consists of intellectually able elderly with autism (*N* = 24), who were diagnosed with autism in their adult life. Individuals were included if they had received a diagnosis of autism via clinical consensus, had a high score (≥ 60) on the Social Responsiveness Scale Adult version (SRS-A; Noens et al., 2012) or if they met both criteria. Most of the participants were included in a previously published study on cognitive functioning in the elderly with autism (Geurts and Vissers, 2012). We included one additional participant who filled out the QoL questionnaire but who was not tested for the original study.

TD controls (*N* = 24) were included if they did not have autism (based on self-report and SRS-A scores < 60) or any other psychiatric disorder, and did not have any relatives diagnosed with autism. For more details about the participants, see Table 2.

Materials

Quality of life. There is currently no instrument available to measure QoL specifically in people with autism. A benefit of a generic measure of QoL, such as the RAND-36 (Van der Zee and Sanderman, 1993), is that it is easier to compare across groups of people. The RAND-36 (also known as SF-36) is a brief and comprehensive 36-item self-report health-related QoL questionnaire. It includes eight of the most frequently measured health concepts: physical functioning, role limitations caused by physical health problems, role limitations caused by emotional problems, social functioning, emotional well-being, energy/fatigue, pain and general health perceptions (Ware, 2000). The Dutch manual of the RAND-36 (Van der Zee and Sanderman, 1993) was used to calculate the total scores. The total score is the mean of the aforementioned eight scale scores.

A higher score indicates a better QoL. Van der Zee and Sanderman (1993) conclude that the RAND-36 is a valid, reliable and sensitive measurement of QoL. Internal consistency for each of the scale scores range between $\alpha = 0.71$ and 0.92 (mean is 0.84).

Verbal intelligence. The Dutch Adult Reading Test (DART; Schmand et al., 1992) is a list of 50 words with irregular spelling that are to be read aloud. The number of correctly pronounced words was used to calculate a DART-IQ score. It is relatively insensitive to cognitive impairment caused by neurological disorders and is a strong predictor of intelligence (Schmand et al., 1998). The psychometric properties are excellent as the interrater reliability is 0.96 and the internal consistency is 0.91 (Schmand et al., 1992).

Symptom severity. The SRS-A (Noens et al., 2012) is a 64-item self-report rating scale that serves as an index of severity of deficits in the autism spectrum. Five domains were assessed: social awareness, social information processing, capacity for reciprocal social communication, social anxiety/avoidance and autistic preoccupations and mannerisms. This scale has excellent internal consistency, $\alpha = 0.94$ – 0.95 (Constantino and Todd, 2005).

Psychological problems. The symptom checklist (SCL-90; Arrindell and Ettema, 1986) is a widely used multidimensional 90-item self-report symptom inventory that screens for a broad range of psychological problems. Subscales are anxiety, agoraphobia, depression, somatisation, insufficiency of thought and behaviour, distrust and interpersonal sensitivity, hostility and sleeping problems. The SCL-90 has good test–retest reliability, internal consistency and concurrent and discriminant validity (Derogatis, 1977).

Cognitive problems. The Cognitive Failures Questionnaire (CFQ; Broadbent et al., 1982; Ponds et al., 1997) is a 25-item self-report questionnaire that assesses common types of cognitive mistakes in perception, memory and motor function (e.g. misplacing one's keys or forgetting appointments). Internal consistency is 0.91 , and test–retest reliability over a 2-month period is 0.82 (Vom Hofe et al., 1998).

Of the aforementioned materials, the SRS-A was specifically designed for an autism population, and the SCL-90 (Spek et al., 2013) has been used in an autism population before. The RAND-36, DART and CFQ have not previously been used in an autism population.

Data analyses

First, a univariate analysis of variance was conducted to determine the difference in QoL between people with and without autism. Next, we combined the autism and TD

groups in order to conduct a linear regression analysis. Age, IQ and symptom severity were tested as potential predictors of QoL. Exploratory regression analyses on the combined group were performed to test interaction effects of age, IQ and symptom severity with group, as well as the predictive powers of psychological and cognitive problems on QoL.

A boxplot revealed three outliers in the TD control group that had low scores on the RAND-36 (more than $1.5\times$ the interquartile range (IQR)). All analyses were performed with and without these outliers. When there was no difference in the pattern of findings, we reported the results based on the full sample. When a difference was found, we reported the results of both analyses.

Results Study 2

Main outcome

The autism group reported a lower QoL ($M = 66.7$, $SD = 16.6$) compared to the TD group ($M = 82.0$, $SD = 13.2$), $F(1,46) = 12.5$, $p = 0.001$, $\eta^2 = 0.21$. To test whether age, IQ and symptom severity were predictors of QoL, a regression analysis was conducted. Results showed that the model was significant, $F(3,43) = 4.06$, $p = 0.013$, $\eta^2 = 0.17$. Age and IQ were not significant, but symptom severity did significantly predict QoL, $\beta = -0.44$, $p = 0.002$, with more symptoms relating to a lower QoL. However, all the individuals in the autism group had more severe autism symptoms than the TD control group, and so this seems to be an effect of group. An additional regression analysis was run on only the autism group; this showed that for people with autism, symptom severity did not predict QoL, $F(1,22) = 0.044$, $p = 0.836$.

Exploratory regression analyses

Exploratory regression analyses were performed to determine whether age, IQ and symptom severity had a different impact on QoL within the two groups. There were neither interaction effects nor main effects of age, IQ or symptom severity. The effect of group was present in most models (results can be obtained from the first author).

Furthermore, we explored whether cognitive problems and psychological problems were potential predictors of QoL. The direct group comparison showed that the autism group reported more cognitive problems ($M = 45.1$, $SD = 13.8$) than the TD group ($M = 30.9$, $SD = 11.5$), $F(1,46) = 15.1$, $p < 0.001$, $\eta^2 = 0.25$. Moreover, the autism group reported more psychological problems ($M = 166.3$, $SD = 36.7$) than the TD group ($M = 120.2$, $SD = 42.5$), $F(1,46) = 16.2$, $p < 0.001$, $\eta^2 = 0.26$. The regression analysis showed that there were main effects for cognitive problems and psychological problems, but these effects disappeared when outliers were removed. This suggests that these effects were due to influential outlying data points.

Inclusion of Study 2 in the meta-analysis

As this study provides unique information about QoL in the elderly with autism (Cohen's $d = -1.02$, variance 0.10), it was also included in the meta-analysis; this did not significantly change any of the findings reported in Study 1.

Discussion Study 2

This study shows a large difference in QoL between people with and without autism. Factors that are likely to have an effect on QoL – intelligence, symptom severity and language development – could not be studied in the meta-analysis due to the lack of information in the included studies. Fortunately, some insight into these factors could be gained from Study 2. Intelligence – as estimated by the DART-IQ – was not found to have an effect on QoL. It should be noted that all participants were intellectually able (IQ range 81–130), and the effect of intelligence may be more pronounced in the lower ranges (Howlin et al., 2004; McGovern and Sigman, 2005). Severity of symptoms did not influence QoL, which is contradictory to previous reports (Eaves and Ho, 2008; Kuhlthau et al., 2010). This might be due to the small sample size that resulted in insufficient power to detect such an effect. However, an alternative interpretation could be that among the elderly, symptom severity may no longer be a relevant factor. Unfortunately, there was no information on language development in Study 2. For the elderly, it is often not feasible to recover information about language development in childhood. However, this is still a factor that, when feasible, should be studied. The exploratory analysis showed that psychological problems or cognitive problems did not contribute to QoL over and above the effect of group.

General discussion

Our main aim was to test if there was a difference in QoL between people with and without autism and to explore the factors that influence QoL. The meta-analysis shows that there is a large difference in QoL between people with and without autism. This effect was observed in children, adolescents, adults and the elderly.

The hypothesis that age affects QoL is not supported by the present results. This indicates that individual differences seem to be more prominent than an overall trend. This is in line with previous research that also found no associations between QoL and age (Kamio et al., 2012; Kamp-Becker et al., 2011). To the contrary, Bennet et al. (2005) reported that increasing age was associated with decreasing QoL. However, they did not include a subjective QoL assessment, and their results could reflect deteriorating circumstances, and not necessarily life satisfaction. When focussing solely on relationships with friends (one aspect of QoL), QoL did seem to improve with age in a

sample aged 10 to 19 years old (Cottenceau et al., 2012). This coincides with the finding that adolescents improve in the reciprocal social interaction domain (Seltzer et al., 2003). However, overall there is no substantial evidence of QoL improving or deteriorating over the lifespan. This lack of an age effect should be interpreted with caution; the number of included studies in the meta-analysis is relatively small, and the distribution of ages is skewed towards young people. While Study 2 was a good addition to the meta-analysis, the number of studies concerning adults and the elderly remained small, and thus the effect of age could not be studied optimally.

Several limitations might complicate the interpretation of our findings. The participants in Study 2 had an estimated IQ in the normal range and received the autism diagnosis in their adult life. Therefore, they may not be representative of the whole autism population. Another caveat is the small amount of studies in the meta-analysis. There was a sufficient amount of studies to conduct a meta-analysis, but unfortunately, another 17 studies with valuable information about QoL in autism could not be included because they did include a control group. These studies describe QoL, or they use QoL to measure treatment efficacy. Including a control group in these types of studies would generate a wealth of relevant information regarding QoL in people with autism. We also recommend including measurements that assess variables that might influence QoL because in the current meta-analysis much heterogeneity was left unexplained. As IQ, symptom severity and language development are thought to be important factors in adult outcome, it is recommended that future studies on QoL at least take these into account, so it can be determined whether these factors indeed explain some of the heterogeneity. Moreover, a longitudinal study to examine the age factor would be ideal, but a well-balanced cross-sectional study could already improve our understanding of the developmental trajectory of QoL in people with autism.

The large difference in QoL between people with and without autism warrants that much work needs to be done to help people with autism reach a higher level of QoL. Environmental factors and treatment can contribute to this. A higher QoL is related to early diagnosis (Kamio et al., 2012) and a larger amount of perceived support (Renty and Roeyers, 2006). QoL improves with supported employment (García-Villamizar et al., 2002), residential (Gerber et al., 2011) and leisure programmes (García-Villamizar and Dattilo, 2010). Hence, it is important that organizations that care for people with autism take all of the above into account so that the QoL of people with autism can be improved.

The meta-analysis also shows that the focus of research is on young people (see also Mukaetova-Ladinska et al., 2012). For a comprehensive picture of autism, more attention should be paid to adults and the elderly. In return, this can also lead to valuable information for young people with autism. A

better understanding of the developmental trajectory of autism means that interventions can be adjusted to the specific needs of people with autism across the lifespan.

Acknowledgements

We would like to thank Anika Bexkens and Laura Ruzzano for their statistical advice regarding the meta-analysis. Moreover, we also thank Laura Ruzzano for her helpful grammar-related comments.

Funding

This work is part of the research program “Autism and Aging: A Double Jeopardy”, which is financed (VIDI grant number 452-10-003) by the Netherlands Organization for Scientific Research (NWO).

Note

1. Autism is used here as a term for the entire autism spectrum, consisting of autistic disorder, Asperger syndrome and pervasive developmental disorder—not otherwise specified (PDD-NOS).

References

- Arrindell WA and Ettema JHM (1986) *SCL-90: Handleiding bij een multidimensionele psychopathologie-indicator*. Lisse: Swets & Zeitlinger BV.
- Barnard J, Harvey V, Potter D, et al. (2001) *Ignored or Ineligible? The Reality for Adults with Autism Spectrum Disorders*. London: NAS Publications.
- Bárrios H, Narcico S, Guerreiro M, et al. (2013) Quality of life in patients with mild cognitive impairment. *Aging & Mental Health* 17: 287–292.
- Bastiaansen D, Koot HM, Ferdinand RF, et al. (2004) Quality of life in children with psychiatric disorders: self-, parent, and clinician report. *Journal of the American Academy of Child and Adolescent Psychiatry* 43: 221–230.
- Bennet HE, Wood CL and Hare DJ (2005) Providing care for adults with autistic spectrum disorders in learning disability services: needs-based or diagnosis-driven? *Journal of Applied Research in Intellectual Disabilities* 18: 57–64.
- Bernardi S, Faraone SV, Cortese S, et al. (2012) The lifetime impact of attention deficit hyperactivity disorder: results from the national epidemiologic survey on alcohol and related conditions (NESARC). *Psychological Medicine* 42: 875–887.
- Billstedt E, Gillberg C and 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: 351–360.
- Broadbent DE, Cooper PF, Fitzgerald P, et al. (1982) The Cognitive Failures Questionnaire (CFQ) and its correlates. *The British Journal of Clinical Psychology* 21: 1–16.
- Brod M, Schmitt E, Goodwin M, et al. (2012) ADHD burden of illness in older adults: a life course perspective. *Quality of Life Research* 21: 795–799.
- Burgess AF and Gutstein SE (2007) Quality of life for people with autism: raising the standard for evaluating successful outcomes. *Child and Adolescent Mental Health* 12: 80–86.
- Constantino JN and Todd RD (2005) Intergenerational transmission of subthreshold autistic traits in the general population. *Biological Psychiatry* 57: 655–660.
- Cottenceau H, Roux S, Blanc R, et al. (2012) Quality of life of adolescents with autism spectrum disorders: comparison to adolescents with diabetes. *European Child & Adolescent Psychiatry* 21: 289–296.
- Derogatis LR (1977) *SCL-90: Administration, Scoring and Procedure Manual-I for the R (Revised) Version*. Baltimore, MD: The Johns Hopkins University School of Medicine.
- Eaves LC and Ho HH (2008) Young adult outcome of autism spectrum disorders. *Journal of Autism and Developmental Disorders* 38: 739–747.
- Eiser C and Morse R (2001) Can parents rate their child’s health-related quality of life? Results of a systematic review. *Quality of Life Research* 10: 347–357.
- Esbensen AJ, Seltzer MM, Lam KSL, et al. (2009) Age-related differences in restricted repetitive behaviors in autism spectrum disorders. *Journal of Autism and Developmental Disorders* 39: 57–66.
- García-Villamizar D, Wehman P and Navarro MD (2002) Changes in the quality of autistic people’s life that work in supported and sheltered employment: a 5-year follow-up study. *Journal of Vocational Rehabilitation* 17: 309–312.
- García-Villamizar DA and Dattilo J (2010) Effects of a leisure programme on quality of life and stress of individuals with ASD. *Journal of Intellectual Disability Research* 54: 611–619.
- Gerber F, Bessero S, Robbani B, et al. (2011) Comparing residential programmes for adults with autism spectrum disorders and intellectual disability: outcomes of challenging behaviour and quality of life. *Journal of Intellectual Disability Research* 55: 918–932.
- Gerhardt PF and Lainer I (2011) Addressing the needs of adolescents and adults with autism: a crisis on the horizon. *Journal of Contemporary Psychotherapy* 41: 37–45.
- Geurts HM and Jansen MD (2012) A retrospective chart study: the pathway to a diagnosis for adults referred for ASD assessment. *Autism* 16: 299–305.
- Geurts HM and Vissers ME (2012) Elderly with autism: executive functions and memory. *Journal of Autism and Developmental Disorders* 42: 665–675.
- Happé F and Charlton RA (2012) Aging in autism spectrum disorders: a mini-review. *Gerontology* 58: 70–78.
- Higgins JPT and Deeks JJ (2011) Chapter 7. Selecting studies and collecting data. In: Higgins JPT and Green S (eds) *Cochrane Handbook for Systematic Reviews of Interventions Version 5.1.0*. paragraph 7.7.3.8, table 7.7.a. Available at: <http://handbook.cochrane.org/>.
- Hobson PR, Chidambi G, Lee A, et al. (2006) Foundations for self-awareness: an exploration through autism. *Monographs of the Society for Research in Child Development* 71: vii–166.
- Howlin P, Goode S, Hutton J, et al. (2004) Adult outcome for children with autism. *Journal of Child Psychology and Psychiatry* 45: 212–229.
- Howlin P, Savage S, Moss P, et al. (2013) Cognitive and language skills in adults with autism: a 40-year follow-up. *Journal of Child Psychology and Psychiatry*. Epub ahead of print 15 July. DOI: 10.1111/jcpp.12115.
- Jennes-Coussens M, Magill-Evans J and Koning C (2006) The quality of life of young men with Asperger syndrome: a brief report. *Autism* 10: 403–414.
- Johnson SA, Filiter JH and Murphy RR (2009) Discrepancies between self- and parent- perceptions of autistic traits and empathy in high functioning children and adolescents on

- the autism spectrum. *Journal of Autism and Developmental Disorders* 39: 1706–1714.
- Kamio Y, Inada N and Koyama T (2012) 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, Schröder J, Muehlan H, et al. (2011) Health-related quality of life in children and adolescents with autism spectrum disorder. *Zeitschrift für Kinder- und Jugendpsychiatrie und Psychotherapie* 39: 123–131.
- Kamp-Becker I, Schröder J, Remschmidt H, et al. (2010) Health-related quality of life in adolescents and young adults with high functioning autism-spectrum disorder. *GMS Psycho-Social-Medicine* 7: Doc03.
- Kern JK, Trivedi MH, Garver CR, et al. (2006) The pattern of sensory processing abnormalities in autism. *Autism* 10: 480–494.
- Kuhlthau K, Orlich F, Hall TA, et al. (2010) Health-related quality of life in children with autism spectrum disorders: results from the autism treatment network. *Journal of Autism and Developmental Disorders* 40: 721–729.
- Limbers CA, Heffer RW and Varni JW (2009) Health-related quality of life and cognitive functioning from the perspective of parents of school-aged children with Asperger's syndrome utilizing the PedsQL™. *Journal of Autism and Developmental Disorders* 39: 1529–1541.
- Lombardo MV, Barnes JL, Wheelwright SJ, et al. (2007) Self-referential cognition and empathy in autism. *PLoS One* 2: e833.
- McGovern CW and Sigman M (2005) Continuity and change from early childhood to adolescence in autism. *Journal of Child Psychology and Psychiatry* 46: 401–408.
- Mukaetova-Ladinska EB, Perry E, Baron M, et al. (2012) Ageing in people with autistic spectrum disorder. *International Journal of Geriatric Psychiatry* 27: 109–118.
- Noens I, De la Marche W and Scholte E (2012) *SRS-A: Screeningslijst voor autismspectrumstoornissen bij volwassenen Handleiding*. Amsterdam: Hogrefe.
- Ponds RW, Commissaris KJ and Jolles J (1997) Prevalence and covariates of subjective forgetfulness in a normal population in The Netherlands. *International Journal of Aging and Human Development* 45: 207–221.
- Renty J and Roeyers H (2006) Quality of life in high-functioning adults with autism spectrum disorder: the predictive value of disability and support characteristics. *Autism* 10: 511–524.
- Rosenthal R (1979) The “file drawer problem” and tolerance for null results. *Psychological Bulletin* 86: 638–641.
- Schmand B, Geerlings MI, Jonker C, et al. (1998) Reading ability as an estimator of premorbid intelligence: does it remain stable in emergent dementia? *Journal of Clinical and Experimental Neuropsychology* 20: 42–51.
- Schmand B, Lindeboom J and Van Harskamp F (1992) *Nederlandse leestest voor Volwassenen*. Lisse: Swets & Zeitlinger BV.
- Seltzer MM, Krauss MW, Shattuck PT, et al. (2003) The symptoms of autism spectrum disorders in adolescence and adulthood. *Journal of Autism and Developmental Disorders* 33: 565–581.
- Seltzer MM, Shattuck P, Abbeduto L, et al. (2004) Trajectory of development in adolescents and adults with autism. *Mental Retardation and Developmental Disabilities Research Reviews* 10: 234–247.
- Shattuck PT, Seltzer MM, Greenberg JS, et al. (2007) Change in autism symptoms and maladaptive behaviors in adolescents and adults with an autism spectrum disorder. *Journal of Autism and Developmental Disorders* 37: 1735–1747.
- Sheldrick RC, Neger EN, Shipman D, et al. (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: 53–57.
- Shipman DL, Sheldrick RC and Perrin EC (2011) Quality of life in adolescents with autism spectrum disorders: reliability and validity of self-reports. *Journal of Developmental and Behavioral Pediatrics* 32: 85–89.
- Skevington SM, Loftly M and O'Connell KA (2004) The World Health Organization's WHOQOL-BREF quality of life assessment: psychometric properties and results of the international field trial – a report from the WHOQOL Group. *Quality of Life Research* 13: 299–310.
- Spek AA, Van Ham NC and Nyklíček I (2013) Mindfulness-based therapy in adults with an autism spectrum disorder: a randomized controlled trial. *Research in Developmental Disabilities* 34: 246–253.
- StatLine (2010) *Gezondheid en Aandoeningen: generieke gezondheidsmaat. Gezondheid, leefstijl, zorggebruik; t/m 2009*. Den Haag/Heerlen: CBS. Available at: <http://statline.cbs.nl/StatWeb/publication/?DM=SLNL&PA=03799&D1=152-153,241&D2=0-17&D3=0&D4=a&HDR=G2,T,G3&STB=G1&P=P&VW=T>
- Szatmari P, Bryson SE, Boyle MH, et al. (2003) Predictors of outcome among high functioning children with autism and Asperger syndrome. *Journal of Child Psychology and Psychiatry* 44: 520–528.
- Tavernor L, Barron E, Rodgers J, et al. (2012) Finding out what matters: validity of quality of life measurement in young people with ASD. *Child: Care, Health and Development*. Epub ahead of print 20 April. DOI: 10.1111/j.1365-2214.2012.01377.x.
- Thalheimer W and Cook S (2002) How to calculate effect sizes from published research articles: a simplified methodology. Available at: http://education.gsu.edu/coshima/EPRS8530/Effect_Sizes_pdf4.pdf
- Turner HM and Bernard RM (2006) Calculating and synthesizing effect sizes. *Contemporary Issues in Communication Science and Disorders* 33: 42–55.
- Upton P, Lawford J and Eiser C (2008) Parent-child agreement across child health-related quality of life instruments: a review of the literature. *Quality of Life Research* 17: 895–913.
- Van der Zee KI and Sanderman R (1993) *Het meten van de gezondheidstoestand met de RAND-36: Een handleiding*. Groningen: Noordelijk Centrum voor gezondheidsvraagstukken.
- Viechtbauer W (2010) Conducting meta-analyses in R with the metafor package. *Journal of Statistical Software* 36: 1–48.
- Vom Hofe A, Mainemarre G and Vannier L (1998) Sensitivity to everyday failures and cognitive inhibition: are they related? *European Review of Applied Psychology* 48: 49–55.
- Wang WC, Yao G, Tsai YJ, et al. (2006) Validating, improving reliability, and estimating correlation of the four subscales in the WHOQOL-BREF using multidimensional Rasch analysis. *Quality of Life Research* 15: 607–620.
- Ware JE (2000) SF-36 health survey update. *Spine* 25: 3130–3139.
- World Health Organization (WHO) (1995) World Health Organization Quality of Life Assessment (WHOQOL): position paper from the World Health Organization. *Social Science & Medicine* 41: 1403–1409.