Critical Review:

Do Deaf children with Autism Spectrum Disorder differ in their use or comprehension of signed communication from those without ASD?

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ABSTRACT

Background: Suprasegmental elements of signed languages are conveyed through body movement and facial expression (von Agris, Knorr, & Kraiss, 2008). Children with Autism Spectrum Disorder (ASD) are known to have difficulties with social communication that include, but are not limited to, eye contact, facial expression, and gestures. It would therefore be expected that children with ASD who learn a signed language would have difficulties with the nonverbal communicative behaviours that serve a suprasegmental purpose.

Purpose: This critical review explored the differences between Deaf native signers with and without ASD in their use and understanding of signed communication.

Method: A computerized search of the literature included terms such as "Deaf", "autism", and "sign language" or "ASL", among others. Articles included for review compared Deaf children with ASD to typically developing Deaf children on expressive and receptive aspects of sign language.

Results: Overall, there appears to be a difference in sign language use and understanding by Deaf children with ASD, compared to typically developing (TD) Deaf children. The groups differed in number and types of: formational errors produced, emotion recognition, incidence of echolalia, pronoun use, and praxis errors.

Clinical Implications: The differences between groups appear analogous to deficits in social communication observed in hearing children with ASD. Speech-Language Pathologists need to be aware of these differences when planning assessment and intervention. More research is needed in this population, particularly in regards to diagnosis and intervention

Introduction

In signed languages¹, there is more to the language than the individual signs themselves. Suprasegmental elements of the language are conveyed through body movement and facial expression (von Agris, Knorr, & Kraiss, 2008). A phrase becomes a question with an upward inflection of the eyebrows; peoples' emotion in a story is communicated through facial expression. Spatial information is conveyed through the signer's position and use of their physical space. Finally, some signs may be differentiated by palm orientation, as in

the case of TUESDAY and TOILET/BATHROOM in American Sign Language (ASL).

Communication deficits associated with Autism Spectrum Disorder (ASD) make it a population that often seeks out the expertise of Speech-Language Pathologists. The DSM-5 (American Psychiatric Association, 2013) lists the diagnostic criteria for ASD as a deficit in social communication that includes, but is not limited to: "Deficits in nonverbal communicative behaviors used for social interaction, ranging, for example, from poorly integrated verbal and nonverbal communication; to abnormalities in eye contact and body language or deficits in understanding and use of gestures; to a total lack of facial expressions and nonverbal communication." (DSM-5, 2013, section 299.00 F84.0). It would thus be expected that children with ASD who learn a signed language would have

¹ The use of "signed language" is to account for the fact that some papers included in this study explore the use or understanding of American Sign Language (ASL), while others discuss British Sign Language (BSL).

difficulties with the nonverbal communicative behaviours that serve a suprasegmental purpose.

Objectives

The purpose of this critical review was to explore the differences between Deaf² native signers with and without ASD in their use and understanding of signed communication.

Methods

Search Strategy

Articles were found by an online search of the computerized databases PubMed and JSTOR. Keywords used were as follows:

[(deaf) (autism) (ASL)]

[(deaf) (autism) (sign language)]

[(deaf) (autism) (facial expression)]

The computerized database CINAHL was searched using the MeSH terms: [Autistic Disorder AND Deafness] A fourth search of Google Scholar using [("deaf" OR "HH" OR "HI") AND ("autism" OR "ASD") AND ("facial expression" OR "facial cues") AND ("signed language" OR "signing" OR "manual language")] yielded no results.

Selection Criteria

Articles selected for inclusion compared Deaf children with ASD as the experimental group and typically developing (TD) Deaf children as a control group. Duplicate studies were discarded.

Data Collection

Results of the literature search yielded six articles that met the selection criteria. Five of the articles (Bhat et al, 2018; Denmark et al, 2014; Denmark et al 2018; Shield et al, 2015; and Shield, Cooley & Meier, 2017) included Level 2b research evidence studies: between groups designs. The sixth article (Shield & Meier, 2012) consisted of a pilot study that included Level 4 research evidence (a series of observational case studies), as well as a Level 2b between groups design. Four papers

included Aaron Shield as one of the authors (Bhat et al, 2018; Shield & Meier, 2012; Shield et al, 2015; and Shield, Cooley & Meier, 2017), and the other two studies were completed by the same research team (Denmark et al 2014; Denmark et al 2017).

Results

Appropriate statistical analyses by the papers included in this review revealed that — with the exception of Denmark, Atkinson, Campbell, and Swettenham (2018) — there appears to be a difference in the way Deaf children with ASD use and understand signed communication, as compared to TD Deaf children.

In all studies, the children's ASD diagnosis was confirmed using formal (professional assessment, often Diagnostic Interview for Social Communication Disorders, Autism Diagnostic Observation Schedule-2, Social Responsiveness Scale, Social Communication Questionnaire) and informal methods (investigator observation). Not all measures were used in all studies, but all studies used at least two methods of confirmation, with the exception of one study: Shield, Cooley, & Meier (2017) confirmed the ASD diagnosis of 16/17 children using only the ADOS-2.

All authors remark that there are no standardised tests for diagnosing Autism in Deaf children. They use the "gold standards" of their countries, but note the limitations of test items such as "responds to name being called", which relies on hearing.

Observational Study

Shield &Meier (2012)

Shield & Meier's 2012 article marked an early instance of a study of Deaf children with ASD. The importance is compelling: the authors found in previous research that gesture imitation by hearing children with ASD often contained reversal errors. It was the authors' intention to explore whether this reversal would be found in children whose first language is a signed language.

As one of the first studies to look at Deaf children with ASD, Shield & Meier (2012) included a preliminary study of three native-signing male children with ASD (age 4;6 to 7;5). This study was conducted by videotaping the children in naturalistic settings for 20 to 60 minutes, and coding the number of formational errors (location in space, hand shape, movement, and palm reversal errors), as a percentage of overall number of signs produced. The three participants produced formational errors on 72.3%, 34.1%, and

² The use of "Deaf" is to signify the culturally Deaf population, whose primary language tends to be a signed language. It is differentiated from "deaf" which is a designation related to level of hearing loss. Deaf people may be hearing children of Deaf parents, as in the case of some of the participants (i.e. Shield & Meier, 2012)

59.3% of their signs, respectively. Palm orientation made up an average of 48.3% of all errors, and of that percentage, 78% related to inward-outward palm reversal errors.

Because there was no control group in this study, the validity of the evidence is merely suggestive.

Between-Groups Studies

Shield & Meier (2012, continued)

To further explore their question, Shield & Meier (2012) conducted a second study, reported in the same article, of five signing male children with ASD (age 5;8 to 7;5), and included twelve TD Deaf children (age 3;7 to 6;9) as a control group. It should be noted that the groups were *not* matched for chronological age or language age, and that these differences were significant. This brings into question the validity of the comparison.

This study was conducted by presenting ten words written in English orthography on index cards, and asking the children to fingerspell them. The cards were presented in random order. Because there may be a difference in performance between children who can read English and those who cannot, the validity of this method is equivocal.

The authors report that 3/4 the Deaf children with ASD who responded (a fifth child who did not respond to stimuli was excluded from discussion) produced palm reversal errors, while no TD Deaf child did. This evidence would appear to be suggestive of difference, but one should not draw firm conclusions, given that the validity of the method was questionable.

Denmark, Atkinson, Campbell, & Swettenham (2014)

Denmark and colleagues (2014) examined how the face is used by Deaf children to understand emotion in signed language. The authors outlined two main questions of interest: do TD Deaf children use the face in ways similar to Deaf adults? And: do Deaf children with ASD show impaired emotion recognition in language processing analogous to that of their hearing counterparts?

Denmark and colleagues (2014) studied 12 TD Deaf children and 13 Deaf children with ASD, matched for chronological age, nonverbal IQ, and BSL receptive and productive language skills. Groups had similar levels of technology use (cochlear implants [CI], hearing aids [HA], or unaided). There were no significant differences reported between groups.

Denmark and colleagues based much of their study on a previous article by Reilly et al (1992), which used "masked" (hands only) and "unmasked" (hands and face visible) conditions, designed to measure comprehension of facial emotions. In the current study, one adult signer was videotaped in all conditions, for consistency. The children were presented with one of eight neutral sentences, produced using one of eight emotional conditions: ("surprise" "happy", "sad", "angry", "neutral", "disgust", "annoyance", and "mischief"). Each emotion was paired with 3 sentences (24 items), and presented in semi-random order in unmasked and masked conditions, for a total of 48 test items per child (masking type and emotion items were semi-randomised so that they would not appear more than twice consecutively).

The results of the study showed that TD Deaf children were better able to recognise emotion in both conditions than Deaf children with ASD. Masking had a greater effect on the TD deaf group, although both groups' performance was significantly impacted. "Our findings suggest that deaf children with ASD are less accurate in their judgements of emotion compared with TD deaf children." (p. 7). This study is of suggestive importance, and highly suggestive validity.

Denmark, Atkinson, Campbell & Swettenham (2018)

In a study of 12 TD Deaf children and 13 Deaf children with ASD, Denmark and colleagues (2018) examined the use of the face for emotional expression in a BSL narrative retelling. Groups were matched for chronological age, non-verbal intellectual ability, and BSL comprehension. Two raters coded the children's use of sign language, in reference to two Deaf adult native signers' model retellings.

The results showed no significant difference between the groups of Deaf children in terms of story comprehension, length of retelling (although the group with ASD was shorter), and number of facial action targets. The TD group produced more identical expressions to the adult models than the group with ASD. Significant differences were found in the number of productions of "demand" and "mischief", but not in other emotions. No differences were found between groups for narrative content, narrative structure, or grammar.

This study presents equivocal evidence for a difference in use of facial emotional expression during signed communication between Deaf children with ASD and TD Deaf children.

Shield, Meier, & Tager-Flusberg (2015)

Subsequent to their initial study, Shield and Meier teamed up with Tager-Flusberg to examine how use of pronouns differed between native signing children with ASD as compared to TD Deaf children. 15 Deaf children with ASD and 18 TD Deaf children were recruited for

this study. The groups were matched for chronological age and non-verbal intelligence. The groups were not matched for language on the basis that language and communication are known deficit in children with ASD. The TD group scored significantly higher on a sign language comprehension test than the ASD group.

A picture identification task was modified to be conducted in ASL, whereby the participant would see a picture of themselves or of the experimenter and asked THAT+WHO. Informal observation of pronoun use during ADOS-2 evaluation of both groups was also coded. Finally, the parents of children with ASD were given a questionnaire to determine if the pronouns elicited were typical for their child.

A significant between-groups difference was found in use of pronouns (vs. signed names). There was a significant positive correlation between pronoun production and ASL comprehension, as well as a moderate correlation with nonverbal intelligence. No significant correlation with chronological age was found.

Because language level was not matched between groups, there is a possibility that that, independent of ASD diagnosis, may have contributed to the use of a name rather than pronoun. As such, the validity of this study is suggestive. Overall, there was a difference in pronoun use by TD Deaf children and Deaf children with ASD in a picture identification task.

Shield, Cooley, and Meier (2017)

One phenomenon that is often observed in verbal children with ASD is echolalia, or the repetition - in whole or in part - of utterances other people have produced. Shield, Cooley, and Meier (2017) examined the presence of echolalia in the sign language production of Deaf children with and without ASD. 17 Deaf children with ASD and 18 TD Deaf children were videotaped in assessment and naturalistic settings, and instances of echoes were noted. The two groups did not differ significantly in chronological or mental age. However, on average, TD Deaf children had higher standard language scores than Deaf children with ASD. TD Deaf children did not echo any signs, whereas 7/17 (41%) of Deaf children with ASD produced echoes about one quarter of the time. Those children who produced echoes had lower receptive language skills than non-echoic children, and age-matched TD Deaf children. They also tended to be younger than nonechoic children.

The authors claim that this study confirms that echolalia is a feature of language of children with ASD, regardless of modality. While the results of this study may support previous research, the limited sample size

indicates highly suggestive – rather than compelling – evidence.

Bhat, Srinivasan, Woxholdt, & Shield (2018)

Bhat and colleagues (2018) investigated praxis performance and sign comprehension of Deaf children with and without ASD. 11 TD Deaf children and 11 Deaf children with ASD were recruited for this study. The groups were comparable on chronological age and nonverbal intelligence. Similar to Shield & Meier (2012), the authors videotaped the participants fingerspelling 15 words presented with English orthography, and coded movement errors.

Deaf children with ASD had significantly lower receptive skills, and a greater number of total praxis errors than TD Deaf children. The ASD group was also found to be slower fingerspellers. There was a significant negative correlation between praxis errors and receptive language scores for the group of Deaf children with ASD only. This evidence appears to be highly suggestive of a difference between TD Deaf children and Deaf children with ASD in their use of ASL. The authors noted that one of the limitations of their study was that they attempted to control for overall language exposure by including children whose native language was ASL. However, they could not control for fingerspelling experience.

Discussion

Across all the studies, a trend has begun to emerge: there appears to be a difference in the way Deaf children with ASD use and understand signed languages, as compared to TD Deaf children. In particular, many of the hallmarks of Autism – echolalia, gestural deficits, and difficulties with eye contact – appear to have analogous impairments in a manual language modality.

It would seem that Deaf children with ASD differ from TD Deaf children in their exhibition of characteristics of echolalia. It is unclear, however, if the echoic signs are produced with or without comprehension (see Roberts, 2014, for a study comparing echolalia in children with ASD to children with Specific Language Impairment). There also appeared to be a difference between the aforementioned groups in the incidence of palm reversal errors. A mentioned earlier, palm reversal can play an important role in distinguishing signs. Shield and colleagues (2012) report that there is little or no documentation of palm reversal errors in TD Deaf sign language acquisition beyond two years of age. Shield & Meier (2012) indicated that there may be a delay in

outgrowing of this type of error for Deaf children with ASD.

In the studies that reported a gender breakdown of participants (Bhat et al, 2018; Shield & Meier, 2012; Shield et al, 2015; and Shield, Cooley & Meier, 2017), there is a higher number of males in the ASD group than the TD Deaf group. This is to be expected, as: "Autism spectrum disorder is diagnosed four times more often in males than in females." (DSM-5, 2013, section 299.00 F84.0). The control groups had a more evenly divided sample. While this would appear representative, some caution in interpretation is recommended with regards to the use and comprehension of signed communication of female Deaf children with ASD, as there may be differences not accounted for in the current studies.

Limitations and Future Research

One limitation that was remarked upon in five of the studies included in this review (Bhat et al, 2018; Denmark et al, 2014; Denmark et al 2018; Shield et al, 2015; and Shield, Cooley & Meier, 2017) is the lack of a gold standard method of diagnosing ASD in the Deaf population. The authors note that the existing tools are designed for use with hearing children with ASD, and had to be modified to be used with the population under investigation. This highlights the need for future research to develop a sensitive and specific instrument for detecting ASD in Deaf children.

A second limitation that was noted is the conspicuous lack of tested interventions for this population. As Speech-Language Pathologists will likely have a role in supporting communication development in Deaf children with ASD, future research identifying interventions that are effective for this population are required.

Clinical Implications

It is common practice to use sign language to supplement the language repertoire of nonverbal children with ASD (Bonvillian, Nelson, & Rhyne, 1981). The suggestive evidence brought forth by this review would indicate that this may not be the most efficacious for this population. That is, the differences in sign language use and understanding by Deaf children with ASD appear analogous to the deficits in social communication observed in hearing children with ASD. As such, the use of sign language to complement the expressive and receptive language of children with ASD should be implemented judiciously.

While the validity of the studies included in this review are at best suggestive, overall this evidence indicates that there is likely a difference in the way Deaf children with ASD use and understand signed languages, when compared with TD Deaf children. As Speech-Language Pathologists may work with both ASD and Deaf populations, it is important to be aware of this difference when planning assessment and intervention with this population.

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