



Objective and Subjective Measurement of Alexithymia in Adults with Autism

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Abstract

High rates of alexithymia, a condition characterised by difficulties identifying and describing emotions, are frequently reported in both children and adults with autism spectrum disorder (ASD). However, the dilemma of measuring alexithymia via self-report has rarely been addressed. In this study, we compared objective and subjective measures of alexithymia in adults with ASD and typically developing adults. We found significantly higher levels of alexithymia in the ASD sample as measured by both self-report on the Toronto Alexithymia Scale (TAS-20) and by the observer rated Alexithymia Provoked Response Scale (APRQ). However, the two measures did not correlate with each other. We explore the different facets of the alexithymia construct that these two measures may be distinguishing.

Keywords Autism spectrum disorder · Alexithymia · Subjective · Self-report · Objective · Observer-rated

Introduction

Alexithymia, a term first coined by Sifneos in 1973, literally means a lack of words for emotions. Subsequent studies have expanded and elaborated the alexithymia construct as being multi-faceted (Bagby et al. 2009), encompassing a reduced ability to consciously experience, identify, label and describe one's emotional experiences and to discriminate between internal states and bodily sensations. It also includes a reduced fantasy life and an externally orientated cognitive style (Apfel and Sifneos 1979; Hobson et al. 2019; Krystal 1979).

Early work on alexithymia focused on its manifestation in somatic difficulties (Taylor et al. 1992), reactions to trauma (Krystal and Krystal 2015) and substance misuse (Haviland et al. 1994). However, Bird and Cook (2013) proposed that underlying alexithymia may play an explanatory role in the inconsistent findings observed in participants with autism spectrum disorder (ASD) in studies of emotion recognition and emotion processing. Evidence has mounted

that alexithymia is a common co-occurring, sub-clinical condition, with high prevalence rates among children and adults with ASD (Berthoz and Hill 2005; Hill et al. 2004; Milosavljevic et al. 2016; Tani et al. 2004) and as an alternative explanation for the emotion processing difficulties, it has gained some empirical support (Bothe et al. 2019; Cook et al. 2013; Heaton et al. 2012; Kinnaird et al. 2019; Trevisan et al. 2016). Indeed, a recent study by Desai et al. (2019) suggests that alexithymia and autism may contribute to different difficulties in the neural processing of facial expressions, with autistic traits associated with the perceptual encoding of facial features, while alexithymia was associated with higher order emotion decoding. If some of the emotional processing difficulties seen in people with ASD are due to undiagnosed comorbid alexithymia, this has important implications for assessment and treatment (Bird and Cook 2013).

Alexithymia refers to a sub-clinical difficulty with four key features: (1) difficulties identifying one's own emotional states and distinguishing these from physical or bodily sensations, (2) difficulties describing or communicating one's emotions to others, (3) an absence of imaginative or fantasy life, and (4) an externally-oriented cognitive style, focusing on external events and associated actions, with little insight into one's own inner world (Kooiman et al. 2002). It may seem somewhat paradoxical that the most commonly used measure of alexithymia, the Toronto Alexithymia Scale

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(TAS), is a self-report measure, which attempts to assess difficulties in reflecting on the self, by asking the person to self-reflect. To some extent the TAS attempts to mitigate this apparent shortcoming by asking questions designed to tap the observable impacts of alexithymia. For instance, item 3 asks the person to report whether “I have physical sensations that even doctors don’t understand”. This particular item also illustrates something of the history of alexithymia as a disorder that emerged from work on somatisation and post-traumatic stress (Krystal 1979). The original TAS-26 attempted to measure all four features of alexithymia, however the third factor, reduced daydreaming, correlated negatively with the first factor – ‘difficulties identifying emotions’, and was found to have low item-total correlations with the full score on the TAS, so all these items were dropped in the subsequent development of the 20-item version (Parker et al. 1993). An alternative five component model of alexithymia was proposed by Vorst and Bermond (2001) which in addition to the four features mentioned above, included a reduced capacity to experience emotions, sometimes labelled as emotionalizing or emotional arousability. They also distinguished between two types of alexithymia – Type 1 or affective alexithymia consisting of reductions in emotionalizing and in emotion-accompanying cognitions, whereas Type 2 or cognitive alexithymia is characterised by the normal presence of emotions, but with severe reductions in cognitions associated with emotions (Bermond et al. 2007). Though they retained all of the features measured by the TAS-26, one of the key differences was the introduction of a measure of difficulty ‘emotionalizing’. This was defined as a reduced degree to which emotions were induced by stimuli (Bermond et al. 2007).

Despite these short-comings, the TAS-20 is the most widely used measure of alexithymia (Sekely et al. 2018). It has demonstrated convergent validity with other self-report measures of alexithymia including with the Bermond-Vorst Alexithymia Questionnaire (Zech et al. 1999), with the Emotional Intelligence Scale (Besharat 2007; Onur et al. 2013) and continues to discriminate between clinical and non-clinical samples (Goerlich 2018). The TAS-20 has been used with ASD participants (Bird et al. 2010; Cook et al. 2013; Gökçen et al. 2016; Milosavljevic et al. 2016) however, disquiet has been expressed about the use of a self-report measure for people who, by definition, may not be very self-reflective (Kooiman et al. 2002; Ricciardi et al. 2015). Hill et al. (2004) have argued that their study demonstrated that adults with ASD are capable of completing self-report questionnaires about their own emotions. Likewise, Gaigg et al. (2018) found that self-report alexithymia questionnaires produced qualitatively similar data on the emotional difficulties in those with and without a diagnosis of ASD. However, some have suggested that the TAS-20 may underestimate the severity of alexithymia in ASD (Bird and Cook 2013).

One might ask, how do subjective measures of alexithymia, such as the TAS-20, compare with objective measures of the same phenomenon? A few informant-based or clinician-rated scales have been developed to measure alexithymia, though as a number of authors have pointed out, independent ratings of alexithymia are, by their nature, more time-consuming. For instance, in a research context, the preferred method is for the observer to carry out interviews with the participant and several of the participant’s friends and acquaintances (Haviland et al. 2002). The Beth Israel Hospital Psychosomatic Questionnaire (BIPQ—Apfel and Sifneos 1979) is a 17-item clinical interview, with 8 of the items scored as indicators of alexithymia. There is a lack of normative data for the measure (Sriram et al. 1988) and it has been criticized for unacceptably low inter-rater reliability (Lolas et al. 1980; Taylor et al. 1981). The modified version of the Beth Israel Questionnaire includes 12 items, some original and some new items, and relies on trained clinicians scoring each item on a 7-point Likert scale rather than a dichotomous format (Haviland et al. 2002). It was reported to have relatively good inter-item reliability, but data on inter-rater reliability was not available. Another measure, the Alexithymia Provoked Response Questionnaire (APRQ), takes a radically different approach to assessing alexithymia: rather than relying on self-report, or the clinicians interpretations of an unstructured interview, participants are given a series of imaginary scenarios to consider and asked to describe their hypothetical responses. The APRQ derived from a self-report prototype of the Beth Israel Psychosomatic Questionnaire which was the second of three instruments reported by Apfel and Sifneos (1979) and was subsequently renamed as the APRQ by Krystal et al. (1986) to emphasise the use of the items as a structured interview. Responses are judged for their emotional content, and a binary score is assigned to each response, which are summed to give a rating of the overall level of alexithymia. Krystal et al. (1986) recommends scoring responses as alexithymic if they describe, without mentioning affect, either actions, or detailed descriptions of the imagined scenario, or descriptions of physical sensations. Ambiguous responses that contain both alexithymic and non-alexithymic elements are judged to be non-alexithymic. The APRQ was found to correlate highly with the BIPQ (Krystal et al. 1986), but given its limited use in research to date, which aspects of the alexithymia construct it is measuring is less certain. While the APRQ is an objective measure in the sense that it relies on an objective rater of the participant’s response, it does not have the objectivity of measures of alexithymia that gauge physiological arousal and interoception (Gaigg et al. 2018). It was described as having promise as a measure of alexithymia in these early studies, since it is relatively easy and quick

to administer (Kosten et al. 1992), had high test–retest reliability (Kosten et al. 1992) and had excellent inter-rater reliability for the total score (Krystal et al. 1986).

The APRQ provides an overall score of alexithymia, but also offers the potential for content analysis. Each interview can be transcribed and analyzed for emotional word use. A number of studies have indicated that individuals with alexithymia produce fewer affective words in verbal tasks (Friedman et al. 2003; Roedema and Simons 1999). One strategy is to investigate alexithymia by exposing participants to strong emotional stimuli, then having them describe the experience. The number of emotion words used in these verbal accounts was then treated as an index of emotionality (Luminet et al. 2004). Similarly, in this study, we plan to treat the linguistic content of the responses to each item on the APRQ as additional data.

Further studies aimed at comparing objective measures of alexithymia with self-report measures such as the TAS-20, have been recommended (Ricciardi et al. 2015). Very few such studies have been carried out; however, Lumley et al. (1997) found that the total score on the TAS-20 was significantly and negatively associated with the total score on the APRQ in patients experiencing chronic pain. This negative correlation is expected given that higher scores on the TAS-20 and lower scores on the APRQ are both indicators of higher levels of alexithymia on each scale. However, to our knowledge, such comparisons have very rarely been made, and more importantly, given the nature of ASD, not in this specific population.

Alexithymia is an increasingly important construct in ASD research (Berkovits et al. 2017; Gaigg et al. 2018; Kinnaired et al. 2019; Trevisan et al. 2016). Given the lack of studies investigating the intersection between self-report and observer-report measures of alexithymia in the wider population, and more significantly, in the ASD population, with their unique difficulties in self-reflection and self-awareness, this study will add to our understanding of both alexithymia in ASD and how to measure it more effectively.

We examined both measures of alexithymia in a sample of adults with ASD and a control group of TD individuals. The primary aim of this research was to test if the TAS-20, a subjective measure and the APRQ, an objective measure of alexithymia, correlate with one another, indicating that they measure the same construct in both groups. Secondly, we predicted that higher alexithymic scores on both measures would be found for the ASD participants compared with the TD participants. We also predicted that the measure of ASD symptomatology across both samples would be strongly correlated with both measures of alexithymia. Finally, given the sparsity of data around the use of the APRQ, we also predicted that a measure of affective language use, created using linguistic analysis software (Linguistic Inquiry and Word Count—LIWC; Pennebaker et al. 2015) would be

strongly correlated with levels of alexithymia measured by the APRQ.

Method

Participants

Sixty-four adult participants were recruited, 32 with a diagnosis of ASD (mean age = 26.5 years) and 32 without ASD (mean age = 24.5). The ASD and TD groups did not differ significantly in age ($t(59.58) = 1.06, p = 0.295, d = 0.27$). All of the adults with ASD attend a support service that requires a full diagnostic work-up using either the Autism Diagnosis Interview-Revised (ADI-R) (Lord et al. 1994) or the Diagnostic Interview for Social and Communication Disorders (DISCO, Wing et al. 2002), and Autism Diagnostic Observation Schedule (ADOS; Lord et al. 2000) by a clinical team, before accessing the service. The 32 non-ASD participants were recruited through advertising in the university, social media and word of mouth, and had the same sex distribution as the ASD sample (25 male, 7 female). In both samples 31 out of 32 participants identified their nationality as Irish. The levels of education varied significantly between samples $\chi^2(4) = 10.11, p = 0.039$, with the non-ASD sample having a higher proportion of graduates (66% to 41%).

Measures

Autism Quotient (AQ-10)

Autism traits were measured using the self-report Autism Quotient (AQ-10 Allison et al. 2012;). This is a brief questionnaire based on the full-length 50-item Autism Quotient (Baron-Cohen et al. 2001) and is the selection of the 10 items from the original measure that showed the highest levels of discrimination between participants with and without ASD. Each question has four response categories ('definitely agree', 'slightly agree', 'slightly disagree', 'definitely disagree'), however, items are scored as one or zero by collapsing each of the two levels of agree and disagree, resulting in a maximum score of 10, with scores of 6 or above being indicative of ASD. The AQ-10 had very good internal consistency in our samples ($\alpha = 0.96$) even higher than the value ($\alpha = 0.85$) reported in the original AQ-10 study by Allison et al. (2012).

Toronto Alexithymia Scale (TAS-20)

The Toronto Alexithymia Scale (TAS-20) is a twenty item self-report scale designed to measure alexithymia, developed by Bagby et al. (1994). The items are rated on a five-point Likert scale from strongly disagree to strongly agree.

Five items are reverse scored to reduce response bias. While it is recommended that TAS-20 scores be analysed as a continuous variable, cut-off points for the presence and severity of alexithymia were identified as: ≤ 51 = non-alexithymia, $52\text{--}60$ = possible alexithymia and ≥ 61 = clinically significant alexithymia (Taylor et al. 1992). The TAS-20 assesses three factors of alexithymia, namely (1) Difficulty Identifying Feelings (DIF), containing seven items, an example of which is “I am often confused by what emotion I am feeling”, (2) Difficulty Describing Feelings (DDF), containing five items, an example of which is “It is difficult for me to find the right words for my feelings” and (3) Externally-Oriented Thinking (EOT), containing eight items, an example of which is “I prefer to talk to people about their daily activities rather than their feelings”. In the original study, the TAS-20 had demonstrated acceptable internal consistency with Cronbach’s α estimate of 0.81 for the total score, and 0.78, 0.75 and 0.66 for the DIF, DDF and EOT subscales respectively. In our samples, the overall Cronbach’s α estimate was 0.81 for the total score, and alpha coefficients of 0.86, 0.70 and 0.60 for the DIF, DDF and EOT.

Alexithymia Provoked Response Questionnaire (APRQ)

The APRQ is a 17-item structured interview, in which participants are asked to imagine how they would feel in a range of hypothetical scenarios. For example, item 2 is: “how would you feel if a policeman arrested you for a crime you did not commit?” The responses to the APRQ are scored by the interviewer. We utilized the guidelines of Kosten et al. (1992), evaluating the participant’s ability to identify and describe emotions and emotional experiences. A higher score on the APRQ indicates a lower degree of alexithymia, as a score of zero is attributed to an alexithymic response and a score of one attributed to a non-alexithymia response. Responses that described an intended action, detailed descriptions of a situation or physical/bodily sensations, without any mention of affect, were classified as alexithymic and thus, given a score of zero. Responses to all 17 items were summated to give a total score and the measure does not have subscales. The APRQ interviews were scored independently by the third and fourth authors, following which discrepancies or disagreements in scores were discussed between authors. In the event that these discrepancies were not resolved between authors, the principal researcher was further consulted to resolve disagreement in scoring. Cohen’s kappa was calculated to determine the level of agreement in scoring of the APRQ across both samples, confirming a high level of agreement, $k = 0.97$, $p < 0.001$. The overall Cronbach’s α estimate for the APRQ was 0.71.

Mean Affective Vocabulary

As a further validation check on the use of the APRQ, we analyzed transcripts of all the interviews using computerized text analysis. This allows researchers to extract psychological terms from verbal material in a systematic and objective manner. The computer software Linguistic Inquiry and Word Count (LIWC; Pennebaker et al. 2015) searches for words and word stems in texts and assigns them to a range of linguistic categories, including words associated with emotion and affect. The percentage of words in each category is reported. Our analysis focused on one of these categories: affect. We treated each item on the APRQ as a separate text and calculated a mean affect score for each participant, which represents the average percentage of emotion words used across the 17 questions.

Statistical Analysis

All the statistical analyses were undertaken in R (R Core Team 2017). Results were analysed using ANOVA, independent samples t-tests, and bivariate correlations. Cohen’s d and partial Eta-Squared are reported as effect size measures. The Shapiro–Francia test has been identified as the best test statistic in detecting deviation from normality among all the commonly used measures (Mbah and Paothong 2015). Neither the TAS-20 nor the APRQ showed significant deviations from normality on the Shapiro–Francia test.

Given this significant difference between the samples on education, we ran a series of ANOVAs on education against the key variables of interest, to check whether to include them as potential confounds in later analysis. We found no significant differences between education levels and scores on the APRQ ($F(4, 59) = 0.94$, $p = 0.447$, $\eta_p^2 = 0.06$), on the TAS-20 total score ($F(4, 58) = 0.63$, $p = 0.641$, $\eta_p^2 = 0.04$), on the Autism Quotient ($F(4, 59) = 0.54$, $p = 0.707$, $\eta_p^2 = 0.04$), on affective word use ($F(4, 59) = 0.36$, $p = 0.839$, $\eta_p^2 = 0.02$) or on word count ($F(4, 59) = 1.09$, $p = 0.372$, $\eta_p^2 = 0.07$).

Results

ASD Symptomatology—AQ-10

As expected, the ASD participants had much higher scores on the AQ-10 ($M = 6.78$, $SD = 2.01$) than the TD participants ($M = 3.25$, $SD = 1.32$), $t(53.51) = 8.30$, $p < 0.001$, $d = 2.08$. The clinical cut-off for the AQ-10 is 6, with 78% of the ASD participants scoring in the clinical range, and none of the TD group scoring 6 or above.

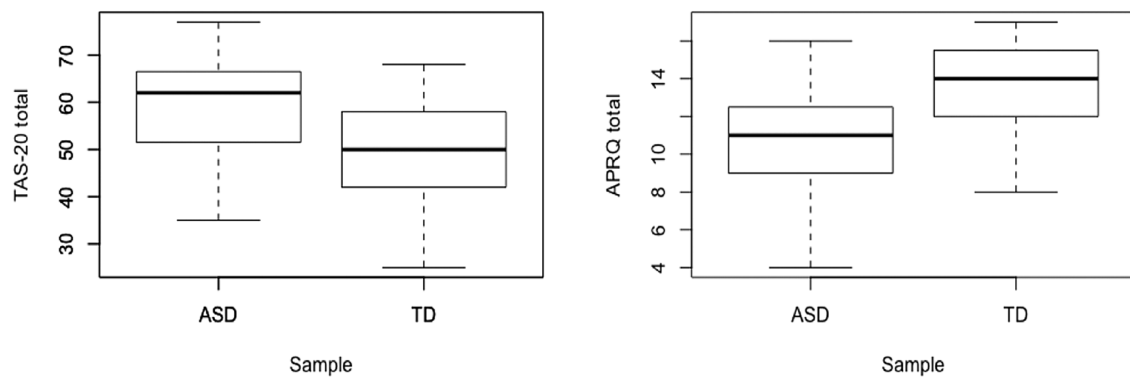


Fig. 1 Mean scores on TAS-20 and APRQ

Group Comparisons of Alexithymia Measures

In line with our prediction, the ASD and TD samples differed significantly in their mean scores on the TAS total (see Fig. 1). Results indicated a significantly higher mean alexithymia TAS total score for ASD participants ($M = 59.55$, $SD = 10.56$) compared with TD participants ($M = 48.94$, $SD = 11.24$), $t(60.94) = 3.86$, $p < 0.001$, with a large effect size (Cohen's $d = 0.97$). The ASD group also scored significantly higher on each of the three subscales of the TAS-20, with higher scores on *Difficulty Identifying Feelings* subscale $t(59.79) = 2.94$, $p = 0.005$, $d = 0.74$, higher scores on the *Difficulty Describing Feelings* subscale $t(56.44) = 2.84$, $p = 0.006$, $d = 0.72$ and higher scores on the *Externally Oriented Thinking* subscale $t(60.00) = 2.34$, $p = 0.022$, $d = 0.59$. Likewise the ASD group scored lower (indicating greater alexithymia) on the APRQ ($M = 10.81$, $SD = 2.76$) than the TD participants ($M = 13.49$, $SD = 2.55$), $t(61.27) = -4.16$, $p < 0.001$, with a large effect size (Cohen's $d = -1.04$).

TAS-20 scores of around 48 are common for typical populations (Franz et al. 2008), indicating that our TD sample scored in the expected range. In the TD sample, 5 participants scored above the cut-off of ≥ 61 for identifying alexithymia, representing 15.6% of the TD group. In the ASD sample, 18 of the participants or 56.2% were in the alexithymic range. With regard to the APRQ, though the authors did not establish cut-off scores, a healthy control group in a study by Legorreta et al. (1988), were reported to have a mean 13.13 ($SD 2.08$) with clinical samples scoring 10.81 ($SD 3.20$). Similarly, a chronic somatic pain group scored a mean of 10.4 ($SD 2.9$) in a study by Lumley et al. (1997). In our ASD sample the mean was comparable to previously examined clinical samples ($M = 10.81$, $SD = 2.76$).

Correlation Analysis

In order to explore whether AQ-10 scores are associated with severity of alexithymia, we examined Pearson

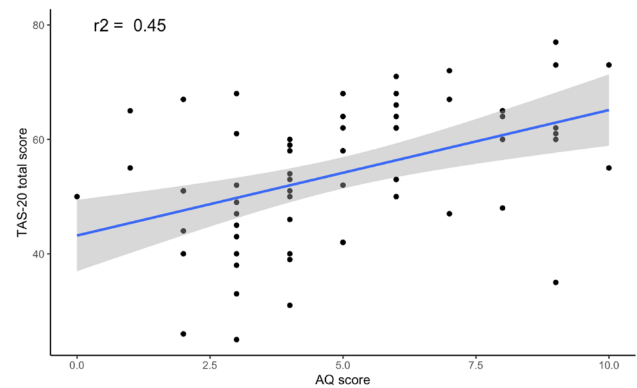


Fig. 2 Correlation between TAS-20 and AQ10

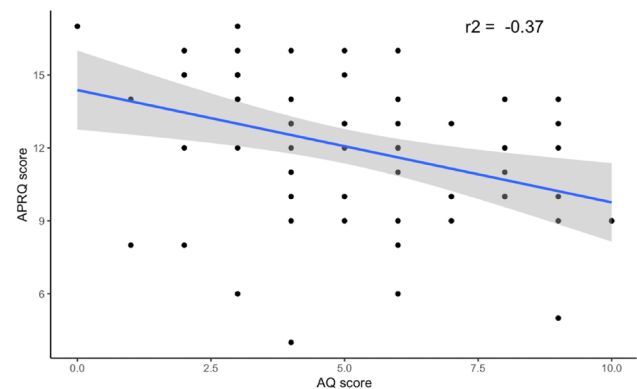


Fig. 3 Correlation between APRQ and AQ10

product-moment correlations between AQ and TAS total scores, and AQ and APRQ scores. As can be seen in Fig. 2. There was a strong correlation between AQ score and TAS total score, with increased ASD severity being associated with increased alexithymia on the TAS-20 ($r(61) = 0.45$, $p < 0.001$).

Likewise, we examined the influence of ASD trait severity on APRQ scores. Figure 3 illustrates the significant

negative correlation between scores on the AQ and APRQ, ($r(62) = -0.37, p = 0.002$).

Relationship Between Measures of Alexithymia

To examine whether the objective measure of alexithymia, the APRQ and the subjective measure of alexithymia, the TAS-20 are measuring the same, or similar constructs, we calculated Pearson product-moment correlations between TAS total and APRQ scores. The correlation was not significant ($r(61) = -0.10, p = 0.439$). We also calculated the correlation coefficients between these variables by sample, in case the relationship held within samples but not across them. Rather than correct for the familywise error rate, we calculated these correlations as Bayes Factors (Dienes 2016) using the software JASP (Version 0.11.1; JASP Team 2020). The advantage of this technique is that it avoids the potential in frequentist statistics for arbitrary specification of the family of comparison, allowing for too much flexibility in defining relevant families (Dienes 2016). By convention Bayes Factors between 0.33 and 3 are considered inconclusive evidence for or against a hypothesis, and a guideline suggested by Held and Ott (2018) for interpreting Bayes Factors is that values between 10 and 100 suggest strong evidence in favour of H1. The correlation was not significant in the TD sample ($r(30) = -0.13, p = 0.462, BF_{10} = 0.285$, with the Bayes factor less than 0.3 suggesting evidence in favour of the null hypothesis, whereas the correlation in the ASD sample was $r(29) = 0.39, p = 0.028, BF_{10} = 2.24$, with the Bayes factor being inconclusive evidence of a correlation. Furthermore, to check for a difference between these correlations we compared them using the cocor package in R (Diedenhofen and Musch 2016). This returned a Fisher's $z = 2.0868, p = 0.037$ suggesting a significant difference between the two groups in their correlation coefficients.

Relationship Between Mean Affective Vocabulary and Alexithymia Measures

Mean Affective Vocabulary (MAV) scores were calculated for all of the APRQ interviews. We would not expect perfect correspondence between the computerised word counting approach to emotion word use by participants and the clinical judgements made in scoring the APRQ, however, a reasonable level of correlation between the two measures was expected. The APRQ total score and MAV calculated from LIWC were correlated, ($r(62) = 0.49, p < 0.001$) with a medium effect size, however the MAV did not correlate with the total TAS scores ($r(61) = -0.13, p = 0.312$). ASD participants used significantly less affective words (mean = 11.25, $SD = 6.88$) than TD participants (mean = 16.53, $SD = 9.62$), $t(56.12) = -2.53, p = 0.014, d = -0.63$.

Discussion

The purpose of this study was to examine whether subjective and objective measures of alexithymia are assessing the same construct, by comparing the self-report measure of alexithymia, the TAS-20 (Bagby et al. 1994) with an objective, interviewer rated measure, the APRQ (Krystal 1979). We also wanted to test whether participants with ASD would differ in their responses to these two measures, given that ASD has been characterised as having reduced psychological self-awareness (Frith and de Vignemont 2005). This view has been challenged recently by researchers who argue that self-related processing in autism is complex and some areas such as awareness of one's body and actions may be unimpaired (Williams 2010), whilst other areas such as psychological awareness of the self may be altered (Nijhof and Bird 2019).

As predicted, the participants with ASD scored significantly higher on both self-report and observer-rated measures of alexithymia, both measures indicating significantly elevated levels of alexithymia in the ASD sample when compared to a TD group. In the ASD sample, 56% of the participants scored above clinical cut-off of > 61 on the TAS-20. This result is consistent with findings by Berthoz and Hill (2005) and Hill et al. (2004) who reported prevalence rates of alexithymia in people with ASD as varying between 40 and 65%. This is in keeping with the growing body of literature, using self-report measures alone, which has identified a strong co-occurrence between ASD and alexithymia (Bird and Cook 2013; Shah et al. 2016; Trevisan et al. 2016). However, this is the first study to replicate this finding using an objective measure of alexithymia in the form of the APRQ. Various researchers have argued that alexithymia self-reports should be supplemented with other measures, including observer accounts (Berthoz and Hill 2005; Kooiman et al. 2002). Our finding supports the notion that observer-based alexithymia measures also corroborate the higher levels of alexithymia in individuals with ASD than those without.

Secondly, we found that not only did the ASD participants score significantly higher on both measures of alexithymia than the TD participants, levels of ASD traits, as measured by the AQ-10 were significantly correlated with each of the measures of alexithymia. There was a strong positive correlation between scores on the AQ-10 and TAS-20, with increased ASD severity being associated with increased alexithymia. Likewise, there was a strong negative correlation between the AQ-10 and scores on the APRQ. There was also a strong negative relationship between affective word use and scores on the APRQ. The association between ASD traits and both measures of alexithymia was notable as this is contrary to a previous study

carried out by Milosavljevic et al. (2016) who found no relationship between alexithymia and the SRS as a measure of ASD severity. However, in their study they only compared scores on the SRS across dichotomized groups of high and low alexithymia within their ASD sample. It is perfectly possible that some individuals score highly on ASD traits but have low alexithymia, while there still being a significant correlation between the two measures. How these two conditions interact requires further attention and may generate better insights into how to intervene more effectively to support people with ASD in their social and emotional well-being.

Perhaps most surprisingly, the two measures of alexithymia did not correlate with each other. This would imply that that the measures, despite the robust relationship with underlying ASD traits, are actually measuring different aspects of the alexithymia construct. In the abstract from a conference paper by Pierce et al. (1990), they reported that scores on the TAS-20 did not correlate significantly with the APRQ, which they argued was due to these different assessment methods measuring “related but distinct constructs”. What different aspects of alexithymia might these two measures be tapping?

The TAS-20 is considered the gold standard measure of alexithymia due to its excellent reliability and construct validity (Bagby et al. 1994), but as discussed previously, this reliability was partly achieved at the expense of excluding the items relating to reduced daydreaming and an impoverished fantasy life, one of the four key features of alexithymia in the original definition (Sifneos 1973). Some have argued that the TAS-20 may also fail to identify other features of alexithymia. As we saw, Bermond (1997) distinguished between two types of alexithymia: Type 1 characterized by a lack of awareness of emotional arousal and low levels of emotion accompanying cognitions and Type 2 characterized by normal degrees of awareness of emotional arousal but with low levels of accompanying cognitions. Their measure of alexithymia, the Bermond-Vorst Alexithymia Questionnaire (BVAQ) examines both the emotional component and the cognitive component of alexithymia (Larsen et al. 2003). They have shown total TAS-20 scores only correlate with the cognitive component (represented by the subscales identifying, analysing and verbalizing emotions) of the BVAQ, whereas the emotional component (which captures difficulty fantasizing and difficulties emotionalizing) is statistically uncorrelated with the total TAS-20 scores. However, the idea of dividing alexithymia into two types has not met with universal support and has been criticized on the grounds that the sub-type characterization does not match the performance of the BVAQ in cluster analytic studies (Bagby et al. 2009). The alternative perspective, that alexithymia

is a multi-dimensional construct, remains the dominant model.

It is possible that the APRQ is measuring this *difficulty emotionalizing* component of alexithymia, which would explain the lack of correlation between the two measures. Emotionalizing is defined as the degree to which someone is aroused by emotionally stimulating events and the APRQ is designed to present just such events—the imaginary scenarios read to participants—and the scoring method assesses the degree to which the participants report emotional responses. The APRQ scores in this study also correlated highly with the mean affective vocabulary used by participants. Early work on the analysis of speech of people with alexithymia had focused on pauses and silences in conversations with therapists (Overbeck 1977), and the use of pronouns and auxiliary verbs, but more significantly, they showed that those believed to have alexithymia used less affect-laden words (von Rad et al. 1977). This finding was replicated by Taylor and Doody (1985), who argued that affective vocabulary score is a valid measure of alexithymia. Similarly, Allen et al. (2013) found reduced levels of verbal emotional responsiveness to short clips of emotional music in adults with ASD compared to a control group and this was mediated by their level of comorbid alexithymia. But it may also be useful to distinguish between two levels of emotionality in this context. For instance, Allen et al. (2009) found normal degrees of arousal induced through listening to music in participants with ASD, but this arousal was not labeled as emotional valence by participants. To what degree can we say that someone is experiencing an emotion (emotionalizing) if the experience is one of undifferentiated arousal?

It is important to acknowledge several limitations to the study. The APRQ remains an under-researched psychometric measure with limited data available on its psychometric properties. Secondly the APRQ is designed to tap emotional reactions to imaginary events—how this might generalize to real events is unknown. Though the sample was representative of the gender balance within the ASD community, the small number of female participants means that further studies are necessary to establish whether this pattern of responding to alexithymia measures is robust in women with ASD. Finally, we cannot rule out verbal IQ as a potential confound.

In summary, the APRQ has promise as an additional measure of alexithymia, suitable for adults with ASD, with the potential to enhance our understanding of different aspects of the alexithymia construct, particularly with regard to the emotionality dimension. In addition, the use of computerised linguistic analysis on the transcripts of the APRQ interviews offers further potential for exploring the presentation of alexithymia and to differentiate it from aspects measured through self-report, in addition to

the validation of observer ratings. The use of a subjective measure such as the TAS-20 is an effective and quick way of identifying emotional self-awareness. The benefit of an objective measure such as the APRQ, is it allows for the gathering of rich personal experiences across both positive and negative emotions, potentially providing the interviewer with an insight into the relationship between alexithymia and emotion processing difficulties. A measure such as the APRQ also provides a novel way of addressing difficulties in reporting specific types of emotion, as self-report measures are based on the assumption that all emotions are equally difficult to recognize and describe (Ricciardi et al. 2015). Recent work in ASD has highlighted the importance of emotion processing difficulties, the reduced ability to recognise emotion in oneself and in others (Bird and Cook 2013), and the high frequency with which alexithymia co-occurs with ASD. Further research into the interaction between alexithymia symptoms and ASD holds the potential to increase our ability to improve the social relationships of people with ASD, that depend on appropriate emotional response to other people's emotional state (Luminet et al. 2018).

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Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical Approval All procedures were in accordance with the ethical standards of the local ethics committee guidelines and with the 1964 Helsinki declaration and its later amendments. The study was approved by the appropriate local ethics committee.

Informed Consent Informed consent was obtained from all participants of the study.

References

- Allen, R., Davis, R., & Hill, E. (2013). The effects of autism and alexithymia on physiological and verbal responsiveness to music. *Journal of Autism and Developmental Disorders*, 43(2), 432–444. <https://doi.org/10.1007/s10803-012-1587-8>.
- Allen, R., Hill, E., & Heaton, P. (2009). 'Hath charms to soothe ...': An exploratory study of how high-functioning adults with ASD experience music. *Autism*, 13(1), 21–41. <https://doi.org/10.1177/1362361307098511>.
- Allison, C., Auyeung, B., & Baron-Cohen, S. (2012). Toward brief "red flags" for autism screening: The short autism spectrum quotient and the short quantitative checklist in 1,000 cases and 3,000 controls. *Journal of the American Academy of Child & Adolescent Psychiatry*, 51(2), 202–212.e7. <https://doi.org/10.1016/j.jaac.2011.11.003>.
- Apfel, R. J., & Sifneos, P. E. (1979). Alexithymia: Concept and measurement. *Psychotherapy and Psychosomatics*, 32(1–4), 180–190. <https://doi.org/10.1159/000287386>.
- Bagby, R. M., Parker, J. D. A., & Taylor, G. J. (1994). The twenty-item Toronto Alexithymia scale—I. Item selection and cross-validation of the factor structure. *Journal of Psychosomatic Research*, 38(1), 23–32. [https://doi.org/10.1016/0022-3999\(94\)90005-1](https://doi.org/10.1016/0022-3999(94)90005-1).
- Bagby, R. M., Quilty, L. C., Taylor, G. J., Grabe, H. J., Luminet, O., Verissimo, R., et al. (2009). Are there subtypes of alexithymia? *Personality and Individual Differences*, 47(5), 413–418. <https://doi.org/10.1016/j.paid.2009.04.012>.
- Baron-Cohen, S., Wheelwright, S., Skinner, R., Martin, J., & Clubley, E. (2001). The autism-spectrum quotient (AQ): Evidence from Asperger syndrome/high-functioning autism, males and females, scientists and mathematicians. *Journal of Autism and Developmental Disorders*, 31(1), 5–17. <https://doi.org/10.1023/A:1005653411471>.
- Berkovits, L., Eisenhower, A., & Blacher, J. (2017). Emotion Regulation in Young Children with Autism Spectrum Disorders. *Journal of Autism and Developmental Disorders*, 47(1), 68–79. <https://doi.org/10.1007/s10803-016-2922-2>.
- Bermond, B. (1997). Brain and alexithymia. In A. Vingerhoets, F. Bussel, & J. Boelhouwer (Eds.), *The (non)expression of emotions in health and disease* (pp. 115–130). Tilburg: Tilburg University Press.
- Bermond, B., Clayton, K., Liberova, A., Luminet, O., Maruszewski, T., Ricci Bitti, P. E., et al. (2007). A cognitive and an affective dimension of alexithymia in six languages and seven populations. *Cognition and Emotion*, 21(5), 1125–1136. <https://doi.org/10.1080/02699930601056989>.
- Berthoz, S., & Hill, E. L. (2005). The validity of using self-reports to assess emotion regulation abilities in adults with autism spectrum disorder. *European Psychiatry*, 20(3), 291–298. <https://doi.org/10.1016/j.eurpsy.2004.06.013>.
- Besharat, M. A. (2007). Reliability and factorial validity of a farsi version of the 20-item Toronto Alexithymia Scale with a sample of Iranian students. *Psychological Reports*, 101(1), 209–220. <https://doi.org/10.2466/pr0.101.1.209-220>.
- Bird, G., & Cook, R. (2013). Mixed emotions: The contribution of alexithymia to the emotional symptoms of autism. *Translational Psychiatry*, 3(7), e285–e285. <https://doi.org/10.1038/tp.2013.61>.
- Bird, G., Silani, G., Brindley, R., White, S., Frith, U., & Singer, T. (2010). Empathic brain responses in insula are modulated by levels of alexithymia but not autism. *Brain*, 133(5), 1515–1525.
- Bothe, E., Palermo, R., Rhodes, G., Burton, N., & Jeffery, L. (2019). Expression recognition difficulty is associated with social but not attention-to-detail autistic traits and reflects both alexithymia and perceptual difficulty. *Journal of Autism and Developmental Disorders*, 49(11), 4559–4571. <https://doi.org/10.1007/s10803-019-04158-y>.
- Cook, R., Brewer, R., Shah, P., & Bird, G. (2013). Alexithymia, not autism, predicts poor recognition of emotional facial expressions. *Psychological Science*, 24(5), 723–732.
- Desai, A., Foss-Feig, J. H., Naples, A. J., Coffman, M., Trevisan, D. A., & McPartland, J. C. (2019). Autistic and alexithymic traits modulate distinct aspects of face perception. *Brain and Cognition*, 137, 103616. <https://doi.org/10.1016/j.bandc.2019.103616>.
- Diedenhofen, B., & Musch, J. (2016). cocron: A web interface and R package for the statistical comparison of cronbach's alpha coefficients. *International Journal Internet Science*, 11, 51–60.

- Dienes, Z. (2016). How Bayes factors change scientific practice. *Journal of Mathematical Psychology*, 72, 78–89. <https://doi.org/10.1016/j.jmp.2015.10.003>.
- Franz, M., Popp, K., Schaefer, R., Sitte, W., Schneider, C., Hardt, J., et al. (2008). Alexithymia in the German general population. *Social Psychiatry and Psychiatric Epidemiology*, 43(1), 54–62. <https://doi.org/10.1007/s00127-007-0265-1>.
- Friedman, S. R., Rappaport, L. J., Lumley, M., Tzelepis, A., VanVoorhis, A., Stettner, L., et al. (2003). Aspects of social and emotional competence in adult attention-deficit/hyperactivity disorder. *Neuropsychology*, 17(1), 50–58. <https://doi.org/10.1037/0894-4105.17.1.50>.
- Frith, U., & de Vignemont, F. (2005). Egocentrism, allocentrism, and Asperger syndrome. *Consciousness and Cognition*, 14(4), 719–738. <https://doi.org/10.1016/j.concog.2005.04.006>.
- Gaigg, S. B., Cornell, A. S., & Bird, G. (2018). The psychophysiological mechanisms of alexithymia in autism spectrum disorder. *Autism*, 22(2), 227–231. <https://doi.org/10.1177/1362361316667062>.
- Goerlich, K. S. (2018). The multifaceted nature of Alexithymia—A neuroscientific perspective. *Frontiers in Psychology*. <https://doi.org/10.3389/fpsyg.2018.01614>.
- Gökçen, E., Frederickson, N., & Petrides, K. V. (2016). Theory of mind and executive control deficits in typically developing adults and adolescents with high levels of autism traits. *Journal of Autism and Developmental Disorders*, 46(6), 2072–2087. <https://doi.org/10.1007/s10803-016-2735-3>.
- Haviland, M. G., Hendryx, M. S., Shaw, D. G., & Henry, J. P. (1994). Alexithymia in women and men hospitalized for psychoactive substance dependence. *Comprehensive Psychiatry*, 35(2), 124–128. [https://doi.org/10.1016/0010-440X\(94\)90056-N](https://doi.org/10.1016/0010-440X(94)90056-N).
- Haviland, M. G., Warren, W. L., Riggs, M. L., & Nitch, S. R. (2002). Concurrent validity of two observer-rated alexithymia measures. *Psychosomatics*, 43(6), 472–477. <https://doi.org/10.1176/appi.psy.43.6.472>.
- Heaton, P., Reichenbacher, L., Sauter, D., Allen, R., Scott, S., & Hill, E. (2012). Measuring the effects of alexithymia on perception of emotional vocalizations in autistic spectrum disorder and typical development. *Psychological Medicine*, 42(11), 2453–2459. <https://doi.org/10.1017/S0033291712000621>.
- Held, L., & Ott, M. (2018). On *p*-values and Bayes factors. *Annual Review of Statistics and Its Application*, 5(1), 393–419. <https://doi.org/10.1146/annurev-statistics-031017-100307>.
- Hill, E., Berthoz, S., & Frith, U. (2004). Cognitive processing of own emotions in individuals with autistic spectrum disorder and in their relatives. *Journal of Autism and Developmental Disorders*, 34(2), 229–235. <https://doi.org/10.1023/B:JADD.0000022613.41399.14>.
- Hobson, H., Brewer, R., Catmur, C., & Bird, G. (2019). The role of language in alexithymia: Moving towards a multiroute model of alexithymia. *Emotion Review*, 11(3), 247–261. <https://doi.org/10.1177/1754073919838528>.
- JASP, T. (2020). JASP (Version 0.12.2) [Computer software]. Retrieved from <https://jasp-stats.org>
- Kinnaird, E., Stewart, C., & Tchanturia, K. (2019). Investigating alexithymia in autism: A systematic review and meta-analysis. *European Psychiatry*, 55, 80–89. <https://doi.org/10.1016/j.eurpsy.2018.09.004>.
- Kooiman, C. G., Spinhoven, P., & Trijsburg, R. W. (2002). The assessment of alexithymia. *Journal of Psychosomatic Research*, 53(6), 1083–1090. [https://doi.org/10.1016/S0022-3999\(02\)00348-3](https://doi.org/10.1016/S0022-3999(02)00348-3).
- Kosten, T. R., Krystal, J. H., Gillier, E., Frank, J., & Dan, E. (1992). Alexithymia as a predictor of treatment response in post-traumatic stress disorder. *Journal of Traumatic Stress*, 5, 563–573.
- Krystal, H. (1979). Alexithymia and psychotherapy. *American Journal of Psychotherapy*, 33(1), 17–31. <https://doi.org/10.1176/appi.psychotherapy.1979.33.1.17>.
- Krystal, H., & Krystal, J. H. (2015). *Integration and self healing: Affect, trauma, alexithymia*. <https://search.ebscohost.com/login.aspx?direct=true&scope=site&db=nlebk&db=nlabk&AN=1131720>
- Krystal, J. H., Giller, E. L., & Cicchetti, D. V. (1986). Assessment of alexithymia in posttraumatic stress disorder and somatic illness: Introduction of a reliable measure. *Psychosomatic Medicine*, 48(1), 84–94. <https://doi.org/10.1097/00006842-198601000-00007>.
- Larsen, J. K., Brand, N., Bermond, B., & Hijman, R. (2003). Cognitive and emotional characteristics of alexithymia. *Journal of Psychosomatic Research*, 54(6), 533–541. [https://doi.org/10.1016/S0022-3999\(02\)00466-X](https://doi.org/10.1016/S0022-3999(02)00466-X).
- Legorreta, G., Bull, R. H., & Kiely, M. C. (1988). Alexithymia and symbolic function in the obese. *Psychotherapy and Psychosomatics*, 50(2), 88–94. <https://doi.org/10.1159/000288105>.
- Lolas, F., de la Parra, G., Aronsohn, S., & Collin, C. (1980). On the measurement of alexithymic behavior. *Psychotherapy and Psychosomatics*, 33(3), 139–146. <https://doi.org/10.1159/000287424>.
- Lord, C., Risi, S., Lambrecht, L., Cook, E. H., Leventhal, B. L., DiLavore, P. C., et al. (2000). The autism diagnostic observation schedule—generic: A standard measure of social and communication deficits associated with the spectrum of autism. *Journal of Autism and Developmental Disorders*, 30(3), 205–223. <https://doi.org/10.1023/A:1005592401947>.
- Lord, C., Rutter, M., & Le Couteur, A. (1994). Autism diagnostic interview—revised: A revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. *Journal of Autism and Developmental Disorders*, 24(5), 659–685. <https://doi.org/10.1007/BF02172145>.
- Luminet, O., Bagby, R. M., & Taylor, G. J. (Eds.). (2018). *Alexithymia: Advances in Research, Theory, and Clinical Practice* (1st ed.). Cambridge: Cambridge University Press. <https://doi.org/10.1017/9781108241595>
- Luminet, O., Rimé, B., Bagby, R. M., & Taylor, G. (2004). A multimodal investigation of emotional responding in alexithymia. *Cognition and Emotion*, 18(6), 741–766. <https://doi.org/10.1080/02699930341000275>.
- Lumley, M. A., Asselin, L. A., & Norman, S. (1997). Alexithymia in chronic pain patients. *Comprehensive Psychiatry*, 38(3), 160–165. [https://doi.org/10.1016/S0010-440X\(97\)90069-9](https://doi.org/10.1016/S0010-440X(97)90069-9).
- Mbah, A. K., & Paothong, A. (2015). Shapiro-Francia test compared to other normality test using expected-value. *Journal of Statistical Computation and Simulation*, 85(15), 3002–3016. <https://doi.org/10.1080/00949655.2014.947986>.
- Milosavljevic, B., Carter Leno, V., Simonoff, E., Baird, G., Pickles, A., Jones, C. R. G., et al. (2016). Alexithymia in adolescents with autism spectrum disorder: Its relationship to internalising difficulties, sensory modulation and social cognition. *Journal of Autism and Developmental Disorders*, 46(4), 1354–1367. <https://doi.org/10.1007/s10803-015-2670-8>.
- Nijhof, A. D., & Bird, G. (2019). Self-processing in individuals with autism spectrum disorder. *Autism Research*, 12(11), 1580–1584. <https://doi.org/10.1002/aur.2200>.
- Onur, E., Alkin, T., Sheridan, M. J., & Wise, T. N. (2013). Alexithymia and emotional intelligence in patients with panic disorder, generalized anxiety disorder and major depressive disorder. *Psychiatric Quarterly*, 84(3), 303–311. <https://doi.org/10.1007/s1126-012-9246-y>.
- Overbeck, G. (1977). How to operationalize Alexithymic phenomena—Some findings from speech analysis and the Giessen test (GT). *Psychotherapy and Psychosomatics*, 28(1–4), 106–120. <https://doi.org/10.1159/000287051>.
- Parker, J. D. A., Michael Bagby, R., Taylor, G. J., Endler, N. S., & Schmitz, P. (1993). Factorial validity of the 20-item Toronto

- Alexithymia Scale. *European Journal of Personality*, 7(4), 221–232. <https://doi.org/10.1002/per.2410070403>.
- Pennebaker, J. W., Booth, R. J., Boyd, R. L., & Francis, M. E. (2015). *Linguistic inquiry and word count: LIWC2015*.
- Pierce, M., Krystal, J. H., Faryna, A., Markert, R., & Davidson, A. (1990). A comparison of alexithymia measures. In Proc. American Psychiatric Association, Vol. 59
- Ricciardi, L., Demartini, B., Fotopoulou, A., & Edwards, M. J. (2015). Alexithymia in neurological disease: A review. *The Journal of Neuropsychiatry and Clinical Neurosciences*, 27(3), 179–187. <https://doi.org/10.1176/appi.neuropsych.14070169>.
- Roedema, T. M., & Simons, R. F. (1999). Emotion-processing deficit in alexithymia. *Psychophysiology*, 36(3), 379–387. <https://doi.org/10.1017/S0048577299980290>.
- Sekely, A., Bagby, R. M., & Porcelli, P. (2018). Assessment of the Alexithymia Construct. In O. Luminet, R. M. Bagby, & G. J. Taylor (Eds.), *Alexithymia* (1st ed., pp. 17–32). Cambridge: Cambridge University Press.
- Shah, P., Hall, R., Catmur, C., & Bird, G. (2016). Alexithymia, not autism, is associated with impaired interoception. *Cortex*, 81, 215–220. <https://doi.org/10.1016/j.cortex.2016.03.021>.
- Sifneos, P. E. (1973). The prevalence of 'Alexithymic' characteristics in psychosomatic patients. *Psychotherapy and Psychosomatics*, 22(2–6), 255–262. <https://doi.org/10.1159/000286529>.
- Sriram, T. G., Pratap, L., & Shanmugham, V. (1988). Towards enhancing the utility of Beth Israel Hospital psychosomatic questionnaire. *Psychotherapy and Psychosomatics*, 49(3–4), 205–211. <https://doi.org/10.1159/000288085>.
- Tani, P., Lindberg, N., Joukamaa, M., Nieminen-von Wendt, T., von Wendt, L., Appelberg, B., et al. (2004). Asperger syndrome, alexithymia and perception of sleep. *Neuropsychobiology*, 49(2), 64–70. <https://doi.org/10.1159/000076412>.
- Taylor, G., Doody, K., & Newman, A. (1981). Alexithymic characteristics in patients with inflammatory bowel disease. *The Canadian Journal of Psychiatry*, 26(7), 470–474. <https://doi.org/10.1177/070674378102600706>.
- Taylor, G. J., & Doody, K. (1985). Verbal measures of alexithymia: What do they measure. *Psychotherapy and Psychosomatics*, 43(1), 32–37. <https://doi.org/10.1159/000287855>.
- Taylor, G. J., Parker, J. D. A., Bagby, M. R., & Acklin, M. W. (1992). Alexithymia and somatic complaints in psychiatric out-patients. *Journal of Psychosomatic Research*, 36(5), 417–424. [https://doi.org/10.1016/0022-3999\(92\)90002-J](https://doi.org/10.1016/0022-3999(92)90002-J).
- Team, R. C. (2017). *R: A language and environment for statistical computing*. <https://www.R-project.org/>.
- Trevisan, D. A., Bowering, M., & Birmingham, E. (2016). Alexithymia, but not autism spectrum disorder, may be related to the production of emotional facial expressions. *Molecular Autism*, 7(1), 46. <https://doi.org/10.1186/s13229-016-0108-6>.
- von Rad, M., Lalucat, L., & Lolas, F. (1977). Differences of verbal behaviour in psychosomatic and psychoneurotic patients. *Psychotherapy and Psychosomatics*, 28(1–4), 83–97. <https://doi.org/10.1159/000287047>.
- Vorst, H. C. M., & Bermond, B. (2001). Validity and reliability of the Bermond-Vorst alexithymia questionnaire. *Personality and Individual Differences*, 30(3), 413–434. [https://doi.org/10.1016/S0191-8869\(00\)00033-7](https://doi.org/10.1016/S0191-8869(00)00033-7).
- Williams, D. (2010). Theory of own mind in autism: Evidence of a specific deficit in self-awareness? *Autism*, 14(5), 474–494. <https://doi.org/10.1177/1362361310366314>.
- Wing, L., Leekam, S. R., Libby, S. J., Gould, J., & Larcombe, M. (2002). The diagnostic interview for social and communication disorders: Background, inter-rater reliability and clinical use. *Journal of Child Psychology and Psychiatry*, 43(3), 307–325. <https://doi.org/10.1111/1469-7610.00023>.
- Zech, E., Luminet, O., Rimé, B., & Wagner, H. (1999). Alexithymia and its measurement: Confirmatory factor analyses of the 20-item Toronto Alexithymia Scale and the Bermond-Vorst alexithymia questionnaire. *European Journal of Personality*, 13(6), 511–532. [https://doi.org/10.1002/\(SICI\)1099-0984\(199911/12\)13:6<511::AID-PER347>3.0.CO;2-0](https://doi.org/10.1002/(SICI)1099-0984(199911/12)13:6<511::AID-PER347>3.0.CO;2-0).

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