Clinical Use of the Autism Diagnostic Observation Schedule–Second Edition with Children Who Are Deaf

Deborah Mood, Ph.D.[1] and Aaron Shield, Ph.D.[2]

ABSTRACT

The Autism Diagnostic Observation Schedule–Second Edition (ADOS-2) was administered to eight children who are deaf and who are native American Sign Language (ASL) users with previous autism spectrum disorder (ASD) diagnosis. Classification on two different module selection criteria was compared based on: (1) standardized administration rules (signs not counted as equivalent to words) and (2) commonly utilized clinical administration (sign language complexity treated equivalently to spoken language complexity). Differential module selection resulted in discrepant classification in five of the eight cases (63%) and suggests that ADOS-2 via standardized test administration may result in a failure to identify autism among children who are deaf with primary communication in ASL. Two of the eight children (25%) did not exceed the cutoff for an ASD classification on either module administered despite previous ASD diagnosis. Overall results suggest that caution should be used when utilizing the ADOS-2 with children who are deaf who primarily communicate using ASL.

KEYWORDS: Autism, deaf, ADOS-2, assessment

Learning Outcomes: As a result of this activity, the reader will be able to (1) discuss issues that arise in the evaluation of children who are deaf or hard of hearing referred for suspected autism spectrum disorder; (2) discuss the use of the Autism Diagnostic Observation Schedule–Second Edition when applied to children who are deaf.

[1]Child Development Unit, Children's Hospital Colorado, Aurora, Colorado; [2]Department of Psychology, Boston University, Boston, Massachusetts.

Address for correspondence: Deborah Mood, Ph.D., Child Development Unit, Children's Hospital Colorado, 13123 E. 16th Avenue, Box 140, Aurora, CO 80045 (e-mail: Deborah.Mood@childrenscolorado.org).

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An autism spectrum disorder (ASD) is defined as significant impairment in social communication in addition to the presence of repetitive behaviors and restricted interests.[1] When deficits in social communication are coupled with hearing loss, this complex comorbidity can significantly complicate language development. Being able to identify autism among children who are deaf or hard of hearing (D/HH) is necessary to inform treatment, which may take a markedly different course than typical D/HH intervention.

Just as increasing rates of ASDs have been reported in the general population, increasing rates of ASD have been identified among children who are D/HH.[2] Furthermore, research has indicated that when compared with estimates among the general population (CDC 1:68)[3] a greater than expected number of children who are D/HH are also reported to have an ASD. In looking at rates of hearing loss among children with autism, Rosenhall and colleagues reported a prevalence of profound hearing loss to be 10 times greater than the 0.1 to 0.2% commonly reported within the general population.[4] Conversely, Jure reported 4% of their sample of students who are deaf also had an ASD.[5] Most recently, Szymanski and colleagues reported a rate of 1 in 59 (1.6%) 8-year-old children receiving school-based services for hearing loss also had an additional educational classification of autism.[2]

Age of autism diagnosis tends to occur significantly after the identification of the child's hearing loss[6] and later than in the hearing population.[7] The advent of newborn infant hearing screening procedures and specialized early intervention services for children who are D/HH should allow deafness professionals to tease apart developmental issues from hearing-related issues at a much earlier age, in theory allowing for earlier identification of ASD among this population. However, several factors complicate early diagnosis. On a surface level, symptoms of autism and hearing loss may appear to overlap. For example, similar to children with autism, children who are D/HH may demonstrate language delays,[8] theory of mind delays,[9] and most obviously may not respond to their name depending on their degree of hearing loss. Despite this, professionals familiar with hearing loss recognize several other symptoms unique to autism as distinct from those associated with typically developing children with hearing loss. For example, deficits in preverbal communication skills (i.e., eye contact, joint attention, gesture use) are not typically impacted by hearing loss.[10] These are reviewed in more detail in additional articles within this special issue (see Szarkowski et al[11] and Shield,[12] both in this issue).

Currently, distinguishing symptoms of autism as distinct from features commonly associated with hearing loss depends largely on the clinical knowledge of professionals due to limited research describing this complex comorbidity. With few providers trained in both autism and deafness and several potential overlapping symptoms, it is not surprising that research suggests limited diagnostic agreement among professionals when diagnosing ASD among children who are D/HH.[13] Unfortunately, to further complicate diagnosis, there are no ASD screening or diagnostic tools that have been validated for use with children who are D/HH,[14] and there is limited research describing the use of existing ASD diagnostic tools with this population. However, in clinical practice, professionals seeking to identify autism with children who are D/HH commonly rely on ASD assessment tools with considerable variability in how the tools are applied. Lack of dually trained professionals coupled with the unavailability of validated tools for assessing ASD among children who are D/HH likely contributes to misdiagnosis. In this article, we describe an exploratory study using one of the gold-standard tools for diagnosing autism, the Autism Diagnostic Observation Schedule–Second Edition (ADOS-2), with a novel research population: children who are deaf, who have been diagnosed with ASD, and who have been exposed to American Sign Language (ASL) by their parents who are deaf. Because these children have had optimal sign language exposure from birth, we investigate how the ADOS-2 might best be administered and scored when a signed language is the primary or only means of communication. The ADOS-2 was selected because it has strong empirical evidence for use with hearing children. Although children who are D/HH are not

included in the ADOS-2 standardization sample, and strict adherence to the manual is not possible for the reasons described below, our intention was to use ADOS-2 activities as a standardized barometer to explore the usefulness of the ADOS-2 in differential diagnosis of ASD among children who are D/HH.

METHODS

Participants

This study describes analysis of a subset of data collected for a larger parent project exploring the effects of autism on the sign language development of children who are deaf. Subjects for the parent project were recruited nationally from a project Web page and social media. Subjects in that project were administered the ADOS-2 to gather data to confirm subjects' prior ASD diagnosis. Participants in the current study were those tested by the lead author as part of the parent project. Although 20 children in the parent sample completed the ADOS-2, only data collected by the lead author were utilized to ensure consistency across clinical administration and scoring. Participants were eight children, ages 5 to 12, who met the inclusion criteria of severe to profound bilateral sensorineural hearing loss, at least one parent who is deaf, primary exposure to ASL since birth, and previously diagnosed with an ASD. ASD was identified via an educational classification process for each of the eight children. Diagnosis was additionally provided by a psychologist or physician for four of the eight children, although a physician provided diagnosis based on a review of educational data rather than direct assessment in one of the four cases. Age of ASD diagnosis/educational classification ranged from 2 to 8 years. All children demonstrated average nonverbal intelligence, as measured by the Test of Nonverbal Intelligence–Fourth Edition (TONI-4) (mean = 97.25 [SD = 5.1], range 88 to 104).

Measures

AUTISM DIAGNOSTIC OBSERVATION SCHEDULE–SECOND EDITION

The ADOS-2 is a semistructured standardized assessment that uses developmentally appropriate social and toy-based interactions in a 30- to 45-minute interaction to elicit symptoms of autism in four areas: social interaction, communication, play, and repetitive behaviors.[15] The ADOS-2 provides cutoff scores on the classification algorithm for autism and autism spectrum; scores that do not exceed the cutoff are classified as non-ASD. The ADOS-2 consists of a toddler module and four additional modules. Per the manual's guidelines, module selection is based on the child's particular level of language ability: module 1 for children who are preverbal or at the single-word level, module 2 for children with phrase speech, module 3 for children with fluent speech, and module 4 for adolescents with fluent speech.[15] For hearing children, module selection is straightforward. However, for children who communicate primarily using sign language, module selection can take one of two routes: strict adherence to the ADOS-2 manual, which dictates that signs not be considered equivalent to words (thus any signing, nonspeaking child would be administered a module 1), or a less "by-the-book" approach, in which module selection is guided by the child's overall sign language ability. We must stress that the authors of the ADOS-2 explicitly indicate that the instrument was not designed for use with children who are D/HH. However, we consider the ADOS-2 a useful tool in the diagnosis of ASD and there is evidence that clinicians are in fact using it to diagnose ASD in children who are D/HH in clinical practice.

In the present study, participants completed one or more modules of the ADOS-2. First, all children were administered module 1, for children who are minimally verbal or at the single word level, regardless of the complexity of their ASL. Notably, none of the children in this study also communicated using spoken language beyond the single word level. Thus, in the absence of spoken language, for the purpose of the ADOS-2 administration, their language was considered minimally verbal. This was considered "standardized" module selection based on strict adherence to the manual's instructions. If children's sign productions exceeded the production of single signs, then a "clinical" module was administered based on ASL complexity. For example, subjects who

combined signs into simple signed phrases were also administered module 2. Those who demonstrated more advanced sign language (e.g., the ability to combine two different thoughts in ASL) were administered module 3. For subjects whose sign language was at the single sign level, only module 1 was administered. Finally, the scoring algorithm was applied in two different ways: according to standardized administration (e.g., "few to no words") as well as clinical administration (e.g., "some words").

PROCEDURES

This secondary data review was performed under Institutional Review Board approval. ADOS-2 data, collected as part of the parent project protocol, were completed in the homes of the subjects or in one case, at the child's school, per the parent request. Although administered in nonclinical settings, standardized procedures per ADOS-2 manual for setting up materials (i.e., table available, removal of non-ADOS toys) were followed to the extent possible. Data were collected by the first author, who is experienced in working with children who are D/HH and who had obtained reliability on the ADOS-2 through a postdoctoral psychology training program. The examiner communicated with the subjects and their parents directly using ASL.

Several tasks were modified for clinical administration. Modifications were based on literature review and the evaluator's clinical opinion about use of the instrument with children who primarily communicate using sign language. Every effort was made to make consistent modifications in administration and scoring across subjects. Table 1 outlines modifications that were made to standardized by-the-book administration of particular tasks, as well as the justification for these modifications. In addition, some module 1 and 2 tasks were modified to be more developmentally appropriate for children who demonstrated advanced language skills in ASL. Modifications followed guidelines noted in the ADOS-2 manual and elsewhere (i.e., use of more developmentally appropriate free play items, modifications to social routine task).[15,16] Modifications were minimized but were utilized when deemed necessary for valid administration per the ADOS-2 manual.[15]

Likewise, for the purposes of clinical scoring, effort was made to consistently apply modified item scoring criteria for use with this population. Although we cannot capture the full complexity of translating a test for use with native ASL users, modifications consisted largely of interpreting "sign" as equivalent to spoken language. This change was adopted universally for clinical scoring for the purposes of identifying discrepancies in usefulness between standardized and clinical interpretation. In some cases, particular scores were modified to take the unique features of ASL into account (Table 2). Where possible, this decision was empirically supported. Gesture use provides a good example of this process. Although distinguishing gestures from formal signs can be difficult and is theoretically disputed, especially for pointing signs, inflecting (agreement) verbs, and classifier constructions, for the purpose of this study, gestures were differentiated from signs in the following ways.[17–19] First, lexical signs (i.e., nouns, verbs, and other signs that one would normally find in a dictionary) were considered signs. Classifier signs (such as the index finger to represent a person, or the three-hand shape to represent a vehicle) were considered both signs and gestures; the hand shape component (which is lexically determined and varies across signed languages) was considered linguistic, but the movement of the classifier (e.g., to describe the size, shape, or movement of an object) was considered gestural, because this type of description cannot be listed in a dictionary or mental lexicon. Thus, when scoring for Gesture Use, a subject who used formal signs (e.g., BOOK,* CHURCH, SCORPION) would score a 0 when standardized procedures were used (where signs are considered equivalent to gestures rather than words). However, they would only receive a 0 when clinical scoring procedures were used if they also used classifiers in a descriptive manner (e.g., using their pointer fingers to describe the size and shape of a mirror).

* *As is conventional in the literature, we denote signs by their English translations in small caps.*

Table 1 Task Modifications

Task	Modification	Clinical Justification
Response to Name	1. Standardized procedures for responding to spoken name were first presented. 2. If the examinee did not respond to spoken presses presented by the examiner, the examiner presented the examinee's name sign within their peripheral vision (×3). 3. If the examinee did not respond, attention-getting procedures not involving touch typically used within deaf culture were administered (e.g., tapping on the ground, waving within the individual's line of sight). 4. If the examinee did not respond, standardized directions for attempting to get the examinee's attention first by implying they would be tickled and then by tickling them were administered (either by the examiner or parent if available). 5. For the purposes of standardized scoring, Response to Name was scored as a 9.	Sample included children who had a severe to profound bilateral sensorineural hearing loss. Therefore, many of the children would not be expected to respond to their name when presented verbally. However, it is reasonable that typically developing deaf children would respond to culturally accepted norms for attention getting.
Joint Attention	1. Modifications to standardized statements included using the sign for SEE or LOOK + a head turn, without the directional element of that sign, during initial presses. 2. The sign for SEE was then paired with a point (versus directional sign LOOK leading to point) on the last press.	In ASL, the sign for LOOK typically includes a directional gestural element that involves making the sign for SEE (a V shape by the eye and moving it in the direction of what the signer wants the observer to notice). This modifies the task when administered to deaf children, as the signed directions essentially take on a similar function to a pointing gesture used by hearing individuals. Therefore, the directional element of the sign was not included in early presses.
Demonstration Task	1. Elements of directions were finger spelled rather than using signs that were iconic in nature, and/or 2. An alternative task whose directions involved less iconic sign was used.	Signed directions for demonstration task can be iconic in nature. For example, to sign *"brush your teeth,"* one must make movements indicative of brushing teeth, that a hearing child would be expected to spontaneously produce, rather than imitate.
Functional Symbolic Imitation	1. Modifications incorporating appropriate ASL classifiers to maintain the task's intention (e.g., PLANE + 5 hand shape in a forward, flying motion) were used. The object then was used to mimic the motion of the presented signed directions.	This modification was made to ensure that focus was placed on labeling the object and identifying its movement (rather than confusing movement and object's sign).

Abbreviation: ASL, American Sign Language.

Table 2 Scoring Modifications

Overall Level of Non-Echoed Spoken Language	Scored as complexity of signed phrases (e.g., 3 or more signs per utterance would count as phrase speech) and equivalent of grammar in ASL (hand shape, position, movement), use of facial grammatical markings for adjectives/questions, plurals, tense (e.g., FINISH).
Frequency of Spontaneous Vocalization Directed Toward Others	Interpreted as frequency of spontaneous signing *or* vocalizations (if any) directed toward others.
Intonation of Speech	Interpreted with regard to ASL fluency, attending to difficulties with rate (e.g., signing too quickly or in a choppy manner), precision of signs, mixed hand dominance with signs, errors of location or movement.
Echolalia	Echolalia for signed language was scored when there was an exact duplication/repetition of the examiner's (or parent's) sign.
Gestures	Pointing was considered a conventional gesture; descriptive gestures were scored if ASL classifiers or other hand shapes/movements were used in a descriptive way (e.g., to describe the shape of a mirror) ; formal signs were not counted as gestures (e.g., BLUE, DOG) even when there was a descriptive quality about them (e.g., generic sign BIG would not be considered a descriptive gesture necessarily, but using other classifiers to more precisely indicate the size of an object may be counted as descriptive gesture use).
Facial Expressions Directed Toward Others	Emotional facial expressions were scored based only on communication of affect rather than ASL linguistic features (e.g., furrowing brow when asking a question, use of facial features to denote topic identification, etc. were not scored as affective expression unless also paired with emotion signs such as PUZZLED + puzzled expression).
Spontaneous Initiation of Joint Attention	Followed general scoring procedures but also took into consideration cultural norms for initiating joint attention such as signing within the examiner/parent's line of sight between the parent and object of interest. Signing within parent's visual space without eye contact was scored as 1.
Intonation of Vocalizations/ Verbalizations	Scored as 9 with notations made to assist in interpretation regarding ASL fluency attending to difficulties with rate, position/location of signs, mixed hand dominance.
Idiosyncratic/Stereotyped Language	Scored for frequency of the occurrence of any of the following: scripted/overly formal language, palm rotation errors,[18] persistent use of a gesture when a known sign was within the examinee's repertoire or exposure (based on parent/teacher report), unusual pronoun use.

Abbreviation: ASL, American Sign Language.

Additional data collected as part of the parent project (e.g., Social Communication Questionnaire [SCQ], TONI-4) were collected by the second author, the primary investigator of the parent project, or his research assistants via a site visit (in the case of the TONI-4). The SCQ was accessed online, provided as a hard paper copy, or discussed in a Skype (videophone) interview conducted in ASL. A deidentified data set containing data administered as part of the parent project (e.g., ADOS-2, SCQ, TONI-4) was provided to the first author for data analysis.

Because ASD assessments were not standardized on children who are D/HH, qualitative and quantitative data from the assessments were used in conjunction with *Diagnostic and Statistical Manual of Mental Disorders*, Fourth

Edition criteria.[27] The two authors arrived at a consensus opinion regarding diagnosis based on the available data. In the case of two children who did not appear to meet criteria based on the ADOS-2 in the opinion of the first author, the two authors and an additional reviewer, trained in deafness and ASD, arrived at consensus opinion about an ASD diagnosis based on a review of all available information including previous evaluation reports.

Data analysis was completed to address the following questions:

1. Does differential module selection (e.g., standardized versus clinical) result in differential classification (autism versus autism spectrum versus nonautism)?
2. How does administering the ADOS-2 to children whose native language is ASL impact scoring?
3. How does administering the ADOS-2 to ASL users impact algorithm use?
4. What symptoms of ASD are demonstrated on the ADOS-2 when administered to children who are deaf?

RESULTS

Differential Classification across Modules

As described above, the ADOS-2 provides classifications based on cutoff criteria for the overall algorithm scores (e.g., autism, autism spectrum, non-ASD). Of the eight subjects included in this study, a "standardized" versus "clinical" interpretation of the child's language level resulted in the selection of more than one module for five of the eight subjects (see Table 3). The three other subjects completed only module 1. For hearing children, clinicians must choose between two different module 1 scoring algorithms: "few to no words" for children with fewer than five words and "some words" for children with more than five words. Thus, all children were scored under the "few to no words" if they exclusively used sign language and less than five spoken words. The algorithm "some words" was also scored as a "clinical" administration if the child used more than five signs. Each of the three subjects administered a module 1 used more than five signs; they were therefore scored using both the module 1 algorithm for children with "few to no words" as well as the algorithm for children with "some words." Thus, every subject in the entire study was scored on a different algorithm, if not a different module, when clinical rather than standardized procedures were followed for module selection and scoring.

Notably, when *standardized* module selection and scoring procedures were followed, only one child was classified as scoring within the "autism" range and one additional child scored within the "autism spectrum range." The remaining six children did not meet cutoff criteria for ASD. By contrast, when *clinical* module

Table 3 Clinical versus Standard Module Selection

Sex	Age	Standard Module	Clinical Module	Standard Module Classification	Clinical Module Classification	Discrepancy
M	5;1	1	2	Non-ASD	Autism spectrum	Yes
M	5;3	1—few words	1—some words	Non-ASD	Non-ASD	No
F	7;1	1—few words	1—some words	Autism	Autism	No
M	8;5	1	2	Non-ASD	Autism	Yes
M	9;0	1	3	Non ASD	Non-ASD	No
M	11;0	1	3	Non ASD	Autism spectrum	Yes
F	11;1	1	3	Non ASD	Autism spectrum	Yes
M	12;7	1—few words	1—some words	Autism spectrum	Autism	Yes

Abbreviation: ASD, autism spectrum disorder.

selection and scoring was utilized, three children met criteria for autism and three additional children met cutoff criteria for an ASD.

Two children (25%) did not meet an ASD classification under either administration/scoring technique. This is striking given that all of the children included in the sample were previously diagnosed with an ASD. For the purposes of the parent project, a psychologist who was knowledgeable in deafness, a native signer of ASL, and trained on the ADOS-2 reviewed cumulative data of the subjects in the study who did not meet cutoff for an ASD based on the ADOS-2 administration. Information available to the blind reviewer included the child's SCQ, nonverbal IQ and language scores, and previous reports documenting the ASD diagnosis, which were provided by the families in addition to videotapes of the ADOS-2 administration. The independent reviewer indicated that in each of the two cases included in this study that did not meet cutoff for an ASD classification, cumulative data were supportive of an ASD classification.

Usefulness of Item Scores with Children Who Are Deaf

Analyzing ratings on individual items in the ADOS-2 provides information about the challenges of particular scoring codes when administered to children who are D/HH. As indicated above and outlined in Table 2, all codes were scored based on standardized procedures, with some codes modified for "clinical" administration to reflect a clinical interpretation of the intention of the code, translated for application with ASL users. Individual codes that resulted in inconsistencies between a standardized versus clinical administration were primarily the Language and Communication (A) codes, with the exception of Pointing, Use of Another's Body, and Gesture Use. Pointing and Use of Another's Body did not need to be modified for clinical versus standardized interpretation. Although Gesture Use was interpreted differently in light of standardized versus clinical administration, only one case resulted in a scoring difference. This finding may be because all children who communicated using sign language used signs that could be considered descriptive gestures under standardized procedures. Likewise, with the exception of one case, all subjects used gestures in conjunction with formal ASL signs.

Items that were unscorable when applied to children who were deaf in a standardized manner in this sample included: Frequency of Spontaneous Vocalization Directed to Others (four of eight); Immediate (Vocal) Echolalia (eight of eight); Stereotyped/Idiosyncratic Use of Words/Phrases (eight of eight); Speech Abnormalities Associated with Autism (eight of eight); (Verbal) Conversation (eight of eight), Response to Name (eight of eight). Items that were unscorable even when presented in a clinical manner included: Intonation of Vocalizations or Verbalizations (seven of eight), Response to Name (four of eight), Amount of Social Overtures: Parent (three of eight). This last item was due to the fact that in some cases parents were not present during the ADOS-2 administration.

Algorithm Level Interpretation

Comparing overall algorithms scored in a standardized versus clinical manner yielded interesting results. Specifically, just a few item scores were responsible for the majority of differences in classification and scores on algorithms. For example, in the case of three children who received module 1 and were scored on the algorithm for "few to no words" under a standardized administration but also received a module 3 under clinical administration, differences in scoring algorithms were largely accounted for only by differential scores on Pointing, Response to Joint Attention, and Stereotyped/Idiosyncratic Language. Notably, when comparing children who were administered a module 1 as well as a module 3, none of the three children demonstrated Idiosyncratic Language on module 1 when administered in a standardized manner, because strict interpretation of this item (i.e., referring to spoken language only) rendered this item unscorable. However, two of the three children demonstrated unusual/idiosyncratic language on module 3 when scored in a clinical manner. These were the two children who scored within the ASD classification range on module 3, but not

on module 1. Thus, scoring Stereotyped/Idiosyncratic Use of Words clinically contributed meaningful information in discriminating an ASD among this population.

When administered in a standardized fashion, 2 of the 16 items on the module 1 "few to no words" algorithm were not interpretable in a standardized administration and ultimately had no contribution to the final score. Likewise, 2 of the 16 items on the "some words" algorithm were not interpretable. For module 2, for a child over the age of 5, 2 of 14 items on the algorithm are not interpretable when administered in a standardized manner. Three of 14 algorithm items on module 3 were not interpretable under a standardized administration.

Descriptive Data of Symptoms

The ADOS-2 was able to detect some of the behavioral symptoms of autism in the children who are D/HH in this sample, even under a standardized interpretation. The following items yielded scores greater than 0: Use of Another's Body (three of eight); Pointing (six of eight); Unusual Eye Contact (six of eight); Facial Expressions Directed to Others (three of eight); Language Production and Linked Nonverbal Communication (five of eight); Giving (seven of eight) and Showing (four of eight); Quality of Social Overtures (seven of eight); Unusual Sensory Interests (two of eight); Hand and Finger and Other Complex Mannerisms (two of eight); and Unusually Repetitive Interests/Stereotyped Behaviors (three of eight).

In addition to the symptoms above, a nonstandard, clinical administration and interpretation of the ADOS-2 yielded additional information about language/communication autism symptoms in this small sample. Specifically, five of eight children showed sign echolalia, four of eight children demonstrated idiosyncratic sign language, and three of eight children showed "speech" (i.e., sign language) abnormalities associated with autism. These features, which are described in more detail below, would be missed by a strict standardized administration of the ADOS-2.

Symptoms of echolalia entailed repeating signs used by the examiner immediately after the examiner presented the signs. Idiosyncratic language use included scripted/overly formal language, palm rotation errors,[20] persistent use of a gesture or finger spelling when a known sign was within the examinee's repertoire or exposure (e.g., signing DOG by gesturing to describe dogs' ears), or repeating a particular phrase over and over. In one case, insertion of nonsensical signing (e.g., finger spelling nonwords) was observed. Although several subjects used the examiner's hands to manipulate objects, one subject also manipulated the examiner's hands into precisely formed hand shapes (e.g., requiring that the examiner's thumb and middle finger form an exactly round hole when forming the letter D).

Several children in this sample (five of eight) demonstrated markedly reduced giving (e.g., score of 2) or limited giving (two of eight). Likewise, several children demonstrated reduced showing (four of eight scoring 1 or greater). Furthermore, six of the eight subjects demonstrated reduced pointing for drawing another's attention to an object or requesting. A reasonable hypothesis is that children who are deaf and sign fluently rely less on these earlier preverbal communication skills for communicating once they have acquired more formal language. Therefore, administration of a module that contains these developmentally easier items may inadvertently penalize children who are deaf.

Analysis of item-level data also revealed that only one child did not demonstrate suitable descriptive gestures resulting in a score of "0" on the item, even when gestures were interpreted clinically (e.g., subjects must demonstrate descriptive gestures above and beyond formal signs). The finding that the majority of subjects in this study utilized gestures effectively, when gesture use is often reduced among hearing children with ASD, suggests that use of a visual-spatial language such as ASL could help attenuate a gesture deficit through consistent, lifelong, repeated practice with gesture use.

DISCUSSION

Our results show that a child who is deaf who has significant, obvious symptoms of ASD and

limited sign language is likely to exceed an ASD cutoff on any ADOS-2 module, whether interpreted in a standardized or clinical fashion. Module selection and interpretation, although important, is less likely to result in a different ADOS-2 classification for these children. It is important to note, however, that this was the case for only two of the eight subjects we describe, suggesting that assessment of ASD in children who are D/HH is often not a clear-cut case and requires clinical judgment and training in both deafness and ASD to maximize the usefulness of the ADOS-2. The results also suggest that discrepancies across module 1 and 2 are less apparent (i.e., less of a difference in scoring between the two administrations), likely due to overlapping tasks administered and greater number of overlapping items on the algorithms. However, the social/communication skills of fluent signers (i.e., those who received a module 3 per clinical procedures but a module 1 per standardized administration) are most likely to be misrepresented by administration of the ADOS-2 according to standard protocol. Therefore, fluent signers are most likely to be misdiagnosed if the ADOS-2 is used according to its authors' standard administration protocol. Our results suggest that standardized administration and interpretation of the ADOS-2 with these fluent signers may underidentify ASD in children who are D/HH and whose native language is ASL. Comparison of differential scoring obtained from clinical administration and scoring of modules 1 versus 3 suggests that a potential danger of administering a module 1 (per standardized instructions) to a fluent ASL user is the risk of administering tasks that may be too developmentally easy or that may not elicit enough language for the child to demonstrate symptoms of ASD (e.g., idiosyncratic language).

The ADOS-2 manual clearly indicates that the validation sample did not include children who are D/HH and cautions against use of standardized algorithms with children with sensory impairments.[15(p.207)] It is important to highlight that our results confirm that use of standardized ADOS-2 algorithms is inappropriate with this population. However, administration of the ADOS-2 when used in a clinical manner may yield useful information that can assist with differential diagnosis. Current results suggest that many of the core symptoms of ASD can be identified among children who are deaf native ASL users via administration of the ADOS-2. However, much of this clinical information would be missed if communication items (e.g., A codes) were entirely dismissed. Many items are unscorable when administered in a standardized rather than a clinical manner, which supports the idea that the ADOS-2 may yield more clinically useful information when modifications in scoring are made. To use this information effectively across providers, it will be necessary to standardize modifications for consistent administration and interpretation of results.

The challenges of administering an assessment to child who is D/HH is akin to assessing other individuals representing a cultural and linguistic background that differs from the test's standardization sample. *The Standards for Educational and Psychological Testing* developed jointly by the American Educational Research Association, the American Psychological Association, and the National Council on Measurement in Education, provide best practice guidelines for assessing individuals from diverse cultural/linguistic backgrounds, which are highly applicable[21] and discussed in more detail in the summary article included in this special edition (see Szarkowski et al, in this issue).[11] The ADOS-2 manual clearly indicates that even when strict standardized administration procedures are followed, evaluation of ASD requires evaluation of multiple functioning domains from multiple sources and should never be used in isolation.[15(p.6)] Notably, two of the children in the sample did not meet ASD criteria based on the ADOS-2 administration but a consensus diagnosis of ASD was arrived at based on review of all available information, supporting the need for careful review of multiple sources of information rather than relying on the ADOS-2 alone.

Although this study provided data-driven descriptions regarding the use of the ADOS-2 with children who are deaf and communicate using ASL, we cannot address additional questions that point to the reliability and validity of the ADOS-2 when applied to this population.

It is too simplistic an approach to adapt the current ADOS-2 for children who are deaf merely by equating sign language complexity as equivalent to spoken language. Research is needed to clarify how specific tasks (such as Response to Name) and scoring items (such as Intonation and Gesture Use) should be adapted for signing children, given the visual-gestural modality of sign and the grammatical features of ASL. To give just one example, facial expressions in ASL can serve grammatical purposes (such as marking questions or negation) as well as affective purposes. It is thus an open question whether a distinction should be made formally in scoring the ADOS-2 for signing children between these two different types of facial expressions. A further complication is represented by the iconic nature of some signs (i.e., the fact that some signs look like their referents, such as the ASL sign for TREE in which the forearm represents the tree trunk and the fingers its branches). If iconic signs are used in the administration of the ADOS-2 to signing children, then this shifts the task demands. For example, the Demonstration Task requires examiners to ask children to show them how they brush their teeth using gesture and pantomime, but in ASL it is impossible to sign BRUSH-TEETH without enacting the gestural mimicry to be elicited. In contrast with the entirely verbal nature of the task directions for hearing children, children who are deaf and rely on sign would be inadvertently presented with a model of the task, therefore shifting the task demands partially to motor imitation rather than spontaneous gesture use. The ADOS-2 also contains items that are inappropriate or at best complicated with children who are deaf due to the impact of hearing loss on access to the skill assessed (e.g., Response to Name) or because materials may not be as user-friendly for deaf children who are not as drawn to musical toys, or for whom using figurines in play while signing is a more complex task than use of the same toys for hearing children.

Simply translating the ADOS-2 to ASL also fails to capture unique features of ASL or the deaf cultural experience that are not addressed in current scoring approaches, such as giving credit for perspective-taking demonstrated in ASL storytelling features in which one may need to assume the role of a character,[22] or taking into consideration typical methods of establishing joint attention between children who are deaf and their parents, which may look different than joint attention in hearing dyads.[23,24]

Finally, the number of communication A items that are unscorable under a standardized administration suggests that these item codes and the ADOS-2 algorithm are, at best, complicated when used with children who are deaf. These codes could be particularly difficult to interpret if the ADOS-2 is administered to a child who is deaf via an interpreter by an examiner unfamiliar with the nuances of ASL. Our results suggest that the ADOS-2 should be used with extreme caution in such cases. Further research is necessary to examine the impact of administering the ADOS-2 via an interpreter as well as to investigate the importance of an appropriate communication match between the examiner and child/adolescent.

LIMITATIONS

This study provides a somewhat limited view of the use of the ADOS-2 with children who are deaf because it only captures the relatively substantially smaller subset of the overall D/HH population that communicates using sign language and have parents who are also deaf (5 to 10%).[25] Thus, the results may not be easily translatable across all members of the D/HH community.

Procedures in this study are likely also unique because the ADOS-2 was administered directly by a professional trained in both hearing loss and ASD, rather than via an interpreter or by a professional trained in one but not both special populations. A dually trained examiner is unlikely to be available in many areas of the country. Research suggests that there is considerable diagnostic variability when attempting to diagnose ASD among children who are D/HH.[26] This may be due in part to the variability in agreement among professionals who are trained in autism but not hearing loss, compared with the greater diagnostic agreement among professionals who are trained

in hearing loss.[13] It also seems reasonable to hypothesize that variability in diagnostic agreement likely stems from the use of tools for diagnosing ASD that were not developed for children who are D/HH and for whom no clinical best practice agreement currently exists. Current results, which indicate that several communication items are not scorable when administered in a standardized manner, also suggest that these same items are likely problematic when administered and interpreted via an interpreter without the ability of the examiner to recognize unusual language features in ASL or the training of the interpreter to recognize and convey these features.

Because this study represented secondary analysis of data obtained from the parent project's protocol, certain methodological issues may have impacted results. Specifically, it was not possible to control for order effects of module administration. Most importantly, best practice to inform clinical opinion may have been for the evaluator to complete the ADOS-2, in addition to gathering additional data sources such as a comprehensive developmental interview assessing for symptoms of ASD (i.e., completion of the ADI-R), as well as record review. In the case of children who are D/HH, although previous evaluation reports may document symptoms that are consistent with ASD, evaluations conducted by professionals who lack specific training in deafness and ASD may contain inherent misinterpretations of behavior and thus misrepresent clinical symptoms.

An obvious limitation of this research is its small sample size. However, this represents the largest descriptive data set of the use of the ADOS-2 with children who are deaf to date. It is especially important to note that all children described in this study were exposed natively to ASL, and thus represent ideal test subjects for understanding the manifestation of ASD in signing children. A major limitation of this study was that interpretation of findings was based on one clinician's administration of the ADOS-2 and interpretation of data. Greater professional consensus is necessary to determine clinical best practice in utilizing the ADOS-2 with children who are D/HH to assess for a possible ASD.

FUTURE DIRECTIONS

This article highlights the need for additional research to be conducted to formulate a "best clinical estimate" approach for diagnosing ASD among children who are D/HH. The summary article included within this journal issue outlines combined clinical opinion among professionals trained in both deafness and ASD regarding possible red flags of ASD and current best practices in assessing ASD within this population. This article may serve as the foundation for further research. Additional research with broader samples of children who are D/HH with a variety of communication preferences is necessary to determine which ADOS-2 tasks/scores are most useful in distinguishing ASD and to further clarify algorithm use. This line of research may also be helpful in further describing what aspects of the phenotype in children who are D/HH are similar or different to idiopathic autism as described in the current literature.

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