# Adapting and Validating the Autism Diagnostic Interview – Revised for Use with Deaf Children and Young People

Barry Wright1,2, Helen Phillips2, Victoria Allgar1,Jennifer Sweetman2, Rachel Hodkinson2, Emily Hayward2, Amelia Ralph-Lewis2, Catarina Teige2 Martin Bland4 and Ann Le Couteur3

1Hull York Medical School, University of York, York, UK

2Leeds and York Partnership NHS Foundation Trust, Leeds, UK

3Newcastle University, Newcastle, UK

4University of York, York, UK

# Author Note

Barry Wright  <https://orcid.org/0000-0002-8692-6001>

Helen Phillips  <https://orcid.org/0000-0001-9376-9502>

Victoria Allgar  <https://orcid.org/0000-0002-5228-2623>

Jennifer Sweeman  <https://orcid.org/0000-0003-196-4586>

Rachel Hodkinson  <https://orcid.org/0000-0002-3972-2890>

Emily Hayward  <https://orcid.org/0000-0002-2413-0104>

Amelia Ralph-Lewis  <https://orcid.org/0000-0003-4474-6288>

Catarina Teige  <https://orcid.org/0000-0002-8958-6487>

Ann Le Couteur  <https://orcid.org/0000-0001-9991-3608>

Martin Bland https://orcid.org/0000-0002-9525-5334.

Correspondence concerning this article should be addressed to Professor Barry Wright; Child Oriented Mental health Intervention Centre (COMC), IT Centre building, Innovation way, Heslington, York, YO10 5NP. Email: barry.wright1@nhs.net

# Abstract

A Delphi consensus methodology was used to adapt the Autism Diagnostic Interview – Revised (ADI-R) for the assessment of deaf children with suspected Autism Spectrum Disorder (ASD). Each ADI-R item was considered by a panel of nine international experts in terms of relevance and acceptability. Modifications were proposed and agreed by the expert panel for 45% of items. The pre-specified criterion for agreement between experts was set at 80% for each item. A first validation of the revised version, adapted for deaf children (ADI-R Deaf Adaptation) , was undertaken with a United Kingdom (UK) sample of 78 parents/carers of deaf children with ASD and 126 parents/carers with deaf children without ASD. When compared to NICE guideline standard clinical assessments, the ADI-R Deaf Adaptation diagnostic algorithm cut-off/threshold scores achieved a sensitivity of 89% (79% – 96%) and specificity of 81% (70%-89%) for ASD. The alpha coefficients for each algorithm symptom domain ranged from 0.80 to 0.91, suggesting that the items had high internal consistency. Our findings indicate that the ADI-R Deaf Adaptation is likely to be a useful measure for the assessment of deaf children with suspected ASD, although further research is needed.

## Lay Abstract

Autism assessment processes need to improve for deaf children as they are currently being diagnosed later than their hearing counterparts and misdiagnosis can occur. We took one of the most commonly used parent developmental interviews for ASD the Autism Diagnostic Interview – Revised (ADI-R) and adapted it using international expert advice. Modifications were proposed and agreed by the expert panel for 45% of items; the remaining 55% of items were unchanged. We then tested the revised version, adapted for deaf children (ADI-R Deaf Adaptation) , in a United Kingdom sample of 78 parents/carers of deaf children with ASD and 126 parents/carers with deaf children without ASD. When compared to NICE guideline standard clinical assessments, the ADI-R Deaf Adaptation diagnostic algorithm threshold scores could identify those deaf children with a definite diagnosis (true ASD positives) well (sensitivity of 89% (79% – 96%)) and those deaf children who did not have ASD ( true ASD negatives) well (specificity of 81% (70%-89%)). Our findings indicate that the ADI-R Deaf Adaptation is likely to prove a useful measure for the assessment of deaf children with suspected ASD and that further research would be helpful.

## Keywords

Autism Spectrum Disorder, ADI-R Deaf Adaptation, ADI-R, Deaf, modified, Delphi consensus methodology, children, British Sign Language, gesture, language, deaf culture.

# Introduction

Autism Spectrum Disorder (ASD) is a globally recognised neurodevelopmental condition, with considerable impact on the affected individual, their family and wider society (Buescher et al., 2014; Elsabbagh et al., 2012; Saccá et al., 2019; Sim et al., 2018). The behavioural profile of strengths and difficulties (Baird et al., 2006) is associated with differences in brain neural architecture and connectivity compared with the general population (O’Reilly et al., 2017). These differences may be responsible for social communication difficulties (Bishop et al., 2016) and increased intense or unusual preoccupations and repetitive actions (Bishop et al., 2006; Goldman et al., 2009)- the required criteria for an ASD diagnosis (APA, 2013) . Some deaf children without ASD with limited exposure to language in early life present with social communication differences and empathy skill developmental delay (Schick et al, 2007), which has some similarities to hearing children with ASD (Baron-Cohen, 2008; Hayes & Watson, 2013). Individuals are also predisposed to a range of co-occurring physical and mental health conditions and difficulties (Gurney et al., 2006; Lai et al., 2019) and some children with ASD are also deaf (Beers et al, 2014). It is important for clinicians and researchers to accurately identify patterns of development when diagnosing ASD within a broad range of differential diagnoses to inform individual intervention and family support planning. Children with an ASD (including those with an intellectual disability) benefit from early social communication interventions, appropriately adapted education and additional support (Bond et al., 2016; Kasari et al., 2015; Pickles et al., 2013).

Clinicians assessing deaf children for ASD report difficulties in the diagnostic assessment process: concerns about overlapping symptoms and behaviours with ASD in deaf children as a result of lack of exposure to early language learning experiences (Hall et al, 2017), those with social and emotional developmental delay (Wright & Oakes, 2012) and risk of misinterpretation of behaviours (e.g. visual sensory interests). There are concerns that diagnostic uncertainties such as these delay the assessment pathway (Brenman et al., 2017). Parents of deaf children experience delay in assessment and diagnosis, diagnostic uncertainty, lack of understanding of the deaf experience and problems establishing effective communication with the child from assessing clinicians (Roper et al., 2003; Wright & Oakes, 2012;Young et al., 2019).

There are over 45,000 deaf children living in the UK (CRIDE, 2016); with approximately 40% of these children having additional needs (Fortnum & Davis, 1997). The prevalence of deaf children with ASD is unknown, although many of the neurological, perinatal and syndromic causes of being deaf (Morzaria et al., 2004) also carry a higher risk of ASD (Chess, 1977; Jiang et al., 2016; Wachtel et al., 2007). It has been estimated that approximately 1.6-1.7% of deaf children have a reported ASD (Gallaudet Research Institute, 2008; Szymanski et al, 2012).

Relatively little is known about differences in the way ASD presents in deaf children outside small cohort studies. These show some interesting findings such as the presence of reverse palm orientation for signs that have a specific inward/outward orientation, (Shield & Meier, 2012), the absence of sign pronominal reversal and limited use of pronouns in sign with a preference for names (Shield et al, 2015) and that echolalia presents in sign language (Shield et al, 2017). The importance of facial expressions in emotional understanding has also been demonstrated in deaf people with ASD (Denmark et al, 2014). The research on prevalence of being deaf in ASD is greatly variable between studies (Beers et al, 2014).

The median age of all ASD diagnoses in hearing children in UK is 55 months (Brett et al., 2016) with the mean age in the US varying between studies between 38 and 120 months (Daniels and Mandell, 2014). Whilst there is limited research in both countries for deaf children the age of diagnosis is purported to be comparatively late (Jure et al., 1991; Roper et al., 2003) with one recent study in the US reporting a median diagnosis of ASD of 66.5 months (Meinzen-Derr et al, 2014). These differences may be related to an overlap, confusion or misattribution of symptoms (Szymanski & Brice, 2008; Wright & Oakes, 2012; Schick et al., 2007). Most deaf children are born to hearing parents who are not expecting a deaf child (Vaccari & Marschark, 1997), and often seek information and advice(Szarkowski & Brice, 2016) and report challenging experiences when having to consider their understanding of the development of a child who is deaf or is deaf and has ASD (Young et al., 2019). Another contributory factor to delay may include: a limited number of professionals with knowledge or experience in the assessment of deaf children with/or without suspected ASD, (Syzmanski et al., 2012; Shield et al., 2015). There is a clear benefit to early identification and intervention for ASD in hearing children (Warren et al., 2011) and this is likely to pertain also to deaf children. Finding new and better ways of assessing deaf children could lead to earlier and better identification both of which would benefit those children and their families. No existing ASD diagnostic assessment tools have been validated for the assessment of deaf children (Syzmanski & Brice, 2008). In one of the most widely used parent semi-structured interviews (the Autism Diagnostic Interview-Revised (ADI-R)), (Rutter et al., 2003) a number of the questions are not suitable for the assessment of deaf children. There is an urgent need to adapt and validate diagnostic measures for use in the deaf population. We report the adaptation and first validation of the ADI-R for use with deaf children (hereafter termed the ADI-R Deaf Adaptation).

**Methods**

## Permissions

Positive ethical opinions were obtained from the National Research Ethics Service (NRES) Committee Yorkshire & the Humber - South Yorkshire (REC Reference: 15/YH/0093) for the Delphi Consensus (22/05/2014) and validation study (17/04/2015). The study was undertaken with the agreement of the ADI-R original authors (Rutter et al., 2003) and relevant permissions were obtained from the publishers Western Psychological Services (WPS).

## The Instrument

The ADI-R is a semi-structured interview administered by a trained interviewer to an informant (usually parent(s) or caregiver(s)) (Lord et al., 1994*;* Rutter et al., 2003)*.* The interview framework provides sufficient information to identify the presence of neurodevelopmental disorder diagnoses (World Health Organization International Classification of Diseases (ICD-10)) (WHO, 1993) and for a DSM-5 ASD diagnosis (American Psychiatric Association, 2013). The ADI-R focuses on key domains of functioning: Background (including family, educational and medical history); Current concerns; Early Development; Acquisition and Loss of Language/Other Skills; Language and Communication Functioning; Social Development and Play; Interests and Behaviors; and General Behaviours.

The scores for selected items are combined into an ADI-R algorithm with an ADI-R diagnostic threshold for autism based on scores exceeding the specified cut-offs in the three sections (Reciprocal Social Interaction; Language/Communication; and Repetitive and Stereotyped Patterns of Behaviours), together with evidence of abnormality in development at or before 36 months of age. The ADI-R algorithm threshold is not a clinical diagnosis but can contribute to the diagnostic process.

## Adaptation of ADI-R using the Delphi Consensus Methodology

An online Delphi method was used where internationally recruited experts in deaf children with ASD examine each item of the ADI-R and make comments sharing their opinions to seek a shared consensus (Gibson, 1998; McKenna, 1994; Sharkey & Sharples, 2001). Experts comment independently and their opinions are collated by an expert panel . A number of rounds were undertaken (details described below) (Beech, 1999; Sharkey & Sharples, 2001; Sheikh et al., 2008).

## Community and Patient and Public Involvement (PPI)

During the study a hearing parent of a deaf child with ASD and a deaf parent of a child with ASD supported our research attending meetings, checking materials for use with participants and advising on a arrange of issues. At all stages of the research we have involved people with lived experience of being deaf including deaf participants, deaf parents, a number of deaf clinicians/professionals including those trained within the ten centres of the national deaf child and adolescent mental health service in England and several members of the research team are profoundly deaf.

## Delphi International Expert Panel members (DIEP)

International experts in ASD and deafness were identified through: publications, specialist contacts, conferences and relevant organisations. Identification and selection criteria (see figure 1 for methodology flow).

## Independent Research Review Team (IRRT)

An independent research review team (IRRT) was assembled with representatives including parents of deaf children with autism, educational professionals with experience working with deaf children with autism and deaf and hearing clinical and academic research practitioners in child mental health and linguistics. We prioritised the inclusion of deaf professionals to ensure that the discussions incorporated deaf cultural perspectives. At the end of each Delphi round the IRRT reviewed a summary of the collated DIEP feedback. The IRRT then agreed whether items were accepted or modified and re-circulated to the DIEP for the next round (see figure 1 and description below).

**Figure 1**

*Flowchart Illustrating the ADI-R Delphi International Expert Panel (DIEP) Consensus Process for ADI-R-Deaf Adaption*

Letters to recognised international experts

Potential participants were requested to return their expression of interest and eligibility criteria

Potential participants then completed a demographic questionnaire

Participants eligible if met minimum criteria: experience with deaf children with ASD; trained in ADI-R(or equivalent) and had used to assess deaf children in last 2 years.

 Participants were excluded if they did not meet the eligibility criteria:

Have worked with deaf children with ASD

Have used the ADI-R

ADI-R-Deaf Adaption Delphi International Expert Panel (DIEP)

## Delphi Consensus Procedure

Full details are previously reported (Phillips et al., 2021; Wright et al., 2020). The DIEP members were presented with each ADI-R item and coding definitions and asked to select one of three options: 1) agree the item is acceptable for use with deaf participants; 2) decide item should be discarded; or 3) recommend modification (suggest wording for modification of the individual item in the ADI-R booklet with relevant evidence).

A pre-specified rate of 80% agreement (to bank an item as accepted or rejected) was used in line with previous Delphi Consensus literature (Beattie & Mackway-Jones, 2004; Brown et al., 2006; Crawford et al., 2004).

One of the pre-specified goals for the IRRT was to ensure that the modifications of the ADI-R items, including item and coding definitions, were kept to a minimum. This aimed to ensure as much conceptual integrity of the original assessment measure and coding systems to be retained as possible.

Following completion of the DIEP procedure, the modified ADI-R was referred to as the Autism Diagnostic Interview- Revised Deaf Adaption (ADI-R Deaf Adaption).

## Translation

To enable those parents (including deaf parents) whose communication preference might be a signed language and as this study was undertaken in England, there was a need to make the items in the ADI-R Deaf Adaptation to be available in British Sign Language (BSL). To achieve this and in line with previous research, a robust forward and back translation methodology was employed to translate all written mandatory questions (modified and unmodified) into British Sign Language (Moore et al., 2013). This involved 2 bi-lingual forward translators, 2 bi-lingual back translators and an expert panel including expert linguistics, clinicians, psychiatrists and consultants. The translation iteratively went between groups until 100% accuracy was achieved.

## Validation Study

### Participants and Recruitment

Parents of children and young people aged 2-18 years with a bilateral hearing status of at least 40dBHL (i.e. this is classified as ‘moderate’ to ‘profound’ by the main internationally agreed hearing classification systems) (Wright et al, 2018) included those using a range of audiological devices such as hearing aids and cochlear implants, were invited to participate. There were no study exclusions based on language preference or the presence of child intellectual disability or other health or mental health co-morbidities. Recruitment was undertaken across England. The 10 National Deaf Child and Adolescent Mental Health Services (NDCAMHS) and other CAMHS services across England worked with the study team to identify and circulate details of the study to potentially eligible children and their families. Schools for the deaf, mainstream schools with specialist resource bases for deaf children, special educational needs schools and teachers with case lists for all deaf children (<40dbHL) in England were contacted. Recruitment information stressed that we were recruiting parents of deaf children who did not have an existing clinical diagnosis of ASD; deaf children with a diagnosis of ASD; and children and young people for whom parents were seeking assessment or teachers had concerns about a possible ASD. Members of the National Autistic Society, National Deaf Children’s Society and the national ASD-UK and Daslne (Database of Children Living with Autism Spectrum Disorder in the North East) research databases were also contacted.

### ADI-R Deaf Adaptation Interviews

The ADI-R Deaf Adapted interviews were conducted by experienced NDCAMHS clinicians who had received training in the ADI-R Deaf Adaptation delivered at a series of training events across the country. Twenty parents/carers were deaf and met with a deaf clinician who communicated in the parent/carer’s preferred language (e.g. BSL) and carried out the ADI-R Deaf adaptation. These national deaf child and adolescent mental health trained clinicians attended additional training and had access to BSL videos of the mandatory questions in the ADI-R Deaf adaptation, which they could show to parents/carers. All parents completed both the ADI-R Deaf Adaptation and the NICE guideline standard clinical assessment using their preferred language on separate days. When working with families with both a deaf and hearing parent/carer together we allowed parents to choose their choice of language but also provided a qualified BSL/English interpreter familiar with the guidance video. Throughout the duration of the study the same two interpreters were used to conduct the ADI-R Deaf adapted interviews. Both were very knowledgeable having interpreted preliminary discussions.

### Diagnostic Procedure

Twenty two clinicians, who currently or have recently worked for NDCAMHS were involved in the assessments. Ten clinicians were trained to use the ADI-R Deaf Adaptation research assessments and 17 conducted the National Institute for Health and Care Excellence (NICE) guideline standard clinical assessments (NICE, 2017). Five clinicians were involved in both types of assessment but never with the same child. The assessments were completed at different times, blind to each other and diagnostic status. The order of assessments was scheduled for the convenience of the family and clinicians. The NICE guideline standard clinical assessment was based on WHO Research Diagnostic Criteria for autism, atypical autism or Asperger Syndrome (referred to hereafter as ASD) (WHO, 1993) (Wright et al., 2012). Clinicians met with parents and children to gather a comprehensive child history, observed and interacted with children and young people at home or school, and viewed additional professional reports where available (e.g. teacher, speech and language therapy and educational psychology). The clinicians also accessed the Social Communication Questionnaire (SCQ) results (Chandler et al., 2007) filled in by all parent participants using the written form and used the responses as a discussion prompt. All this information was collated using a reporting matrix (WHO, 1993).

The NICE guideline standard clinical assessment(described above) was used to confirm a clinical ASD diagnosis. In the event that the NICE guideline clinical assessment did not take place, children were classified using the parent report of an ASD diagnosis from a previous professional NHS clinical assessment or a score above the threshold (≥ 15) of the Social Communication Questionnaire (SCQ) (Chandler et al., 2007; Rutter et al., 2003). This created a ‘diagnostic group’ categorisation that was used as the basis for validation.

### Sample Size Calculation for the Validation Study

A sample size of 65 per group was based on estimating the mean difference in the ADI-R Deaf adaptation algorithm domain scores of the ADI-R between deaf children with ASD and deaf children without ASD to within ±0.34 standard deviations (95% confidence interval on each side of the estimate).

### Analysis

Descriptive statistics are presented as mean (sd) or number (percentage). To compare deaf children with ASD and deaf children without ASD, a t- test or a chi-square test was used for mean and number values respectively. Diagnostic accuracy refers to the degree of agreement between the results from the ADI-R Deaf Adaptation and those from a reference test (Bossuyt, 2015). To validate the ADI-R Deaf Adaptation, we compared the classification from the ADI-R Deaf Adaptation diagnostic algorithm (referred to in this study as ‘ADI-R Deaf Adaptation positive’) against the established diagnostic groups. We also conducted a sensitivity analysis based on the NICE guideline standard clinical assessment only. To determine the optimal ADI-R Deaf Adaptation algorithm cut-off value, the value with highest Youden Index, which determines the highest Sensitivity + Specificity scores, was used. To explore the reliability of the ADI-R Deaf Adaptation scoring algorithms in deaf children, Cronbach’s alpha was used (Tavakol & Dennick, 2011) to explore internal consistency. A “high” value for alpha does not imply that the measure is unidimensional (Tavakol & Dennick, 2011), so an exploratory factor analysis was undertaken as a method of checking dimensionality. For the factors loadings, a cut-off of 0.5 assures that items were clearly related to the factor. Analysis was undertaken on STATA/SE 14.2 (StataCorp). 2015. Stata Statistical Software: Release 14. College Station, TX: StataCorp LP).

# Results

## Delphi Consensus

Forty five international experts responded to the invitation letter sent to 150 people identified as potentially suitable. Forty professionals completed the demographic questionnaire; of these 17 had experience using the ADI-R or equivalent. Nine experts were available to complete the Delphi Consensus process; one expert reported their hearing status as deaf.

The professional groups involved are shown in Table 1.

**Table 1**

*Demographic Characteristics for Delphi Consensus Experts*

|  |  |  |
| --- | --- | --- |
|  | **Completed Demographic Questionnaire** | **Reviewed****ADI-R** |
|  | **Participants****(%)** | **Participants****(%)** |
| **Total** | 40 | 9 |
| **Background** |  |  |
| Parent/Carer | 1 (2.5) | - |
| Professional | 39 (97.5) | 9 (100) |
| **Gender** |  |  |
| Female | 34 (85) | 7 (78 ) |
| Male | 6 (15) | 2 (22 ) |
| **Hearing status** |  |  |
| Deaf | 4 (10) | 1 (11 ) |
| Hearing | 36 (90) | 8 ( 89 ) |
| **Preferred Language ᵅ** |  |  |
| English | 38 | 9 (100) |
| Australian Sign Language (AUSLAN) | 3 | 1 (11 ) |
| American Sign Language (ASL) | 2 | 1 (11 ) |
| British Sign Language (BSL) | 2 | - |
| Dutch | 1 | - |
| **Country** |  |  |
| England | 15 | 4 ( 44 ) |
| Australia | 14 | 3 (33 ) |
| United States  | 8 | 2 ( 22 ) |
| Netherlands | 2 | - |
| Russia | 1 | - |
| **Occupation** |  |  |
| Psychologist  | 11 (27.5) | 2 (22 ) |
| Psychiatrist  | 8 (20) | 5 ( 56 ) |
| Speech and Language therapist or pathologist  | 5 (12.5) | 1 (11 ) |
| Teacher of the Deaf  | 4 (10) | - |
| Sign Language Model (teacher Aide) | 3 (7.5) | 1 (11 ) |
| Professor or academic researcher | 2 (5) | - |
| Developmental Paediatrician  | 2 (5) | - |
| Educational Advisor  | 2 (5) | - |
| Deaf Service consultant  | 1 (2.5) | - |
| Program Manager  | 1 (2.5) | - |
| Trainee clinical psychologist  | 1 (2.5) | - |

*ᵅ 2 members selected 2 languages as their preferred language.*

One hundred and seventy four items were reviewed in round one, 78 items in round two and 10 items in round three. Seventy seven ADI-R items were recommended for modification. No items were deleted. DIEP consensus was achieved for all items (mandatory questions and coding definitions) after three rounds.

## DIEP Recommended Modifications

No changes were made to the overall structure of the key domains of functioning in the ADI-R (Rutter et al., 2003). Modifications were made in several areas (detailed in supplementary materials table A-C and Figures A-F for details):

### 1) The General Layout

#### **Background: Family History; and the Deaf Child’s Social, Education and Medical History**.

Prompts were added to capture information about the deaf child’s current and past health, including the cause (if known) and time course of being deaf; other physical and mental health problems; and any technical aids and/or resources the child, family or school were accessing. Further detail about the child’s language and communication history, their education setting(s) and any other support received were also added.

#### Acquisition and Loss of Language/Level of Communication

The questions to parents about the development of their child’s language (Items 9 and 10) were expanded to capture the development of spoken (new Item 9a and 10a) *and* signed languages (Items 9b and 10b) (see table A and B supplementary materials section).

#### Additional Item and Adaptation of Items

Item 37 Pronominal reversal was modified (Item 37a) based on research in deaf children (Shield et al., 2012) and a new item to consider sign reversal (Item 37b) was created with suggested prompts and coding. This item will enable clinicians and researchers to capture new information (Shield et al., 2015) (see figure A in supplementary material section).

Item 39 Verbal/Signing Rituals was modified to include non- linguistic verbal utterances (see figure F in supplementary material section). The question has been expanded to enable the assessor to gain information about verbal or signing deaf children/young people.

The DIEP agreed to retain the Undue general sensitivity to noise but recommended an additional item (72b) Undue general sensitivity to sensations (that the child might see, hear or feel such as perfume, certain food textures, movements or appliances). The modified wording was agreed following Round 2 (see figures B + C in supplementary material section).

The title for item 40 has been modified to the term Prosody (previously termed Intonation/volume/rhythm/rate). The revised wording is to include the alternative expression of prosody in signed languages alongside verbal prosody (for example facial expression, hand shapes and signing space usage) (Corina et al., 1999; Dachkovsky & Sandler, 2009).

### 2. Language and Communication Functioning

The inclusion of use of signs and visual language as well as words and spoken language throughout the ADI-R Deaf Adaptation is integrated into the coding. Some additional prompts also gather information about the child’s social communication interactions with deaf and hearing individuals separately.

The DIEP highlighted the importance of Deaf culture as well as language (e.g. how to gain attention from a deaf person). These modifications mainly apply to item definitions and questions rather than wording changes to coding. The definitions for gestures and signs have been expanded for Item 45 Conventional /instrumental gestures to clarify the difference between some gestures used by hearing individuals and the use of some iconic linguistic signs in children using a signed language (Wright et al., 2020). (See figure E in supplementary materials section).

***3. The algorithm***

The items included in the algorithm remain unchanged and the items (some reworded) included within it are detailed in supplementary table C.

## Validation Results

Two hundred and four parents with a deaf child were recruited from across England. 139 completed the ADI-R Deaf Adaptation, with one child having an incomplete ADI-R Deaf Adaptation so final diagnosis was not possible using the threshold for autism based on scores exceeding the specified cut-offs in all three sections, together with evidence of abnormality in development at or before 36 months of age (see Figure 2). Blinded NICE guideline standard clinical assessment was undertaken to determine whether deaf children/young people had ASD or not. This was missing for 4 children/young people and so the SCQ higher validated threshold (15) score was used as defined in the protocol (3 children scored in the ASD range and 1 scored below this threshold and so was placed in the non-ASD group).

**Figure 2**

*STARD Flowchart for ADI-R Deaf Adaptation*



The characteristics of the children in the total eligible sample compared to those whose parents completed the ADI-R Deaf Adaptation were similar in terms of age, gender, ethnic group and language used (see Table 2). In those taking part (and completing the ADI-R Deaf adaptation), the deaf children with ASD group had an older age profile, a higher proportion of males (85% vs. 74%) and were comparable based on ethnicity compared to the deaf without ASD. Methods of communication used by Deaf children and young people, parents and guardians included: spoken English, Sign Supported English (SSE) or British Sign Language (BSL) (see Table 2). Supplementary Table 1 shows the parent reported co-morbid diagnoses of the children and Supplementary Table D shows the assistive devices used). A lower proportion of deaf children with ASD used BSL (37% vs. 66%) and Other communication (19% vs. 3%) compared to deaf children without ASD but similar proportions used spoken English and sign supported English.

 **Table 2**

*Demographic Characteristics by Diagnostic Group.*

|  |  |  |  |
| --- | --- | --- | --- |
|  |  | **Total eligibleᵅ**n=204 | **ADI\_R-Deaf Adaption**n=139 |
|  |  | **Deaf with ASD**n=78 | **Deaf without ASD**n=126 | **Deaf with ASD**n=65 | **Deaf without ASD**n=74 |
| **Gender** | Male | 65 (83%) | 92 (74%) | 55 (85%) | 55 (74%) |
| Female | 13 (17%) | 33 (26%) | 10 (15%) | 19 (26%) |
| **Age** | 2-3 | 6 (8%) | 16 (13%) | 4 (6%) | 11 (15%) |
| 4-9 | 29 (37%) | 61 (49%) | 25 (39%) | 37 (50%) |
| 10+ | 43 (55%) | 48 (38%) | 38 (55%) | 26 (35%) |
| **Ethnicity** | White | 60 (77%) | 106 (85%) | 49 (75%) | 61 (82%) |
| Black | 2 (3%) | 5 (4%) | 1 (2%) | 4 (4%) |
| Asian | 8 (10%) | 8 (6%) | 8 (12%) | 5 (7%) |
| Mixed | 7 (9%) | 5 (4%) | 6 (9%) | 3 (6%) |
| Other | 1 (1%) | 1 (1%) | 1 (2%) | 1 (1%) |
| **Languages** Used (yes) | Use BSL | 31 (40%) | 82 (66%) | 24 (37%) | 49 (66%) |
| Spoken English | 48 (62%) | 71 (57%) | 41 (63%) | 27 (50%) |
| Sign supported English | 26 (33%) | 33 (26%) | 22 (34%) | 19 (26%) |
| Other spoken language | 1 (1%) | 3 (2%) | 1 (2%) | 1 (1%) |
| Other | 15 (19%) | 4 (3%) | 12 (19%) | 1. (3%)
 |

ᵅMissing data for 1 deaf child without ASD

There were statistically significant differences in scores between deaf children with and without ASD for all the three ADI-R algorithm behavioural domain scores (p<0.001) (see Table 3).

The items included in the ADI-R published algorithms for each domain have not changed in the ADI-R Deaf Adaptation (Rutter et al., 2003).

**Table 3** *Mean Scores for ADI-R-Deaf Adaptation Algorithm Domains by Diagnostic Group*

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
|  | Deaf with ASD | Deaf without ASD | Mean difference (SE), (95% CI) | p value |
| Mean (SD) | n | Mean (SD) | n |
| A: Reciprocal Social Interaction | 23.7 (4.3) | 65 | 8.5 (8.1) | 73 | 15.2 (1.1),(13.0, 17.5) | p<0.001 |
| B: Communication – verbal/signing \* | 16.5 (4.4) | 48 | 6.5 (6.5) | 59 | 10.0 (1.1).(7.8, 12.1) | p<0.001 |
| B: Communication – non verbal/non signing\* | 11.9 (2.6) | 17 | 4.8 (4.5) | 14 | 7.2 (1.3).(4.5, 9.8) | p<0.001 |
| C: Restricted, repetitive and stereotyped patterns ofbehaviour | 6.7 (3.0) | 65 | 2.5 (2.7) | 74 | 4.1 (0.5).(3.2, 5.1) | p<0.001 |

\*‘Verbal/signing’ defined as functional use of 3 word / sign phrases (see item 30 ADI-R -deaf adaptation (2020)

The sensitivities and specificities for each of the domain scores for the existing published and optimal domain cut-off scores are presented in Table 4. In deaf children, the optimal cut-off scores were higher than the published cut-off scores for all the domain scores. For participants whose scores exceeded the specified cut-offs in all three algorithm content areas and for whom the onset of disorder was evident by 36 months of age, the sensitivity was 89% (95 CI: 79%, 96%) and specificity 81% (95% CI: 70%, 89%). The sensitivity analysis using only the NICE guideline standard ASD clinical assessment data gave a sensitivity of 88% (77%, 95%) and a specificity of 81% (70%, 90%). These perform as well as recent published systematic review data from the ADI-R, which quote overall figures of 75% and 82% respectively (Lebersfeld et al, 2021).

**Table 4**

*ADI-R Deaf Adaptation Algorithm Domain Scores against Diagnostic Group*

|  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- |
|  | **AUC (95% CI)** | **N****(Deaf with ASD/Deaf without ASD)** | **Existing Cut-offᵅ** | **Sensitivity** | **Specificity** | **Optimal** **cut-off using highest Youden Index** | **Sensitivity** | **Specificity** |
| **Aspirational success****criteria** |  |  |  | 70% | 60% |  |  |  |
| **A: Reciprocal Social Interaction** | 0.921(0.874, 0.968) | 138 (65/73) | =10 | 100% (95%, 100%) | 65%(54%, 76%) | =20 | 85% (74%, 92%) | 89% (80%, 95%) |
| **B: Communication -** verbal/signing \* | 0.870(0.801,0.939) | 107 (48/59) | =8 | 100% (93%, 100%) | 66% (53%, 78%) | =11 | 92% (80%, 98%) | 73% (60%, 84%) |
| **B: Communication –** non verbal/non signing\* | 0.917(0.813,1.000) | 31 (17/14) | =7 | 94% (71%, 100%) | 57% (29%, 82%) | =10 | 88% (64%, 99%) | 86% (57%, 98%) |
|  **B: Communication combined** |  |  |  | 98% (92%, 100%) | 64% (52%, 75%) |  | 91% (81%, 97%) | 75% (64%, 85%) |
| **C: Restricted, repetitive****and stereotyped patterns of behaviour** | 0.843(0.777,0.908) | 139 (65/74) | =3 | 91% (81%, 97%) | 58% (46%, 69%) | =5 | 80% (68%, 89%) | 77% (66%, 86%) |
| **Scores in all three content areas exceed the specified cut-offs, and onset of the disorder is evident by 36 months of age.** |  | 138 (65/73) |  | 89% (79%, 96%) | 81% (70%, 89%) |  |  |  |

ᵅ Published cut-offs scores for the ADI-R (Rutter et al., 2003)

\*‘Verbal/signing’ defined as functional use of 3 word / sign phrases (see item 30 ADI-R -deaf adaptation (2020)

## Reliability of the ADI-R Deaf Adaptation Scoring Algorithm

### Section A: Reciprocal Social Interaction (n=138)

The alpha coefficient was 0.91, showing that the items have high internal consistency. The factor analysis revealed only one factor, explaining 85% of the variation, suggesting that these items are unidimensional. Comparing the factor scores between deaf children with and without ASD showed a significant difference between groups (p=<0.001).

### Section B: Qualitative Abnormalities in Communication (n=138)

For children with language the alpha coefficient was 0.81 (n=106), suggesting that the items have high internal consistency. The factor analysis revealed only one factor (explaining 67% of the variation), suggesting that these items are unidimensional. Comparing the factor scores between deaf children with and without ASD showed a significant difference between groups (p=<0.001).

For non-linguistic children, the alpha coefficient is 0.80 (n=31), suggesting that these items also have high internal consistency. The factor analysis revealed only one factor, explaining 85% of the variation, suggesting that these items are unidimensional. Comparing the factor scores between deaf children with and without ASD showed a significant difference between groups (p=<0.001).

### Section C: Restricted, Repetitive or Stereotyped Patterns of Behaviour (n=139)

The alpha coefficient was 0.81, suggesting that the items have a high internal consistency. The factor analysis revealed two factors. The eigenvalue for the first factor is distinctly larger than the eigenvalue for the second factor (3.437 versus 1.264), explaining 59% of the variation. Only 2 items fall into the second factor (Verbal/signing rituals and Hand and finger mannerisms; [Table](#_bookmark29) 5). Comparing the factor scores between deaf children with ASD and deaf children without ASD shows a significant difference for Factor 1 (p=<0.001), but not Factor 2 (p=0.226).

##### **Table 5**

##### *Component Matrix for ADI-R-ADS Section C: Restricted, Repetitive or Stereotyped Patterns of Behaviour*

|  |  |
| --- | --- |
|  |  **Factor loading** |
| 1 | 2 |
| **Section C1** |  |  |
| Unusual preoccupations | 0.701 |  |
| Circumscribed interests | 0.618 |  |
| **Section C2** |  |  |
| Verbal/signing rituals |  | -0.539 |
| Compulsions/rituals | 0.680 |  |
| **Section C3** |  |  |
| Hand and finger mannerisms |  | 0.708 |
| Other complex mannerisms or stereotyped body movements | 0.713 |  |
| **Section C4** |  |  |
| Repetitive use of objects or interest in parts of objects | 0.782 |  |
| Unusual sensory interests | 0.745 |  |

# Discussion

The findings suggest that the ADI-R Deaf Adaptation is a valid and useful measure for the assessment of deaf children with suspected ASD. This includes children who are verbal or non-verbal and those using signing or spoken language.

Some key changes have been made that address concerns raised by parents and clinicians about the ASD diagnostic assessment process (Brenman et al 2017; Young et al 2019). For example, information about the deaf child’s language and communication history and their environment (including family communication and educational environment), sign language milestones together with new ADI-R scoring code descriptors to allow equivalence of signed and spoken languages in assessment and rating are included in the ADI-R Deaf Adaptation. The new version also includes peer-reviewed research findings, including sign reversal seen in deaf children with autism (Shield & Meier, 2012) and problems using sign language prosody (as distinct from spoken language prosody) in deaf children. Changes also recognise the emerging research literature that proposes that, from linguistic and neuroimaging perspectives, spoken language/gesture has equivalence to sign language/gesture (Goldin-Meadow & Brentari, 2017).

The existing published ADI-R algorithm was found to be reliable and able to discriminate well (with good enough sensitivity and specificity) between deaf children with and without ASD in this community sample. Retaining the existing algorithm thresholds has the advantage that it avoids confusion across ADI-R versions and enables comparisons across different studies. Interestingly the Youden Index for optimal cut-offs in the ROC curve suggests that when used with deaf children the thresholds are higher in all domains compared to the existing algorithm. This finding is in keeping with the observation that deaf children without ASD have some behaviours that score on the ADI-R but deaf children with ASD show many more of these types of behaviours. Previous studies highlight that these overlapping behaviour profiles can lead to diagnostic uncertainty during assessment (Wright & Oakes, 2012). Clinicians need to undertake a detailed developmental history which includes detail about being deaf as part of the multisource assessment process to minimise errors in diagnosis (Phillips et al, 2021).

The algorithm items in the reciprocal social communication and qualitative abnormalities of communication domain showed high reliability and internal consistency with one main factor identified - replicating previous studies {Rutter et al, 2003). Interestingly in our population of deaf children for the Restricted, Repetitive or Stereotyped Patterns of Behaviour (RRB) domain there is a significant difference for Factor 1 (p=<0.001), between those with ASD and those without ASD but not for Factor 2 (p=0.226). Factor 2 includes hand and finger mannerisms and verbal/signing rituals. Developing deaf children without ASD engage in many varied hand and finger movements as part of language exploration (Roos, 2013) and communicative non-linguistic verbal utterances (Lederberg & Everhart, 1998). Thus scoring on these items has little/no discriminative diagnostic value. The additional questions included in the ADI-R deaf adaptation provide a useful framework for this part of the assessment of deaf children. High scores in some domains for some deaf children could be related to a range of co-morbidities seen in deaf children (see supplementary table E).

It is of note that lower proportions of deaf children with ASD use BSL as their main language than deaf children without ASD. This may reflect additional challenges for a deaf child with ASD accessing learning and using a signed language; although there is no research we are aware of confirming this belief. It is also possible that common misunderstandings about language acquisition for deaf children (Humphries et al, 2016) may mean some parents are advised not to use sign language with their deaf children despite evidence that this may be harmful (Hall, 2017).

The ADI-R Deaf Adaptation mandatory questions were translated into BSL using a robust forward and backwards translation process (Moore et al., 2013) so that for this first validation study, the interview was available in BSL and in English. In addition for parents (informants) who were deaf and whose preferred language was BSL, our study team recommended that a deaf clinician who communicated using BSL administered the interview (or worked with a qualified interpreter if this was not possible) (Ackroyd & Wright, 2018). For this subset the deaf parents reported that this process worked well but further analysis was not possible due to the small sample.

## Strengths and Limitations

Parents of deaf children were recruited from a variety of community and referral sources. As such the sample is likely to be representative of both deaf children requiring an ASD diagnostic assessment and deaf children without ASD. Our sample included some older children, more males and children with a range of co-occurring conditions as expected in a sample of deaf children (Brett et al, 2016; Fortnum et al, 2002). However the comorbidity data needs to be treated with caution as it is based on parent report and despite the sample size (relatively large for an ASD study) a larger validation study would be needed to investigate specific subgroups such as children of different ages or levels of learning disability (Havdahl et al, 2016). Our sample also included children using a range of assistive devices for hearing or none at all and represent a wide range of child and family backgrounds.

A further strength is the composition of the research team which included specialist clinical academics supported by the NDCAMHS all with expertise in ASD and deafness. However this also highlights a national challenge, namely that these skills are not more widely available in community child health services. The team also emphasised the important contribution of people with deaf lived experience (deaf researchers and deaf professionals). Anecdotally it is encouraging to note that NDCAMHS is now receiving more requests to train deaf clinical staff in the ASD diagnostic assessment process.

A limitation of the research is that despite wide international recruitment strategies the Delphi Consensus process identified just 17 experts who met the criteria to take part in our study and due to different time commitments and other pressures only 9 experts (one of whom was deaf) were able to complete the Delphi process. We used a number of strategies with the aim of mitigating this issue including time extensions with the support of our funder. The study has benefitted from the participation of many people with perspectives on deaf experiences including the deaf PPI members , deaf parents of participants, members of the IRRT and the valued contributions of the deaf researchers within the research team - the trial co-ordinator and 5 research assistants. Despite this we believe we could have improved on this believing that it is essential to have deaf perspectives at the heart of research of this nature.

A further limitation facing many research groups is the difficulty finding a gold standard assessment for deaf people to compare the new assessments with, given that there were no available validated measures in deaf children. We addressed this by using the best current practice namely the NICE guideline recommended assessment process. Several children were given a new diagnosis of ASD as a result of these NICE guideline assessments (but not as a result of the ADI-R Deaf adaptation which was being researched).

## Further Research

Whilst this new tool was developed for use with informants about the development and current functioning of deaf children whether they used spoken or signed language we researched this in a context where spoken English and British Sign Language were the main languages being used in our recruited families. A future step will be to ascertain whether the findings can be replicated in other settings (in the UK and further afield) including the feasibility and validity in different countries with different linguistic and cultural backgrounds.

It will also be interesting to investigate whether the use of standardised instruments for the assessment of suspected ASD in deaf children can contribute to improving standards and service provision and whether this will lead to a reduction in average age of assessment and diagnosis in deaf children.

A further key area for future research is whether this instrument can differentiate between ASD and those deaf children who have had limited early life exposure to language learning (some have termed this ‘language deprivation’) (Hall et al, 2017). Our new adaptations included a range of questions about language learning experiences at home and at school, which the DIEP highlighted as an important consideration. However this study was not designed to compare findings between deaf children who have experienced language deprivation and those without. This is an important area for future research.

##  Implications for Policy and Practice

Our findings are encouraging and certainly if replicated indicate that the ADI-R Deaf Adaptation could be considered for use in the assessment of deaf children with suspected ASD. This research contributes to the emerging body of work recommending that validated ASD diagnostic assessment tools may be helpful when assessing deaf children (Syzmanski & Brice, 2008). Improved training in the use of validated tools should enable more community child health services to provide ASD diagnostic services appropriately modified for deaf children/young people.

The National Deaf Child and Adolescent Mental Health Service (found in ten centres across England) has made the strategic decision through its clinical network to have improved autism assessment processes that include using these new tools and to expand the training of clinical staff in the use of ASD assessment and diagnostic measures specifically adapted for use with deaf children. There are inevitably resource implications for this endeavour.

# Conclusion

This is the first validation of a parent semi-structured interview to assess for ASD in a deaf population. Against a backdrop of limited research in this field (Hansen & Scott, 2017) this provides a very welcome addition for clinical practice. Our findings indicate that the ADI-R Deaf Adaptation is a valid measure for use in the deaf child population in the UK context, no matter what languages or communication methods they use. Further validation work is necessary in other linguistic and cultural contexts, as is further research into the presentation of ASD in deaf people. The improved use of validated ASD diagnostic assessment processes may help reduce inequities in diagnostic age and accuracy in deaf children, which could in turn facilitate improved planning of appropriate educational provision and early parental support.

# Acknowledgements

We would like to express our gratitude to the international experts who contributed to the Delphi consensus. We are also grateful to the children and families that took part and to the clinicians who trained and conducted the ADI-R Deaf Adaptation interviews and carried out the NICE standard guideline clinical assessments. We would also like to give our thanks to the members of the IRRP including Professor Helen McConachie, Dr Hannah George, Ms Rachael Hayes, Professor Richard Ogden, Dr Katie Rowley and Ms Josie Mulloy. We especially thank Prof Cathy Lord and Prof Sir Michael Rutter for their support for this development work and acknowledge the advice and enthusiasm of Western Psychological Services, the publishers of ADI-R who hold the copyright for the measure. We thank Prof Alys Young and Emma Ferguson-Coleman for involvement in the qualitative work with parents and Dr Natassia Brenman involved in qualitatively work with clinicians published separately.

# Declaration of Conflicting Interest

The final author of this manuscript is one of the original authors of the ADI-R. None of the other authors have declared any potential conflicts of interest with respect to the research, authorship and/or publication of this article.

# Funding

This research was funded by Medical Research Council - MR/K015435/1. The sponsor and host Trust was the Leeds and York Partnership NHS Foundation Trust.

# References

Ackroyd, V., & Wright, B. (2018). Working with British Sign Language (BSL) interpreters: lessons from child and adolescent mental health services in the UK. *Journal of Communication in Healthcare*, *11*(3), 195-204.

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

Baron-Cohen, S. (2008). Theories of the autistic mind. *The Psychologist*.

Batten, G., Oakes, P. M., & Alexander, T. (2014). Factors associated with social interactions between deaf children and their hearing peers: A systematic literature review. *Journal of deaf studies and deaf education*, *19*(3), 285-302.

Beattie, E. M. J. K., & Mackway-Jones, K. (2004). A Delphi study to identify performance indicators for emergency medicine. *Emergency Medicine Journal*, *21*(1), 47-50.

Beech, B. (1999). Go the extra mile-use the Delphi Technique. *Journal of nursing management*, *7*(5), 281-288.

Bishop, S. L., Richler, J., & Lord, C. (2006). Association between restricted and repetitive behaviors and nonverbal IQ in children with autism spectrum disorders. *Child neuropsychology*, *12*(4-5), 247-267.

Bishop, S. L., Havdahl, K. A., Huerta, M., & Lord, C. (2016). Subdimensions of social‐communication impairment in autism spectrum disorder. *Journal of Child Psychology and Psychiatry,* 57(8), 909-916.

Bond, C., Symes, W., Hebron, J., Humphrey, N., & Morewood, G. (2016). Educating persons with autistic spectrum disorder–A systematic literature review. *The National Council for Special Education,[NCSE]*, *20*.

Bossuyt, P. M., Reitsma, J. B., Bruns, D. E., Gatsonis, C. A., Glasziou, P. P., Irwig, L., ... & Kressel, H. Y. (2015). STARD 2015: an updated list of essential items for reporting diagnostic accuracy studies. *Clinical chemistry*, *61*(12), 1446-1452.

Brenman, N. F., Hiddinga, A., & Wright, B. (2017). Intersecting cultures in deaf mental health: An ethnographic study of NHS professionals diagnosing autism in D/deaf children. *Culture, Medicine, and Psychiatry*, *41*(3), 431-452.

Brett, D., Warnell, F., McConachie, H., & Parr, J. R. (2016). Factors affecting age at ASD diagnosis in UK: no evidence that diagnosis age has decreased between 2004 and 2014. *Journal of autism and developmental disorders*, *46*(6), 1974-1984.

Brown, N., Crawford, I., Carley, S., & Mackway-Jones, K. (2006). A Delphi-based consensus study into planning for biological incidents. *Journal of Public Health*, *28*(3), 238-241.

Buescher, A. V., Cidav, Z., Knapp, M., & Mandell, D. S. (2014). Costs of autism spectrum disorders in the United Kingdom and the United States. *JAMA pediatrics*, *168*(8), 721-728.

Chandler, S., Charman, T., Baird, G., Simonoff, E., Loucas, T. O. M., Meldrum, D., ... & Pickles, A. (2007). Validation of the social communication questionnaire in a population cohort of children with autism spectrum disorders. *Journal of the American Academy of Child & Adolescent Psychiatry*, *46*(10), 1324-1332.

Chess, S. (1977). Follow-up report on autism in congenital rubella. *Journal of autism and childhood schizophrenia*, *7*(1), 69-81.

Dade, P. (2013). Encyclopedia of Autism Spectrum Disorders. *Reference Reviews*.

Corina, D. P., Bellugi, U., & Reilly, J. (1999). Neuropsychological studies of linguistic and affective facial expressions in deaf signers. *Language and Speech*, *42*(2-3), 307-331.

Consortium for Research in Deaf Education. (2015). CRIDE Report on 2015 Survey on Educational Provision for Deaf Children.

Crawford, I. W. F., Mackway-Jones, K., Russell, D. R., & Carley, S. D. (2004). Delphi based consensus study into planning for chemical incidents. *Emergency medicine journal*, *21*(1), 24-28.

Dachkovsky, S., & Sandler, W. (2009). Visual intonation in the prosody of a sign language. *Language and speech*, *52*(2-3), 287-314.

Daniels, A. M., & Mandell, D. S. (2014). Explaining differences in age at autism spectrum disorder diagnosis: A critical review.*Autism*, 18(5), 583-597.

Denmark, T., Atkinson, J., Campbell, R., & Swettenham, J. (2014). How do typically developing deaf children and deaf children with autism spectrum disorder use the face when comprehending emotional facial expressions in British sign language?. *Journal of autism and developmental disorders*, *44*(10), 2584-2592.

Fortnum, H., & Davis, A. (1997). Epidemiology of permanent childhood hearing impairment in Trent Region, 1985–1993. *British journal of audiology*, *31*(6), 409-446.

Fortnum, H. M., Marshall, D. H., & Summerfield, A. Q. (2002). Epidemiology of the UK population of hearing-impaired children, including characteristics of those with and without cochlear implants—audiology, aetiology, comorbidity and affluence. *International Journal of Audiology*, 41(3), 170-179.

Gallaudet Research Institute. (2011). Regional and national summary report of data from the 2009–10 annual survey of deaf and hard of hearing children and youth.

Gibson, J. M. (1998). Using the Delphi technique to identify the content and context of nurses' continuing professional development needs. *Journal of clinical nursing*, *7*(5), 451-459.

Goldin-Meadow, S., & Brentari, D. (2017). Gesture, sign, and language: The coming of age of sign language and gesture studies. *Behavioral and Brain Sciences*, 40.

Goldman, S., Wang, C., Salgado, M. W., Greene, P. E., Kim, M., & Rapin, I. (2009). Motor stereotypies in children with autism and other developmental disorders. *Developmental Medicine & Child Neurology*, *51*(1), 30-38.

Guardino, C., & Cannon, J. E. (2015). Theory, research, and practice for students who are deaf and hard of hearing with disabilities: Addressing the challenges from birth to postsecondary education. *American Annals of the Deaf*, *160*(4), 347-355.

Gurney, J. G., McPheeters, M. L., & Davis, M. M. (2006). Parental report of health conditions and health care use among children with and without autism: National Survey of Children's Health. *Archives of pediatrics & adolescent medicine*, *160*(8), 825-830.

Hall, W. C. (2017). What you don’t know can hurt you: The risk of language deprivation by impairing sign language development in deaf children. *Maternal and Child Health Journal*, 21(5), 961-965.

Hall, W. C., Levin, L. L., & Anderson, M. L. (2017). Language deprivation syndrome: A possible neurodevelopmental disorder with sociocultural origins. *Social Psychiatry and Psychiatric Epidemiology,*52(6), 761-776.

Havdahl, K.A., Bal, V.H., Huerta, M., Pickles, A., Øyen, A.S., Stoltenberg, C., Lord, C. and Bishop, S.L., (2016). Multidimensional influences on autism symptom measures: implications for use in etiological research.*Journal of the American Academy of Child & Adolescent Psychiatry,* 55(12), 1054-1063.

Hayes, S. A., & Watson, S. L. (2013). The impact of parenting stress: A meta-analysis of studies comparing the experience of parenting stress in parents of children with and without autism spectrum disorder. *Journal of autism and developmental disorders*, *43*(3), 629-642.

Humphries, T., Kushalnagar, P., Mathur, G., Napoli, D.J., Padden, C., Rathmann, C. and Smith, S., (2016). Language choices for deaf infants: Advice for parents regarding sign languages.*Clinical Pediatrics*, 55(6), 513-517.

Jiang, H. Y., Xu, L. L., Shao, L., Xia, R. M., Yu, Z. H., Ling, Z. X., ... & Ruan, B. (2016). Maternal infection during pregnancy and risk of autism spectrum disorders: a systematic review and meta-analysis. *Brain, behavior, and immunity*, *58*, 165-172.

Jure, R., Rapin, I., & Tuchman, R. F. (1991). Hearing‐impaired autistic children. *Developmental Medicine & Child Neurology*, *33*(12), 1062-1072.

Kasari, C., Gulsrud, A., Paparella, T., Hellemann, G., & Berry, K. (2015). Randomized comparative efficacy study of parent-mediated interventions for toddlers with autism. *Journal of consulting and clinical psychology*, *83*(3), 554.

Kelly, B., Williams, S., Collins, S., Mushtaq, F., Mon-Williams, M., Wright, B., ... & Wright, J. (2019). The association between socioeconomic status and autism diagnosis in the United Kingdom for children aged 5–8 years of age: Findings from the Born in Bradford cohort. *Autism*, *23*(1), 131-140.

Knapp, M., Romeo, R., & Beecham, J. (2009). Economic cost of autism in the UK. *Autism*, *13*(3), 317-336.

Lebersfeld, J. B., Swanson, M., Clesi, C. D., & O’Kelley, S. E. (2021). Systematic review and meta-analysis of the clinical utility of the ADOS-2 and the ADI-R in diagnosing autism spectrum disorders in children. *Journal of Autism and Developmental Disorders*, 1-14. 21st January 2021. https://doi.org/10.1007/s10803-020-04839-z

Lederberg, A. R., & Everhart, V. S. (1998). Communication between deaf children and their hearing mothers: The role of language, gesture, and vocalizations. *Journal of Speech, Language, and Hearing Research,* 41(4), 887-899.

Lord, C., Rutter, M., & Le Couteur, A. (1994). Autism Diagnostic Interview-Revised: a revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. *Journal of autism and developmental disorders*, *24*(5), 659-685.

McKenna, H. P. (1994). The Delphi technique: a worthwhile research approach for nursing?. *Journal of advanced nursing*, *19*(6), 1221-1225.

Meinzen-Derr, J., Wiley, S., Bishop, S., Manning-Courtney, P., Choo, D. I., & Murray, D. (2014). Autism spectrum disorders in 24 children who are deaf or hard of hearing. *International Journal of Pediatric Otorhinolaryngology*, 78(1), 112-118.

Moore, K., Wright, B., Moore, D., Ogden, R., & Rogers, K. (2013). Overcoming the challenges of translating mental health instruments into sign languages. *International Journal on Mental Health and Deafness*, *3*(1).

Morzaria, S., Westerberg, B. D., & Kozak, F. K. (2004). Systematic review of the etiology of bilateral sensorineural hearing loss in children. *International journal of pediatric otorhinolaryngology*, *68*(9), 1193-1198.

National Institute for Health and Care Excellence (NICE) (2017, December 20) *Diagnosing Autism Spectrum Disorder in Under 19s.* <https://www.nice.org.uk/Guidance/CG128>

O’Reilly, C., Lewis, J. D., & Elsabbagh, M. (2017). Is functional brain connectivity atypical in autism? A systematic review of EEG and MEG studies. *PloS one*, *12*(5), e0175870.

Phillips, H.,Wright, B., Allgar, V., McConachie, H., Sweetman, J., Hargate, R.,Hodkinson, R., Bland, M., George, H., Hughes, A., Hayward, E., Fernandez Garcia De las Heras, F., Le Couteur, A. (2021) Adapting and validating the Autism Diagnostic Observation Schedule Version 2 for use with deaf children and young people. *Journal of Autism Developmental Disorders* (2021). 24th March 2021 online. https://doi.org/10.1007/s10803-021-04931-y

Roos, C. (2013). Young deaf children's fingerspelling in learning to read and write: An ethnographic study in a signing setting. *Deafness & Education International*, 15(3), 149-178.

Roper, L., Arnold, P., & Monteiro, B. (2003). Co-occurrence of autism and deafness: diagnostic considerations. *Autism*, *7*(3), 245-253.

Rutter, M., Bailey, A., & Lord, C. (2003). *The social communication questionnaire: Manual*. Western Psychological Services.

Rutter, M., Le Couteur, A., & Lord, C. (2003). Autism diagnostic interview-revised. *Los Angeles, CA: Western Psychological Services*, *29*(2003), 30.

Saccà, A., Cavallini, F., & Cavallini, M. C. (2019). Parents of Children with Autism Spectrum Disorder: a systematic review. *Journal of Clinical & Developmental Psychology*, *1*(3).

Schick, B., De Villiers, P., De Villiers, J., & Hoffmeister, R. (2007). Language and theory of mind: A study of deaf children. *Child development*, *78*(2), 376-396.

Sheikh, A., Major, P., & Holgate, S. T. (2008). Developing consensus on national respiratory research priorities: key findings from the UK Respiratory Research Collaborative's e-Delphi exercise. *Respiratory medicine*, *102*(8), 1089-1092.

Shield, A., & Meier, R. P. (2012). Palm reversal errors in native-signing children with autism. *Journal of Communication Disorders*, *45*(6), 439-454.

Shield, A., Meier, R. P., & Tager-Flusberg, H. (2015). The use of sign language pronouns by native-signing children with autism. *Journal of Autism and Developmental Disorders*, *45*(7), 2128-2145.

Shield, A., Cooley, F., & Meier, R. P. (2017). Sign language echolalia in deaf children with autism spectrum disorder. *Journal of Speech, Language, and Hearing Research*, 60(6), 1622-1634.

Sharkey, S. B., & Sharples, A. Y. (2001). An approach to consensus building using the Delphi technique: developing a learning resource in mental health. *Nurse education today*, *21*(5), 398-408.

Sim, A., Vaz, S., Cordier, R., Joosten, A., Parsons, D., Smith, C., & Falkmer, T. (2018). Factors associated with stress in families of children with autism spectrum disorder. *Developmental neurorehabilitation*, *21*(3), 155-165.

Szymanski, C. A., Brice, P. J., Lam, K. H., & Hotto, S. A. (2012). Deaf children with autism spectrum disorders. *Journal of autism and developmental disorders*, *42*(10), 2027-2037.

Szymanski, C., & Brice, P. J. (2008). When Autism and Deafness Coexist in Children: What We Know Now. *Odyssey: New Directions in Deaf Education*, *9*(1), 10-15.

Szarkowski, A., & Brice, P. J. (2016). Hearing parents’ appraisals of parenting a deaf or hard-of-hearing child: Application of a positive psychology framework. *Journal of deaf studies and deaf education*, *21*(3), 249-258.

Tavakol, M., & Dennick, R. (2011). Making sense of Cronbach's alpha. *International journal of medical education*, *2*, 53.

Vaccari, C., & Marschark, M. (1997). Communication between parents and deaf children: Implications for social‐emotional development. *Journal of Child Psychology and Psychiatry*, *38*(7), 793-801.

Warren, Z., McPheeters, M. L., Sathe, N., Foss-Feig, J. H., Glasser, A., & Veenstra-VanderWeele, J. (2011). A systematic review of early intensive intervention for autism spectrum disorders. *Pediatrics*, *127*(5), e1303-e1311.

Wachtel, L. E., Hartshorne, T. S., & Dailor, A. N. (2007). Psychiatric diagnoses and psychotropic medications in CHARGE syndrome: A pediatric survey. *Journal of Developmental and Physical Disabilities*, *19*(5), 471-483.

World Health Organization. (2011). *The ICD-10 International Classification of mental and behavioural disorders: diagnostic criteria for research* (Vol. 2). World Health Organization.

Wright, B., & Oakes, P. (2012). Does socio-emotional developmental delay masquerade as autism in some deaf children?. *International Journal on Mental Health and Deafness*, *2*(1).

Wright, B., Collingridge-Moore, D., Smith, J., & Richardson, T. (2018). The Use of Audiological

Classification Systems. *International Journal on Mental Health and Deafness*, 4(1), 59-64.

Wright, B., Phillips, H., Le Couteur, A., Sweetman, J., Hodkinson, R., Ralph-Lewis, A., Hayward, E.,

Brennan, A., Mulloy, J., Day, N., Bland, M., & Allgar, V. (2020). Modifying and validating the social responsiveness scale edition 2 for use with deaf children and young people.*PloS one*,15(12), e0243162. <https://doi.org/10.1371/journal.pone.0243162>

Young, A., Ferguson-Coleman, E., Wright, B., & Le Couteur, A. (2019). Parental Conceptualizations of Autism and Deafness in British Deaf Children. *The Journal of Deaf Studies and Deaf Education*, *24*(3), 280-288.