An Investigation into the Relationship Between Uncertainty and Negative Affect in Individuals with Autism Spectrum Disorder

By:

Richard Jenkinson

A thesis submitted in partial fulfilment of the requirements for the award of
Doctor of Clinical Psychology at the University of Sheffield

The University of Sheffield
Faculty of Science
Clinical Psychology Unit, Department of Psychology

Submission Date: June 2019

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Declaration

I declare that this work has not been submitted for any other degree at the University of Sheffield, or any other institution. The work presented is original and all other sources have been references accordingly.

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Word Counts

Section One: Literature Review

Excluding references and tables ……..8015

Including references and tables ……...11737

Section Two: Research Report

Excluding references and tables ……..7942

Including references and tables……10836

Total word count

Excluding references and tables ……..15957

Including references and tables ……..……..22573

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Lay Summary (Targeted Towards Research Participants)

People who find it especially hard to cope with the unexpected or unknown are said to have an intolerance of uncertainty. Individuals with autism often report a preference for certainty and experience levels of anxiety that can interfere with their daily life. Understanding more about the link between the intolerance of uncertainty and anxiety in people with autism might lead to better treatments being developed. Therefore, the first part of this thesis aimed to review previous research in order to explore this link.

Twelve studies were found and their results compared and contrasted. In general, people with autism showed very high levels of anxiety and intolerance of uncertainty. Out of ten studies that used relevant statistics, nine found a statistically-significant link between anxiety and intolerance of uncertainty. In general, the strength of the link was about the same as previous research found in people without autism. A person's age and gender did not change the strength of the link, but it appeared slightly stronger in people with autism who scored higher on intelligence tests. There were limitations with this part of the thesis and these are discussed below, together with the implications of this work.

Some adults report being dissatisfied with the assessment process they went through when they were diagnosed with autism; particularly because they felt stressed and anxious due to not knowing what to expect beforehand. Therefore, in part two of this thesis, an intervention was created to address this. The intervention was essentially a short story (with accompanying photographs), describing what it was like to attend an assessment. It was written in a way that research has suggested is helpful for people with autism. Interventions of this type are known as Social Stories.

The people who took part were adults awaiting an assessment at one of two NHS services in the UK. They were divided at random into two groups; those in the first group read a standard leaflet about what to expect, whereas those in the second group read both the Social Story and the

leaflet. People in both groups completed questionnaires at home and on arrival at their assessment. It was expected most would experience an increase in unpleasant emotions (such as anxiety, fear and frustration) on arrival at their assessment, compared to how they felt at home. The results showed that, on average, people who read the Social Story reported significantly less of an increase in unpleasant emotions than those who only read the leaflet. This suggested the intervention was effective. However, the results from a different questionnaire suggested that, in general, the Social Story and the leaflet were equally effective at helping people know what to expect. People in both groups were also equally satisfied with the assessment. The limitations and implications associated with this study are discussed below.

Acknowledgements

I would like to express my gratitude to the people who gave up their time to participate in this research and to those who helped develop the intervention. I wanted to create something that would potentially be beneficial for people with autism and it is my sincere hope that reading the Social Story might make the process of attending a diagnostic assessment a little easier for some.

I would like to thank my research supervisors, Dr Elizabeth Milne and Dr Andrew Thompson. Dr Milne's approach was ideal for me as she provided invaluable guidance, feedback and support; whilst allowing me enough autonomy to conduct a piece of work in my own style. Dr Thompson was influential in the decision to submit an ethical amendment that resulted in a considerable improvement in recruitment. Without this amendment, I honestly do not think the study would have been successful.

I would also like to thank all the staff at the collaborating services which hosted the clinical trial. I was touched by the degree to which staff at the two busy NHS services were so keen to help develop and test an intervention of potential benefit to their clients. I would like to especially acknowledge the efforts of the two collaborating clinicians, Dr Richard Smith and Dr Charmian Round. Both are passionate about offering the best possible service for their clients; and this helped me find the motivation to continue during the over-whelming stages of the thesis. Dr Smith dedicated a great deal of time to ensuring this project was a success and I am extremely grateful. Similarly, Dr Round was essential in helping me open up recruitment to a second service which meant I achieved the required sample size.

Finally, I would like to thank my wife, Liz. Your support and encouragement was invaluable and I intend to make up for all the weekends when I disappeared to the study. And to my two daughters, you helped ensure I maintained perspective about what really matters and stopped me being consumed by this project (after all it's very hard to think about statistics when you're being chased around the garden with a hosepipe).

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Section One

The Relationship Between Intolerance of Uncertainty and Anxiety in People with Autism Spectrum Disorder: A Systematic Literature Review and Meta-Analysis

Prior to formal commencement of the study, a protocol was published on the Prospero database (http://www.crd.york.ac.uk/PROSPERO/display_record.php?ID=CRD42019125315).

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Abstract

Objectives. A systematic review and meta-analysis was conducted of the extant literature to investigate the association between *intolerance of uncertainty* (IoU) and anxiety in people with Autism Spectrum Disorder (ASD). In neurotypical populations, this association has proved robust and has led to effective interventions targeting IoU.

Methods. Scopus, Web of Science, PsycINFO, MEDLINE, White Rose Online and Proquest databases were searched from database inception to 1st March 2019 for relevant articles and dissertations, using search terms related to IoU, anxiety and ASD. A total of 12 studies (comprising 656 high-functioning participants) were included in a systematic review; ten of which were included in a meta-analysis (comprising 562 high-functioning participants). The ages of participants were variable; ranging from 4-years to 70-years (5-years to 24-years in the meta-analysis).

Results. Examining the correlation between IoU and anxiety, the meta-analysis found a large sample-weighted effect size, $r = .62$ [95% CI = .52, .71], $p < .001$. Meta-regression suggested full-scale IQ accounted for a small proportion of the heterogeneity. Subgroup-analyses suggested the association was not significantly impacted by data-informant; but was impacted by the particular research team conducting the study. The systematic review found anxiety and IoU were consistently elevated in individuals with ASD.

Conclusions. IoU and anxiety appear elevated in people with ASD. A large, significant correlation between the two constructs was found; the strength of which was comparable to meta-analyses conducted on neurotypical populations.

Practitioner Points

- IoU and anxiety appear elevated in high-functioning individuals with ASD.

- There is a strong correlation between IoU and anxiety in children and young adults with ASD; the strength of which is consistent with neurotypical populations.
- It does not appear that age or gender impacts on the relationship. IQ appears to have a small moderating effect.
- IoU may be an appropriate target for intervention in this population, but conclusions are limited by the quality of the research.

Limitations

- Only a small number of relevant studies have been conducted, and there are issues with methodological quality.
- The majority of studies have been conducted by a particular research team, and they tended to find stronger correlations than external researchers.
- There is an absence of longitudinal studies meaning the direction of the relationship between IoU and anxiety cannot be fully established.
- Given the observed heterogeneity, it is likely there are more moderators of the relationship that require investigation.

Introduction

Diagnostically, autism spectrum disorder (ASD) is characterised by significant difficulties with social communication/interaction and restricted, repetitive behaviours (American Psychiatric Association, 2013). As it is a neurodevelopmental diagnosis, difficulties are required to have been present during the individual's early life, even though they may become more pronounced as demands and expectations increase with age.

Anxiety and ASD

Approximately 50% of people with ASD have a co-morbid anxiety disorder (Lugnegård, Hallerbäck, & Gillberg, 2011). Anxiety amplifies difficulties with social functioning in this population and is predictive of poorer quality of life (van Steensel, Bögels, & Dirksen, 2012; White, Oswald, Ollendick, & Scahill, 2009). Furthermore, research into the effectiveness of treatments for anxiety have shown high non-response rates in people with ASD (e.g. Storch et al., 2013; Storch et al., 2015; Wood et al., 2015). Therefore, as White et al. (2009) argues, a more thorough understanding of the mechanism(s) underpinning anxiety in this population is required to inform targeted treatments.

Intolerance of Uncertainty

Intolerance of uncertainty (IoU) is a trait characterised by the overvaluation of predictability and the tendency to become overwhelmed by the unexpected or the unknown (Birrell, Meares, Wilkinson, & Freeston, 2011; Carleton, 2016; Koerner & Dugas, 2006). In neurotypical populations, IoU is recognised as a dispositional risk factor in the development of generalised anxiety disorder (Carleton et al., 2012; Freeston, Rhéaume, Letarte, Dugas, & Ladouceur, 1994) and has also been suggested to play a role in social anxiety (Boelen & Reijntjes, 2009), obsessive-compulsive disorder (Holaway, Heimberg, & Coles, 2006) and depression (Carleton et al., 2012). Across diagnostic groups, meta-analytic studies have revealed a robust association between IoU and anxiety in children (Osmanağaoğlu, Creswell, & Dodd, 2018) and in adults (Gentes & Ruscio,

2011). Increased understanding of the association has led to interventions that have aimed to increase tolerance of uncertainty and these have demonstrated effectiveness in the treatment of anxiety (e.g. Dugas et al., 2003; Ladouceur et al., 2000).

Uncertainty and Anxiety in ASD

For individuals with ASD, even slight uncertainty is reported to lead to distress and anxiety; which exacerbates difficulties with social interaction (Ashburner, Bennett, Rodger, & Ziviani, 2013; Bogdashina & Casanova, 2016; Trembath, Germano, Johanson, & Dissanayake, 2012). These qualitative accounts are supported by a limited number of empirical studies. For example, Ivey, Heflin, and Alberto (2004) found that children with ASD showed increased participation in novel social events when they knew what to expect beforehand; and Ferrara and Hill (1980) demonstrated that children with ASD were more likely to interact with toys if they could predict when the toys would be revealled to them.

It has been suggested that people with ASD often lack a theory of mind (meaning individuals are less able to recognise and interpret the behaviour of others and their associated internal states); and that executive difficulties make it more challenging to adapt flexibly to uncertainty (Baron-Cohen, Leslie, & Frith, 1985; Frith, 2003). It would be understandable, therefore, that the social world appears more uncertain for indiviudals with ASD, and that such uncertainty is overwhelming at times. Similarly, other features associated with ASD, such as sensory sensitivites, might motivate a need for predictability in order that aversive stimuli can be avoided (Ashburner, Bennett, Rodger, & Ziviani, 2013). In line with this, some authors (e.g. Joosten, Bundy, & Einfeld, 2009) suggest that insistence on sameness, a core feature of ASD, might function to reduce the anxiety associated with uncertainty.

Recently, the construct of IoU has been investigated in samples of individuals with ASD. A seminal study by Boulter, Freeston, South, and Rodgers (2014) demonstrated that IoU and anxiety were significantly elevated in the group of young people with ASD in the sample (compared with a

neurotypical group). However, once IoU was controlled for, the variance in anxiety accounted for by diagnosis was no longer significant; suggesting IoU might mediate the association between ASD and anxiety. In addition to increased IoU potentially accounting for the elevated anxiety commonly observed in individuals with ASD, results from additional analyses conducted by the study authors suggested that the relationship between IoU and anxiety functioned similarly in individuals with and without ASD. This might mean that individuals with ASD who experience debilitating anxiety could benefit from interventions targeting IoU.

The Current Review

The research into the association between IoU and anxiety in ASD is still in its infancy and, to the best of the author's knowledge, there has not been an associated systematic review or meta-analysis. However, a scoping search of the literature suggested there is a growing evidence-base. Furthermore, as researchers have begun piloting anxiety interventions that target IoU in people with ASD (e.g. Rodgers et al., 2017), it seems warranted the current knowledge is collated and analysed, to potentially help inform this work. Therefore, this systematic review and meta-analysis has the primary aim of examining the strength and pattern of the association between IoU and anxiety in children and adults with ASD.

A secondary aim will be to explore the variability in the research and the effect of potential moderators such as age and IQ. There is reason to suspect the association might present differently in people of different ages and abilities because, in typical development, the cognitive faculties required to detect and reflect upon uncertainty are likely to mature with age (Osmanağaoğlu et al., 2018). Gender will also be explored given mixed findings were reported by Boulter et al. (2014). Although the meta-analysis will focus on the association between IoU and anxiety, the narrative synthesis will summarise the broader collection of empirical studies investigating IoU and anxiety in people with ASD, to shed further light on the association.

As there is little consensus in regards to behavioural or physiological measures that are valid for assessing anxiety in this population (Lydon et al., 2016; Vasa et al., 2016), this review will parallel meta-analyses conducted with neurotypical populations (e.g. Gentes & Ruscio, 2011; Osmanağaoğlu et al., 2018) by including only questionnaire measures. Where studies report both self- and other-reported versions (e.g. child- and parent-reported measures), self-reported data will always take precedence. This is because research has suggested there is often a discrepancy between self-reported and proxy-reported data in relation to people with ASD; and that individuals are often better-informants of their IoU (Comer et al., 2009). This parallels the approach taken by Osmanağaoğlu et al. (2018).

Research Questions

- What is the strength and pattern of the association between IoU and anxiety in individuals with ASD?
- How does the association compare to that observed in individuals without ASD?
- Is the relationship moderated by age, gender , IQ or informant-type (self-report versus proxy-report)?

Method

Search Strategy

Prior to formal commencement of the study, a protocol was published on the Prospero database (http://www.crd.york.ac.uk/PROSPERO/display_record.php?ID=CRD42019125315). Four electronic databases (Scopus, Web of Science, PsycINFO and MEDLINE) were searched from database inception (to 1st March 2019). Two electronic research repositories (White Rose Online and Proquest) were searched to retrieve unpublished studies, in order to reduce publication bias. The Cochrane Library was searched to identify existing reviews on this research topic. Cited references from eligible articles were searched manually.

The titles, abstracts and keywords of databases were searched using terms related to ASD, anxiety and IoU. Table 1 gives an overview of the strategy (database-specific search strings are presented in Appendix A).

Table 1

Overarching search strategy

"anxiety" "fear" "GAD" "OCD" "compulsive disorder" "panic"	AND	"autism" "ASD" "ASC" "PDD" "Asperg*" "pervasive developmental disorder" "Pathological Demand" "PDA"	AND	"Intolerance"	AND	"uncertainty"

Note. OR used as operator between items in each column

Screening

The search retrieved 405 articles. There were 113 duplicates removed and the remaining 292 articles were screened for relevance. After 219 irrelevant records were excluded, the full text of the remaining 73 articles was reviewed and examined using a priori inclusion and exclusion criteria. To be included, articles were required to have been available in English and to have included original research in which questionnaire measures of both IoU and anxiety were used to report on individuals with autism (either via self-report or via proxy). To be included in the meta-analysis, studies were required to have reported the correlation between IoU and anxiety. However, if this data was not available, studies were still included in the narrative synthesis if they made comparisons between an ASD group and a neurotypical group (on IoU and anxiety). Studies were excluded if they used data from an earlier published study, or if they used single-case designs. After the inclusion and exclusion criteria were applied, 12 studies remained and were included in the narrative review. Ancestry searches were conducted but no additional articles meeting the inclusion/exclusion criteria were discovered. Additional data was requested from authors where necessary. Ten out of the 12 studies

were included in the meta-analysis (two on the basis of data supplied via email). A diagrammatic representation of the process can be seen in Figure 1.

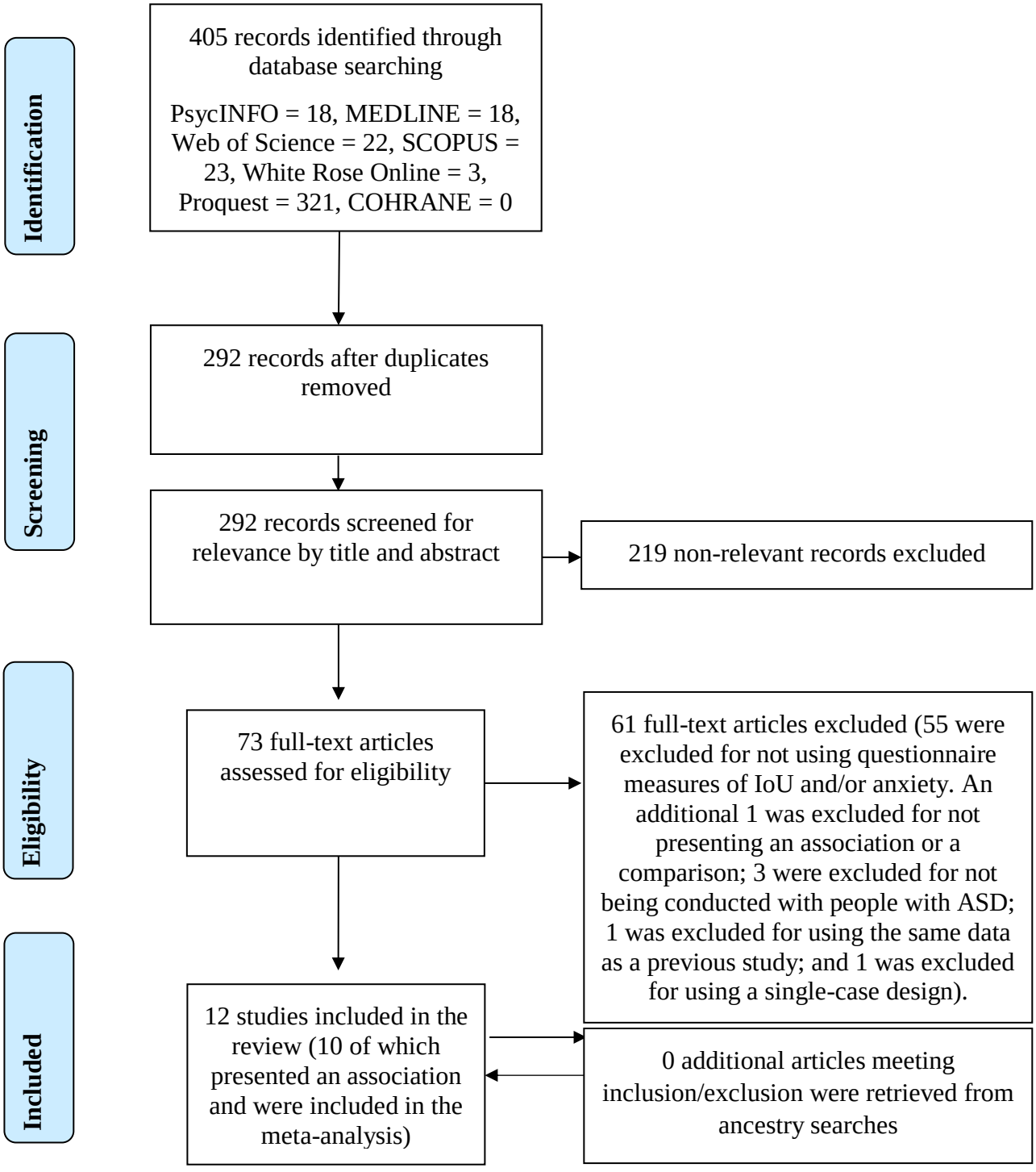

Figure 1 PRISMA flow diagram representing the selection of studies included in the review

Quality Appraisal

Studies were appraised using a quality appraisal checklist for correlational studies (The National Institute for Health and Clinical Excellence, 2012). This tool was chosen as the majority of its items were relevant to this review and enabled the assessment of internal and external validity. Quality was denoted with a double cross (++) next to a study that fully met the criteria for an item; a single cross (+) if criteria were partially-met, and a minus sign (-) if criteria were not met. The checklist included two summary items in which an overall rating of the study's internal and external validity was made (using the same scoring metric). Please see Appendix B for a copy of the checklist. The ratings from this checklist, together with the reasoning, was integrated into the narrative of the review. In addition, each study was assigned an overall quality score (calculated as a percentage) to aid inter-study comparisons. Please see Appendix C for full details.

The wording of checklist item 2.3 was changed as it pertained to potential contamination between an exposure and comparison group (which was not relevant to the present review). To fulfil a similar criterion, the revised item specified whether a diagnosis of ASD was confirmed independently by the researchers, as this minimised bias by ensuring the study only included participants who had autism. Modifying checklists in the manner described above consistent is consistent with guidance from the Centre for Reviews and Dissemination (2008).

Statistical Analyses

Pearson's product-moment correlation-coefficient (*r*) was selected as the effect size for the meta-analysis, due it being easily interpretable and a popular choice for meta analyses between IoU and anxiety conducted with neurotypical populations (e.g. Gentes & Ruscio, 2011; Osmanağaoğlu et al., 2018); facilitating comparisons. Analyses were performed using the software package, Comprehensive Meta Analysis (Borenstein, Hedges, Higgins, & Rothstein, 2005). A random-effects model was selected due to anticipated heterogeneity between studies and because it permits results to more readily be generalised (Chen & Peace, 2013). To interpret the correlations, guidelines by

Cohen (1988) were used to define small, moderate and large effects (r = .10, r = .30, r = .50 respectively) and 95% confidence intervals were calculated. To correct for skewed sampling distribution when population values of r move further from zero, correlations were transformed to Fisher's Z for meta-analytic computations (Cooper & Hedges, 1993).

To aid visual inspection of the data, funnel plots and forest plots were produced. A regression test (Egger, Davey Smith, Schneider, & Minder, 1997) was also used to assess publication bias. Fail-safe analysis (Rosenthal, 1979) was conducted to aid this assessment by quantifying the number of studies that would be required to invalidate the effect (Borenstein, Hedges, Higgins, & Rothstein, 2011).

In order to assess heterogeneity, the Q and I^2 statistics were used. Significant results indicate heterogeneity. Higgins, Thompson, Deeks, and Altman (2003) suggest that I^2 percentages of 25%, 50% and 75% can be interpreted as representing low, moderate and high heterogeneity, respectively.

Heterogeneity was explored using potential moderators specified a priori. Meta-regression was planned for numerical moderators (age, percentage male, IQ). Sub-group analyses were planned to examine the effect of informant-type and instrument-selection on the relationship between IoU and anxiety. However, the latter analysis was not conducted given it was specified a priori that there needed to be at least four studies in each subgroup (Bakermans-Kranenburg, Van Ijzendoorn, & Juffer, 2003).

Results

Of the twelve studies included in the narrative review; two (Chamberlain et al., 2013; Maisel et al., 2016) were excluded from the meta-analysis as a correlation was not available. Of the 12, there were nine that were cross-sectional. There were two (Chamberlain et al., 2013; Damiano, 2015) that measured neuro-physiological measurements during conditions of artificially-induced uncertainty and one (Keefer et al., 2017) that used a pre-post, controlled-experimental design to measure the

effectiveness of a cognitive-behavioural intervention. However, in these three studies, questionnaire data were available at baseline and so, for the purposes of this review, all studies were considered cross-sectional. Between-group (ASD; neurotypical) comparisons were made at baseline in six of the studies.

Participants

As can be seen from Table 2, the 12 included studies comprised 656 participants (562 in the meta-analysis). Three studies (Keefer et al., 2017; Vasa, Kreiser, Keefer, Singh, & Mostofsky, 2018; Wigham, Rodgers, South, McConachie, & Freeston, 2015) reported being embedded in larger studies. This was investigated by retrieving further details about the three larger studies (contacting authors where necessary) and it was confirmed there was not participant-overlap. One study (Boulter et al., 2014) reported combining archival data (from a separate study) with primary data. However, it was confirmed through investigation that the archival data was not from another study in the review. Therefore, it was assumed all studies used independent participants.

The ages of participants were variable; ranging from 4 years to 70 years (4 years – 24 years in the meta-analysis). There were nine studies with samples comprising child and adolescent participants (with ages that ranged from 4 years to 18 years), one of which was excluded from the meta-analysis. There was one study that used adult participants (but was excluded from the meta-analysis), and two studies that included both teenagers and young adults (with ages which ranged from 13 – 24). The samples of all studies comprised predominately males (ranging from 70.5% to 94.4%). Dates of publication were all within the last seven years.

All studies used participants recruited from Western, English-speaking countries (one from Australia, four from the USA, four from the UK and three from both the UK and USA). The most popular method of recruitment was via a research database, with nine studies mentioning this formed at least part of their recruitment strategy (Boulter et al., 2014; Cai, Richdale, Dissanayake, & Uljarević, 2018; Damiano, 2015; Joyce, Honey, Leekam, Barrett, & Rodgers, 2017; Maisel et al.,

2016; Neil, Olsson, & Pellicano, 2016; Rodgers et al., 2017; Vasa et al., 2018; Wigham et al., 2015).

Additional sources of recruitment included schools (Cai et al., 2018; Glod, 2017; Joyce et al., 2017; Neil et al., 2016; Vasa et al., 2018); clinicians (Boulter et al., 2014; Cai et al., 2018; Keefer et al., 2017; Vasa et al., 2018); the internet (Joyce et al., 2017; Neil et al., 2016); GPs and community events (Vasa et al., 2018); and local newsletters (Glod, 2017). Owing to the origins of the IoU research in people with ASD, there were seven studies which included at least a partial collaboration with the research team at Newcastle University (Boulter et al., 2014; Chamberlain et al., 2013; Glod, 2017; Joyce et al., 2017; Maisel et al., 2016; Rodgers et al., 2017; Wigham et al., 2015)

Authors of eleven of the studies reported using participants diagnosed with ASD, although one of these (Boulter et al., 2014) reported their sample also included participants with Asperger's Syndrome and one (Cai et al., 2018) reported including participants with Asperger's Syndrome and Autistic Disorder. The remaining study (Neil et al., 2016) did not specify and referred to the participants as "autistic children" (p. 1964).

Eight studies measured full-scale IQ. Participants had a combined mean of 105.4 (SD = 15.2). The six studies measuring IQ in the meta-analysis had a combined mean of 103.5 (SD = 15.3). An additional study had non-verbal and verbal IQ scores within one standard deviation of the general-population mean and the remaining three reported excluding participants with intellectual disability.

Instruments and Data Analysis

As can be seen from Table 1, ten studies used a variant of the Intolerance of Uncertainty Scale (Buhr & Dugas, 2002); with seven of these opting to use the 12-item version of the original, 27-item scale (five of which were included in the meta-analysis). The remaining two studies used the IoU

subscale of the Anxiety Scale for Children-ASD (ASC-ASD; Rodgers et al., 2016). They met inclusion criteria because both studies included a separate anxiety measure.

In terms of anxiety, six studies used the Spence Children's Anxiety Scale (SCAS; Spence, 1998), although one was excluded from the meta-analysis. Glod (2017) combined SCAS *T*-scores with *T*-scores from the pre-school version of the same measure (PAS; Edwards, Rapee, Kennedy, & Spence, 2010) which the authors used to measure anxiety in the younger participants in the sample. Four studies used the Screen for Child Anxiety Related Emotional Disorders (SCARED; Birmaher et al., 1997), one used the Dimensional Anxiety Scales (DAS; American Psychiatric Association, 2013) and one (excluded from the meta-analysis) used the State-Trait Anxiety Inventory (STAI; Spielberger, 2010). All studies used trait measures of IoU and anxiety.

Studies used a variety of data-analytic techniques. Between-group (ASD; neurotypical) differences in IoU, anxiety and demographic variables (age, gender, IQ, country) were assessed using independent/paired samples *t*-tests (or Mann-Whitney *U* tests) and Analysis of Variance (ANOVA). Correlation analyses (Pearson *r*) were used to explore associations between IoU and anxiety, in addition to associations between these variables and demographic variables, and their relationship with the core features of ASD. Regression, structural equation modelling and mediator analyses were used to explore the relationship between IoU and anxiety and the role of ASD core features and demographics.

Quality Assessment Summary

Overall quality ratings for included studies was variable, with scores ranging from 30% to 82% (38% to 82% in the meta-analysis). Given the limited number of studies available, none were excluded on the basis of quality. Please see Appendix B for full details.

In general, studies scored better on items pertaining to internal validity than external validity. Internal validity, as assessed by the checklist, concerned the way the study was conducted (within the limitations of correlational research). All studies fully met criteria (++) for item 2.2 that specified

authors must have provided sound, theoretical reasons for their selection of variables. There were eleven out of twelve studies that confirmed participants had a valid diagnosis of ASD (item 2.3), either using independent gold-standard diagnostic tools (five studies, ++; three in the meta-analysis) or by confirming self-reports utilising established ASD screening tools and excluding participants who did not meet clinical thresholds (six studies, +). One study (Joyce et al., 2017) used a screening tool but did not exclude participants with sub-threshold scores (-).

None of the studies fully met criteria for item 3.1, which pertained to whether outcome measures were reliable. This was primarily because none of the studies used an IoU measure that had been validated for people with ASD (several commented that one was not available). There were seven studies that checked the internal consistency for the IoU measure (values ranged from acceptable to excellent). All seven were included in the meta-analysis. The remaining five did not check. Only one study (Boulter et al., 2014) had missing data that was not accounted for (item 3.2). There were only two studies (Boulter et al., 2014; Rodgers et al., 2016) that reported making an a priori power calculation and achieving adequate power so these were the only two that met full criteria (++) for item 4.1. The precision of the studies (item 4.6) was highly variable, with only three (Boulter et al., 2014; Neil et al., 2016; Rodgers et al., 2016) meeting full criteria. The analytical methods used in the data analysis (item 4.3) were generally appropriate, with all studies meeting at least partial criteria.

A common external validity issue was a lack of detail about the source population (item 1.1.), the detail and representativeness of the eligible population (item 1.2), and how the clinical and demographic characteristics of these populations compared with the participants in the sample (item 1.3). Only one study (Rodgers et al., 2016) met full criteria for these items. There was generally lack of detail about recruitment and the details of those who were eligible but declined. All studies used a convenience sample. External validity was also informed by item 2.3 (discussed above) and item 2.5, pertaining to whether the setting was applicable to the UK. Given the population demographics, all studies met at least partial (+) criteria for item 2.5.

Levels of IoU and Anxiety

Of the six studies that drew comparisons between participants with ASD and a neurotypical group, all six reported finding the ASD group had significantly higher IoU and anxiety (with the majority reporting a large effect size). However, there was some discrepancy when researchers made these comparisons with both child-reported and parent-reported data (see below). There were three studies which cited the percentages of ASD participants who scored above cut-offs for clinically-significant anxiety. Cai et al. (2018) reported a figure of 43%, Joyce et al. (2017) reported 46% and Boulter et al. (2014) 60%. Boulter reported this compared with only 12% of the neurotypical group.

Discrepancies Between Informants

When comparing self- and parent-reported measures, Joyce et al. (2017) analysed data using intraclass correlation-coefficients and found a high level of agreement on anxiety (.66) but not IoU (.12). Vasa et al. (2018) and Chamberlain et al. (2013) both found ASD and neurotypical groups differed significantly on anxiety according to parent-report, but not according to self-report. Chamberlain et al. found the same pattern for IoU. Damiano (2015) found a significant correlation between parent and self-reported IoU in the neurotypical-group, but not in the ASD-group. Keefer et al. (2017) did not find a significant difference between parental- and self-reported IoU.

The Impact of Participant Demographics on IoU and Anxiety.

Gender. Cai et al. (2018) found anxiety was significantly higher in females with ASD than in males, with a medium effect size. Across a sample of ASD/ neurotypical participants, Boulter et al. (2014) found parents of girls reported significantly more anxiety than parents of boys. However, there were no gender differences in self-reported data. Results from ANOVAs showed there was not a significant main effect of gender on IoU. Similarly, three studies (Glod, 2017; Neil et al., 2016; Wigham et al., 2015) examining parent-data did not find a significant correlation between gender and IoU or anxiety. However, Cai et al. (2018) found females with ASD self-reported significantly more IoU than males.

Age. Wigham et al. (2015) found age was not significantly correlated with IoU, but was inversely correlated with anxiety, such that younger children scored higher. However, Glod (2017) and Neil et al. (2016) did not find age correlated with IoU or anxiety. All of these studies used parental reports. Similarly, Damiano (2015) found age was not significantly correlated with IoU when using parental data, but when examining self-reported data, he found age was significantly positively correlated with IoU in the ASD group (but not in the neurotypical group). However, Damiano scored 38% (- +) for quality; the study lost marks on item 4.3 which pertained to whether the analytical methods were appropriate. This was primarily because he examined 44 correlations but did not apply a Bonferroni correction (or similar) to minimise the risk of making a Type 1 error.

IQ. Three studies (Glod, 2017; Neil et al., 2016; Wigham et al., 2015) examining parent-data found IQ was not significantly correlated with anxiety or IoU. Damiano (2015) found a positive correlation between IQ and IoU in the neurotypical group, but not the ASD group when using parent-reported data. When he used self-reported data, however, there was not a significant correlation in either group.

The Relationship between IU and Anxiety in People with ASD

Keefer et al. (2017) used combined self-reported and parent-reported data to group children with ASD into a high and low IoU group. All participants in the high IoU group were found to have clinically-significant anxiety at baseline; compared with 65% in the low IoU group.

Self-reported data. Five studies (Boulter et al., 2014; Cai et al., 2018; Joyce et al., 2017; Rodgers et al., 2016; Vasa et al., 2018) reported finding a large, significant association between self-reported anxiety and IoU.. The quality of these studies was variable; from Joyce et al. (2017), who scored 46% (-, -); to Rodgers et al. (2016), who scored 82% (NA, ++). Rodgers et. al was the only study to receive the overall ++ rating for external validity. The researchers recruited participants from two UK databases so, arguably, a limited pool. However, they cited research that had compared children and families on the databases to the source population and found they were comparable in

terms of gender and socioeconomic status. They also provided comprehensive demographic and clinical data on participants and compared participants to those children whose families did not respond to the invitation to participate. The researchers found no significant differences in gender, age, type of diagnosis, age at diagnosis or anxiety and, therefore, the reader could be reasonably confident that participants were representative of the eligible population (people on the database) and the source population (UK children with ASD between 8-years and 15-years, without speech difficulties or a co-morbid diagnosis). Unfortunately, as the study was designed to test a new measure (rather than the association between IoU and anxiety), it was not appropriate to subject it to same criteria for internal validity assessment as the other studies because the design was significantly different (data on the association was supplied on request by the author). However, it was one of the only studies to conduct an a priori power analysis and used a large sample size (N = 112). Its primary limitation in relation to internal validity was that the IoU measure was a subscale of a new questionnaire. Although, the researchers found good internal consistency of the 8-item uncertainty subscale (and established excellent test-retest reliability and convergent validity of the overall measure), the psychometric properties of the subscale as a measure of IoU are unknown and it might represent a different construct to that captured by the primary IoU measure used in the review (the IUS).

Keefer et al. (2017), 38%; ++ reported finding a moderate, significant association between self-reported IoU and anxiety in their USA-sample of children with ASD aged 8-years to 14-years. Keefer et al. recruited child and adolescent participants who had been diagnosed utilising the gold-standard ADOS-assessment, from three University-clinics in the USA. The researchers excluded those with intellectual disability and provided adequate demographic data on participants for making comparisons with the source population. In terms of internal validity, the biggest limitation was that the researchers modified the IUS for their study, by changing the language to make it more suitable for people with ASD. Whilst this was not necessarily a weakness, it potentially invalidated the psychometric properties of the measure. However, this was arguably less of an issue considering the

lack of research that has validated the measure in children with ASD. Furthermore, the researchers' modifications were approved by the developer of the original scale and they checked internal consistency and found it to be good.

Damiano (2015) (38%; - +) did not find a significant association in the ASD group, nor in the neurotypical group. As described above, the study had limited external validity and some limitations with internal validity.

Parent-reported data. Contrary to the findings obtained using self-reported data, Damiano (2015) found a large, significant association between IoU and anxiety whilst utilising parent-reported data. Two studies (Joyce et al., 2017; Vasa et al., 2018) presented correlations using parent-data to supplement the self-reported data. Both found significant correlations; Joyce et al. (46%; - -) observed a large effect size, whereas Vasa et al. (69%; + +) reported a moderate effect size. Joyce et al. lost marks for internal and external validity due to potential concerns around the diagnostic purity of the sample. The researchers relied on self-reports and teacher-reports for some of their participants and, although they used an established screening tool, they included three participants who failed to meet the clinical threshold. Therefore, this introduced a potential bias into the study by including participants who might not have had a valid diagnosis.

Three other studies (Glod, 2017; Neil et al., 2016; Wigham et al., 2015) also found a large, significant association utilising parent data. Neil et al. (73%; ++, +) was a UK-based study and, although a convenience sample was used, the diversity of recruitment methods used might have feasibly increased the representativeness of the eligible population. Furthermore, the researchers independent validated the ASD diagnosis by administering the ADOS and SCQ and excluded two participants for scoring below the diagnostic threshold. They also excluded those with intellectual disability after administering a standardised intelligence test. Established measures of IoU and anxiety were used and data analytic methods were appropriate (e.g. conducting tests of normality on the data and using a significance level of .01 to account for the number of correlations conducted), increasing

confidence in internal validity. Wigham et al. 58% (+, +) recruited participants from two University databases in the UK and USA, which arguably reduced external validity as people expressing strong willingness to participate in research studies (by consenting to inclusion on the databases) might not be representative of the wider population. Acceptable demographic data were provided on participants. Although they did not use a gold-standard measure to validate the ASD diagnosis, they did administer the SRS screening tool (although did not specify thresholds for inclusion in the study). They also assessed IQ independently, although this too, was only used descriptively. Data were analysed appropriately (e.g. conducting tests of normality on the data; using t-tests to examine differences in sample characteristics between countries). The researchers were also transparent about how they handled missing data and outliers.

Comparisons with Neurotypical Individuals

Three studies reported correlations between the primary measures in both ASD and neurotypical groups. Damiano (2015) compared SCARED scores and self-reported IoU and found precisely the same non-significant correlation (r = .16) in both the neurotypical and ASD groups. However, when parent-reported IoU was used, significant correlations with SCARED scores were found in both groups; but the ASD group was of a much larger effect size (r = .79, p < .01) than the neurotypical group (r = .44, p < .05). Neil et al. (2016) also correlated anxiety (SCAS) with parent-reported IoU and, like Damiano, found a larger correlation in the ASD group (r = .74, p < .01) than the neurotypical group (r = .59, p < .01). Furthermore, in a regression model, IoU directly predicted anxiety (b = 0.67, p < .001) in both the neurotypical group and the ASD group (b = 1.35, p < .001), but the beta-coefficient was considerably larger in the latter group. Vasa et al. (2018) found significant correlations between parent-reported anxiety (SCARED) and self-reported IoU, but the effect size was considerably larger in neurotypical group (r = .71, p = .005) than in the ASD group (r = .40, p = .005). However, self-reported anxiety and self-reported IoU were only correlated significantly in the ASD group (r = .46, p = .005).

ASD as a Predictor

Vasa et al. (2018) found a diagnosis of ASD was predictive of IoU and that this was not fully accounted for by the effect of anxiety. Neil et al. (2016) found a large, significant, indirect-effect of a diagnosis of ASD on anxiety, through IoU (without an accompanying direct effect). Maisel et al. (2016) used structural equation modelling to investigate this relationship, with scores from the Autism Spectrum Quotient (Baron-Cohen, Wheelwright, Skinner, Martin, & Clubley, 2001) representing ASD-severity. Similarly to Vasa et al. and Neil et al., the researchers found severity predicted IoU and that IoU partially-mediated the association between severity and anxiety (accounting for 36% of the effect). When IoU was controlled for, severity did not predict anxiety.

Two studies (Glod, 2017; Wigham et al., 2015) found sensory hyper-responsiveness correlated significantly with IoU. Furthermore, Wigham et al. (2015) conducted a regression-analysis that revealed a significant, serial, indirect effect from sensory-responsiveness through IoU and anxiety to insistence on sameness.

Table 2

Summary of findings from primary outcome measures

Study	Country	ASD sample size, gender, IQ (SD)	Age (SD), range	IoU measure	Anxiety measure	Correlation	Additional Findings	Quality
Boulter et al. (2014)	UK, USA	N = 114 88% male IQ = 108.5 (13.8)	12.7 (2.9) 8 - 18	IUS-12 (Child); IUS-12 (parent)	SCAS (child); SCAS (parent)	0.70** (data from author via email)	ASD group had significantly higher anxiety than TD group (60 % of the had clinically-significant anxiety, versus 12% of the TD group). ASD group had significantly higher IoU than TD group, with a medium effect size. No significant main effect of gender on IoU. No effect of Country on anxiety or IoU, or interaction with diagnosis.	62% (+,+)
Cai et al. (2018)	Australia	N = 61 70% male IQ = NR	18.2 (2.2) 14 - 24	IUS-12	DSM-5 DAS	0.63**	43% of participants scored above threshold for clinically-significant anxiety. Females had significantly higher anxiety and IoU scores, with a moderate effect size.	46% (+,+)
Chamberlain et al. (2013)	USA	N = 18 94% male IQ = 104.8 (13.3)	16.6 (1.0) 15 - 18	IUS-12 (Child); IUS-12 (parent)	SCAS (child); SCAS (parent)	Not available so excluded from meta-analysis	Based on parental report, ASD group had significantly higher anxiety and IoU than TD group. However, no significant differences found on child data.	30% (NA, -)
Damiano (2015)	USA	N = 26 92% male IQ = 105.69 (17.5)	14.1 (3.2) 9 - 18	IUS-27 (child); IUS-27 (parent)	SCARED (parent)	0.16	ASD group had significantly higher anxiety and IoU than TD group. Significant correlation found in TD group between parent and child-reported IoU, but not in ASD group. Parents reported significantly more IoU than children in ASD group; the opposite pattern was found in TD group. Based on self-report data, IoU scores were significantly correlated with age in the ASD group, but not in the TD group. Based on parental-reported data, IoU was not significantly associated with age in either group. Based on self-report data, there were no significant correlations between IoU and IQ in either group. Parent data showed IoU was significantly correlated with IQ in the TD group, but not in the ASD group. No correlations with ASD severity were found.	38% (-, +)

Glod (2017)	UK	N = 19 (parents) 84% male Verbal IQ = 89.7 (13.6)	7.21 (1.8) 4 – 9	IoU subscale of ASC-ASD (parent)	T-scores from the SCAS/PAS (parent)	0.83*	IoU and anxiety were not significantly correlated with age, gender or IQ. IoU significantly correlated with sensory sensitivity and insistence on sameness, but not with repetitive motor/sensory behaviour.	42% (+, -)
Joyce et al. (2017)	UK	N = 13 84% male IQ = NR	16.8 (2.4) 13 - 20	IUS-12 (Child); IUS-12 (parent)	SCAS	0.82**	46.2 % of sample had clinically-significant anxiety. High level of agreement between parent and child reported anxiety but not IoU. However, a significant correlation was found between parent-reported IoU and anxiety (as it was using self-report data). Significant correlation between parent-reported IoU and repetitive motor/sensory behaviour, and with rigidity/routines/restricted interests. However, no significant correlations were found on self-reported measures.	46% (-, -)
Keefer et al. (2017a)	USA	N = 43 81% male IQ = 102.6 (14.7)	11.2 (2.0) 8 - 14	IUS-27 (child, modified language); IUS-27 (parent)	SCARED (child); SCARED (parent)	0.36*	No significant correlation been child and parent reported IoU. Grouping participants by IoU severity, 100% of the high IoU group had clinically-significant levels of anxiety (compared with 65% of the low IoU group). A brief CBT intervention did not significantly improve IoU or anxiety. IoU predicted the effectiveness of the intervention on anxiety.	38% (+, +)
Maisel et al. (2016)	UK, USA	N = 76 78% male IQ = 111.2 (15.0)	33.8 (14.9) 17 - 70	IUS-12	STAI-T (Form-Y).	Not available so excluded from meta-analysis	Based on self-report, ASD group had significantly higher anxiety and IoU than TD group. Across the sample (combining the ASD and TD groups), the UK site had significantly higher anxiety than the US site but there was not a significant difference in IoU. Using Structural Equation Modelling on a combined sample of participants (with and without ASD), ASD severity predicted more IoU, but not anxiety (whilst controlling for IoU). IoU predicted anxiety and partially mediated the relationship between ASD severity and anxiety (accounting for 36% of the variance).	55% (NA, -)
Neil et al. (2016)	UK	N = 64 (parents) 86% male IQ = 98.6 (14.9)	10.4 (2.4) 6 - 14	IUS-12 (parent)	SCAS (parent)	0.74**	ASD group had significantly higher anxiety and IoU than TD group. In the ASD group, there was a significant correlation between IoU and ASD severity (based on SCQ scores but not ADOS scores). IoU fully mediated the relationship between ASD diagnosis (yes or no) and anxiety.	73% (++, +)

							In the ASD group, IoU explained 45% of the variance in sensory sensitivity (IoU did not have a direct effect on TD sensory sensitivity scores). The relationship between IoU and sensory sensitivity was partly mediated by anxiety, with a large effect size. IoU and anxiety were not correlated with gender, age or IQ.	
Rodgers et al. (2016)	UK	N = 112 77% male IQ = NR	11.1 (2.1) 8 - 15	IoU subscale of ASC-ASD (child) IoU subscale of ASC-ASD (parent)	SCARED (child); SCARED (parent)	0.72* (data from author via email)		82% (NA, ++)
Vasa et al. (2018)	USA	N = 57 83% male IQ = 100.9 (14.9)	10.9 (2.0) 7 - 16	IUS-27 (child, modified language) IUS-27 (parent)	SCARED (child); SCARED (parent)	0.46*	No significance between self-reported anxiety in ASD and TD groups. However, parents reported significantly more anxiety in ASD group than TD group. Based on both self-reported and parent-reported data, ASD group had significantly higher IoU than TD group. Across the sample, children reported more IoU than their parents. In addition to finding a significant association in the ASD using self-reported data, a significant correlation was found between parent-reported IoU and anxiety, of a moderate – high effect size. Based on parental data, the correlation between IoU and anxiety was stronger in the TD group than the ASD group. However, based on self-report data, there was not a significant association. A regression analysis showed that, when controlling for anxiety, ASD severity significantly predicted IoU (but when emotional dysregulation was added to the model, only this variable significantly predicted IoU). In the ASD group, significant relationships were found between IoU and repetitive behaviour, but not with social communication difficulties.	69% (+, +)
Wigham et al. (2015)	USA, UK	N = 53 89% male	12.50 (2.3) 8 - 16	IUS-12 (parent)	SCAS (parent)	0.57*	IoU was not significantly correlated with age, gender, IQ or Country. IoU was not significantly correlated with social responsiveness or sensory hypo-responsiveness. IoU was significantly correlated with	58% (+, +)

	IQ = 106.2 (14.8)	sensory hyper-responsiveness, insistence on sameness and repetitive motor behaviour. Anxiety inversely correlated with age and sensory over-responsiveness and positively correlated with social responsiveness and insistence on sameness and repetitive motor behaviours. Anxiety was not significantly associated with Country, gender or IQ. Mediation analyses provided evidence of a serial path from sensory responsiveness, through IoU and anxiety, to insistence on sameness behaviours (but not to repetitive motor behaviours).

Note: SD, Standard deviation; NR, Not reported; *, significant at $p < .05$ level; **, significance at $p < .01$ level; ASD, Autism Spectrum Disorder; (Child), Child-informant version of the measure; (Parent), Parent-informant version of the measure; TD, typical development; IoU, Intolerance of uncertainty; IoUS-12, Intolerance of Uncertainty Scale (12 item version); IoUS-27, Intolerance of Uncertainty Scale (27 item version); DSM-5 DAS, Diagnostic and Statistical Manual of Mental Disorders (5th Edition) Dimensional Anxiety Scales; SCAS, Spence Children's Anxiety Scale; PAS, Preschool Anxiety Scale; ASC-ASD, Anxiety Scale for Children- ASD; SCARED, Screen for Child Anxiety Related Disorders; STAI-T (Form Y), State-Trait Anxiety Inventory (trait version); SCQ, Social Communication Questionnaire; ADOS, Autism Diagnostic Observation Schedule.

Meta-Analysis

The sample-weighted effect size was r = .62, 95% CI [.52, .71] and significant ($p < .001$); which suggested a large, positive correlation between IoU and anxiety. The Q statistic was significant $Q(9) = 28.84$, $p = .001$, and the I^2 (69%) statistic was moderate-high; suggestive of heterogeneity in the data. The corresponding Forest Plot is shown in Figure 2.

	Correlation	Lower limit	Upper limit	Z-Value	p-Value
Boulter et al. (2014)	0.70	0.59	0.78	9.14	.00
Cai, Richdale, Dissanayake, & Uljarević (2018)	0.63	0.45	0.76	5.68	.00
Damiano (2015)	0.16	-0.24	0.52	0.77	.44
Glod (2017)	0.83	0.60	0.93	4.70	.00
Joyce, Honey, Leekam, Barrett, & Rodgers (2017)	0.82	0.48	0.94	3.63	.00
Keefer et al. (2017)	0.36	0.07	0.60	2.38	.02
Neil, Olsson, & Pellicano (2016)	0.74	0.60	0.83	7.42	.00
Rodgers et al. (2016)	0.72	0.62	0.80	9.52	.00
Vasa, Kreiser, Keefer, Singh, & Mostofsky (2018)	0.46	0.23	0.64	3.65	.00
Wigham, Rodgers, South, McConachie, & Freeston (2015)	0.57	0.35	0.72	4.53	.00
	0.62	0.52	0.71	8.85	.00

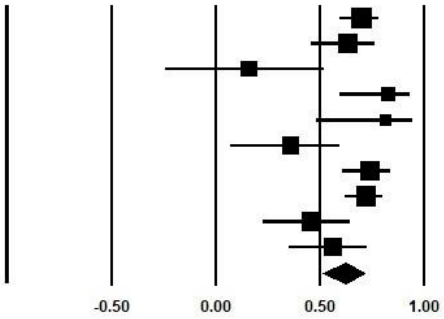

Figure 2 Meta-Analysis Forest Plot

A funnel plot was produced, with the effect size from each study on the x axis, and a measure of study precision – indicated by standard error of effect size on the y axis (see Figure 3). Because greater variability is expected in less precise studies, the dots (representing the individual studies) were expected to have shown greater dispersion at the bottom of the graph; with increased clustering around the mean effect size (vertical line) as precision increased.. Although sample size has often been used on the y axis in funnel plots, Sterne and Egger (2001) recommend that standard error now be used because the plots it produces facilitate the assessment of bias (due to the probability that the dispersion will approximate an inverse funnel shape in the absence of bias, that diagonal lines can be added to indicate 95% confidence limits, and that the plot emphasises smaller studies with a greater risk of bias).

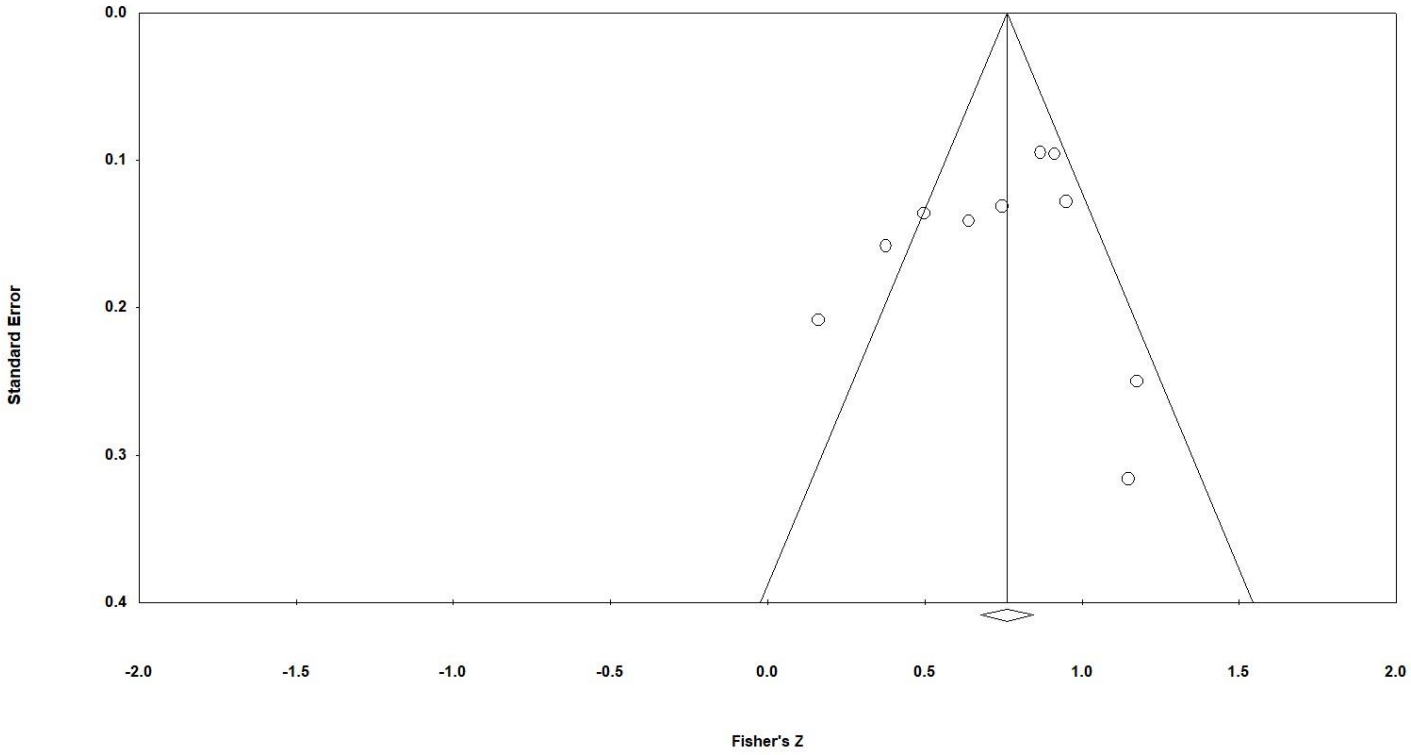

Figure 3 Funnel Plot

The asymmetry of the funnel plot indicated evidence of potential publication bias and two studies fell outside the 95% confidence limits. However, the low number of studies included in this meta-analysis clearly limit interpretation of this plot. Examining the symmetry statistically via a regression test indicated there was not significant evidence of publication bias ($t(8) = 0.62$, $p = .56$). Furthermore, the fail-safe analysis indicated that 679 missing studies would be required to bring the p-value to > .05.

Heterogeneity was explored using subgroup analyses and meta-regression. For numerical variables, meta-regression analysis revealed there was not a significant effect of age ($Q(1) = 0.27$, $p = .61$) or gender ($Q(1) = 0.41$, $p = .52$). Both of these analyses included all studies. For the six studies that provided a full-scale IQ score, meta-regression analysis indicated a significant effect ($Q(1) = 6.91$, $p = .001$. A scatterplot of the regression of Fisher's Z on IQ is shown in Figure 4.

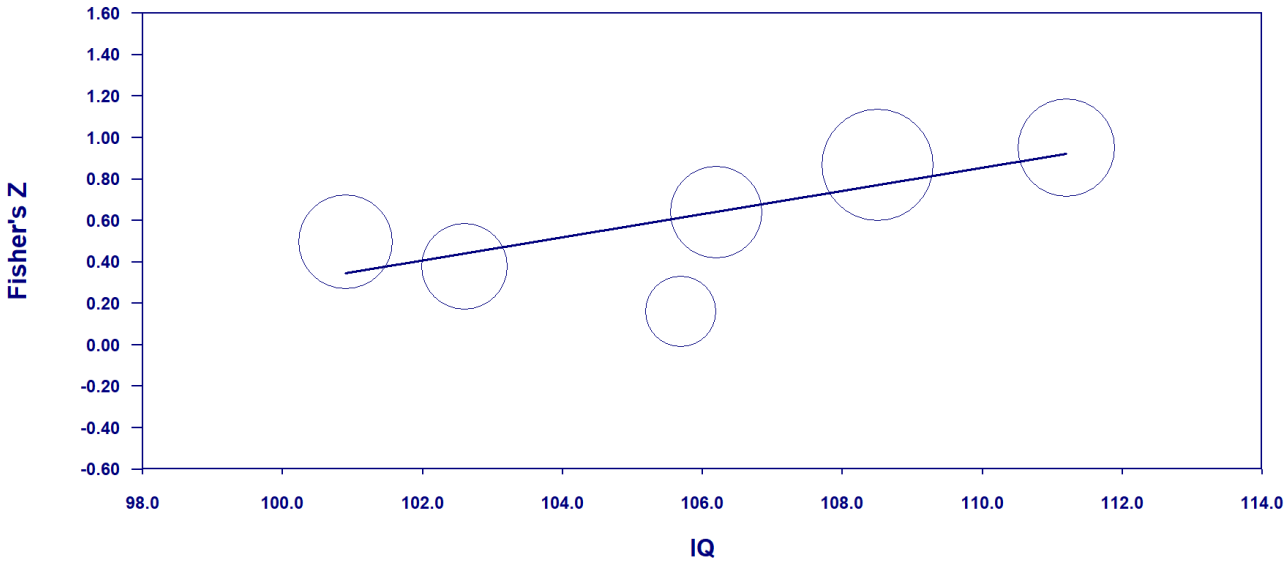

Figure 4 Scatterplot of the regression of Fisher's *Z* on IQ

Categorical moderators were explored using subgroup analyses (see Table 3). Two subgroup analyses met the criterion specified a priori of comprising at least four studies. The first explored the effect of informant on the association between IoU and anxiety. Data from studies that was exclusively self-reported (n = 6) yielded a pooled-effect size (r = .62, p < .001) that was virtually identical to the pooled-effect of studies (n = 4) where the data on at least one measure was parent-reported (r = .63, p < .001).

The second subgroup-analysis was not planned a priori. It was conducted because, unexpectedly, half of the studies reported at least a partial collaboration with the University of Newcastle research team. These studies were combined as a sub-group and yielded a large, pooled effect size (r = .71, p < .001). The other five who did not have links with Newcastle were included a second sub-group. They also yielded a large, pooled effect size (r = .53, p < .001), albeit less strong than the Newcastle sub-group. There was significant between-groups heterogeneity (Q_{bet} (1) = 4.19, p < .041), suggesting that the particular research team did have a significant impact on the relationship between IoU and anxiety.

Table 3

Summary of subgroup analyses

	Number of studies	Correlation	Lower CI	Upper CI	Significance	Q Between
Informant						
Correlation based on data which included parent-report	4	.62	.42	.77	.000	
Correlation based on data which was exclusively self-reported	6	.63	.48	.74	.000	
Between						$Q(1) = 0.001$, $p = .97$
Overall	9	.65	.56	.73	.000	
Research group						
Newcastle	5	.71	.59	.80	.000	
Other	5	.53	.36	.66	.000	
Between						$Q(1) = 4.19$, $p = .041$
Overall	10	.63	.41	.78	.000	

Discussion

This was the first time the research on the association between anxiety and IoU in people with ASD was synthesised and analysed in a systematic review and meta-analysis. Of the ten studies included in the meta-analysis, a significant correlation was found in nine (seven finding a large effect; two finding a moderate effect). All effects were in the same direction, indicating a positive association between IoU and anxiety, that is, higher anxiety was generally found in participants who were more intolerant of uncertainty (and vice-versa). There was only one study that did not find a significant correlation and this was an unpublished dissertation and among the weakest in terms of quality; suggesting caution was advisable when interpreting the result.

The meta-analysis showed a mean effect size that was large and suggested that IoU was associated with 38% of the variance in anxiety amongst the participants (with ages ranging from 4-years to 24-years). This result was consistent with a recent meta-analysis (Osmanağaoğlu et al.,

2018) conducted in a neurotypical, Western-population with ages ranging from 3-years to 20-years, and in which the majority of studies utilised identical or very similar outcome measures to the present review. The researchers found IoU explained 36% of the variance in anxiety. Therefore, the primary conclusion from this review is that the strength of the association between IoU and anxiety in children and young adults with ASD is comparable to that found in the neurotypical population.

Significant heterogeneity was found in the present meta-analysis and potential moderating variables were explored. Using meta-regression analyses, age and gender did not appear to have a significant impact on the effect. The same result was found in the meta-analysis by Osmanağaoğlu et al. (2018). However, as the researchers argued, there are theoretical reasons why it might be expected that the relationship changes as cognitive abilities develop. Although the non-significant effect of age went against this prediction, this review differed from Osmanağaoğlu's as IQ score was also explored as a potential moderator.

Although individual studies did not find an association between IQ and IoU or anxiety, the present meta-regression analysis found a significant result that suggested the association between IoU and anxiety strengthened mildly as IQ increased. This finding is relevant in the context of a recent meta-analysis by van Steensel and Heeman (2017), as this analysis found that anxiety levels were elevated in children with ASD (compared with neurotypical children) and that this difference widened as IQ increased. The authors suggested it was plausible that increased cognitive functioning in children with ASD meant they had more insight into their difficulties and the demands upon them; leading to anxiety.

The present review included studies that suggested IoU partially-mediated the association between the core features of ASD and anxiety, and one study found a significant, serial, indirect-effect from sensory-responsiveness through IoU and anxiety to insistence on sameness. This is in line with theories that suggest sameness behaviours may function to reduce short-term anxiety by avoidance of uncertain situations that provoke distress (Joosten, Bundy, & Einfeld, 2009).

Therefore, perhaps high-functioning individuals with ASD have greater insight into their difficulties and this motivates the need for predictability and raises anxiety about the potential impact their difficulties can have on meeting uncertain demands. To reduce this anxiety, individuals may insist on sameness (resulting in a vicious cycle).

Subgroup analyses were conducted to examine the influence of categorical variables on the association between IoU and anxiety. Studies that used exclusively self-reported data were found not to differ significantly from studies that included parental-reported data. This was surprising, given half of the studies included in the systematic review reported finding at least one significant inconsistency between parent- and child-reports. One possible explanation for this is that the degree of inconsistency was consistent across measures of IoU and anxiety. Therefore, although parents might have scored individual measures of IoU and anxiety differently to their children, it did not have a significant impact on the *relationship* between them. However, it is also plausible that the small number of studies included in this review meant it was inadequately powered to detect an effect.

The review revealed that half the studies reported at least a partial collaboration with the research team at Newcastle University and, therefore, it was decided this would be explored tentatively. This was not planned a priori and the results, therefore, are exploratory. The subgroup analysis suggested the studies that had links with Newcastle reported slightly stronger correlations than studies conducted independently. This could just be a chance finding. However, it could be that there is a uniqueness about the research conducted at Newcastle that inflates the correlation. For example, the majority of the studies that had links reported recruiting participants from a University database and perhaps these individuals differed from the source population in important ways. Rodgers et al. (2016) cited research that suggested children and families on the database were comparable to the source population. However, IoU was not examined and this review suggests it might be prudent for future research to compare people on databases to the source population (in terms of IoU) as a validity check (or alternatively to use a more varied selection of recruitment and

sampling methods to ensure participants are representative). Although none of the authors reported it, it also raises the possibility that some individuals participated in more than one study included in the review (which is acknowledged as a significant limitation).

Across the six studies that compared people with ASD with a neurotypical group, those with ASD were shown consistently to have significantly higher IoU. There was only one study that did not find this. It should be noted that this study had the lowest quality rating in the review and used the smallest sample size. Therefore, it was likely the study was not sufficiently powered. However, given this review's focus was on IoU and anxiety, it is plausible that studies were excluded that explored IoU in isolation, or with variables other than anxiety (and these might have had contradictory findings).

All six of the studies making between-group comparisons also found anxiety was significantly elevated in participants with ASD. However, the elevation of anxiety in ASD is not a new finding and so this review is broadly consistent with the wider literature (e.g. van Steensel & Heeman, 2017).

Additional Limitations

The Intolerance of Uncertainty Scale was by far the most popular measure of IoU in the studies included in this review. Although both the 27-item and 12-item version have been found to have be reliable and valid for use with neurotypical individuals (Khawaja & Ngo Heidi Yu, 2010), neither has been validated in the ASD population. The other uncertainty measure included in the review (a subscale of the ASC-ASD) also lacked sufficient psychometric data. This is, therefore, an important limitation as it is plausible the IoU measures were unreliable for use with people with ASD, or that they were not measuring what they were intended to measure. Further research is necessary to ascertain whether the measures are valid and reliable, before the results from studies using them can be interpreted with confidence. It would also be useful to conduct further research into measuring the impact of uncertainty using neuro-physiological measurements. Although it was

outside the scope of the review, it was noted in one study (Damiano, 2015) that children with ASD showed different fMRI neural-activation patterns to experimentally-induced uncertainty than neurotypical children did.

In addition to the reliability/validity of the measures, the quality of the studies was compromised on a number of variables (e.g. the representativeness of the participants; the precision of the effect sizes reported etc.). It is therefore a limitation that all studies were included in the review, irrespective of their quality.

Although this review aimed to synthesise data across the life-span, only one study used adult participants exclusively, and this was excluded from the meta-analysis. Subsequently, it was not possible to make inferences about how the relationship between IoU and anxiety develops into adulthood. The findings from this analysis are only relevant to children and young adults therefore. There was also a high percentage of males in the included studies, although this proportion was consistent with prevalence estimates (Whiteley, Todd, Carr, & Shattock, 2010).

Another limitation was the sample size. Despite using a comprehensive search strategy and broad inclusion/exclusion criteria, there were only ten studies included in the meta-analysis. Some statisticians (e.g. Field, 2001) caution against conducting correlational meta-analyses with such a limited number of studies. There were a number of features (e.g. the type of outcome measures used, the exclusive use of correlation-coefficients, the gender balance of participants) that were very similar between the different studies. This arguably made the analysis more appropriate to conduct with such a small sample size. However, statistically-significant between-study heterogeneity was still found, which means the results should be interpreted with caution.

Finally, the cross-sectional nature of the studies included in the review limited causal inferences being made. Longitudinal research examining the developmental course of IoU and anxiety would be beneficial.

Clinical Implications

Boulter et al. (2014) found evidence that suggested IoU may mediate the relationship between ASD and anxiety and that the relationship between IoU and anxiety is similar in individuals with and without ASD. In their discussion, the researchers subsequently recommended that anxiety interventions be developed that target IoU in individuals with ASD. The current review adds to the growing evidence base in support of this proposal by demonstrating that IoU and anxiety are consistently elevated in ASD and that the strength of the relationship is comparable to neurotypical populations.

It is encouraging that IoU interventions are currently being developed for people with ASD and that preliminary data is promising (e.g. Rodgers et al., 2017). However, this review has highlighted steps that would potentially strengthen the quality of work in this field (e.g. the validation of IoU measures).

Conclusions

IoU and anxiety appear to be elevated in youth with ASD, but the strength of the relationship between them seems to be comparable to the neurotypical population. This might mean IoU is an appropriate target for intervention in people with ASD, as it is in neurotypical populations.

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Appendix A

Search Strategy

PsycINFO

(("anxiety" or "fear" or "GAD" or "OCD" or "compulsive disorder" or "panic") and ("autism" or "ASD" or "ASC" or "PDD" or "Asperg*" or "pervasive developmental disorder" or "Pathological Demand" or "PDA") and "intolerance" and "uncertainty").ab. or (("anxiety" or "fear" or "GAD" or "OCD" or "compulsive disorder" or "panic") and ("autism" or "ASD" or "ASC" or "PDD" or "Asperg*" or "pervasive developmental disorder" or "Pathological Demand" or "PDA") and "intolerance" and "uncertainty").ti. or (("anxiety" or "fear" or "GAD" or "OCD" or "compulsive disorder" or "panic") and ("autism" or "ASD" or "ASC" or "PDD" or "Asperg*" or "pervasive developmental disorder" or "Pathological Demand" or "PDA") and "intolerance" and "uncertainty").id.

MEDLINE (include related terms)

(("anxiety" or "fear" or "GAD" or "OCD" or "compulsive disorder" or "panic") and ("autism" or "ASD" or "ASC" or "PDD" or "Asperg*" or "pervasive developmental disorder" or "Pathological Demand" or "PDA") and "intolerance" and "uncertainty").ab. or (("anxiety" or "fear" or "GAD" or "OCD" or "compulsive disorder" or "panic") and ("autism" or "ASD" or "ASC" or "PDD" or "Asperg*" or "pervasive developmental disorder" or "Pathological Demand" or "PDA") and "intolerance" and "uncertainty").ti. or (("anxiety" or "fear" or "GAD" or "OCD" or "compulsive disorder" or "panic") and ("autism" or "ASD" or "ASC" or "PDD" or "Asperg*" or "pervasive developmental disorder" or "Pathological Demand" or "PDA") and "intolerance" and "uncertainty").id.

SCOPUS (title, abstract, keywords)

("anxiety" OR "fear" OR "GAD" OR "OCD" OR "compulsive disorder" OR "panic") AND (“intolerance of uncertainty”) AND ("autism" OR "ASD" OR "ASC" OR "PDD" OR "Asperg*" OR "pervasive developmental disorder" OR "Pathological Demand" OR "PDA")

WEB OF SCIENCE

TOPIC: ("anxiety" or "fear" or "GAD" or "OCD" or "compulsive disorder" or "panic") AND TOPIC: ("autism" or "ASD" or "ASC" or "PDD" or "Asperg*" or "pervasive developmental disorder" or "Pathological Demand" or "PDA") AND TOPIC: ("intolerance of uncertainty")

Timespan: All years. Databases: WOS, BCI, BIOSIS, CCC, DRCI, DIIDW, KJD, MEDLINE, RSCI, SCIELO, ZOOREC. Search language=Auto

White Rose

Title (any of): autism, ASD, ASC, PDD, Asperg*, pervasive developmental disorder, Pathological Demand, PDA

+ Abstract (all of):intolerance of uncertainty

PROQUEST

IN anywhere:

("anxiety" OR "fear" OR "GAD" OR "OCD" OR "compulsive disorder" OR "panic") AND (“intolerance of uncertainty”) AND ("autism" OR "ASD" OR "ASC" OR "PDD" OR "Asperg*" OR "pervasive developmental disorder" OR "Pathological Demand" OR "PDA")

Appendix B

Quality Checklist

Quality appraisal checklist – quantitative studies reporting correlations and associations

- Checklist

A correlates review (see section 3.3.4) attempts to establish the factors that are associated or correlated with positive or negative health behaviours or outcomes. Evidence for correlate reviews will come both from specifically designed correlation studies and other study designs that also report on correlations.

This checklist[15] has been developed for assessing the validity of studies reporting correlations. It is based on the appraisal step of the 'Graphical appraisal tool for epidemiological studies (GATE)', developed by Jackson et al. (2006).

This checklist enables a reviewer to appraise a study's internal and external validity after addressing the following key aspects of study design: characteristics of study participants; definition of independent variables; outcomes assessed and methods of analyses.

Like GATE, this checklist is intended to be used in an electronic (Excel) format that will facilitate both the sharing and storage of data, and through linkage with other documents, the compilation of research reports. Much of the guidance to support the completion of the critical appraisal form that is reproduced below also appears in 'pop-up' windows in the electronic version[16].

There are 5 sections of the revised GATE. Section 1 seeks to assess the key population criteria for determining the study's **external validity** – that is, the extent to which the findings of a study are generalisable beyond the confines of the study to the study's source population.

Sections 2 to 4 assess the key criteria for determining the study's **internal validity** – that is, making sure that the study has been carried out carefully, and that the identified associations are valid and are not due to some other (often unidentified) factor.

Checklist items are worded so that 1 of 5 responses is possible:

++	Indicates that for that particular aspect of study design, the study has been designed or conducted in such a way as to minimise the risk of bias.
+	Indicates that either the answer to the checklist question is not clear from the way the study is reported, or that the study may not have addressed all potential sources of bias for that particular aspect of study design.

–	Should be reserved for those aspects of the study design in which significant sources of bias may persist.
Not reported (NR)	Should be reserved for those aspects in which the study under review fails to report how they have (or might have) been considered.
Not applicable (NA)	Should be reserved for those study design aspects that are not applicable given the study design under review (for example, allocation concealment would not be applicable for case–control studies).

In addition, the reviewer is requested to complete in detail the comments section of the quality appraisal form so that the grade awarded for each study aspect is as transparent as possible.

Each study is then awarded an overall study quality grading for internal validity (IV) and a separate one for external validity (EV):

- ++ All or most of the checklist criteria have been fulfilled, where they have not been fulfilled the conclusions are very unlikely to alter.
- + Some of the checklist criteria have been fulfilled, where they have not been fulfilled, or not adequately described, the conclusions are unlikely to alter.
- – Few or no checklist criteria have been fulfilled and the conclusions are likely or very likely to alter.

Checklist

Section 1: Population		
1.1 Is the source population or source area well described? • Was the country (e.g. developed or non-developed, type of health care system), setting (primary schools, community centres etc), location (urban, rural), population demographics etc adequately described?	++ + – NR NA	Comments:
1.2 Is the eligible population or area representative of the source population or area?	++	Comments:

• Was the recruitment of individuals, clusters or areas well defined (e.g. advertisement, birth register)? • Was the eligible population representative of the source? Were important groups underrepresented?	+ – NR NA	
1.3 Do the selected participants or areas represent the eligible population or area? • Was the method of selection of participants from the eligible population well described? • What % of selected individuals or clusters agreed to participate? Were there any sources of bias? • Were the inclusion or exclusion criteria explicit and appropriate?	++ + – NR NA	Comments:
Section 2: Method of selection of exposure (or comparison) group		
2.1 Selection of exposure (and comparison) group. How was selection bias minimised? • How was selection bias minimised?	++ + – NR NA	Comments:
2.2 Was the selection of explanatory variables based on a sound theoretical basis? • How sound was the theoretical basis for selecting the explanatory variables?	++ + – NR NA	Comments:
2.3 Was the contamination acceptably low?	++	Comments:

• Did any in the comparison group receive the exposure? • If so, was it sufficient to cause important bias? AMENDED TO: **2.3 Was the diagnosis of autism confirmed by the researchers ?** • Did they report a gold standard measure to confirm diagnosis or rely on self-report? • Did they specify the type of autism diagnoses in the sample? • Did they use a diagnostic/screening tool to double-check?	+ – NR NA	
2.4 How well were likely confounding factors identified and controlled? • Were there likely to be other confounding factors not considered or appropriately adjusted for? • Was this sufficient to cause important bias?	++ + – NR NA	Comments:
2.5 Is the setting applicable to the UK? • Did the setting differ significantly from the UK?	++ + – NR NA	Comments:
Section 3: Outcomes		
3.1 Were the outcome measures and procedures reliable? • Were outcome measures subjective or objective (e.g. biochemically validated nicotine levels ++ vs self-reported smoking −)? • How reliable were outcome measures (e.g. inter- or intra-rater reliability scores)?	++ + – NR NA	Comments:

• Was there any indication that measures had been validated (e.g. validated against a gold standard measure or assessed for content validity)?		
3.2 Were the outcome measurements complete? • Were all or most of the study participants who met the defined study outcome definitions likely to have been identified?	++ + – NR NA	Comments:
3.3 Were all the important outcomes assessed? • Were all the important benefits and harms assessed? • Was it possible to determine the overall balance of benefits and harms of the intervention versus comparison?	++ + – NR NA	Comments:
3.4 Was there a similar follow-up time in exposure and comparison groups? • If groups are followed for different lengths of time, then more events are likely to occur in the group followed-up for longer distorting the comparison. • Analyses can be adjusted to allow for differences in length of follow-up (e.g. using person-years).	++ + – NR NA	Comments:
3.5 Was follow-up time meaningful? • Was follow-up long enough to assess long-term benefits and harms? • Was it too long, e.g. participants lost to follow-up?	++ + – NR NA	Comments:

Section 4: Analyses		
4.1 Was the study sufficiently powered to detect an intervention effect (if one exists)? • A power of 0.8 (i.e. it is likely to see an effect of a given size if one exists, 80% of the time) is the conventionally accepted standard. • Is a power calculation presented? If not, what is the expected effect size? Is the sample size adequate?	++ + – NR NA	Comments:
4.2 Were multiple explanatory variables considered in the analyses? • Were there sufficient explanatory variables considered in the analysis?	++ + – NR NA	Comments:
4.3 Were the analytical methods appropriate? • Were important differences in follow-up time and likely confounders adjusted for?	++ + – NR NA	Comments:
4.6 Was the precision of association given or calculable? Is association meaningful? • Were confidence intervals or p values for effect estimates given or possible to calculate? • Were CIs wide or were they sufficiently precise to aid decision-making? If precision is lacking, is this because the study is under-powered?	++ + – NR NA	Comments:

Section 5: Summary		
5.1 Are the study results internally valid (i.e. unbiased)? • How well did the study minimise sources of bias (i.e. adjusting for potential confounders)? • Were there significant flaws in the study design?	++ + –	Comments:
5.2 Are the findings generalisable to the source population (i.e. externally valid)? • Are there sufficient details given about the study to determine if the findings are generalisable to the source population? • Consider: participants, interventions and comparisons, outcomes, resource and policy implications.	++ + –	Comments:

[15] Appraisal form derived from: Jackson R, Ameratunga S, Broad J et al. (2006) The GATE frame: critical appraisal with pictures. Evidence Based Medicine 11: 35–8.

[16] Available from CPHE on request.

Appendix C

Quality Table

	Boulter, Freeston, South, & Rodgers (2014)	Cai, Richdale, Dissanayake, & Uljarević (2018)	Chamberlain et al. (2013)	Damiano (2015)	Glod (2017)	Joyce, Honey, Leekam, Barrett, & Rodgers (2017)	Keefer et al. (2017)	Maisel et al. (2016)	Neil, Olsson, & Pellicano (2016)	Rodgers et al. (2016)	Vasa, Kreiser, Keefer, Singh, & Mostofsky (2018)	Wigham, Rodgers, South, McConachie, & Freeston (2015)
1.1 Is the source population or source area well described? Was the country (e.g. developed or non-developed, type of health care system), setting (primary schools, community centres etc), location (urban, rural), population demographics etc adequately described?	+	+	-	+	+	+	+	+	+	++	+	+
1.2 Is the eligible population or area representative of the source population or area? Was the recruitment of individuals, clusters or areas well defined (e.g. advertisement, birth register)? Was the eligible population representative of the source?	+	+	-	-	+	+	+	+	+	++	+	+

Were important groups underrepresented**?**												
1.3 Do the selected participants or areas represent the eligible population or area? Was the method of selection of participants from the eligible population well described? What % of selected individuals or clusters agreed to participate? Were there any sources of bias? Were the inclusion or exclusion criteria explicit and appropriate?	-	-	-	-	-	-	-	-	-	++	+	-
2.1 **Selection of exposure (and comparison) group. How was selection bias minimised?**	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2.2 Was the selection of explanatory variables based on a sound theoretical basis?	++	++	++	++	++	++	++	++	++	++	++	++
2.3 Was the diagnosis of autism confirmed by the researchers ? Did they use a gold-standard diagnostic measure?	+	+	++	++	+	-	+	++	++	+	++	+
2.4 How well were likely confounding factors identified and controlled? Were there likely to be other confounding factors not considered or appropriately adjusted for? Was this sufficient to cause important bias?	+	+	NA	+	-	-	-	NA	+	NA	++	+

2.5 Is the setting applicable to the UK? Did the setting differ significantly from the UK?	+	+	+	+	++	++	+	+	++	++	+	+
3.1 Were the outcome measures and procedures reliable? Were outcome measures subjective or objective (e.g. biochemically validated nicotine levels ++ vs self-reported smoking −)? How reliable were outcome measures (e.g. inter- or intra-rater reliability scores)? Was there any indication that measures had been validated (e.g. validated against a gold standard measure or assessed for content validity)?	+	+	-	-	-	+	-	-	+	+	-	-
3.2 Were the outcome measurements complete? Were all or most of the study participants who met the defined study outcome definitions likely to have been identified?	-	+	+	+	+	++	++	+	++	+	++	++
3.3 Were all the important outcomes assessed? Were all the important benefits and harms assessed? Was it possible to determine the overall balance of benefits and harms of the intervention versus comparison?	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA

3.4 Was there a similar follow-up time in exposure and comparison groups? If groups are followed for different lengths of time, then more events are likely to occur in the group followed-up for longer distorting the comparison. Analyses can be adjusted to allow for differences in length of follow-up (e.g. using person-years).	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
3.5 Was follow-up time meaningful? Was follow-up long enough to assess long-term benefits and harms? Was it too long, e.g. participants lost to follow-up?	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
4.1 Was the study sufficiently powered to detect an effect (if one exists)? A power of 0.8 (i.e. it is likely to see an effect of a given size if one exists, 80% of the time) is the conventionally accepted standard. Is a power calculation presented? If not, what is the expected effect size? Is the sample size adequate?	++	+	-	-	+	+	+	+	+	++	+	+

4.2 Were multiple explanatory variables considered in the analyses?	++	+	NA	++	-	+	-	++	++	NA	++	++
4.3 Were the analytical methods appropriate? Were important differences in follow-up time and likely confounders adjusted for?	++	+	NA	-	+	+	+	NA	++	+	++	++
4.6 Was the precision of association given or calculable? Is association meaningful? Were confidence intervals or p values for effect estimates given or possible to calculate? Were CIs wide or were they sufficiently precise to aid decision-making? If precision is lacking, is this because the study is under-powered?	++	+	-	-	+	-	-	+	++	++	+	+
OVERALL QUALITY SCORE	62%	46	30%	38%	42%	46%	38%	55%	73%	82%	69%	58%
5.1 Are the study results internally valid (i.e. unbiased)? How well did the study minimise sources of bias (i.e. adjusting for potential confounders)? Were there significant flaws in the study design?	+	+	NA	-	+	-	+	NA	++	NA	+	+
5.2 Are the findings generalisable to the source population (i.e. externally valid)? Are there sufficient details given about the study to determine if the findings are	+	+	-	+	_	_	+	-	+	++	+	+

generalisable to the source population? Consider: participants, interventions and comparisons, outcomes, resource and policy implications.												

Note: Criteria fully met (++) = 2 points, criteria partially met (+) = 1 point, criteria not met (-) = 0 points, not applicable (NA) = 0 points but item not counted in percentage for particular study. Overall quality score is calculated as a percentage by dividing the study's points by the total possible for the particular study and multiplying by 100. Summary items (5.1. and 5.2) are not included in the percentage.

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Section Two

The Effectiveness of a Social Story Intervention for Reducing Negative Affect in Adults Attending a Diagnostic Assessment for Autism Spectrum Disorder

The trial was registered prior to recruitment commencing. This can be viewed online by visiting http://www.ClinicalTrials.gov (identifier: NCT03372421).

Abstract

Objectives. An intervention was developed based on the Social Story literature. It was designed to increase the predictability of a novel social situation (a diagnostic assessment for Autism Spectrum Disorder [ASD]). A clinical trial was conducted to evaluate the effectiveness of the intervention on negative affect, predictability and satisfaction.

Methods. The intervention was developed in collaboration with service-users and a randomised, controlled trial was subsequently conducted at two NHS diagnostic services for adults. Included participants were allocated to experimental (n = 27) and control (n = 24) conditions and completed a measure of negative affect approximately one week before their assessment (time one) and again upon arrival (time two). The effectiveness of the intervention on negative affect was examined with an analysis of variance. Bespoke outcome measures were used to make between-group comparisons on predictability (time two) and satisfaction (post-assessment; time three). Participants who did not receive a diagnosis of ASD were excluded from data-analysis.

Results. A statistically-significant interaction between time and group was found on negative affect, such that participants who read the Social Story reported less of an increase in negative affect across time, relative to the control group. There was not a statistically-significant between-group difference in either predictability or satisfaction.

Conclusions. The results suggested the intervention had a salutary impact on negative affect, but not on predictability or satisfaction. Although replication of the results is needed, it may be a simple, low-cost way of improving the experience of attending an assessment for ASD in adulthood.

Practitioner Points

- The results showed that attending a diagnostic assessment for ASD is associated with a significant increase in state negative affect.
- Social Stories require minimal resources for services to develop but may be effective at preventing clients with ASD from experiencing a surge of negative affect on arrival at their first appointment.
- Guidance about how to create Social Stories is freely-available online. A template based on the Social Story used in this study is available on request.
- Although further research is required to understand the mechanism of action of Social Stories, it may be important to seek service-user involvement when developing a Social Story that details what to expect from a particular service.

Limitations

- Owing to recruitment difficulties, deviations from the original protocol were required.
- There were issues with intervention-/procedural-fidelity.
- Participants moved through the study process at different times (data were collected from the first participant approximately ten months before data from the final participant were collected).
- Secondary outcomes were assessed using bespoke measures which may have had issues with reliability and validity.

Keywords: Autism, Social Story, Anxiety, Negative Affect, PANAS

Introduction

Social situations for people with autism spectrum disorder (ASD) can provoke a variety of emotional responses, such as anxiety, fear, stress and frustration (Iseminger, 2009; Trembath, Germano, Johanson, & Dissanayake, 2012; Volkmar, Paul, Klin, & Cohen, 2005). Such emotions are often referred to collectively as *negative affect* (Stringer, 2013; Watson, Clark, & Tellegen, 1988).

Individuals with ASD report that other people tend to respond to overt expressions of negative affect with avoidance, criticism and patronising talk (Lipsky, 2011). These types of social experiences can have a profound psychological impact. As Lipsky and Richards (2009) explain, "After the meltdown phase there are often intense feelings of shame, remorse, and humiliation. There is a frequent fear that relationships have been harmed beyond repair" (p. 22). It is saddening but unsurprising therefore that almost 40% of young adults with ASD report they never socialise and almost half do not receive phone calls or invitations to social events/activities from friends (Orsmond, Shattuck, Cooper, Sterzing, & Anderson, 2013). Furthermore, in a survey of 56 mature students with ASD, Jackson, Hart, Brown, and Volkmar (2018) found that over 75% reported feeling left out, isolated, or lacking companionship; 36% experienced some form of bullying and over half reported experiencing suicidal intent in their lifetime. Therefore, it is plausible that becoming overwhelmed in social situations leads to social isolation for people with ASD; limiting opportunities for social development and impacting on self-esteem.

Dissatisfaction with Healthcare Services

The UK Government published the Fulfilling and Rewarding Lives strategy in 2010, which aimed to improve the lives of adults with ASD who had been socially excluded and "badly let down by public services which have failed to recognise or respond to their needs" (Department of Health, 2010, p. 6). This strategy had a keen focus on helping individuals

with ASD live independently, on overcoming barriers to accessing community services and on ensuring ASD-diagnostic services for adults were available nationwide. Although this led to increased resource for opening new diagnostic services, qualitative research suggests there is still room for improvement.

Jones, Goddard, Hill, Henry, and Crane (2014) surveyed 128 UK adults with ASD about their experience of attending their diagnostic assessment and found 40% were dissatisfied with the overall process. Of relevance to the present study, Jones et. al noted a recurring theme in the written comments of respondents; that the diagnostic process lacked a predictable structure. Similarly, Crane et al. (2018) interviewed ten UK-based adults with ASD and found there was a theme of dissatisfaction in relation to a lack of clarity about what to expect. Furthermore, Trivasse (2019) conducted a qualitative service evaluation at an ASD -diagnostic service in the UK and found that participants remembered feeling anxious prior to their assessment, which they related to their uncertainty about what it would involve. Participants attested to the importance of the service providing clear, practical information about what to expect, in order to mitigate anxiety. This was an area specifically referred to in the Fulfilling and Rewarding Lives strategy as it was noted that people with ASD can "struggle with the formats, language and instructions of forms or standard letters" (Department of Health, 2010, p. 41). The strategy also emphasised that service providers have a legal responsibility to demonstrate reasonable adjustments they have made for adults with ASD, such as initiates that help people better know what to expect from the service.

Predictability

Baron-Cohen (2002) proposed that individuals with ASD seek predictability to prevent becoming overwhelmed in social situations. The utility of this strategy has been documented in qualitative accounts from teachers (e.g. Godfrey & Haythorne, 2013), parents

(e.g. McAuliffe, Thomas, Vaz, Falkmer, & Cordier, 2019) and from individuals themselves (e.g. Trembath et al., 2012).

The effect of increasing predictability has also been tested empirically. In a seminal study, Ferrara and Hill (1980) presented children with social toys (dolls with faces) under a predictable condition (in which the toys consistently became visible after a signalling light) and an unpredictable condition (in which the toys were revealed at random intervals). Children with ASD showed significantly increased interaction with the toys during the predictable condition.

Antecedent Interventions

Therapeutic strategies for increasing predictability and reducing negative affect include avoiding unexpected changes, creating structure and routine, and using antecedent interventions which involve some form of actual or imagined rehearsal prior to an activity or transition (McClean & Grey, 2012; Murin, Hellriegel, & Mandy, 2016; Nason, 2014). In a meta-analysis of 163 studies of interventions for children and adults with ASD, Ma (2009) demonstrated how antecedent interventions such as video priming (involving individuals previewing forthcoming activities or events) and modelling (involving individuals watching other people interacting socially before trying it themselves) had moderate to large effects on increasing social interaction and reducing behaviour that challenged (such as tantrums and aggression). This is relevant to the present study as behaviours that challenge are commonly understood to be expressions of elevated negative affect (Ashburner, Ziviani, & Rodger, 2010; Simonoff et al., 2012).

Social Stories. Among the most researched of the antecedent interventions are Social Stories™, developed by Carol Gray in 1991. As Wright et al. (2016) explain, Social Stories are short, written descriptions of a future situation, event or activity, often accompanied by

illustrations or photographs. They are designed to be read by a person in advance of a social interaction and aim to share accurate, socially-relevant information in a style that is understandable and helpful for people with ASD. Despite being commonly used as an intervention-strategy for reducing overt expressions of negative affect, a directive story that instructs people how to behave would not be considered a Social Story; instead the focus should be on providing supportive information that helps people to know what to expect and to understand what others might expect (Gray, 2018; Wright et al., 2016).

A recent meta-analysis by Wright et al. (2016) synthesised the results from 99 studies on Social Stories and ASD. The authors concluded the research broadly supported the effectiveness of the intervention, but argued interpretation was limited by variation in the quality of the studies. Although the majority used single-case designs, data was sufficient in two between-group studies for Wright et al. to calculate effect sizes. Both of these studies compared a Social Story with a story that had no social loading. The first used social skills as the dependent variable, which researchers measured by observing participants during a game (and rating their social skills based on behaviours such as the number of greeting behaviours they demonstrated). Wright et al. calculated an effect size of 1.21 for this study, demonstrating that the intervention led to improved social skills. The dependent variable in the second study was the learning of facial emotions. In this study, an effect size of 1.38 was calculated for affect-discrimination, 1.73 for emotion-matching and 2.13 for affect-choice; demonstrating that the intervention led to improved learning of facial emotions. Therefore, there is sufficient evidence to justify further investigation into the intervention.

The vast majority of the studies included in meta-analyses of Social Stories and ASD (e.g. Bellini, Peters, Benner, & Hopf, 2007; Kokina & Kern, 2010; Reynhout & Carter, 2006; Test, Richter, Knight, & Spooner, 2011; Wright et al., 2016) have been conducted on USA-based, school-age children in educational settings, using single-case designs and behavioural

outcomes. Of particular relevance to the present study, Cullain (2000) demonstrated that reading a Social Story prior to a social interaction reduced both behavioural expressions of negative affect and self-reported anxiety in five autistic children.

Aims and Hypotheses

The choice of setting for the study was informed by the need to investigate ways of increasing the acceptability of health-care services for people with ASD, coupled with reports of dissatisfaction with the process of diagnostic assessment. Based on the literature outlined above, the primary aim was to explore whether a Social Story could help limit the increase in state negative affect that was anticipated to arise from attending a diagnostic assessment for ASD. A secondary aim was to explore whether the Social Story increased predictability and improved satisfaction. Hypotheses are detailed in Table 1.

Table 1

A priori hypotheses

Number	Category	Hypothesis
1	Primary	Compared with participants who read standard, non-social information prior to attending a diagnostic assessment for ASD (the control group), participants who read a Social Story (the experimental group) will report, on average, less of an increase in negative affect when attending their assessment, as measured by the negative subscale of the Positive and Negative Affect Schedule (Watson et al., 1988)
2	Secondary	Relative to the control group, participants in the experimental group will report, on average, that the assessment was more predictable (measured via a 5-point Likert scale).
3	Secondary	Relative to the control group, participants in the experimental group will report, on average, higher satisfaction (measured via a 5-point Likert scale).

Method

The trial was registered prior to recruitment commencing. The record can be viewed online by visiting http://www.ClinicalTrials.gov (identifier: NCT03372421).

Design

A randomised, controlled trial was conducted. Primary outcomes were assessed between groups and across time. Secondary outcomes were assessed at one time point with between-group comparisons.

Setting

To increase ecological validity, the primary host site was an ASD diagnostic-assessment service in the North of England. A second site was added during the study to increase recruitment. Both were NHS diagnostic-assessment services for adults.

Intervention and Service-User Involvement

For an intervention to qualify as a Social Story (Gray, 2018), there are ten criteria that need to be adhered to. These are provided in Appendix A. In line with these criteria, service-user involvement was sought. Adults with ASD who had recently been diagnosed by the host sites were identified from a University research database of local volunteers keen to participate in research. They were contacted via email and invited to provide feedback about what the intervention should include. Three service-users provided written comments via an online survey. The overarching theme that emerged was the importance of providing detailed instructions about what to expect (including the questions clients are routinely asked, the expectations of the assessor, the structure of the assessment etc.) and the environment (e.g. where to park, what the waiting area looks like, sensory considerations etc.). Similar themes emerged from a qualitative service evaluation undertaken with service-users at the primary host site by an independent researcher (Trivasse, 2019). With in-depth knowledge of the linguistic and cognitive abilities of their clients, clinicians at both sites also provided input into the content and style of the intervention. These were incorporated, as long as they were consistent with service-users' views and the criteria specified by Gray.

A draft was produced which was circulated to clinicians and amended/refined. Refinements included clearer instructions in regards to public transport and revisions to what the assessment would entail to ensure it accounted for variability in the way different clinicians conducted assessments. In accordance with guidance from Gray (2018), the final version of the story had a ratio of descriptive sentences (ones which specify aspects of context) to coaching sentences (ones which direct behaviour) of >2. The intervention was adapted slightly when a second host site was included to increase recruitment. The changes comprised substituting images of the site, directions about how it is accessed, and minor changes to what the assessment involved. The structure, style and format of the intervention

was unchanged and thus was consistent across sites. Excerpts from the Social Stories are provided in Appendix B.

Participants and Recruitment

The eligible population comprised all clients awaiting a diagnostic assessment for ASD at the host sites between July 2018 – April 2019 (recruitment at the second site ran from October 2018 – April 2019). To maximise external validity, only one inclusion criterion was specified; that participants were required to have never visited the sites prior to their first appointment.

The sample size required to achieve 80% power (at a significance level of $p < .05$) was estimated a priori using guidelines from Cohen (1992). In order to use these guidelines, it was necessary to predict whether the effect size would be small, medium or large. This estimation was informed by the meta-analysis conducted by Wright et al. (2016), due to the researchers presenting data that was not available elsewhere: effect sizes (Hedges' g) calculated from studies of Social Stories that used randomised, between-group designs and samples of people with ASD. The effect sizes Wright et al. calculated were unanimously large (1.21, 1.38, 1.73 and 2.13). According to Cohen (1992), a two-group ANOVA (as used in the present study) in which a large effect size ($f = .40$) was anticipated, would require 26 participants per group to limit the risk of type II error. However, to conservatively account for anticipated attrition of 20% (based on data from the host site regarding the percentage of people who do not receive a diagnosis), it was planned that 62 people would be recruited into the present study.

Whilst recruitment was open, an invitation to participate in the study was sent to all clients via post approximately three weeks prior to their diagnostic appointment. During phase one of recruitment, participants were required to respond to the invitation by

completing informed consent online. However, due to a lower than expected recruitment rate, an amendment via NHS ethics later permitted participants to provide informed consent via paper (phase two of recruitment). Invitations, consent forms, participant-information sheets, covering letters and debrief sheets are available in appendices C – M.

During the study, there were 229 invitations posted to participants at site one, and 84 posted to participants as site two. Across sites, there were 58 participants who provided informed consent (n = 10 in phase one; n = 48 in phase two), but seven were excluded from the data analysis for not receiving a diagnosis of ASD. Table 2 shows the characteristics of these participants.

Table 2

Participant characteristics by site

	Included in data analysis (n = 51)		Excluded from the data analysis (n = 7)	
	Site 1 (n = 46)	Site 2 (n = 5)	Site 1 (n = 7)	Site 2 (n = 0)
Gender				
Male, n (% of column)	24 (52.17%)	3 (60.00%)	1 (14.29%)	0
Female, n (% of column)	20 (43.48%)	2 (40.00%)	4 (57.14%)	0
Non-binary, n (% of column)	2 (4.35%)	0	0	0
Non-reported, n (% of column)	0	0	2 (28.57%)	0
Age				
Mean age, years (SD)	34.86 (13.10)	46.75 (20.76)	32.00 (11.63)	NA
Age range, years	17 - 58	27 - 68	22 - 48	NA
Not-reported, n	3	1	3	0

At each of the two sites, there was a higher proportion of males included in the data analysis (compared to females). However, of the seven excluded from the data analysis, the

majority were female. Ages of included and excluded participants were similar in site one. Participants from site two were older than those from site one, on average.

Although data on the characteristics of the eligible population were only available for site one, 90.2% of participants were recruited from this site (facilitating comparisons). At site one, 60.2% of the eligible population identified as male. In comparison, 52.9% of the participants included in the data analysis identified as male. The eligible population was slightly younger (M = 29.0, SD = 9.0) than the included participants (M = 35.9, SD = 14). Therefore, although only 18.5% of the eligible population agreed to take part in the study, their characteristics (in terms of age and gender) were broadly equivalent to the participants included in the study. The 51 participants included in the data analysis were randomised to experimental (n = 27) and control (n = 24) conditions. Their characteristics are shown in table 3 below.

Table 3

Participant characteristics by intervention group

	Experimental (n = 27)	Control (n = 24)
Gender		
Male, n (% of column)	13 (48.15%)	14 (58.33%)
Female, n (% of column)	13 (48.15%)	9 (37.50%)
Non-binary, n (% of column)	1 (3.70%)	1 (4.17%)
Age		
Mean age, years (SD)	34.52 (12.94)	37.41 (15.28)
Age range, years	19 - 61	17 - 68
Not-reported, n	3	1

Average age and age-range were similar between groups. There were equal numbers of males and females in the experimental group. Although there was a very similar number of males in both groups, there was a larger proportion of males in the control group, relative to the experimental group. There was one person in each of the groups who identified as non-binary.

Outcome Measures

The primary outcome measure was the Positive and Negative Affect Scale (PANAS; Watson et al., 1988). It is a widely used self-report measure of positive and negative affect that comprises a list of 20 items that are rated one (very slightly) to five (extremely) on a Likert-scale. The items are adjectives describing different emotional states (e.g. "Distressed"; "Excited"; "Scared"). State and trait versions of the measure are available and are identical, except that the trait-version asks respondents to rate how they felt over the past week, whereas the state-version refers to present-moment affect. Due to the study aims, only the negative-affect subscale was used for data analysis. However, to maintain the integrity of the measure, the full questionnaire was provided to participants (state-version).

Subscale scores are summed and range from 10-50; higher scores on the negative affect scale indicate greater emotional distress. The negative affect subscale has good internal consistency (α = .84 - .87), test-retest reliability (r = .60) and excellent convergent and discriminant validity (Watson et al., 1988).

Potentially owing to the simple, concrete language it uses, the PANAS has been used in previous research to measure negative affect in children and adults with ASD (e.g. Arrowood, Cox, & Ekas, 2017; Brooks, 2014; Buvinger, 2013; Donohue, Darling, & Mitroff, 2012; Kovac, Mosner, Miller, Hanna, & Dichter, 2016; Paul, Corsello, Kennedy, & Adolphs, 2014; Samson, Huber, & Gross, 2012). Samson et al. (2012) administered the PANAS to 27

high-functioning adults with ASD and reported a Cronbach's alpha of .70 for the negative-affect subscale. Buvinger (2013) examined the psychometric properties of the PANAS in a sample of 41 individuals with ASD who had a mean age of 16.1 (*SD* = 1.7). Internal consistency (α = .86) and test-retest reliability (*r* = .82) were good. In terms of convergent validity, the negative affect scale was not significantly correlated with the Beck Depression Inventory total score (*r* = .27), but was significantly positively correlated with the Adult Manifest Anxiety Scale total score (*r* = .51) and the anxiety and depression scales from the Child Behavior Checklist. Regression analyses revealed that PANAS negative affect scores significantly predicted the total score on the Adult Manifest Anxiety Scale and the Child Behavior Checklist anxiety score. Therefore, the PANAS was considered a suitable measure for the present study.

Secondary hypotheses were investigated using measures of satisfaction and predictability. Satisfaction was assessed using a one-item measure comprising a 5-point Likert scale (with options ranging from *very dissatisfied* to *very satisfied*) and the preceding text, "Please rate your overall satisfaction with the assessment". This was adapted from the measure used in the study by Jones et al. (2014). Using the same Likert scale, participants in Jones et al.'s study rated the "diagnostic process" (p. 3037), whereas participants in the present study rated the assessment specifically. This was because Jones et al.'s study revealed satisfaction was predicted by factors such as substantial waiting times and so it was judged that the intervention would be highly unlikely to impact satisfaction with the entire process (but could feasibly have an impact on satisfaction with the assessment itself). No psychometric data is available for this measure.

As there was not a precedent in the literature for assessing predictability in social situations, a simple measure was created specifically for this study in order to gather preliminary data about predictability as the potential mechanism of action of the intervention.

The measure comprised the question, “Has the information we sent you prior to the assessment helped you to know what to expect from today?”, accompanied by a 5-point Likert scale with the options, v*ery much*; *a little*; *somewhat*; *not really*; and *not at all*. The scale was chosen as Windschitl and Wells (1996) suggested that qualitative labels of this nature are preferable to numeric measures in assessing perceived uncertainty in ambiguous social situations. As the psychometric properties of this measure were unknown, the results are exploratory and interpreted with caution. Copies of all measures are available in appendices N - P.

Procedure

Using an online randomisation tool (available at https://www.random.org/sequences), a random sequence was generated which was used to assign participants to condition. During phase one of recruitment, each participant was randomised sequentially, immediately after providing informed consent and completing time one (T1) questionnaires online. In phase two of recruitment, participants completed informed consent and T1 questionnaires on paper and brought them to their assessment. Therefore, all those awaiting an assessment in phase two were required to effectively be randomised to group prior to researcher-confirmation of informed consent (as participants in the experimental group needed to receive the intervention prior to their assessment).

All participants were sent the information about what to expect via post and all completed T1 measures at home and time two (T2) measures on arrival at the host site. As questionnaires were dated, it was possible to retrospectively calculate that T1 and T2 measures were completed by participants one week apart, on average. All participants were contacted post-assessment to complete time 3 (T3) measures.

Participants were blinded to group as they did not know whether the information they were sent had been created specifically for the purposes of the study, or whether it was standard service information. Staff at the host sites were also not informed who was allocated to which group. A diagram of the procedure is shown in Appendix Q.

Validity Checks

The service leaflets (sent to participants in the control group) were audited prior to the study commencing against the Social Story criteria (Gray, 2018). It was confirmed they violated criteria and could not be considered a Social Story (e.g. they had a ratio of descriptive to coaching sentences of < 2 and thus violated the eighth criterion). The degree to which the experimental and control information increased predictability was assessed using a bespoke measure (described above).

There were variables that could have potentially confounded the results of this study (e.g. participants being accompanied by a family member). The effect of these variables was mitigated by randomisation.

Data Analysis

A PANAS questionnaire with >20% missing items was considered incomplete and excluded from data analysis. Participants were not excluded from the primary data analysis if they had missing data on secondary outcomes. All data analysis was conducted using IBM SPSS statistics software, version 25. The data analytic methods are reported in Table 4.

Table 4

Types of data analysis

Hypothesis	Statistical analysis
1	Comparison of means using a 2 (group; experimental, control) X 2 (time; T1, T2) mixed Analysis of Variance (ANOVA; repeated measures on the time factor). The dependent variable was the PANAS score. Additional comparisons of means were made using an independent (between-group) *T*-test at T2, and paired-sample T-tests examining changes in negative affect across time (within each group). Descriptive statistics were also presented.
2	Comparison of means was made using a Mann–Whitney *U* test at T3 (due to the non-parametric nature of data derived from a single Likert scale). The independent variable was group (experimental; control), the dependent variable was satisfaction score. Descriptive statistics were also presented.
3	Comparison of means was made using a Mann–Whitney *U* test at T3 (due to the non-parametric nature of data derived from a single Likert scale). The independent variable was group (experimental; control), the dependent variable was satisfaction score. Descriptive statistics were also presented.

Note. T1; time one (at participant's home pre-assessment); T2; time two (on arrival at diagnostic assessment); T3; time 3 (at participant's home post-assessment).

Although the concept of clinically-significant change is important in intervention research, its calculation relies upon assessing the extent to which the intervention produced within-subject improvements in outcomes. Therefore, it was not meaningful to calculate clinically-significant change in the present study due the prediction that post-intervention scores would be worse than pre-intervention scores (across groups), due to the T2 measurement occurring during exposure to a social stressor.

Ethical Considerations

Following an NHS Research Ethics Committee meeting held on the 5th March 2018, ethical approval was confirmed by the Health Research Authority via a letter dated the 1st of May 2018. Proof of ethical approval is provided in appendices R – Y.

Withholding the intervention from participants in the control group was a necessary but unfortunate aspect of the study design. However, those participants still had access to the same quality of information the service already supplied and, therefore, participation did not disadvantage them.

There are no known hazards or contraindications for using Social Stories (Research Autism, 2017). Therefore, the intervention was assessed as being of low risk.

Participants were given the right to withdraw and have their data destroyed at any point during the study, without having to give a reason. Data were stored securely, confidentially and were anonymised prior to analysis and dissemination.

Results

Participant Flow

The host sites confirmed that none of the participants had accessed the service previously. There were seven participants excluded from the data analysis due to not receiving a diagnosis of ASD on conclusion of their assessment. Two of these seven also met the exclusion criterion of having >20% missing data on a primary measure, but no other participants did. A diagram showing participant flow through the study is provided in figure 2.

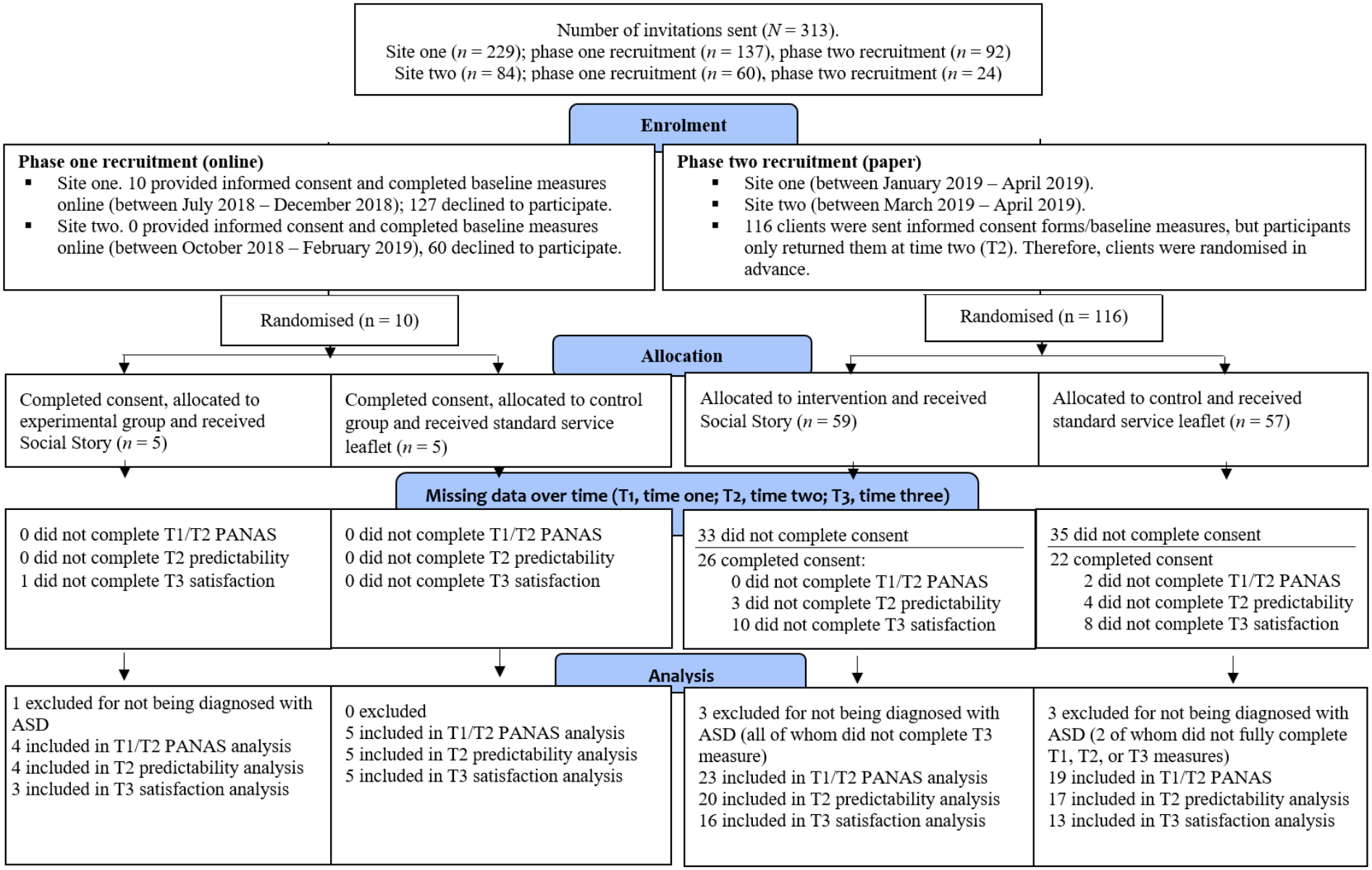

Figure 2 CONSORT Flow diagram

Assumptions of ANOVA

Visual inspection of histograms and Normal Q-Q Plots for T1 and T2 PANAS scores was inconclusive (based on raw data and studentized residuals from each group). However, there was some indication of a positive skew in all graphs. As the sample size was relatively small, additional methods were used to assess normality. T1 and T2 PANAS negative-affect subscale-scores scores were normally distributed for both groups, as assessed by both the Kolmogorov-Smirnov test and the Shapiro-Wilk's test ($p > .05$). Furthermore, Z-scores were calculated by dividing the skewness and kurtosis values by their standard errors. All were less than 1.96, indicating a normal distribution (Kim, 2013).

There were two outliers in the data, as assessed by inspection of a boxplot for values greater than 1.5 box-lengths from the edge of the box. There were no outliers greater than 3 box-lengths. As can be seen from table 7 below, a two-way mixed ANOVA was conducted with and without the outliers, but the results did not differ sufficiently for different conclusions to be drawn from the data. Therefore, the outliers were not removed from the analysis. Furthermore, there were no outliers when assessed by inspection of studentized residuals for values greater than ±3.

There was homogeneity of variances, as assessed by Levene's test of homogeneity of variance ($p > .05$). There was homogeneity of covariances, as assessed by Box's test of equality of covariance matrices ($p > .05$).

Missing Data

Three participants omitted one item from the PANAS negative-affect subscale at T1 and one omitted a single item at T2. In these cases, the missing data points were replaced with the item mean, calculated by dividing the participant's total score on the subscale at the respective time point by the number of items completed (Raymond, 1986). Internal

consistency of the primary measure was high (see below); supporting the appropriateness of this method.

Seven participants were excluded from the data analysis due to not receiving a diagnosis of ASD (their data are provided in the Appendix Z). The low proportion of missing data meant it was not possible to ascertain the reason for items being omitted, or to make meaningful comparisons between participants with complete and incomplete data. It was assumed these data were missing completely at random.

Time One Data

Internal consistency (Cronbach's alpha) was calculated for PANAS scores and the NA subscale was good at T1 (α =.88) and excellent at T2 (α =.91). Table 5 examines descriptive statistics for the primary outcome measure at T1.

Table 5

PANAS negative-affect subscale-scores at T1 (by site, recruitment phase and group)

	Site 1						Site 2					
	Experimental		Control		Combined		Experimental		Control		Combined	
	n	*M (SD)*	*n*	*M (SD)*	*n*	*M (SD)*	*n*	*M (SD)*	*n*	*M (SD)*	*n*	*M (SD)*
Phase one recruitment	4	19.25 (5.68)	5	20.20 (7.66)	9	19.78 (6.46)	0	0	0	0	0	0
Phase two recruitment	19	22.04 (8.31)	16	21.04 (7.98)	35	21.58 (8.06)	3	21.33 (15.50)	2	14.45 (6.29)	5	18.58 (12.01)

Note. n, number*; M,* mean*; SD,* standard deviation

As can be seen from the table, the majority of participants were recruited from site one. This was partly due to the longer window of data collection, and partly due to the site assessing a higher proportion of clients per week than site two. As can be seen from the large standard

deviations, there was considerable variability in the data at site two, most likely due to the small size of the subsample. This made it difficult to make meaningful comparisons across sites.

As can be seen from table 5, the majority of participants were recruited in the second phase of recruitment. This appeared to be because the requirement to visit a website in phase one was a significant barrier for eligible participants.

Primary Analysis

There were no adverse events or side effects reported in either experimental condition. Table 6 shows PANAS negative-affect scores across group and time.

Table 6

Mean scores on PANAS negative affect subscale, by group and time

T1				T2			
Experimental		Control		Experimental		Control	
n	*M (SD)*	*n*	*M (SD)*	*n*	*M (SD)*	*n*	*M (SD)*
27	21.55 (8.58)	24	20.31 (7.72)	27	23.15 (9.07)	24	26.33 (10.54)

Note. n, number*; M,* mean*; SD,* standard deviation; T1, time 1; T2, time 2

A 2 (group; experimental, control) X 2(time; T1, T2) mixed ANOVA revealed there was a statistically significant main effect of time on PANAS negative-affect subscale-scores, F (1, 49) = 13.241, p = .001, of a large effect size (partial η^2 = .213). This suggests that, across groups, participants did experience significantly more negative affect upon attending their assessment (in comparison to T1). There was not a statistically significant main effect of group, suggesting that PANAS scores did not differ between-groups, when compared across time. The mixed ANOVA revealed a statistically significant interaction between group and

time on PANAS negative-affect subscale-scores, F (1, 49) = 4.444, p = .040, with a medium effect size (partial η^2 = .083) and observed power of .543. This interaction is depicted visually in Figure 3 below.

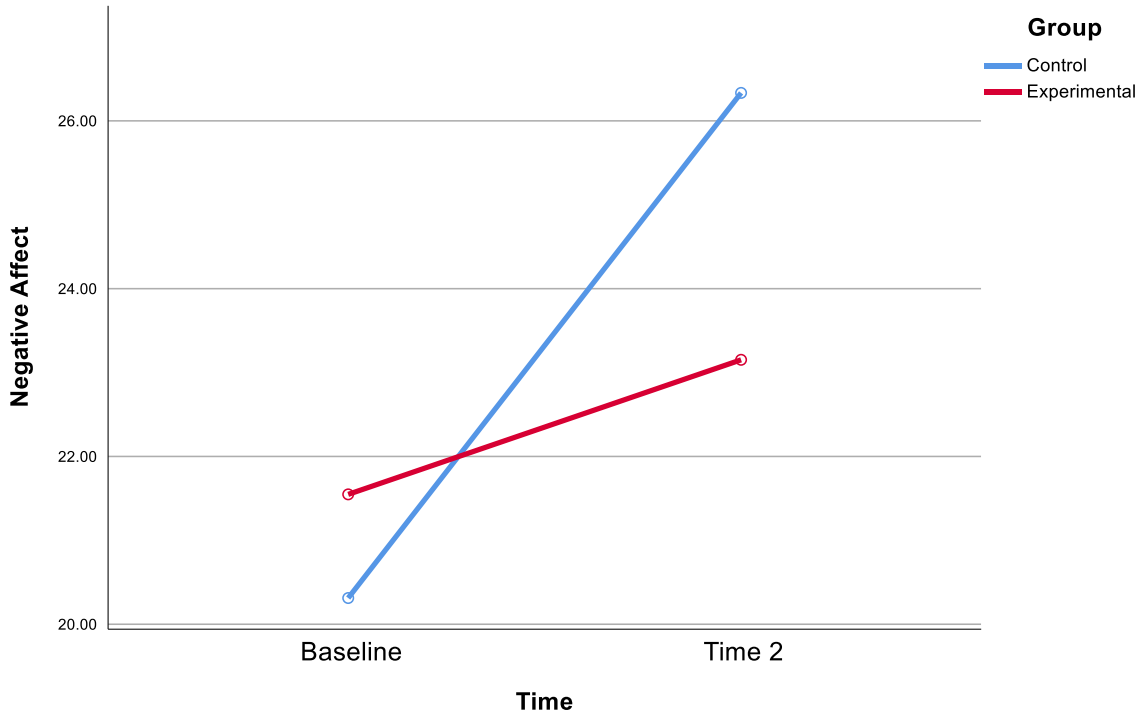

Figure 3 Interaction between time and group

As can be seen, the relationship was characterised by a disordinal interaction. Paired-samples t-tests confirmed that, in the control group, scores worsened significantly across time, t (23) = -3.595, p = .002. However, in the experimental group they did not, t (26) = -1.234, p = .228. Independent samples t-tests on PANAS negative-affect subscale-scores revealed that there was not a significant between-group difference at T1, t (49) = -.538, p = .593, or at T2, t (49) = 1.159, p = .252.

Table 7 shows sensitivity analyses that were conducted to examine the effect of running the primary analysis whilst excluding participants from different sub-groups (which

were formed unintentionally during the study), and which might have feasibly impacted the results.

Table 7

The interaction between group and time on PANAS negative-affect subscale-scores, including sensitivity analyses

	F	*df*	*Sig.*	*Partial Eta Squared*
Excluding phase one participants	3.299	1, 40	.077	.076
Excluding site two participants	4.086	1, 44	.049*	.085
Excluding outliers	4.288	1, 47	.004*	.084
Excluding participants with < 1 day between T1 and T2	2.330	1, 34	.136	.064
Excluding participants with missing data	3.474	1, 45	.069	.072

Note. df, degrees of freedom; *sig*, significance; *, significant at $p < .05$ level; T1, time one; T2, time two

As can be seen from the table, the interaction between time and group was robust to excluding participants with outliers or excluding participants who were recruited from site two. However, when participants were excluded who had less than a day in between completion of their T1 and T1 measures, the interaction failed to reach significance. Excluding participants who had omitted a single item on the PANAS negative-affect subscale-scores (at either T1 or T2) also meant the interaction failed to reach significance, as did excluding participants recruited in phase one.

Secondary Analyses

A Mann-Whitney *U* test indicated that there was not a statistically-significant, between-group difference on the extent to which the information helped participants know what to expect from the assessment, $U = 250.000$ ($z = -0.319$), $p = .750$, $r = -.047$. A Mann-Whitney *U* test also indicated there was not a statistically-significant, between-group

difference on satisfaction with assessment $U = 134.000$ ($z = -1.568$), $p = .186$, $r = -.0254$. Table 8 reports the median values on predictability and satisfaction in both groups.

Table 8

Median scores on secondary outcomes, by group and time

	Experimental		Control	
	n	*Median (IQR)*	*n*	*Median (IQR)*
Predictability	24	4 (3 – 5)	22	4 (3– 5)
Satisfaction	20	5 (4.75 – 5)	18	4.5 (4 - 5)

Note. n, number*; IQR,* interquartile range.

Descriptively, predictability and satisfaction scores were consistently high across groups.

Effect of Age and Gender

Because there were unequal numbers of males and females in each groups, exploratory analyses (not planned a priori) were conducted to examine the effect of gender on PANAS negative-affect scores. An independent-samples *t*-test suggested there was a significant difference between males and females at T1, t (47) = -3.462, $p = .001$, with females having higher scores ($M = 24.713$, $SD = 7.105$) than males ($M = 17.467$, $SD = 7.432$). Therefore, an independent samples t-test was conducted on change scores to examine whether gender impacted the change in negative affect across time. The analysis revealed that female change scores ($M = 4.163$, $SD = 8.262$) and male change scores ($M = 3.473$, $SD = 7.432$) did not differ significantly t (47) = -3.040, $p = .762$. Although age was relatively consistent across groups, a Pearson's correlation was conducted to confirm age was not a

potential confound. This test revealed there was not a significant correlation between age and negative affect change-scores across the sample (r = -.053, p = .725).

Discussion

A clinical trial was conducted to examine whether reading a Social Story would have a salutary impact on negative affect, predictability and satisfaction in relation to attending a diagnostic assessment for ASD. Overall, the results suggested the intervention did improve negative affect, but not predictability or satisfaction.

The primary hypothesis was that participants in the experimental group would report less of an increase in negative affect upon attending the assessment, relative to participants in a control group. It was assessed by comparing groups on how negative affect changed from time one (self-reported at home, approximately one week prior to an assessment) to time two (self-reported upon arrival at the host site for the diagnostic assessment). Results from the primary analysis showed there was a statistically-significant interaction between time and group (such that negative affect in participants in the control group increased at a greater magnitude over time), with a medium effect size observed for the interaction. Therefore, this finding supported the primary hypothesis. It was noted that the primary analysis was inadequately powered (most likely due to a smaller effect size being observed than was anticipated). It is unsurprising the a priori sample size estimate was fallible, given the extremely limited data that was available from previous research to inform the estimate of effect size. A pragmatic approach was therefore adopted and, although the risk of making a type II error was increased by the study being underpowered, this is fortunately inconsequential given a significant result was obtained and the null hypothesis rejected.

Although the primary data-analytic technique was to test the interaction between time and group, additional analyses sought to shed further light. Participants in the control group

reported significantly more negative affect at time two than at baseline; whereas negative affect did not change significantly in participants in the experimental group. Although this finding provided further evidence that the intervention had a positive impact, a between-group comparison of means revealed there was not a significant difference in negative affect at time two. However, this analysis was insufficiently powered and it is possible this is why a significant difference was not found.

There are an abundance of studies synthesised in meta-analytic research (e.g. Kokina & Kern, 2010) that have shown Social Stories can be effective at reducing the frequency and/ or severity of overt expressions of negative affect in people with ASD. Furthermore, Cullain (2000) demonstrated that reading a Social Story prior to a social interaction had a positive impact on self-reported anxiety (as well as behaviour deemed to be an expression of negative affect) in individuals with ASD. The present work supports these findings. It extended the literature by using adult participants in the UK, a self-reported measure of negative affect and a between-groups design (in line with recommendations by Kokina and Kern).

It was theorised that predictability would be the mechanism by which the Social Story would reduce negative affect. The associated hypothesis was that participants in the experimental group would give significantly higher predictability ratings. However, there was not a statistically-significant difference found between-groups. This might be because the Social Story was ineffective at increasing predictability and had a different mechanism of action on reducing negative affect. It is also possible that the Social Story did increase predictability, but that the standard leaflets the host sites sent out were equally effective. Another explanation is that the measure used was not a reliable and/or valid instrument for capturing predictability. This is plausible given it was a one-item measure created specifically for this study. Given predictability was higher than anticipated across groups, there may have been a ceiling effect on the 5-point measure, preventing differences to be detected. Finally,

participants were asked if the information they read helped them to know what to expect from the assessment before they knew the outcome. This might have introduced a bias as some may have answered the question in relation to whether they were any clearer about whether they would receive a diagnosis; something neither the control or intervention information was designed to do.

Quantitative research utilising self-report measures (e.g. Fujino et al., 2019) has highlighted the importance of predictability for people with ASD and qualitative studies (e.g. Trembath et al., 2012) have reported that increasing predictability can reduce negative affect in this population. Furthermore, a qualitative service evaluation conducted at the primary host site by an independent researcher (Trivasse, 2019) found that participants remembered feeling anxious due to uncertainty about what the assessment would involve. For example, several participants described how they had initially been unable to find the correct building and, when they found it, were confused by the procedure for accessing reception; adding to their anxiety and stress. Although the Social Story in the present study included specific guidance to dispel this type of confusion, more research is required as, unfortunately, the results did not shed any light on whether predictability was the mechanism of action by which the Social Story had a salutary impact on negative affect.

The final hypothesis was that participants in the experimental group would report significantly higher satisfaction. This hypothesis was informed by previous researchers (e.g. Crane et al., 2018; Jones et al., 2014) who found that many UK adults were dissatisfied with the diagnostic process they had undergone to receive a diagnosis of ASD; partially due to a lack of knowing what to expect. Therefore, it was theorised that by increasing predictability, the Social Story would increase satisfaction. However, the results did not support this prediction as satisfaction ratings were consistently high across groups (and there was not a

significant between-group difference). It is likely the intervention did not have a significant effect on satisfaction therefore.

It been demonstrated in previous research (e.g. Powell & Acker, 2016) that those who do not receive a diagnosis following an ASD assessment are often left dissatisfied. Because participants in the present study completed the satisfaction question shortly after they had been informed of the diagnostic outcome, it is plausible the outcome inflated their sense of satisfaction (given only those who received a diagnosis were included in the data analysis). Given the diagnostic outcome was uniform across groups, this might be why no between-group differences in satisfaction were found. Furthermore, 25% of participants did not complete the satisfaction measure and so it is possible the results were not representative of the sample.

Strengths, Limitations and Future Directions

The study was designed to run alongside routine clinical practice and only required a small additional investment of time from participants. There were no incentives offered for participation and people were recruited via an invitation to participate; helping to ensure that individuals chose to take part of their own free-will. Participant-burden was also minimised because the social stressor that was anticipated to raise negative affect (attendance at the assessment) would have been experienced by clients, irrespective of whether they chose to participate in the study. The study therefore had high ethical-integrity. Furthermore, as a naturalistic setting was used and inclusion criteria were loose, the results have high ecological validity. The disadvantage of this method, however, was that internal validity was reduced as there was less control over potential confounds.

It was not possible to confirm intervention-fidelity (i.e. that participants read the Social Story). Furthermore, in phase two, baseline questionnaires were sent at the same time

as the intervention. Therefore, it is plausible that some participants could have deviated from the included instructions and read the information prior to completing the baseline measures; possibly increasing their baseline negative affect as they contemplated their forthcoming assessment. Sensitivity analyses were conducted but were based on much smaller sample sizes and so were inadequately powered for the results to be interpreted meaningfully.

Participants in phase two were randomly assigned to group prior to researcher-confirmation of informed consent. The disadvantage of this method of randomisation was that it resulted in slightly uneven group sizes. This was likely to have reduced the statistical power of the analyses (Rusticus & Lovato, 2014). It also raised an ethical question as clients were sent the intervention before they provided informed consent. However, due to the nature of the intervention (written information created with the help of service-users) and people's explicit right to ignore it, the NHS-ethics board approved this procedure.

The amended process in phase two also meant control was lost over procedural fidelity. Participants in phase two were asked to complete baseline measures upon receipt and time two measures upon arrival at the host site, but 11 participants recorded the same date on both measures; evidencing that the instructions were not adhered to. It is possible these participants completed both measures at the same time; impacting the degree to which negative affect appeared to change over time. It is important that the results be replicated in a future study, ideally with tighter control over procedural fidelity.

Another limitation was the length of time the study ran for. The first participant completed the study approximately ten months earlier than the final participant and so the experience of attending an assessment might have differed in subtle ways at different time points. This was mitigated somewhat by randomisation to group and by regular checks at the host sites to ensure there were not any substantial changes that affected the accuracy of the

Social Story. The length of time the study ran for was due to difficulties with recruitment. Although the reasons why clients declined to participate are unknown, the considerable increase in recruitment rate in the second phase suggested the requirement to visit a website to provide informed consent was a barrier (as this was only applicable to phase one).

Commencing recruitment from a second site during the study (requiring slight modifications to the Social Story) was another limitation as it is standard practice in clinical trials to ensure all participants in the experimental group receive exactly the same intervention. However, this did not appear to affect the results as a sensitivity analysis revealed that a significant result and a similar effect size were obtained when using participants exclusively from site one.

As people with ASD can have difficulties with attention and concentration (Southwick et al., 2011), questionnaires were kept brief. It would have been useful to have collected more demographic data so that the results could be generalised more easily. The present study did not find that age or gender had an impact on the interaction between time and negative affect, but it would be useful for this to be replicated.

The use of exploratory measures to investigate secondary hypotheses was a significant limitation. Their psychometric properties were not known and so the results might not be reliable or valid. Because people with ASD often have difficulties with abilities such as interoceptive awareness and alexithymia (Garfinkel et al., 2016), it could be argued that using a self-reported affect measure as the primary dependent variable was also inadvisable. However, previous researchers (e.g. Berthoz & Hill, 2005; Buvinger, 2013) have rebuked this argument by demonstrating that high-functioning individuals with ASD can self-report their emotions reliably.

There might have been participants in phase two who completed informed consent and outcome measures, but then forgot to hand them in. Therefore, although this study appeared to have a low proportion of missing data, participants might have been excluded indirectly; creating a non-representative sample (e.g. people with above-average memory).

Although qualitative feedback was not planned (and thus could not be included in the results), one participant in the experimental group wrote on her time two outcome measure that she found the Social Story patronising. The vast majority of the Social Story literature has been focussed on children (Wright et al., 2016), and, therefore, the intervention might be unsuitable for some adults. Future researchers might wish to investigate this systematically and to measure IQ and linguistic abilities so that more is known about the types of individual the intervention is suited for.

The design of the study did not permit the evaluation of which aspects of the intervention were responsible for the beneficial impact on negative affect. For example, it is plausible that similar results would have been obtained had the photographs and content and been presented in a style that did not adhere to Gray's (2018) Social Story criteria. Further research is required to delineate which components of the intervention are necessary.

Clinical Implications

For the two host sites included in the study, the results suggest that sending the Social Story to clients ahead of their appointment might be a way that the negative affect associated with attending can be reduced. Arguably, this would be consistent with recommendations for services to make reasonable adjustments for clients with ASD (Department of Health, 2010). However, it is important that the limitations discussed above are acknowledged as the intervention might not be beneficial for some and the results might not generalise to other services. Should similar services wish to consider adapting the intervention to their clients, it

is recommended that the criteria specified by Gray (2018) are followed; especially the involvement of service-users in the development of the intervention. It is also recommended that effectiveness and acceptability are evaluated utilising qualitative and quantitative methods.

Although it was not investigated in the present study, previous research (e.g. Ferrara & Hill, 1980; Trembath et al., 2012) has suggested that negative affect often motivates avoidance behaviour in people with ASD. Therefore, the results might have wider implications for reducing non-attendance rates and improving access. One participant in the experimental group wrote on the time 2 outcome measure that, due to anxiety, she would not have attended the diagnostic assessment had she not been sent the information about what to expect beforehand. Although this unprompted feedback cannot be interpreted, it suggests that a potential avenue for future research might be to examine the impact of Social Stories on attendance rates at healthcare services. This could not be investigated here given participants were required to hand their consent form to their clinician (giving the impression of a 100% attendance rate).

In the introduction it was argued that experiencing state negative affect in public might feasibly impact on self-worth and the opportunities individuals with ASD have to develop their social skills. Although further research is needed, it is conceivable that an intervention that lowers state negative affect during social interactions might help people with ASD utilise their social skills and recognise they have more potential at social interaction than they have learnt to give themselves credit for. Arguably, a diagnostic assessment for ASD might represent an opportune time to experience a positive social interaction as it is known to be a pivotal time in a person's life that can determine how he or she begins to reframe and reshape their identity (Hays & Colaner, 2016; Tan, 2018; Webster & Garvis, 2016). It is argued here that researchers and services should therefore be actively

investigating techniques (such as Social Stories) to help make this process as supportive and tailored to individual need as possible.

Conclusions

The results of the present study demonstrated that a simple, low-cost intervention helped reduce negative affect in people attending a diagnostic assessment for ASD. Given methodological rigour was at times subordinated to ethical integrity, there were a number of limitations with the study and caution must be exercised in generalising the findings.

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Appendix A

Social Story 10.2 Criteria

Criterion 1: The Social Story Goal

Authors follow a defined process to share accurate information using a content, format, and voice that is descriptive, meaningful, and physically, socially, and emotionally safe for the Audience.

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Criterion 2: Two-Step Discovery

Keeping the Goal in mind, authors gather relevant information to 1) improve their understanding of the Audience in relation to a situation, skill, or concept and/or 2) identify the specific topic(s) and the most critical information (focus) of each Story. At least 50% of all Social Stories applaud achievements.

Criterion 3: Three-Parts & a Title

A Social Story/Article has a title and introduction that clearly identifies the topic, a body that adds detail, and a conclusion that reinforces and summarizes the information.

Criterion 4: Four·mat Makes it Mine!

The Social Story™ format is tailored to the individual abilities, attention span, learning style and - whenever possible – talents and/or interests of its Audience.

Criterion 5: Five Factors Define Voice & Vocabulary

A Social Story has a patient and supportive "voice" and vocabulary that is defined by five factors:

1) Exclusive use of first- and/or third-person perspective statements (no second person statements);
2) Past, present, or future tense;
3) Positive and patient tone;
4) Literally accurate; and
5) Accurate meaning.

Criterion 6: Six Questions Guide Story Development

A Social Story™ answers relevant 'WH' questions that describe context, including place (WHERE), time-related information (WHEN), relevant people (WHO), important cues (WHAT), basic activities, behaviors, or statements (HOW), and the reasons or rationale behind them (WHY).

Criterion 7: Seven is About Sentences

A Social Story is comprised of Descriptive Sentences and may also have one or more Coaching Sentence(s). Sentences adhere to all applicable 10.2 criteria.

Descriptive Sentences accurately describe relevant aspects of context, including external and/or internal factors while adhering to all applicable Social Story Criteria. They are free of assumption or bias, judgment, devaluation, or unidentified opinion

Coaching Sentences gently guide behavior via descriptions of effective Team or Audience responses, or structured Audience Self-Coaching, adhering to all other applicable Social Story Criteria.

#8 A Gr·eight! Formula

The Social Story™ Formula ensures that every Social Story describes more than directs.

$$\frac{\text{Total \# of Descriptive Sentences}}{\text{Total \# of Coaching Sentences}} \geq 2$$

*If there are no (0) Sentences that Coach, use 1 in the denominator.

Criterion 9: Nine to Refine

The first draft of a story is rarely the final draft. A story draft is always reviewed by relevant caregivers and revised if necessary to ensure that it meets all defining Social Story criteria.

Criterion 10: Ten Guides to Editing and Implementation

The Ten Guides to Implementation ensure that the philosophy and Criteria that guide Story/Article development are consistent with how it is introduced and reviewed with the Audience. They are:

1) Plan for Comprehension
2) Plan Story Support
3) Plan Story Review
4) Plan a Positive Introduction
5) Monitor
6) Organize the Stories
7) Mix & Match to Build Concepts
8) Story Re-runs and Sequels to Tie Past, Present, and Future
9) Recycle Instruction into Applause
10) Stay Current on Social Story Research and Updates

Appendix B

Example Pages from Social Story and Service Leaflets

Site one

Walking through the entrance leads to the reception area.

People can tell the receptionist they are here for an assessment for ASD.

It's a good idea to leave plenty of time for the journey (in case it takes longer than expected).

Therefore, most people arrive a little early and have a short wait before their appointment.

The reception area is usually fairly quiet. However, people can choose to wait outside if they prefer.

Site two

Walking through the entrance leads to the reception area. It is usually fairly quiet.

People can tell the receptionist they are here for an assessment for ASD.

It's a good idea to leave plenty of time for the journey (in case it takes longer than expected).

Therefore, most people arrive a little early and have a short wait before their appointment.

People can wait for their clinician in the seated area.

Site one

Many people find the clinician's questions difficult to answer because they are anxious and in new surroundings. It's okay if people want to take a few moments to think, before answering a question.

People being assessed sometimes force themselves to make eye-contact and stop themselves from fidgeting. This is not necessary and usually increases their anxiety.

The assessment will take approximately 2 – 3 hours. At the start, the clinician will describe what the assessment involves and suggest ways of making the assessment more comfortable (for example, planning comfort breaks and giving out sensory toys).

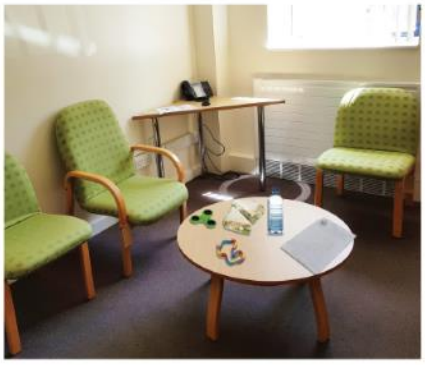
There is a box of sensory toys (including a weighted blanket) that people can use to comfort themselves during the assessment.

The assessment includes a lot of questions and, therefore, it can be emotionally-draining for people.

It's a good idea to rest before and after the assessment.

The assessment does not include a physical examination.

Site two



The assessment will last a minimum of three hours. However, it can last up to six hours. Sometimes it is easier if the person attends two shorter appointments rather than one long one. This will be discussed during the assessment.

At the start, the clinician will describe what the assessment involves and suggest ways of making the person more comfortable (for example, scheduling comfort breaks and offering to close the blinds).

People are welcome to bring their own food / drink to their appointment. They are also welcome to bring sensory items if they wish. Clinicians understand sensory needs.

Many people find the clinician's questions difficult to answer because they are anxious and in new surroundings. It's okay if people want to take a few moments to think, before answering a question.

People being assessed sometimes force themselves to make eye-contact and stop themselves from fidgeting. This is not necessary and usually increases their anxiety.

The assessment includes a lot of questions and, therefore, it can be emotionally-draining for people. It's a good idea to rest before and after the assessment. The assessment does not include a physical examination.

Site one

Topics that might be discussed include:

- Why the person has decided to have an assessment for ASD.
- What sensory issues they find challenging.
- Their thinking-style (e.g. do they often see things in a 'black or white' way).
- Their history of relationships (e.g. describing relationships with friends, family, ex-boyfriends/girlfriends etc.).

- Their strengths/difficulties in conversation and how they understand others.
- Their daily life (e.g. their interests, their routines, and how they cope with change).

- How they used to play as a child.
- Risk issues (e.g. self-harm).

Site two

Topics that might be discussed include:

- Why the person has decided to have an assessment for ASD.
- What sensory issues they find challenging.
- Their thinking-style (e.g. do they often see things in a 'black or white' way).
- Their history of relationships (e.g. describing relationships with friends, family, ex-boyfriends/girlfriends etc.).

- Their strengths/difficulties in conversation and how they understand others.
- Their daily life (e.g. their interests, their routines, and how they cope with change).

- How they used to play as a child.
- Risk issues (e.g. self-harm).

Site one

People often worry about giving the "right" answer to the clinician's questions. Although there are no "right" answers, people can struggle with this concept. There are no right (or wrong) answers because diagnosing ASD in adulthood is not a 'tick box' activity.

The clinician's job is to get to know people and to understand their unique strengths and difficulties and how they experience the world. The clinician then considers whether a diagnosis of ASD is indicated. Because no two people are the same, there are no right or wrong answers.

Towards the end of the appointment the person will be told the outcome of the assessment. There are three potential outcomes:

1. **A diagnosis of ASD is indicated.**
2. **A diagnosis of ASD is not indicated.**
3. **The clinician needs more information before he or she is able to decide.**

If more information is needed before a decision is made, another appointment will be scheduled. However, the clinician will usually able to decide without requiring another appointment.

Site two

People often worry about giving the "right" answer to the clinician's questions. Although there are no "right" answers, people can struggle with this concept. There are no right (or wrong) answers because diagnosing ASD in adulthood is not a 'tick box' activity.

The clinician's job is to get to know people and to understand their unique strengths and difficulties and how they experience the world. The clinician then considers whether a diagnosis of ASD is indicated. Because no two people are the same, there are no right or wrong answers.

Sometimes the clinician will be able to tell whether a diagnosis is indicated at the end of the assessment appointment(s). However, if the decision is more complicated they may need additional time to consider the information and to discuss it with other members of the multidisciplinary team.

The clinician will discuss this with the person at the end of the assessment so that they know what is going to happen next and how long they may need to wait.

Site one

The clinician will tell the person the outcome at the end of the assessment.

If the clinician identifies enough evidence for a diagnosis, this will be provided in a formal report.

The report will be posted to the person shortly after the assessment.

If a diagnosis of ASD is not indicated, the clinician will talk to the person about what else might be causing/maintaining their difficulties.

Site two

If the clinician identifies enough evidence for a diagnosis, this will be provided in a formal report.

Reports take a long time to write and individuals may have to wait for them, but clinicians are happy to write a brief summary of the diagnosis for individuals who prefer to find out sooner.

Individuals can choose who receives a copy of their report (their GP etc.)

Each person is offered a follow-up appointment. Follow-up appointments are sometimes used to discuss the diagnosis or changes individuals wish to make to their report.

Other people use this appointment to find out about the local therapeutic or support services that are available. Clinicians can make referrals on a person's behalf but will only do so if they have the person's consent. Some people prefer to think about their options and contact services themselves.

If the person wishes, the clinician can also discuss strategies for managing their difficulties and how they can build upon their strengths.

Site one

Many people attend an ASD assessment because they are keen to understand themselves better.

People usually acquire a better understanding of themselves from the assessment, even if a diagnosis of ASD is not given..

The clinician will always try their best to think with the person about the type of post-diagnostic support that might benefit them (e.g. counselling; a recommendation to another service; written information about their difficulties etc.).

The clinician will also think with the person about strategies for managing their difficulties and how they can build upon their strengths.

People commonly report finding the assessment a validating and empowering process.

Site two

Many people attend an ASD assessment because they are keen to understand themselves better. People usually acquire a better understanding of themselves from the assessment, even if a diagnosis of ASD is not given.

People commonly report finding the assessment a validating and empowering process.

Site one (control leaflet)

What is an Autism Spectrum Disorder (ASD)?

An Autism Spectrum Disorder (ASD) is a lifelong developmental disability that affects how a person communicates with and relates to other people. It also affects how they make sense of the world around them. The key features of ASD are:

- Difficulties in social communication and social interaction
- Restricted or repetitive patterns of behaviour, interests or activities
- Highly focused interests
- Sensory processing difficulties

Further information is available from the National Autistic Society website: www.autism.org.uk

What is Attention Deficit Hyperactivity Disorder (ADHD)?

ADHD can be associated with ASD. People with ADHD experience hyperactivity, inattention, distractibility and impulsivity which may cause difficulties at work or home.

We are able to offer:

Diagnostic Assessment

How could a diagnosis benefit me?

Receiving a diagnosis of ASD can give individuals, their friends and family a better understanding of the person's needs and abilities.

In the case of ADHD advice and support can be given to help manage the condition.

A diagnosis can also help individuals identify their unique strengths and abilities.

A diagnostic assessment can be a highly emotional experience. The person you meet will support you with this.

Who is this service for?

Our diagnostic service is open to anyone over the age of 16 who lives in Sheffield and has reason to believe they have ASD or ADHD.

A national service is also available subject to funding being agreed.

How do I get a referral to the service?

We are unable to accept self-referrals. Referrals would usually come from your GP.

Please note that if a person has a diagnosis of a Learning Disability they may be seen by the Community Learning Disabilities Team located at Love Street.

What should I expect when I come for an assessment?

You will receive a letter from this service offering you an appointment. We will send you a questionnaire with this letter about your early experiences which would be best filled out by a family member or a person close to you.

On the day of your appointment you will be welcomed into the waiting area where you can buy a drink if you would like to. There may be a quieter waiting area, outside space or sensory space available; please speak to reception if you would like to use this.

Your appointment will last between 2 and 4 hours and there will be the opportunity to have breaks during this time, if you need them. You can go in to the appointment alone but it may be useful to take a friend or family member in with you.

The person you meet will talk to you and ask you different questions about your life – from your childhood through to your experiences as an adult – which will help them to make a decision about the outcome of the appointment. They will share their thoughts with you about the assessment towards the end of your appointment and will support you to understand the outcome.

We also offer:

Reassessment of Needs: where an individual has an existing diagnosis and their support needs have changed.

Support Following Diagnosis

Post Diagnostic Follow Up

Nice Guidelines recommend that you are offered a follow-up appointment to discuss the results of your assessment. If you receive a diagnosis of ASD you will be offered a follow-up appointment a few weeks after your diagnosis (this will be subject to funding for national referrals).

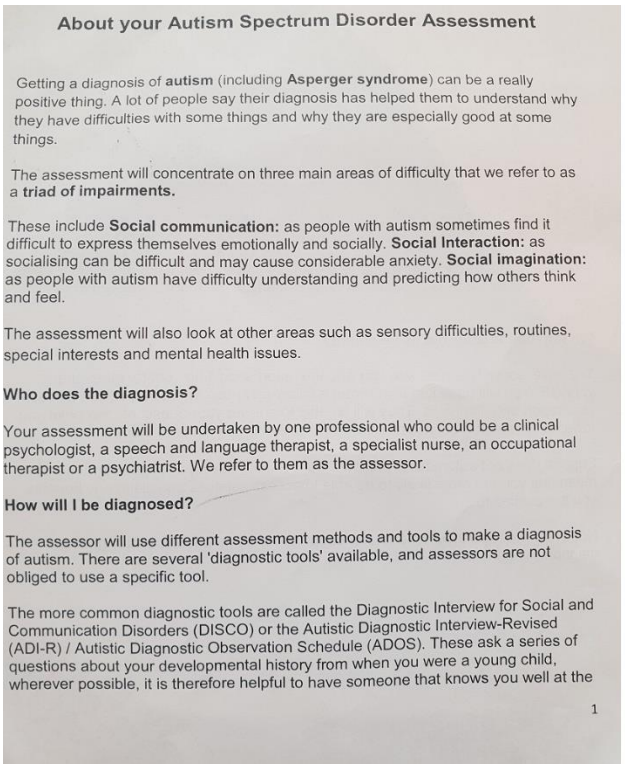
During this appointment you will have the opportunity to talk about your thoughts and feelings following your assessment and to ask any questions you may have about support in the future.

Post Diagnostic Support may include:

- Speech and Language Therapy to support with communication (SLT)
- Occupational Therapy (OT) may include support with Employment/Education, Sensory Processing assessment and Developmental Co-ordination Disorder assessment
- Psychological Support
- ADHD medication advice
- Psychometric Assessment
- Group Support
- Information and Signposting to other support services

Please note: Any support offered will be based on individual need.

Site two (control leaflet)



About your Autism Spectrum Disorder Assessment

Getting a diagnosis of **autism** (including **Asperger syndrome**) can be a really positive thing. A lot of people say their diagnosis has helped them to understand why they have difficulties with some things and why they are especially good at some things.

The assessment will concentrate on three main areas of difficulty that we refer to as a **triad of impairments.**

These include **Social communication:** as people with autism sometimes find it difficult to express themselves emotionally and socially. **Social Interaction:** as socialising can be difficult and may cause considerable anxiety. **Social imagination:** as people with autism have difficulty understanding and predicting how others think and feel.

The assessment will also look at other areas such as sensory difficulties, routines, special interests and mental health issues.

Who does the diagnosis?

Your assessment will be undertaken by one professional who could be a clinical psychologist, a speech and language therapist, a specialist nurse, an occupational therapist or a psychiatrist. We refer to them as the assessor.

How will I be diagnosed?

The assessor will use different assessment methods and tools to make a diagnosis of autism. There are several 'diagnostic tools' available, and assessors are not obliged to use a specific tool.

The more common diagnostic tools are called the Diagnostic Interview for Social and Communication Disorders (DISCO) or the Autistic Diagnostic Interview-Revised (ADI-R) / Autistic Diagnostic Observation Schedule (ADOS). These ask a series of questions about your developmental history from when you were a young child, wherever possible, it is therefore helpful to have someone that knows you well at the

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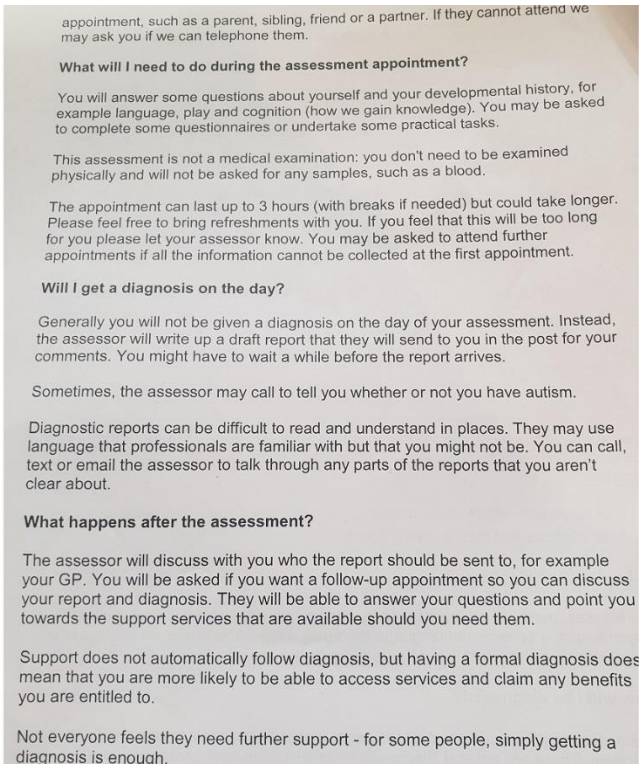


appointment, such as a parent, sibling, friend or a partner. If they cannot attend we may ask you if we can telephone them.

What will I need to do during the assessment appointment?

You will answer some questions about yourself and your developmental history, for example language, play and cognition (how we gain knowledge). You may be asked to complete some questionnaires or undertake some practical tasks.

This assessment is not a medical examination: you don't need to be examined physically and will not be asked for any samples, such as a blood.

The appointment can last up to 3 hours (with breaks if needed) but could take longer. Please feel free to bring refreshments with you. If you feel that this will be too long for you please let your assessor know. You may be asked to attend further appointments if all the information cannot be collected at the first appointment.

Will I get a diagnosis on the day?

Generally you will not be given a diagnosis on the day of your assessment. Instead, the assessor will write up a draft report that they will send to you in the post for your comments. You might have to wait a while before the report arrives.

Sometimes, the assessor may call to tell you whether or not you have autism.

Diagnostic reports can be difficult to read and understand in places. They may use language that professionals are familiar with but that you might not be. You can call, text or email the assessor to talk through any parts of the reports that you aren't clear about.

What happens after the assessment?

The assessor will discuss with you who the report should be sent to, for example your GP. You will be asked if you want a follow-up appointment so you can discuss your report and diagnosis. They will be able to answer your questions and point you towards the support services that are available should you need them.

Support does not automatically follow diagnosis, but having a formal diagnosis does mean that you are more likely to be able to access services and claim any benefits you are entitled to.

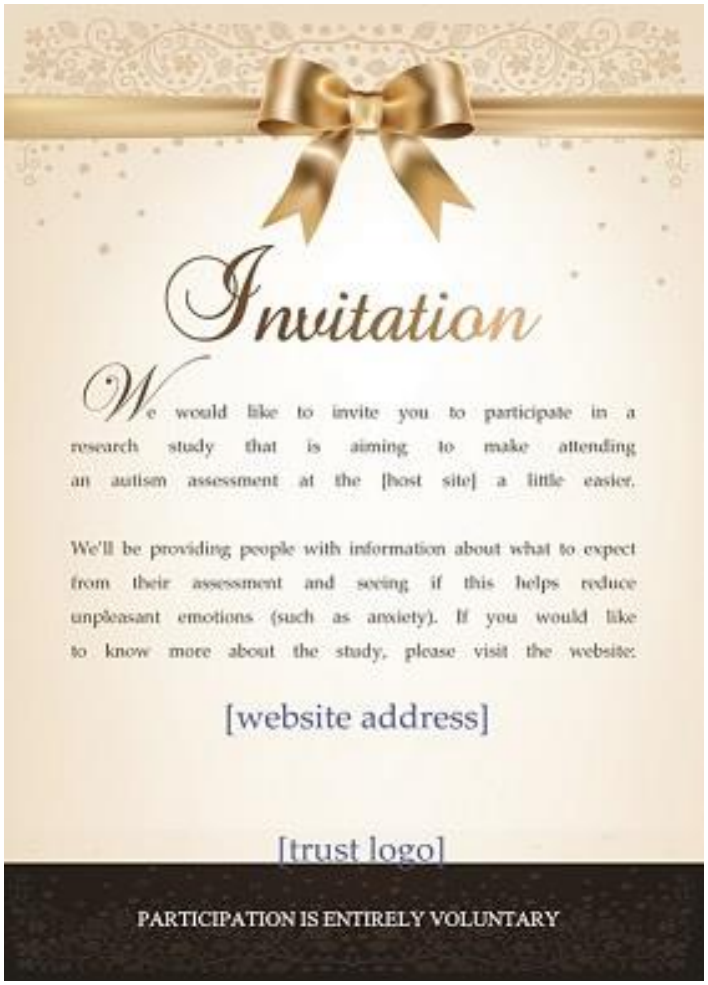
Not everyone feels they need further support - for some people, simply getting a diagnosis is enough.

Appendix C

Postal Invitation (labelled version 5 on IRAS)

Appendix D

Postal Invitation (labelled version 6 on IRAS)



Appendix E

Postal Invitation (labelled cover letter version 3 on IRAS)

[logo]

Invitation

My name's Rich and I'm a Trainee Clinical Psychologist. As part of a research study that's taking place at the [host site]. I'm providing people with information about what to expect from their assessment and seeing if this helps reduce unpleasant emotions (such as anxiety).

If you'd like to take part, please read through the information provided and complete the Research Booklet – **please remember to bring the Research Booklet with you to your appointment.**

Please feel free to contact me if you have any questions about the study. My email address is: rjenkinson2@sheffield.ac.uk. Please direct any questions about your assessment [host site; telephone number].

Yours sincerely,

Richard Jenkinson

Trainee Clinical Psychologist / Lead Researcher

[Trust Name] NHS Foundation Trust / The University of Sheffield

Appendix F

Research Booklet Cover for Amendment 1 (labelled version 1 on IRAS) when Materials were Supplied in Paper Format in a Booklet

[Trust Logo] Iras ID: 239758, V1, 20.08.2018

Research Booklet

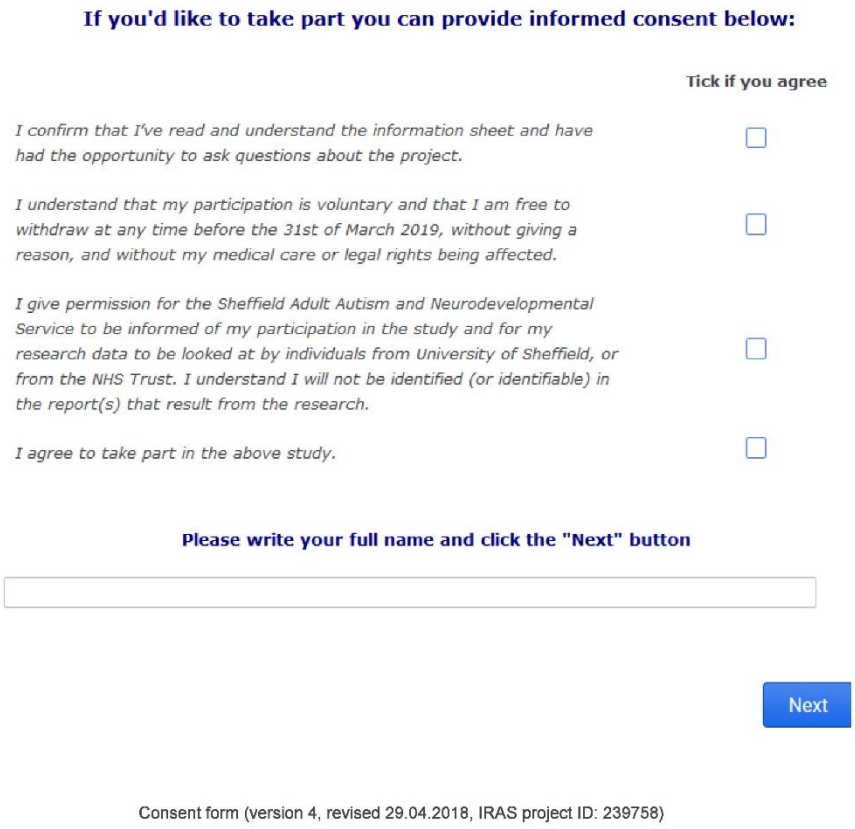
1. Please read the participant information sheet provided.

2. If you wish to participate in the study, please complete the consent form on the next page and the first questionnaire.

3. Please then read through the information provided about what to expect from your assessment. The questionnaire on the back of this booklet is for you to complete when you arrive at your appointment (so please try and remember to bring it with you). Please then give this booklet to your clinician.

Appendix G

Online Consent Form at Start of Trial (labelled version 4 on IRAS)



If you'd like to take part you can provide informed consent below:

	Tick if you agree
I confirm that I've read and understand the information sheet and have had the opportunity to ask questions about the project.	☐
I understand that my participation is voluntary and that I am free to withdraw at any time before the 31st of March 2019, without giving a reason, and without my medical care or legal rights being affected.	☐
I give permission for the Sheffield Adult Autism and Neurodevelopmental Service to be informed of my participation in the study and for my research data to be looked at by individuals from University of Sheffield, or from the NHS Trust. I understand I will not be identified (or identifiable) in the report(s) that result from the research.	☐
I agree to take part in the above study.	☐

Please write your full name and click the "Next" button

Next

Consent form (version 4, revised 29.04.2018, IRAS project ID: 239758)

Appendix H

Paper Consent Form -Substantial Amendment 1 (labelled version 5 on IRAS)

[Logo] Iras ID: 239758, V5, 20.08.2018

Consent Form

If you'd like to take part you can provide informed consent below:

	Please initial
I confirm that I've read the information sheet and have had the opportunity to ask questions about the project.	
I understand that my participation is voluntary and that I am free to withdraw at any time before the 31st of March 2019, without giving a reason, and without my medical care or legal rights being affected.	
I give permission for the [host site] autism service to be informed of my participation and for my research to be looked at by individuals from the University of Sheffield, or from the NHS Trust. I understand I will not be identified (or identifiable) in the report(s) that result from the research.	
I agree to take part in the above study.	

Your full name:…….. Today's date……

Appendix I

Paper Consent Form -Substantial Amendment 2 – to cover host sites can confirm diagnosis of future participants (labelled version 6 on IRAS)

[Logo] Iras ID: 239758, V6, 28.11.2018

Consent Form

If you'd like to take part you can provide informed consent below:

	Please initial
I confirm that I've read the information sheet and have had the opportunity to ask questions about the project.	
I understand that my participation is voluntary and that I am free to withdraw at any time before the 1st June 2019, without giving a reason, and without my medical care or legal rights being affected.	
I give permission for the [host site] autism service to be informed of my participation and for [host site] to confirm the outcome of my assessment with the Lead Researcher. I understand my research data may be looked at by individuals from the University of Sheffield, or from the NHS Trust. I understand I will not be identified (or identifiable) in the report(s) that result from the research.	
I agree to take part in the above study.	

Your full name:…….. Today's date……

Appendix J

Participant Information Sheet at Start of Trial & Unchanged in Substantial Amendment 1

(labelled version 5 on IRAS)

[University Logo]

PARTICIPANT INFORMATION SHEET

Study title: The Use of Social Stories to Reduce Negative Affect and Improve Satisfaction in Adults Attending an Assessment for Autism Spectrum Disorder

What is the purpose of the study? Many people find the unpredictability of social situations emotionally distressing. It is likely that attending a diagnostic assessment for Autism Spectrum Disorder is no exception. This research project is looking at a potential way of making the assessment a little less stressful by helping people know what to expect. They'll be two groups in the study – one group will read standard information about the assessment and the other group will read newly-designed information that is written slightly differently (that research has suggested might be more effective). Each participant will be randomly assigned to one of the two groups. We won't be telling people which group they are in (to prevent potentially biasing the results). However, they'll be told after the study has finished. We want to know if the newly-designed information is more effective than the standard information in terms of helping people know what to expect, increasing satisfaction, and decreasing unpleasant emotions (such as anxiety). If the new information does prove to be more effective, other services might start using the technique to better cater for their clients' needs.

As the principle researcher is currently training to be a Clinical Psychologist at The University of Sheffield, the study is also being conducted for educational purposes.

Why have I been invited? You've been invited as you're awaiting a diagnostic assessment at the [host site]- the place where the research is currently taking place.

Do I have to take part? No- it is entirely voluntary. It's completely up to you and your decision won't affect the level of support you receive from the service.

What will happen if I take part? You'll be asked to fill in a brief questionnaire, online. You'll then be asked to read some information about your forthcoming assessment. When you arrive at your assessment you'll be asked to complete another brief questionnaire. We'll then contact you after

your assessment to ask you two questions about your experience. **What are the benefits of taking part?** You'll have a 50/50 chance of being in the newly-designed information group and, as previous research has suggested this might make the assessment process a little less stressful, it's possible you'll benefit in this way. People in both groups will still receive the standard information the service is currently sending out – so you won't miss out by participating. Another benefit is that you'll be contributing to a research study and, depending on the results, it might lead to more services using the newly-designed information to better cater for their clients' needs.

What are the risks of taking part? There are no known risks of using the newly-designed information. Whenever patient data is collected (whether it's for a research project or part of routine clinical practice), there is always a small risk that data will be accessed by a third-party (e.g. through a cyber attack). To minimise this risk, stringent methods of data collection and protection will be used and data will be anonymised wherever possible.

What if there is a problem? If there's a problem with the research project you can discuss it with the clinician who assesses you or with a member of staff at The University of Sheffield (contact details below).

Can I withdraw at any time? If you agree to participate in the study, you're still free to withdraw from the study at any point, without giving a reason, before the 31st of March 2019 (and your data will be deleted immediately). Shortly after this date your anonymous data will be analysed using statistical software (which is why it will not be feasible to destroy it after 31/03/2019).If you wish to withdraw you can call Richard Jenkinson by phone [telephone number] or by email (rjenkinson2@sheffield.ac.uk).

Will all the information be kept confidential? Study staff will protect your personal information closely so no one will be able to connect your responses and any other information that identifies you. We might be required to show information to University or NHS officials, who are responsible for monitoring the progress of this study. Directly identifying information (e.g. names, addresses) will be safeguarded and maintained under controlled conditions. Your personal data will be stored securely until March 2020 (at the latest). It will then be permanently deleted. Anonymous questionnaire responses will be stored until March 2030. You will not be identified in any publication from this study.

We will inform the team manager at the [host site] of your participation in the study. This is considered good practice for clinical research. It will not affect the care you receive in any way.

What will happen to the results of the study? We aim to get them published so that a wider audience can potentially benefit from the results.

What if I wish to complain about the way the study has been carried out? You can contact Elizabeth Milne, the Principle Researcher's academic supervisor, by phone (0114 22 26558) or by email (E.Milne@sheffield.ac.uk) and/or Andrew Thompson, Director of Research Training, by phone (0114 2226637) or by email (a.r.thompson@sheffield.ac.uk). If you feel that your complaint has not been handled to your satisfaction following this, you can contact the University's Registrar and Secretary Dr Andrew West, by phone (*0114 222 1051*) or by email: (registrar@sheffield.ac.uk).

Contact Information This research is being conducted by Richard Jenkinson**,** Trainee Clinical Psychologist. If you have any questions about the research, you can call Richard Jenkinson by phone [telephone number] or by email (rjenkinson2@sheffield.ac.uk). Please note, he will only be able to talk to you about the research, not your assessment in general.

Appendix K

Participant Information Sheet at Substantial Amendment 2 (labelled version 6 on IRAS)

[University Logo]

PARTICIPANT INFORMATION SHEET

Study title: The Use of Social Stories to Reduce Negative Affect and Improve Satisfaction in Adults Attending an Assessment for Autism Spectrum Disorder

What is the purpose of the study? Many people find the unpredictability of social situations emotionally distressing. It is likely that attending a diagnostic assessment for Autism Spectrum Disorder is no exception. This research project is looking at a potential way of making the assessment a little less stressful by helping people know what to expect. There will be two groups in the study – one group will read standard information about the assessment and the other group will read newly-designed information that is written slightly differently (that research has suggested might be more effective). Each participant will be randomly assigned to one of the two groups. We won't be telling people which group they are in to prevent potentially biasing the results. However, they'll be told after the study has finished. We want to know if the newly-designed information is more effective than the standard information in terms of helping people know what to expect, increasing satisfaction, and reducing unpleasant emotions (such as anxiety). If the new information does prove to be more effective, other services might start using the technique to better cater for their clients' needs.

As the principle researcher is currently training to be a Clinical Psychologist at The University of Sheffield, the study is also being conducted for educational purposes.

Why have I been invited? You've been invited as you're awaiting a diagnostic assessment at the [host site]- the place where the research is currently taking place.

Do I have to take part? No- it is entirely voluntary. It's completely up to you and your decision won't affect the level of support you receive from the service.

What will happen if I take part? You'll be asked to complete the Research Booklet and to read some information about your forthcoming assessment (enclosed). We'll then contact you after your assessment to ask you two questions about your experience.

What are the benefits of taking part? You'll have a 50/50 chance of being in the newly-designed information group and, as previous research has suggested this might make the assessment process a little less stressful, it's possible you'll benefit in this way. People in both groups will still receive the standard information the service is currently sending out – so you won't miss out by participating. Another benefit is that you'll be contributing to a research study and, depending on the results, it might lead to more services using the newly-designed information to better cater for their clients' needs.

What are the risks of taking part? There are no known risks of using the newly-designed information. Whenever patient data is collected (whether it's for a research project or part of routine clinical practice), there is always a small risk that data will be accessed by a third-party (e.g. through a cyber attack). To minimise this risk, stringent methods of data collection and protection will be used and data will be anonymised wherever possible.

What if there is a problem? If there's a problem with the research project you can discuss it with the clinician who assesses you or with a member of staff at The University of Sheffield (contact details below).

Can I withdraw at any time? If you agree to participate in the study, you're still free to withdraw from the study at any point, without giving a reason, before the1st of June 2019 (and your data will be deleted immediately). Shortly after this date your anonymous data will be analysed using statistical software (which is why it will not be feasible to destroy it after 01/06/2019).If you wish to withdraw you can call Richard Jenkinson by phone [telephone number] or by email (rjenkinson2@sheffield.ac.uk).

Will all the information be kept confidential? Study staff will protect your personal information closely so no one will be able to connect your responses and any other information that identifies you. We might be required to show information to University or NHS officials, who are responsible for monitoring the progress of this study. Directly identifying information (e.g. names, addresses) will be safeguarded and maintained under controlled conditions. Your personal data will be stored securely until March 2020 (at the latest). It will then be permanently deleted. Anonymous questionnaire responses will be stored until March 2030. You will not be identified in any publication from this study.

We will inform the team manager at the [host site] of your participation in the study. This is considered good practice for clinical research. It will not affect the care you receive in any way. We

will also ask the service to confirm the outcome of your assessment (i.e. whether you received a diagnosis or not).

What will happen to the results of the study? We aim to get them published so that a wider audience can potentially benefit from the results. If you would like a plain-English summary of the results, please request this via email (rjenkinson2@sheffield.ac.uk) and we will send the summary to you as soon as it is available.

What if I wish to complain about the way the study has been carried out? You can contact Elizabeth Milne, the Principle Researcher's academic supervisor, by phone (0114 22 26558) or by email (E.Milne@sheffield.ac.uk) and/or Andrew Thompson, Director of Research Training, by phone (0114 2226637) or by email (a.r.thompson@sheffield.ac.uk). If you feel that your complaint has not been handled to your satisfaction following this, you can contact the University's Registrar and Secretary Dr Andrew West, by phone (*0114 222 1051*) or by email: (registrar@sheffield.ac.uk).

Contact Information This research is being conducted by Richard Jenkinson**,** Trainee Clinical Psychologist. If you have any questions about the research, please contact Richard via email (rjenkinson2@sheffield.ac.uk). Please note, he will only be able to talk to you about the research, not your assessment in general.

Appendix L

Covering Letter for Participants who Signed up Online (labelled version 1 on IRAS)

[Logo]

Dear [participant name]

Thank you for participating in the research study currently taking place at [host site], it is very much appreciated. Please find enclosed an information booklet about what to expect from your assessment.

If you have any questions about the research study, please contact me via email: rjenkinson2@sheffield.ac.uk or via telephone: [telephone number]. Please direct any questions about your assessment to [host site] (telephone []).

Yours sincerely,

Richard Jenkinson

Trainee Clinical Psychologist / Lead Researcher

SHSC NHS Foundation Trust / The University of Sheffield

Appendix M

Debrief (labelled version 3 on IRAS)

[logo]

DEBRIEF

Study title: The Use of Social Stories to Reduce Negative Affect and Improve Satisfaction in Adults Attending an Assessment for Autism Spectrum Disorder

Thank you for your participation in our research study- we really value your contribution. Please find some additional information about the study below:

What was the purpose of the study? Many people find the unpredictability of social situations emotionally distressing. It is likely that attending a diagnostic assessment for Autism Spectrum Disorder (ASD) is no exception. This research study explored a potential way of making the assessment a little less stressful by helping people know what to expect. There were two groups in the study – one group read standard information about the assessment and the other group read newly-designed information that was written slightly differently. This newly-designed information was based on an intervention technique called Social Stories. You can find out more about Social Stories by visiting: https://carolgraysocialstories.com/social-stories/.

Each participant in our study was randomly assigned to one of the two groups. You were assigned to the [ENTER GROUP] group and so you read the [Social Story / Standard] information. We wanted to know whether the Social Story was more effective than the standard information in terms of helping people know what to expect, increasing satisfaction, and decreasing unpleasant emotions (such as anxiety). We won't know the results until we've had chance to analyse the data but, if you'd like to know the results when we do, please contact the lead researcher (rich Jenkinson-rjenkinson2@sheffield.ac.uk) after June 2019. If the Social Story does prove to be more effective, other services might start using the technique to better cater for their clients' needs.

What if I wish to complain about the way the study has been carried out? You can contact Elizabeth Milne, the Principle Researcher's academic supervisor, by phone (0114 22 26558) or by email (E.Milne@sheffield.ac.uk) and/or Andrew Thompson, Director of Research Training, by phone (0114 2226637) or by email (a.r.thompson@sheffield.ac.uk). If you feel that your complaint has not been handled to your satisfaction following this, you can contact the University's Registrar and Secretary Dr Andrew West, by phone (*0114 222 1051*) or by email: (registrar@sheffield.ac.uk).

Contact Information This research is being conducted by Richard Jenkinson**,** Trainee Clinical Psychologist. If you have any questions about the research, you can call Richard Jenkinson by phone [telephone number] or by email (rjenkinson2@sheffield.ac.uk). Please note, he will only be able to talk to you about the research, not your assessment in general.

Thank you so much for your participation in our research study- we really appreciate it.

Appendix N

PANAS Questionnaire

REMOVED FROM ELECTRONIC VERSION TO PROTECT COPYRIGHT

Appendix O

Predictability Questionnaire

Has the information we sent prior to the assessment helped you to know what to expect from today? (Please circle one option)

Very much A little Somewhat Not really Not at all

Your age……. Your gender……..Today's date……..

Appendix P

Satisfaction Questionnaire

Did you receive a diagnosis of Autism Spectrum Disorder?

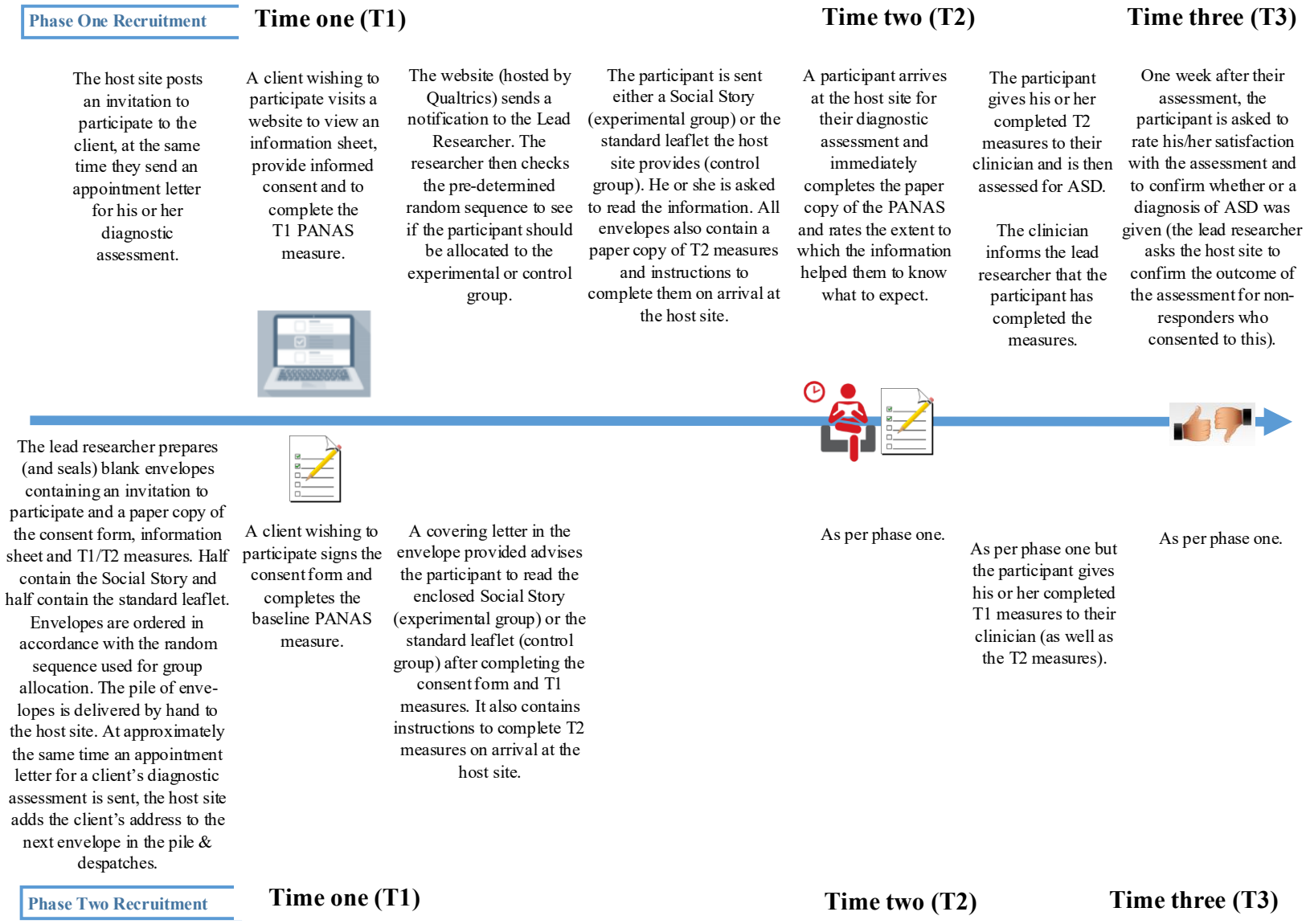
Yes	No

Please rate your overall satisfaction with the assessment:

Very dissatisfied	Quite dissatisfied	Somewhere in-between	Quite satisfied	Very satisfied

Appendix Q

Procedure diagram for both recruitment phases



Appendix R

University Scientific Approval

Department Of Psychology.
Clinical Psychology Unit.

Doctor of Clinical Psychology (DClin Psy) Programme
Clinical supervision training and NHS research training & consultancy.

Clinical Psychology Unit
Department of Psychology
University of Sheffield
Floor F, Cathedral Court
1 Vicar Lane
Sheffield
S1 2LT

Dr A R Thompson, Clinical Training Research Director
Please address any correspondence to Amrit Sinha
Research Support Officer
Telephone: 0114 2226650
Email: a.sinha@sheffield.ac.uk

22nd January 2018
To: Research Governance Office

Dear Sir/Madam,

RE: Confirmation of Scientific Approval and indemnity of enclosed Research Project

Project title: The Use of Social Stories to Reduce Negative Affect and Improve Satisfaction in Adults Attending an Assessment for Autism Spectrum Disorder

Investigators: Richard Jenkinson (DClin Psy Trainee, University of Sheffield); Dr Elizabeth Milne & Dr Andrew Thompson (Academic Supervisors, University of Sheffield).

I write to confirm that the enclosed proposal forms part of the educational requirements for the Doctoral Clinical Psychology Qualification (DClin Psy) run by the Clinical Psychology Unit, University of Sheffield.

Three independent scientific reviewers usually drawn from academic staff within the Psychology Department have reviewed the proposal. Review includes appraisal of the proposed statistical analysis conducted by a statistical expert based in the School of Health and Related Research (ScHARR). Where appropriate an expert in qualitative methods is also appointed to review proposals.

I can confirm that approval of a proposal is dependent upon all necessary amendments having been made to the satisfaction of the reviewers and I can confirm that in this case the reviewers are content that the above study is of sound scientific quality. Consequently, the University will if necessary indemnify the study and act as sponsor.

Given the above, I would remind you that the Department already has an agreement with your office to exempt this proposal from further scientific review. However, if you require any further information, please do not hesitate to contact me.

Yours sincerely

Dr. Andrew Thompson

Director of Research Training

Appendix S

HRA Approval

NHS
Health Research Authority

Mr Richard, P Jenkinson
Trainee Clinical Psychologist
Sheffield Health and Social Care NHS Foundation Trust
Clinical Psychology Unit, University of Sheffield
Cathedral Court, Floor F
1 Vicar Lane, Sheffield.
S1 1HD

Email: hra.approval@nhs.net

01 May 2018

Dear Mr Jenkinson

Letter of HRA Approval

Study title:	**The Use of Social Stories to Reduce Negative Affect and Improve Satisfaction in Adults Attending an Assessment for Autism Spectrum Disorder**
IRAS project ID:	**239758**
Protocol number:	**155578**
REC reference:	**18/YH/0081**
Sponsor	**University of Sheffield**

I am pleased to confirm that **HRA Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further from the HRA.

How should I continue to work with participating NHS organisations in England?
You should now provide a copy of this letter to all participating NHS organisations in England, as well as any documentation that has been updated as a result of the assessment.

Following the arranging of capacity and capability, participating NHS organisations should **formally confirm** their capacity and capability to undertake the study. How this will be confirmed is detailed in the "*summary of HRA assessment*" section towards the end of this letter.

You should provide, if you have not already done so, detailed instructions to each organisation as to how you will notify them that research activities may commence at site following their confirmation of capacity and capability (e.g. provision by you of a 'green light' email, formal notification following a site initiation visit, activities may commence immediately following confirmation by participating organisation, etc.).

It is important that you involve both the research management function (e.g. R&D office) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details of the research management function for each organisation can be accessed here.

IRAS project ID	239758

How should I work with participating NHS/HSC organisations in Northern Ireland, Scotland and Wales?

HRA Approval does not apply to NHS/HSC organisations within the devolved administrations of Northern Ireland, Scotland and Wales.

If you indicated in your IRAS form that you do have participating organisations in one or more devolved administration, the HRA has sent the final document set and the study wide governance report (including this letter) to the coordinating centre of each participating nation. You should work with the relevant national coordinating functions to ensure any nation specific checks are complete, and with each site so that they are able to give management permission for the study to begin.

Please see IRAS Help for information on working with Northern Ireland, Scotland and Wales.

How should I work with participating non-NHS organisations?
HRA Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?

The document "*After Ethical Review – guidance for sponsors and investigators*", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

I am a participating NHS organisation in England. What should I do once I receive this letter?
You should work with the applicant and sponsor to complete any outstanding arrangements so you are able to confirm capacity and capability in line with the information provided in this letter.

The sponsor contact for this application is as follows:

Name: Dr Andrew Thompson
Tel: 0114 2226637
Email: a.r.thompson@sheffield.ac.uk

Who should I contact for further information?
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **239758**. Please quote this on all correspondence.

IRAS project ID	239758

Yours sincerely

Juliana Araujo
Assessor
Email: hra.approval@nhs.net

Copy to: *Sponsor Representative: Dr Andrew Thompson, University of Sheffield*
Lead NHS R&D Office Representative: Ms Michelle Horspool, Sheffield Health & Social Care NHS Foundation Trust

IRAS project ID	239758

List of Documents

The final document set assessed and approved by HRA Approval is listed below.

Document	*Version*	*Date*
Covering letter on headed paper [Cover letter for participants in the experimental group]	1	04 January 2018
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Certificate of insurance]	1	08 January 2018
GP/consultant information sheets or letters [Letter to inform clinic of patient participation]	2	04 January 2018
HRA Schedule of Events [HRA Schedule of Events Validated]		02 February 2018
HRA Statement of Activities [HRA Statement of activities Validated]	2	16 February 2018
Initial Assessment for REC [Initial Assessment for REC]		21 February 2018
IRAS Application Form [IRAS_Form_19022018]		19 February 2018
Letter from funder [Costing Confirmation Email from University of Sheffield]	1	22 January 2018
Letter from sponsor [Email confirming sponsor]	1	12 January 2018
Letter from sponsor [Approval letter stating the "University will if necessary indemnify the study and act as sponsor."]	1	19 January 2018
Letter from statistician [Scientific Approval Letter Confirming Statistical Review]		22 January 2018
Letters of invitation to participant [Invitation to participate]	5	27 April 2018
Non-validated questionnaire [Predictability + Demographic Questionnaire (Added to Validated Paper Version of Positive and Negative Affect Schedule)]	1	19 January 2018
Non-validated questionnaire [Satisfaction Questionnaire - Digital]	1	19 January 2018
Other [University Contract - specifying responsibilities of all parties involved]	1	23 July 2017
Other [Social Story for participants in the experimental group]	1	10 December 2017
Other [Written Debrief Sheet]	3	29 April 2018
Other [Letter to explain changes made since submission]	1	29 April 2018
Participant consent form [Consent Form]	4	29 April 2018
Participant information sheet (PIS) [Participant Information Sheet]	5	29 April 2018
Referee's report or other scientific critique report [Scientific Approval Letter]		22 January 2018
Research protocol or project proposal [Research Protocol]	4	31 December 2017
Response to Additional Conditions Met		
Summary CV for Chief Investigator (CI) [CV]	1	08 December 2017
Summary CV for student [CV]	1	08 December 2017
Summary CV for supervisor (student research) [CV Elizabeth Milne]	1	07 December 2017
Summary of any applicable exclusions to sponsor insurance	1	05 September

IRAS project ID	239758

(non-NHS sponsors only) [Public liability certificate]		2018
Summary, synopsis or diagram (flowchart) of protocol in non technical language [Plain English Summary of Protocol]	1	19 January 2018
Validated questionnaire [Positive and Negative Affect Schedule-Digital Version]	1	19 January 2018
18-YH-0081 239758 Valid Application 19.02.2018.pdf		19 February 2018
18-YH-0081 239758 Favourable Opinion (With Additional Conditions) 20.03.2018.pdf		20 March 2018
18-YH-0081 - 239758 - Conds Met Acknow - 01-05-18.pdf		01 May 2018

IRAS project ID	239758

Summary of HRA assessment

The following information provides assurance to you, the sponsor and the NHS in England that the study, as assessed for HRA Approval, is compliant with relevant standards. It also provides information and clarification, where appropriate, to participating NHS organisations in England to assist in assessing, arranging and confirming capacity and capability.

HRA assessment criteria

Section	HRA Assessment Criteria	Compliant with Standards	Comments
1.1	IRAS application completed correctly	Yes	No comments
2.1	Participant information/consent documents and consent process	Yes	No comments
3.1	Protocol assessment	Yes	No comments
4.1	Allocation of responsibilities and rights are agreed and documented	Yes	The Statement of Activities will form the agreement between the sponsor and the participating NHS organisation. The Schedule of Events was submitted.
4.2	Insurance/indemnity arrangements assessed	Yes	No comments
4.3	Financial arrangements assessed	Yes	No funding application was made for this study.
5.1	Compliance with the Data Protection Act and data security issues assessed	Yes	No comments
5.2	CTIMPS – Arrangements for compliance with the Clinical Trials Regulations assessed	Not Applicable	No comments
5.3	Compliance with any applicable laws or regulations	Yes	No comments
6.1	NHS Research Ethics	Yes	NHS Research Ethics Committee

IRAS project ID	239758

Section	HRA Assessment Criteria	Compliant with Standards	Comments
	Committee favourable opinion received for applicable studies		favourable opinion was confirmed by the by the Yorkshire & The Humber - Sheffield Research Ethics Committee on 01 May 2018.
6.2	CTIMPS – Clinical Trials Authorisation (CTA) letter received	Not Applicable	No comments
6.3	Devices – MHRA notice of no objection received	Not Applicable	No comments
6.4	Other regulatory approvals and authorisations received	Not Applicable	No comments

Participating NHS Organisations in England

This provides detail on the types of participating NHS organisations in the study and a statement as to whether the activities at all organisations are the same or different.
This is a single site study; there is there one site type. The Chief Investigator or sponsor should share relevant study documents with participating NHS organisations in England in order to put arrangements in place to deliver the study. The documents should be sent to both the local study team, where applicable, and the office providing the research management function at the participating organisation. For NIHR CRN Portfolio studies, the Local LCRN contact should also be copied into this correspondence. For further guidance on working with participating NHS organisations please see the HRA website. If chief investigators, sponsors or principal investigators are asked to complete site level forms for participating NHS organisations in England which are not provided in IRAS or on the HRA website, the chief investigator, sponsor or principal investigator should notify the HRA immediately at hra.approval@nhs.net. The HRA will work with these organisations to achieve a consistent approach to information provision.

Principal Investigator Suitability

This confirms whether the sponsor position on whether a PI, LC or neither should be in place is correct for each type of participating NHS organisation in England and the minimum expectations for education, training and experience that PIs should meet (where applicable).
A Principal Investigator should be in place at participating NHS organisations in England. The Chief Investigator will take on this role at the sole participating site. GCP training is not a generic training expectation, in line with the HRA/MHRA statement on training expectations.

IRAS project ID	239758

HR Good Practice Resource Pack Expectations

This confirms the HR Good Practice Resource Pack expectations for the study and the pre-engagement checks that should and should not be undertaken
The research has contractual arrangements in place with the participating NHS organization.

Other Information to Aid Study Set-up

This details any other information that may be helpful to sponsors and participating NHS organisations in England to aid study set-up.
The applicant has indicated that they do not intend to apply for inclusion on the NIHR CRN Portfolio.

Appendix T

HRA Confirmation of conditions met

NHS
Health Research Authority
Yorkshire & The Humber - Sheffield Research Ethics Committee

NHSBT Newcastle Blood Donor Centre
Holland Drive
Newcastle upon Tyne
NE2 4NQ

Tel: 0207 104 8082

01 May 2018

Mr Richard, P Jenkinson
Trainee Clinical Psychologist
Sheffield Health and Social Care NHS Foundation Trust
Clinical Psychology Unit, University of Sheffield
Cathedral Court, Floor F
1 Vicar Lane, Sheffield.
S1 1HD

Dear Mr Jenkinson

Study title:	**The Use of Social Stories to Reduce Negative Affect and Improve Satisfaction in Adults Attending an Assessment for Autism Spectrum Disorder**
REC reference:	**18/YH/0081**
Protocol number:	**155578**
IRAS project ID:	**239758**

The letter sent to you on 20 March 2018 specified that the favourable ethical opinion from the REC was subject to additional conditions being met prior to the start of the study.

The Committee has now received confirmation that these additional conditions have been met.

18/YH/0081	**Please quote this number on all correspondence**

Yours sincerely

Miss Kerry Dunbar
REC Manager

E-mail: nrescommittee.yorkandhumber-sheffield@nhs.net

Appendix U

REC Confirmation of Substantial Amendment 1

NHS
Health Research Authority
Yorkshire & The Humber - Sheffield Research Ethics Committee
NHS Blood and Transplant Blood Donor Centre
Holland Drive
Newcastle upon Tyne
Tyne and Wear
NE2 4NQ

Tel: 0207 104 8079

12 September 2018

Mr Richard P Jenkinson
Clinical Psychology Unit
The University of Sheffield
Cathedral Court, Floor F
S1 1HD

Dear Mr Jenkinson

Study title:	**The Use of Social Stories to Reduce Negative Affect and Improve Satisfaction in Adults Attending an Assessment for Autism Spectrum Disorder**
REC reference:	**18/YH/0081**
Protocol number:	**155578**
Amendment number:	**Substantial Amendment 1**
Amendment date:	**20 August 2018**
IRAS project ID:	**239758**

The above amendment was reviewed by the Sub-Committee in correspondence.

Summary of Amendment

Submission of this amendment was to give participants the option to complete baseline measures (consent, PANAS questionnaire) on paper rather than completing them online. It was proposed that administration staff speaking to clients on the telephone who had a forthcoming appointment would routinely ask if the client received the invitation to participate and whether they would like to find out more about the study by visiting the study's website.

The protocol, consent form and cover letters were amended to reflect the changes.

Ethical opinion

The members of the Committee taking part in the review gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

Yorkshire & The Humber - Sheffield Research Ethics Committee

Attendance at Sub-Committee of the REC meeting via correspondence

Committee Members:

Name	*Profession*	*Present*
Mrs Rhona Bratt	Co-opted Member	Yes
Ms Liz Williams (Chair)	Senior Lecturer in Human Nutrition	Yes

Also in attendance:

Name	*Position (or reason for attending)*
Miss Donna Bennett	REC Assistant

Approved documents

The documents reviewed and approved at the meeting were:

Document	*Version*	*Date*
Letters of invitation to participant [Invitation]	6	20 August 2018
Notice of Substantial Amendment (non-CTIMP)	Substantial Amendment 1	20 August 2018
Other [Booklet Cover]	1	20 August 2018
Other [Covering Letter]	2	20 August 2018
Other [Derby Social Story]		
Participant consent form [Consent Form]	5	20 August 2018
Research protocol or project proposal [Protocol]	5	30 July 2018

Membership of the Committee

The members of the Committee who took part in the review are listed on the attached sheet.

Working with NHS Care Organisations

Sponsors should ensure that they notify the R&D office for the relevant NHS care organisation of this amendment in line with the terms detailed in the categorisation email issued by the lead nation for the study.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

We are pleased to welcome researchers and R & D staff at our Research Ethics Committee members' training days – see details at http://www.hra.nhs.uk/hra-training/

18/YH/0081:	**Please quote this number on all correspondence**

Yours sincerely
Pp

Ms Liz Williams
Chair

E-mail: nrescommittee.yorkandhumber-sheffield@nhs.net

Enclosures: *List of names and professions of members who took part in the review*

Copy to: *Ms Michelle Horspool, Sheffield Health & Social Care NHS Foundation Trust*

Appendix V

HRA Confirmation of Substantial Amendment 1

Dear Mr Jenkinson

IRAS project ID:	239758
REC reference:	18/YH/0081
Short Study title:	Can Social Stories Improve the Experience of an Autism Assessment? V1
Date complete amendment submission received:	21 August 2018
Amendment No./ Sponsor Ref:	Substantial Amendment 1
Amendment Date:	20 August 2018
Amendment Type:	**Substantial**
Outcome of HRA Assessment	**This email also constitutes HRA and HCRW Approval for the amendment, and you should not expect anything further.**

I am pleased to confirm that this amendment has been reviewed by the Research Ethics Committee and has received a Favourable Opinion. Please find attached a copy of the Favourable Opinion letter.

HRA and HCRW Approval Status

As detailed above, **this email also constitutes HRA and HCRW Approval for the amendment**. No separate notice of HRA and HCRW Approval will be issued. You should implement this amendment at NHS organisations in England and/or Wales, in line with the conditions outlined in your categorisation email.

- If this study has HRA and HCRW Approval, this amendment may be implemented at participating NHS organisations in England and/or Wales once the conditions detailed in the categorisation section above have been met
- If this study is a pre-HRA Approval study, this amendment may be implemented at participating NHS organisations in England and/or Wales that have NHS Permission, once the conditions detailed in the categorisation section above have been met. For participating NHS organisations in England and/or Wales that do not have NHS Permission, these sites should be covered by HRA and HCRW Approval before the amendment is implemented at them, please see below;
- If this study is awaiting HRA and HCRW Approval, I have passed your amendment to my colleague and you should receive separate notification that the study has received HRA and HCRW Approval, incorporating approval for this amendment.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/.

If you require further information, please contact hra.amendments@nhs.net

18/YH/0081/AM02 Please quote this number on all correspondence

Kind regards

Donna Bennett

REC Assistant

Health Research Authority

NHS Blood and Transplant Blood Donor Centre | Holland Drive | HRA Newcastle | NE2 4NQ

T. 0207 104 8079

E. nrescommittee.yorkandhumber-sheffield@nhs.net

W. www.hra.nhs.uk

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Appendix W

REC Confirmation of Substantial Amendment 2

NHS
Health Research Authority

Yorkshire & The Humber - Sheffield Research Ethics Committee

NHS Blood and Transplant Blood Donor Centre
Holland Drive
Newcastle upon Tyne
Tyne and Wear
NE2 4NQ

Tel: 02071048026

Please note: This is the favourable opinion of the REC only and does not allow the amendment to be implemented at NHS sites in England until the outcome of the HRA assessment has been confirmed.

18 December 2018

Mr Richard P Jenkinson
Clinical Psychology Unit, The University of Sheffield
Cathedral Court, Floor F
S1 1HD

Dear Mr Jenkinson

Study title:	**The Use of Social Stories to Reduce Negative Affect and Improve Satisfaction in Adults Attending an Assessment for Autism Spectrum Disorder**
REC reference:	**18/YH/0081**
Protocol number:	**155578**
Amendment number:	**Substantial amendment 2, 05-12-18**
Amendment date:	**05 December 2018**
IRAS project ID:	**239758**

The above amendment was reviewed on 12 December 2018 by the Sub-Committee in correspondence.

Ethical opinion

The members of the Committee taking part in the review gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

Approved documents

The documents reviewed and approved at the meeting were:

Document	*Version*	*Date*
Covering letter on headed paper	3.0, Tracked	28 November 2018
Notice of Substantial Amendment (non-CTIMP)	Substantial amendment 2, 05-12-18	05 December 2018
Participant consent form	6.0, Tracked	28 November 2018
Participant information sheet (PIS)	6.0, Tracked	28 November 2018
Research protocol or project proposal	6.0, Tracked	28 November 2018

Membership of the Committee

The members of the Committee who took part in the review are listed on the attached sheet.

Working with NHS Care Organisations

Sponsors should ensure that they notify the R&D office for the relevant NHS care organisation of this amendment in line with the terms detailed in the categorisation email issued by the lead nation for the study.

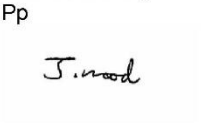
Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

We are pleased to welcome researchers and R & D staff at our Research Ethics Committee members' training days – see details at http://www.hra.nhs.uk/hra-training/

18/YH/0081:	**Please quote this number on all correspondence**

Yours sincerely
Pp



Mr Jacob Wood
REC Assistant
Dr Amaka Offiah
Chair

E-mail: nrescommittee.yorkandhumber-sheffield@nhs.net

Enclosures: *List of names and professions of members who took part in the review*

Copy to: *Michelle Horspool, Sheffield Health & Social Care NHS Foundation Trust*

Yorkshire & The Humber - Sheffield Research Ethics Committee

Attendance at Sub-Committee of the REC meeting on 12 December 2018

Committee Members:

Name	*Profession*	*Present*	*Notes*
Mrs Jacqui Gath	Retired Senior Systems Analyst	Yes	
Dr Amaka Offiah [Chair]	Reader in Paediatric Musculoskeletal Imaging	Yes	

Also in attendance:

Name	*Position (or reason for attending)*
Mr Jacob Wood	REC assistant

Appendix X

HRA Confirmation of Substantial Amendment 2

Dear Mr Jenkinson,

IRAS Project ID:	239758
Short Study Title:	Can Social Stories Improve the Experience of an Autism Assessment? V1
Amendment No./Sponsor Ref:	Substantial amendment 2, 05-12-18
Amendment Date:	05 December 2018
Amendment Type:	Substantial Non-CTIMP

I am pleased to confirm **HRA and HCRW Approval** for the above referenced amendment.

You should implement this amendment at NHS organisations in England and Wales, in line with the conditions outlined in your categorisation email.

The assessment of this amendment noted that the Participant Information sheet for Discussions on data collections needs to be updated to comply with General Data Protection Regulation (GDPR) which applied from 25 May 2018. As such, HRA Approval has been issued subject to regulatory approval and on the basis that the Participant Information Sheet (PIS) is now updated to include the recommended transparency wording which you should use to ensure that your PIS is compliant with the GDPR. Updating the PIS to include the recommended transparency wording is a non-substantial, non-notifiable amendment that can be implemented without needing to submit for approvals

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the

feedback form available on the HRA website: http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/.

Please contact hra.amendments@nhs.net for any queries relating to the assessment of this amendment.

Kind regards

Isobel Lyle

Health Research Authority
Tel 0207 0722496

Ground Floor | Skipton House | 80 London Road | London | SE1 6LH

E.hra.amendments@nhs.net

W. www.hra.nhs.uk

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Non Substantial Amendment 1 - Addition of Site

Dear Mr Jenkinson

IRAS Project ID:	239758
Short Study Title:	Can Social Stories Improve the Experience of an Autism Assessment? V1
Date complete amendment submission received:	15/06/2018
Sponsor Amendment Reference Number:	Non Substantial Amendment 1 - Addition of Site
Sponsor Amendment Date:	15 June 2018
Amendment Type:	Non-substantial
For new sites in Northern Ireland and/or Scotland:	Please start to set up your new sites. Sites may not open until NHS management permission is in place.
For new sites in England and/or Wales:	For studies which already have HRA and HCRW Approval: **This email also constitutes HRA and HCRW Approval for the amendment, and you should not expect anything further**. Please start to set up your new sites. Sites may not open until the site has confirmed capacity and capability (where applicable). For studies which do not yet have HRA and HCRW Approval: **HRA and HCRW Approval for the initial application is pending**. You can start the process of setting up the new site but cannot open the study at the site until HRA and HCRW Approval is in place and the site has confirmed capacity and capability (where applicable). For studies with HRA Approval adding Welsh NHS organisations for the first time. Please take this email to confirm your **original HRA Approval letter is now extended to cover NHS organisations in Wales. You now have HRA and HCRW Approval.** Please start to set up your new sites. Sites may not open until the site has confirmed capacity and capability (where applicable).

Thank you for submitting an amendment to add one or more new sites to your project. This amendment relates solely to the addition of **new sites**.

What should I do next?

Please set up the new site(s) as per the guidance found within IRAS. **Please note** that processes change from time to time so please use the most up to date guidance about site set up.

If your study is supported by a research network, please contact the network as early as possible to help support set up of the new site(s).

If you have listed new sites in any other UK nations **we will** forward the information to the national coordinating function(s) for nations where the new site(s) are being added. In Northern Ireland and Scotland, NHS/HSC R&D offices will be informed by the national coordinating function.

Note: you may only implement changes described in the amendment notice.

Who should I contact if I have further questions about this amendment?

If you have any questions about this amendment please contact the relevant national coordinating centre for advice:

- England – hra.amendments@nhs.net
- Northern Ireland – research.gateway@hscni.net
- Scotland – nhsg.NRSPCC@nhs.net
- Wales – research-permissions@wales.nhs.uk

Additional information on the management of amendments can be found in the IRAS guidance.

User Feedback

We are continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the amendment procedure. If you wish to make your views known please use the feedback form available at: http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/.

Please do not hesitate to contact me if you require further information.

Kind regards

Miss Jade Robinson

Amendment Coordinator

Health Research Authority

Data on Participants Excluded from Data Analysis due to not Receiving ASD Diagnosis

Participants Without ASD

Table 9

Mean PANAS negative affect scores for participants without ASD, by group and time

(Time 1				Time 2			
Experimental		Control		Experimental		Control	
n	*M (SD)*	*n*	*M (SD)*	*n*	*M (SD)*	*n*	*M (SD)*
4	25.50 (12.37)	2	24.50 (4.95)	4	26.75 (11.76)	1	14.00 (N/A)

Note. n, number*; M,* mean*; SD,* standard deviation; N/A, not applicable

Table 10

Median scores on secondary outcomes for participants without ASD, by group and time

	Experimental		Control	
	n	*Median (IQR)*	*n*	*Median (IQR)*
Predictability	4	4.5 (3.75 – 5)	1	5 (N/A)
Satisfaction	0	N/A	1	3 (N/A)

Note. n, number*; IQR,* interquartile range; N/A, not applicable