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Differences in praxis performance and receptive language during fingerspelling between deaf children with and without autism spectrum disorder

Anjana N Bhat[1,2], Sudha M Srinivasan[3], Colleen Woxholdt[2] and Aaron Shield[4]

Abstract

Children with autism spectrum disorder present with a variety of social communication deficits such as atypicalities in social gaze and verbal and non-verbal communication delays as well as perceptuo-motor deficits like motor incoordination and dyspraxia. In this study, we had the unique opportunity to study praxis performance in deaf children with and without autism spectrum disorder in a fingerspelling context using American Sign Language. A total of 11 deaf children with autism spectrum disorder and 11 typically developing deaf children aged between 5 and 14 years completed a fingerspelling task. Children were asked to fingerspell 15 different words shown on an iPad. We coded various praxis errors and fingerspelling time. The deaf children with autism spectrum disorder had greater errors in pace, sequence precision, accuracy, and body part use and also took longer to fingerspell each word. Additionally, the deaf children with autism spectrum disorder had poor receptive language skills and this strongly correlated with their praxis performance and autism severity. These findings extend the evidence for dyspraxia in hearing children with autism spectrum disorder to deaf children with autism spectrum disorder. Poor sign language production in children with autism spectrum disorder may contribute to their poor gestural learning/comprehension and vice versa. Our findings have therapeutic implications for children with autism spectrum disorder when teaching sign language.

Keywords

autism spectrum disorder, children, deaf, fingerspelling, praxis, receptive communication, sign language

Introduction

Autism spectrum disorder: A multisystem disorder

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by social communication impairments such as poor integration of gesture, gaze, and language within social interactions, poor verbal communication, as well as the presence of repetitive behaviors and interests (American Psychiatric Association (APA), 2013). Although not diagnostic, there is extensive evidence for motor comorbidities in 50%–80% of children with ASD (Ament et al., 2015; Bhat et al., 2011; Fournier et al., 2010; Green et al., 2009; McPhillips et al., 2014). Motor impairments in ASD include basic motor skill deficits in reaching and walking (Glazebrook et al., 2006; Jansiewicz et al., 2006; Mari et al., 2003; Vilensky et al., 1981), gross and fine motor incoordination (Ament et al., 2015; Biscaldi et al., 2014; Green et al., 2009; Provost et al., 2007), as well as deficits in praxis/motor planning (Gizzonio et al.,

[1]University of Delaware, USA
[2]University of Connecticut, USA
[3]Indian Institute of Technology Bombay, India
[4]Miami University, USA

Corresponding authors:
Aaron Shield, Department of Speech Pathology and Audiology, Miami University, 2 Bachelor Hall, Oxford, OH 45056, USA.
Email: shielda@miamioh.edu

Anjana Bhat, Department of Physical Therapy, University of Delaware, 540 S College Ave, Newark, DE 19713, USA.
Email: abhat@udel.edu

2015; Mostofsky et al., 2006; Rogers et al., 1996; Smith and Bryson, 1994, 2007). There is growing evidence for the intimate association between motor skills and language skills in infants with older siblings with ASD as well as in children with ASD (Bedford et al., 2016; Bhat et al., 2011; Gernsbacher et al., 2008; Iverson and Wozniak, 2007; LeBarton and Iverson, 2013; Leonard et al., 2015). Therefore, impairments in motor planning and coordination may contribute to the communication difficulties of children with ASD. This article explores links between praxis and sign language skills in a unique group of deaf children with ASD (hereon termed DASD) within the context of fingerspelling, a component of sign language requiring a high degree of motor planning and coordination.

Praxis: Definition and types

Praxis, the ability to perform complex gestures and action sequences, emerges in the second year of life, rapidly improves with development, and becomes adult-like by 12 years of age (Dewey, 1995). Praxis is typically assessed by performing gestures on imitation, on verbal command, and during tool use (Dewey et al., 2007; Mostofsky et al., 2006). A variety of gesture classifications have been reported within the literature, including gestures with and without the use of objects as well as gestures with and without communicative intent/meaning (Dewey et al., 2007; Mostofsky et al., 2006; Smith and Bryson, 1994, 2007). With regard to object use, gestures may be transitive (i.e. involving the use of objects, for example, brushing with a toothbrush) or intransitive (i.e. involving body parts only, without the use of objects, for example, waving bye; Smith and Bryson, 1994). Similarly, gestures can be meaningless (e.g. placing one's hand on the opposite shoulder) or meaningful with a functional or communicative intent (e.g. linguistically complex signed languages of the Deaf[1] such as American Sign Language (ASL) (Klima and Bellugi, 1979; Smith and Bryson, 1994, 2007; Stokoe, 1965). Sign languages such as ASL rely on manual gesture sequences to convey meaning to interlocutors and therefore present a unique opportunity to assess praxis performance.

Developmental dyspraxia: Evidence in ASD

Developmental dyspraxia, a difficulty in performing complex gestures and action sequences, has been widely reported in hearing children with ASD (Gizzonio et al., 2015; Ham et al., 2011; Mostofsky et al., 2006). Children with ASD between 8 and 13 years had greater praxis errors during performance of gestures on imitation, on verbal command, and during tool use compared to age-matched and Intelligence Quotient (IQ)-matched, typically developing (TD) children (Mostofsky et al., 2006). However, a more recent study found that school-age children with ASD make more praxis errors during imitation of actions with imaginary objects (or pantomime actions) compared to actions performed with tools, suggesting that the functional context of the tool may help improve praxis performance (Ham et al., 2011). This is consistent with other reports that children with ASD make more praxis errors during meaningless gestures compared to transitive and communicative gestures (Smith and Bryson, 1994, 2007). Children with ASD typically have greater spatial errors in mirroring, sequencing, and modulation of movement compared to TD children and children with other developmental delays (Dewey et al., 2007; Ham et al., 2011; Mostofsky et al., 2006; Vanvuchelen et al., 2007). Furthermore, gestural performance of children with ASD based on standardized measures correlates with various other skills such as gesture recognition, receptive language, social interactions, and overall autism severity, although not with intelligence (Dowell et al., 2009; Dziuk et al., 2007; Gizzonio et al., 2015; Ham et al., 2011). Dowell et al. (2009) found a strong association between praxis performance measured on the Florida Apraxia Battery (FAB) and social communication skills as measured by the total scores of the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2012), a common tool for ASD diagnosis. Moreover, gesture recognition significantly correlates with gesture performance in older children with ASD, suggesting that gesture perception (i.e. receptive communication) interacts with gesture production (i.e. expressive communication) and vice versa (Dowell et al., 2009; Ham et al., 2011). Overall, these studies suggest that motor impairments may interact with and possibly contribute to some of the social and receptive communication deficits in hearing children with ASD.

Praxis and sign language in ASD: An unexplored link

There are only two previous studies that have specifically examined the relationship between praxis performance and sign language production in children with ASD (Seal and Bonvillian, 1997; Soorya, 2003). Both studies were conducted in hearing children with ASD. The first study found that sign vocabulary size and accuracy significantly correlated with praxis and fine motor scores in children with severe ASD who were learning to sign (Seal and Bonvillian, 1997). Specifically, children had high rates of handshape (incorrect overall shape), movement (incorrect use of joints), and sequence (addition or merging of movements) errors compared to location errors (Seal and Bonvillian, 1997). In the second study, children with ASD assessed on standardized motor measures had greater praxis errors and poorer motor performance compared to age-matched TD children; moreover, there was a strong correlation between motor/praxis performance and the rate of sign production in children with ASD (Soorya, 2003).

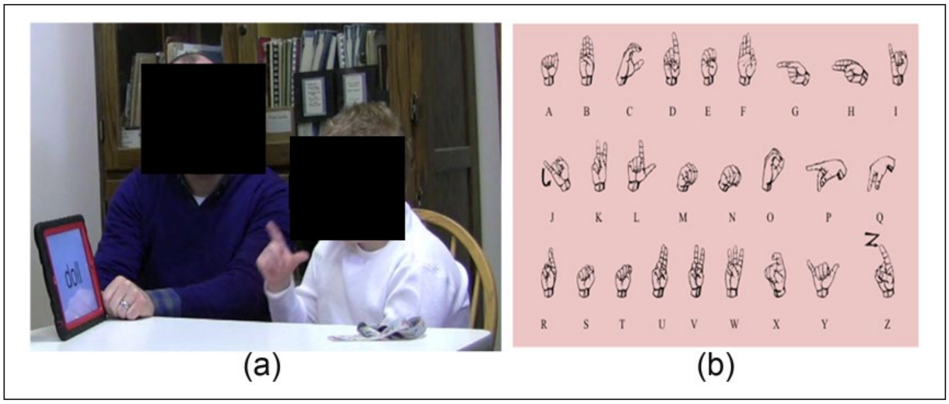

Figure 1. (a) Experimental setup and (b) fingerspelled alphabet of the American Sign Language.

Overall, these studies provide some evidence supporting the influence of praxis performance on sign language acquisition and production in hearing children with ASD.

In spite of considerable research on praxis deficits in hearing children with ASD, there is a dearth of literature evaluating praxis in the DASD population. Moreover, links between praxis performance and sign language comprehension/production have not been examined in this population. ASD is more frequent in the deaf population compared to the hearing population (Szymanski et al., 2012). Although the lack of standardized diagnostic tools specifically designed for deaf children makes ASD diagnosis difficult in this population (Mood and Shield, 2014), current estimates suggest that 1 in every 59 deaf children develops a concurrent ASD diagnosis (Szymanski et al., 2012). DASD acquiring sign language constitute a unique population in which to study the development of praxis and its impact in ASD. Sign language involves a complex series of communicative manual and facial movements. Furthermore, studying deaf children who are exposed to a sign language from birth by their Deaf parents (i.e. native signers) would be informative because such children likely have far more exposure to and practice with gesture imitation than their hearing counterparts.

Fingerspelling: Acquisition and prerequisite skills

The fingerspelling system within ASL could be a particularly rich area to investigate praxis. Fingerspelling refers to a system in which each letter of the English alphabet is represented by a unique manual configuration/handshape (Stokoe, 1978). Figure 1(b) shows the handshapes representing the letters of the Roman alphabet in ASL. Fingerspelling requires high levels of fine motor control for rapid and contrasting movements of individual fingers to form multiple handshapes. In contrast to lexical signs that typically contain only one or two handshapes, a fingerspelled word has as many handshapes as the number of letters in the spelled word, making fingerspelling a highly complex sequence of gestures.

Deaf children acquire a basic understanding of the fingerspelling system by 4 years of age (Padden and LeMaster, 1985). In this process, children need to learn the 26 hand configurations of the manual alphabet, the fixed location where the hand must be held while executing handshapes, as well as the transitional movements to move from one handshape to the next (Padden and LeMaster, 1985). Although the handshapes used in ASL are a representation of the written English alphabet, TD children start to learn fingerspelling prior to the onset of English literacy (Padden, 1991). In fact, deaf children as young as 2 years of age attempt to produce fingerspelled words long before they start learning to read (Erting et al., 2000). Akin to the vocal babbling phase in hearing infants, deaf infants go through a manual babbling phase, wherein they produce meaningless approximations of the target handshapes of the adult ASL lexicon (Petitto et al., 2001; Petitto and Marentette, 1991). Furthermore, similar to hearing infants, deaf infants go through a process of acquiring a phonological inventory, albeit in the manual modality. A longitudinal case study that analyzed articulation of signs over the second year in a deaf child acquiring ASL suggested that handshapes using larger muscle groups, that is, whole hand, forefinger, and thumb, emerged earlier than handshapes involving the middle, ring, and little finger (McIntire, 1977). Over the course of development, deaf children replace easier-to-produce handshapes using larger muscles with more complex handshapes using smaller muscles (McIntire, 1977). Overall, the development of fingerspelling appears to be constrained by the common progression of gross motor and fine motor development. Therefore, it provides a unique context to assess motor functioning, that is, praxis skills in the DASD population.

This study: Need and scope

In spite of the significant motor demands associated with fingerspelling, interestingly, there are no reports of praxis performance during fingerspelling in the DASD group. The only other study that looked at fingerspelling in deaf signing

Table 1. Participant characteristics.

Mean (*SD*)	DASD	Deaf	*t* values	*p* values	SMD	CI (SMD)
Chronological age	10.02 (2.36)	9.81 (1.94)	0.226	0.82	0.09	−0.74 to 0.93
Non-verbal IQ	93.82 (12.91)	98.91 (7.93)	−1.115	0.28	−0.46	−1.30 to 0.39
Receptive ASL	18.73 (11.24)	32.18 (4.35)	−3.70	0.004*	−1.52	−0.57 to −2.47
Total praxis errors	29.73 (3.77)	10.91 (1.56)	4.62	<0.001*	1.89	0.89 to 2.90
Fingerspelling time	2.37 (0.98)	1.23 (0.43)	3.54	0.002*	1.46	0.52 to 2.40
Pace errors	11.09 (2.34)	3.18 (1.34)	2.93	0.008*	3.99	2.55 to 5.44
Accuracy errors	5.36 (1.8)	1.82 (0.38)	1.93	0.08†	2.62	1.48 to 3.76
Sequence precision errors	4.91 (1.25)	1.73 (0.43)	2.40	0.03*	3.27	1.99 to 4.55
Movement modulation errors	5.36 (1.32)	4.09 (0.89)	0.80	0.43	1.09	0.19 to 1.98
Body part errors	3 (1.18)	0.09 (0.09)	2.47	0.03*	3.35	2.05 to 4.64

SD: standard deviation; DASD: deaf children with autism spectrum disorder; SMD: standardized mean difference; CI: confidence interval; IQ: Intelligence Quotient; ASL: American Sign Language.
Non-verbal IQ measured using Test of Nonverbal Intelligence, Fourth Edition (TONI-4), Receptive ASL skills measured using ASL Receptive Skills Test.
*$p < 0.05$; †$p < 0.1$

children with ASD focused on a single articulatory parameter of palm orientation during sign production (Shield and Meier, 2012). Furthermore, this study did not analyze other praxis errors in children nor did the authors examine the relationships between praxis performance, receptive communication, intelligence, and autism severity. Given the previous literature on praxis deficits in hearing children with ASD as well as the significant correlations between praxis performance and measures of gesture recognition, receptive communication, and autism severity, in this study, we aimed to extend these findings to a novel research population (DASD) who are exposed to a sign language from birth by their Deaf parents. The first aim of this study was to compare praxis performance (i.e. sign production during fingerspelling) and receptive communication (i.e. sign recognition as indicated by performance on a standardized comprehension measure, the ASL Receptive Skills Test (ASL RST)) in deaf children with and without ASD. The second aim was to examine the relationship between praxis performance, receptive communication, and non-verbal intellectual ability in deaf children with and without ASD. We hypothesized that children with ASD would exhibit poorer ASL receptive skills as well as more spatio-temporal errors in magnitude, timing, and sequencing during fingerspelling relative to children without ASD. In addition, we hypothesized that praxis performance of children with ASD would correlate with their receptive language skills and autism severity scores but not with their non-verbal IQ.

Method

Participants

We recruited 11 DASD (9 males and 2 females) and 11 deaf children (5 males and 6 females) between 5 and 14 years of age. Of these, 13 children were non-Hispanic Caucasians, 4 were Hispanic, 3 were African-American, 1 was Asian, and 1 was mixed-race. The groups were comparable for chronological age ($t(20)=0.226$, $p=0.82$, *ns*) and non-verbal intelligence ($t(20)=-1.115$, $p=0.28$, *ns*) as assessed using the Test of Nonverbal Intelligence, Fourth Edition (TONI-4; Brown et al., 2010; see Table 1). All children except one were born to at least one Deaf parent, therefore they had been exposed to ASL as their primary language since birth. The exception was a child who had four Deaf grandparents and hearing native-signing parents. None of the children had cochlear implants. Children were recruited through an online video in ASL posted on social media and through schools for the deaf. The last author (A.S.) conducted all research visits in the child's home or school. Children were enrolled in the study following written parental consent. The Institutional Review Board at Boston University prospectively approved the study procedures.

We screened all children for possible ASD using the Social Communication Questionnaire (SCQ)[2] (Rutter et al., 2003). All TD children scored well below the clinical cut-off score of 15 (mean (M)=2.54, standard deviation (SD)=2.38, range: 0–6) and were significantly different than the ASD group ($M=13.45$, $SD=7.71$, range: 4–31) ($t(20)=4.5$, $p<0.001$). To the best of our knowledge, there are currently no instruments available to diagnose ASD in deaf children (Mood and Shield, 2014). Therefore, we confirmed ASD diagnosis in the deaf children using the gold standard, ADOS, Second Edition (ADOS-2; Lord et al., 2012) administered by two ASL-proficient researchers who were research-reliable on the instrument. However, since the ADOS-2 is not primarily designed for deaf children, some of the items of this assessment, such as "Response to name," could not be administered in this sample or were administered with modifications; see a detailed description of all modifications made to the ADOS-2 in Shield et al., 2015. Despite these limitations, 11 of the 12 subjects in the DASD children scored above threshold for ASD classification on the ADOS-2 (ADOS algorithm scores:

Table 2. Error categories for fingerspelling performance.

Error category	Definition
Pace	Slower movements as a result of obvious halts
Accuracy	Errors in spelling words or incorrect orientation of hand during fingerspelling. Commonly seen for words with letters that have similar signs, for example, "a" and "e"
Sequence precision	Errors in the order of the movement sequence including omission of movements or addition of extra movements/letters
Movement modulation	Errors in excursion of joints suggestive of poor control. Movements could be exaggerated or insufficient
Body part	Errors in the use of various body parts/joints/fingers

M=12.73, SD=4.03, range: 4–18; ADOS comparison scores: M=6.09, SD=1.76, range: 2–9). For the one child who scored below the cut-off threshold (ADOS algorithm score=4; ADOS comparison score=2), we relied on the clinical judgment of a native-signing licensed clinical psychologist with training in diagnosing ASD. After reviewing this child's videotaped behavioral data, the clinical psychologist confirmed that the child met clinical criteria for ASD, even with a below-threshold ADOS-2 score. Therefore, we have included this child in our analysis.

Testing protocol

ASL RST. The ASL RST is a reliable test to assess the understanding of ASL grammar in phrases and sentences in children aged between 3 and 13 years (Allen and Enns, 2013; Enns et al., 2013). Prior to administration of this test, a 20-item vocabulary check was conducted to verify that children knew the signs used in the ASL RST. The ASL RST has three practice and 42 test items that were presented on a laptop placed on a table in front of the seated child. For each item, the model signed a sentence in ASL, which was followed by the appearance of four pictures on the screen that depicted the possible meanings of the sentence. Children had to select the picture that best matched the target sentence. Sentence complexity increased as the test progressed. The test was discontinued following five consecutive incorrect responses. We used raw scores on the ASL RST for our analysis.

Fingerspelling test. Children were shown a set of 15 common English words one at a time on an iPad and asked to fingerspell each word (Figure 1(a)). The word list was designed to use common familiar words while still trying to elicit as many different handshapes as possible. The word list included: *ball, paper, girl, school, bird, teach, phone, desk, chair, table, doll, father, mother, van*, and *bug.* We videotaped children as they performed the ASL RST and fingerspelling test for scoring and behavioral coding.

Behavioral coding

We coded the fingerspelling test for errors in the spatial and temporal aspects of each movement using a custom-developed coding scheme based on Dewey's error classification (Dewey et al., 2007). We scored movement errors by comparing children's performance with that of a native-signing adult model. We coded for errors in pace, accuracy, sequence precision, movement modulation, and body part use as children fingerspelled the 15 words (Table 2). Each of the fingerspelled words was coded as correct (0) or incorrect (1) within each error category. If a child made an error while spelling one or more letters of the word, the child was given a score of 1 for that error category. A sum total error was calculated for each error category by summing the error scores across all 15 words. We also calculated a total praxis error score, which was the summed error across all categories for all 15 words. In addition, we coded the fingerspelling time for each word by calculating the time taken in seconds from the start to the end of fingerspelling. Mean fingerspelling time was the average time across all 15 words. The third author (C.W.), trained in ASL, coded all the videos after using 20% of the dataset to establish intra-rater reliability as well as inter-rater reliability with the second author (S.M.S.). Reliability of over 95% was established for all error categories using intra-class correlations (ICCs: pace=0.98, accuracy=0.98, sequence precision=0.99, movement modulation=0.99, body part use=0.96, and total praxis error=0.99).

Statistical analysis

We used independent t-tests to examine group differences in fingerspelling performance and ASL receptive skills. We also conducted correlations using Pearson's r and partial correlations between fingerspelling performance (total praxis errors and mean fingerspelling time), nonverbal intelligence measured on the TONI-4, sign language comprehension using the ASL RST raw scores, and age in both groups. To assess the association between autism severity and praxis performance in the DASD group, we also conducted Pearson's correlations between praxis performance and ADOS-2 scores (restricted and repetitive behavior (RRB) sub-domain total scores, social affect (SA) sub-domain total scores, and overall total scores). We report effect sizes using standardized mean difference (SMD) values (using Hedge's g; Hedges, 1981) or Pearson's r as well as 95% confidence intervals (CIs)

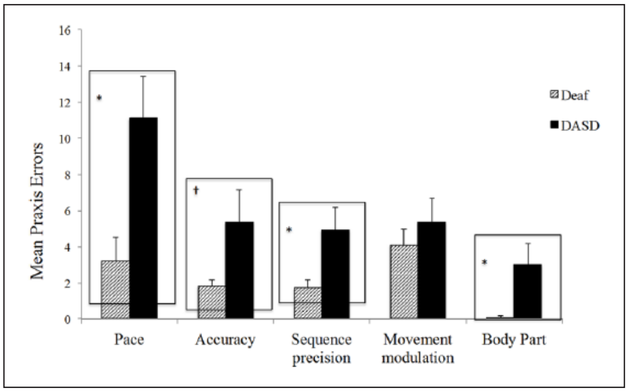

Figure 2. Praxis errors during fingerspelling.
*$p \leqslant 0.05$; †$p \leqslant 0.1$

around the calculated effect sizes. Significance was set at $p \leqslant 0.05$ and statistical trends are reported at $p \leqslant 0.1$.

Results

Group differences

The DASD group showed lower receptive language skills than the deaf children (Table 1). The DASD group also had significantly greater total praxis error scores compared to the deaf children (see Table 1). Specifically, they had greater errors in pace, sequence precision, and body part use compared to the deaf children (see Table 1 and Figure 2). The DASD group also had a trend for greater accuracy errors compared to the deaf children (see Figure 2). In terms of fingerspelling time, the DASD group fingerspelled more slowly than deaf children (see Table 1).

The relationship between praxis, receptive language, non-verbal IQ, and autism severity

There was a significant negative correlation of medium to large effect size between total praxis errors and ASL receptive skills scores in the DASD group but not in Deaf children (see Figure 3 and Table 3). Even after partialling out the effect of chronological age from praxis performance and receptive skills, we found a significant correlation between total praxis errors and raw scores on the ASL RST in the DASD group (see Table 3). Praxis performance was not associated with non-verbal IQ scores in either group. Within the DASD group, there was a trend for an association between total praxis errors and ADOS-2 overall total scores (see Table 3).

Discussion

Summary of results

We present the first study comparing praxis performance and receptive language skills in deaf children with and without ASD who were natively exposed to a signed language by their Deaf parents. We also correlated praxis performance with measures of non-verbal intelligence, receptive communication, and autism severity. We found that the DASD group made more praxis errors and were slower to fingerspell than the deaf children. In addition, the DASD group had significantly lower receptive sign language skills compared to deaf children. In the DASD group only, we found a significant correlation between total praxis errors and receptive language and a trend for an association between praxis errors and autism severity based on total ADOS-2 scores. Overall, these findings highlight the persistence of praxis and receptive communication deficits in the DASD group, despite life-long exposure to and practice with a complex manual-gestural communication system. Our findings are in line with two previous studies (Seal and Bonvillian, 1997; Soorya, 2003), where hearing children with ASD showed greater difficulty with praxis; however, these children had severe ASD and were exposed to sign language as an alternative to speech much later in life. Our results are notable because it appears that whatever benefit early sign language exposure might confer on the DASD, it is not enough to overcome the praxis and receptive language deficits associated with ASD. In the first three sections below, we discuss evidence supporting our findings. The subsequent two sections discuss mechanistic intrinsic and extrinsic factors that could have contributed to our findings. Finally, we discuss the implications of our work and directions for future research.

Praxis errors during fingerspelling by DASD

We found four distinct types of praxis errors during the fingerspelling task: errors in sequencing, body part use, spatial orientation of fingers, and movement pace. Spatial errors such as errors in hand configuration/orientation, movement amplitude, movement force, and movement accuracy as well as body-part-for-tool and movement reversal errors are often described in hearing children with ASD (Dewey et al., 2007; Ham et al., 2011; Mostofsky et al., 2006). Our findings therefore fit with the broader dyspraxia literature in ASD (Dewey et al., 2007; Gizzonio et al., 2015; Miller et al., 2014; Mostofsky et al., 2006; Smith and Bryson, 2007) and further extend it to fingerspelling/signing skills.

We found that the DASD group made more frequent body part errors involving the use of proximal joints than the Deaf children. This phenomenon has been reported previously in young deaf children who employ immature forms of fingerspelling involving gross movements of proximal joints until they improve fine motor control of distal joints to produce precise handshapes using only fingers (McIntire, 1977). In the same vein, Seal and Bonvillian (1997) reported a strong association between fine motor scores and signing accuracy in hearing children with

Table 3. Correlations between fingerspelling performance, non-verbal intelligence, receptive language, and ADOS-2 scores.

Correlations	DASD	Deaf
Total praxis error and TONI-4	−0.20	0.18
Total praxis error and ASL RST	−0.60*	−0.28
Total praxis error and ASL RST after partialling out the effect of chronological age	−0.71*	−0.18
Total praxis error and ADOS-2 total	0.53†	–
Total praxis error and ADOS-2 SA sub-domain	0.31	–
Total praxis error and ADOS-2 RRB sub-domain	0.44	–
Total praxis error and chronological age	0.07	−0.37
Fingerspelling time and TONI-4	0.18	0.09
Fingerspelling time and ASL RST	−0.46	−0.46
Fingerspelling time and ADOS-2 total	0.09	–
Fingerspelling time and ADOS-2 SA sub-domain	0.10	–
Fingerspelling time and ADOS-2 RRB sub-domain	0.03	–
Fingerspelling time and chronological age	−0.55†	−0.46

ADOS-2: Autism Diagnostic Observation Schedule, Second Edition; TONI-4: Test of Nonverbal Intelligence, Fourth edition; ASL RST: American Sign Language Receptive Skills Test; ADOS-2 Total: Overall total scores on the ADOS-2; ADOS SA: Social Affect sub-domain total scores; ADOS RRB: Restricted and Repetitive Behaviors sub-domain total scores.
Raw scores of ASL RST scores have been used for analysis; ADOS-2 was only conducted in the DASD group.
$^{*}p < 0.05$; $^{\dagger}p < 0.1$

severe ASD. Consistent with these findings, difficulty in precise motor control of fingers may have contributed to the fingerspelling inaccuracies in our DASD group. Along these lines, delays in fine motor coordination are frequently reported in school-age children with ASD (Bhat et al., 2011; Fuentes et al., 2009; Provost et al., 2007). Around 68% of preschoolers with ASD performed below average in terms of fine motor skills on the Peabody Developmental Motor Scales (PDMS-2) and qualified for early intervention (Provost et al., 2007). Similar to our findings, other studies have also reported greater movement overflow and incoordination during repetitive hand and foot movements in school-age children and adolescents with ASD compared to TD children (Biscaldi et al., 2014; Jansiewicz et al., 2006). Furthermore, our findings add to the broader literature on dyspraxia as a comorbid and secondary symptom of ASD as well as other developmental disorders and may reflect generalized abnormalities in brain connectivity (Levit-Binnun et al., 2013).

Greater temporal errors such as slowness in fingerspelling were observed in the children with ASD compared to the TD children. Dewey et al. (2007) also reported

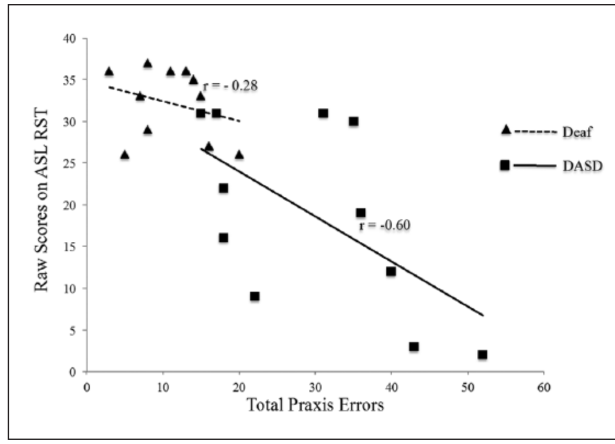

Figure 3. Correlations between praxis performance and sign language comprehension.

delays in completing praxis tasks in hearing children with ASD (Dewey et al., 2007). Moreover, movement slowness across multiple tasks, including reaching (Mari et al., 2003), handwriting (Fuentes et al., 2009; Kushki et al., 2011), button pressing (Biscaldi et al., 2014), and repetitive hand movements (Biscaldi et al., 2014; Jansiewicz et al., 2006), has been reported in ASD. Perhaps by moving slowly, the DASD group improved their visual processing of the displayed word and access to the motor patterns underlying fingerspelling, thereby facilitating the planning and execution of the various gestural sequences. Finally, apart from motor planning, other more cognitive aspects such as short-term memory or working memory deficits often associated with ASD may have also contributed to the poor fingerspelling performance of the DASD group (Rogers et al., 1996).

ASL receptive skill deficits in DASD

Children with ASD had lower scores on the ASL RST compared to the deaf children. Delayed receptive language and poor gestural understanding are often reported in hearing children with ASD (Dowell et al., 2009; Hudry et al., 2010; Kwok et al., 2015; Smith and Bryson, 2007). In fact, studies describing the developmental trajectories of language acquisition in children with ASD reported poor receptive vocabulary compared to expressive vocabulary (Charman et al., 2003; Luyster et al., 2008). A recent meta-analysis of receptive and expressive communication showed that both domains are significantly delayed in children with ASD compared to TD peers (Kwok et al., 2015). Studies on gestural communication also confirm that both gesture recognition and production are worse in hearing children with ASD than TD children (Dowell et al., 2009; Ham et al., 2011; Smith and Bryson, 2007). However, the gesture recognition/discrimination deficits are greater for communicative gestures versus transitive gestures; the objects used during transitive gestures provide a functional context to aid recognition. In line with this evidence, our ASD group may have encountered difficulties in comprehending the communicative signs of fingerspelling thereby contributing to poor receptive language skills.

Relationship between praxis performance, receptive language, and autism severity

Our study found strong correlations between praxis performance and sign language comprehension in the DASD group only (see Figure 3). Prior studies have reported co-occurring deficits in gesture discrimination and production extending across multiple modalities (visual and verbal) and not limited to imitation among older children with ASD compared to age- and IQ-matched TD children (Dewey et al., 2007; Dowell et al., 2009; Ham et al., 2011; Mostofsky et al., 2006; Smith and Bryson, 2007). Children with ASD showed strong correlations between "verbal" gesture recognition and gesture production on "verbal command" for communicative gestures (Smith and Bryson, 2007). Along the same lines, 7–15 year-old children with ASD showed a strong association between pantomime recognition and pantomime imitation skills (Ham et al., 2011). Poor gestural performance could be a result of impairments in storing and accessing the learned motor sequences or in gestural knowledge (Dowell et al., 2009; Smith and Bryson, 2007), beyond the basic motor (Vanvuchelen et al., 2007) or visuo-spatial mapping difficulties in ASD (Ham et al., 2011; Williams et al., 2004). We also found a trend for an association between praxis errors and autism severity, suggesting that dyspraxia may contribute to the symptoms of ASD. This fits with the recent literature reporting a significant association between praxis performance and autism severity (Dowell et al., 2009; Dziuk et al., 2007; Gizzonio et al., 2015; Ham et al., 2011). Overall, these findings support the notion that impaired praxis performance continuously interacts with and is affected by the social communication difficulties of children with ASD. Future studies should further explore this association across various levels of autism severity for praxis to be considered among the diagnostic criteria for ASD.

Proficiency in the use of sign language may depend on various intrinsic child-related factors such as the child's ability to perceive social input through caregiver observation and their levels of fine motor coordination/praxis, as well as extrinsic environmental factors such as the quantity and quality of caregivers' input.

Child-related factors influencing fingerspelling

The social and motor impairments associated with ASD can significantly influence fingerspelling skills. From very early on, hearing infants who later develop ASD show excessive interest in non-social cues, do not tune into social cues, and demonstrate persistent joint attention difficulties such as poor gaze alternation between objects (Bhat et al., 2010; Kaur et al., 2015; Maestro et al., 2002; Osterling and Dawson, 1994; Presmanes et al., 2007; Sullivan et al., 2007). For example, recent prospective studies have shown that infants with older diagnosed siblings with ASD are unable to divide their attention between objects and people and invariably have reduced social attention (Bhat et al., 2012; Sullivan et al., 2007). These early atypicalities in social gaze in young toddlers with ASD may affect their social communication development by limiting their opportunities for shared attention/empathy with others (Charman and Baron-Cohen, 1997; Charman et al., 2003) and language development (Baldwin, 1995; Mundy and Sigman, 2006; Toth et al., 2006). In the DASD group, similar impairments such as reduced social gaze (Bhat et al., 2012; Maestro et al., 2002; Osterling and Dawson, 2002), joint attention difficulties, and reduced shared attention (Bhat et al., 2012; Mundy and Sigman, 2006; Osterling and Dawson, 1994; Srinivasan and Bhat, 2016) may prevent children from observing and reproducing the sign language stimuli offered by caregivers. Moreover, given that deaf children need to look at and attend to a person in order to perceive the sign language signal, the DASD population may suffer from reduced access to language input compared to hearing children because their opportunities for incidental learning (i.e. ability to overhear conversations when not attending to them) are perhaps reduced.

Another factor contributing to poor motor praxis performance could be the early motor delays observed in children with ASD (Bhat et al., 2011; Landa and Garrett-Mayer, 2006; Ozonoff et al., 2008). There is substantial evidence of a variety of fine motor delays in reaching, grasping, showing/pointing, and other complex skills in infants and young hearing children with ASD (Gernsbacher et al., 2008; Kaur et al., 2015; Koterba et al., 2012; LeBarton and Iverson, 2013; Libertus and Landa, 2014; Provost et al., 2007). These early motor delays will directly impact the perception–action couplings of deaf infants/toddlers with ASD as they learn to sign. From an ecological perspective, "we must perceive in order to move effectively and we must move in order to perceive effectively" (Gibson, 1979). At a young age, if children with ASD are unable to produce the correct motor sequences or are not receiving correct visual and proprioceptive information while signing, this will lead to atypical perception–action couplings, ultimately contributing to poor sign language comprehension. Further support for this hypothesis comes from research supporting the cascading adverse effects of fine motor impairments on communication skills in hearing infants with an older sibling with ASD (Bedford et al., 2016; Iverson and Wozniak, 2007; LeBarton and Iverson, 2013; Leonard et al., 2014; McDuffie et al., 2007). Poor fine motor performance correlated with expressive language delays at 3 years of age in infants with an older sibling with ASD (LeBarton and Iverson, 2014). Similarly, infants who later developed language delays or ASD used fewer deictic gestures such as pointing and showing as well as symbolic gestures such as "putting the telephone to the ear" or "eating with a spoon" between 12 and 24 months compared to TD infants (LeBarton and Iverson, 2016; Mitchell et al., 2005; Werner and Dawson, 2005). Overall, there is substantial evidence in hearing infants with ASD that corroborates our findings of fine motor dyspraxia contributing to poor sign language comprehension and production in the DASD group.

Caregiver-related factors influencing fingerspelling

The children in our study grew up in enriched and stimulating language environments, having been exposed to ASL from birth. Therefore, their sign language comprehension impairments cannot be attributed to an impoverished linguistic environment. However, atypical interactions of children with caregivers may influence the amount of sign language input received, which could further contribute to delays in sign language comprehension/production. Multiple studies have reported differences in caregiver behaviors of children with ASD including greater physical proximity, gesture use, and verbal input indicative of a more directive parenting style compared to caregivers of TD children (El-Ghoroury and Romanczyk, 1999; Leezenbaum et al., 2014; Lemanek et al., 1993; Wan et al., 2012). A prospective study of naturalistic toddler–caregiver interactions suggested that at 18 months, infants with an older sibling with ASD engaged in fewer sophisticated gestures like "pointing" and "showing" that serve as ideal "teaching" opportunities for caregivers to facilitate infants' language; therefore, motor and communication delays in high-risk infants can alter the caregiver input received, which can further alter the course of infants' language development (Leezenbaum et al., 2014). In contrast, relatively little is known about the interactions of deaf children with their caregivers (Pizer and Meier, 2008). A case series of three sets of toddler–caregiver interactions suggested that in two out of the three families, Deaf caregivers modified their signing to grab the attention of their toddlers using strategies such as repetition, displacement, lengthening, and so on (Pizer and Meier, 2008). In contrast, the third parent showed low levels of sign language input due to the poor signing abilities of her child (Pizer and Meier, 2008). We hypothesize that similar processes could occur during the development of sign language in the DASD population. If there are difficulties in getting the child's attention or in the child's ability to move their hands/fingers swiftly, caregivers may not persist over the long term and may inadvertently reduce their sign language input. These modifications may perhaps alter the child's understanding of ASL. Overall, several child-related and caregiver-related factors may affect both sign language comprehension and production in children with ASD.

Clinical implications for children with ASD

In terms of assessment, given the motoric complexity of signing, it is important to evaluate praxis performance in DASD. Our study suggests that while communicating with children, parents of DASD should consider their child's motor impairments along with their social communication deficits. While teaching strategies for sign language, they should be sensitive to the perceptuo-motor demands of sign language learning. Given the tight relationship we find between motor skills and language development, motor interventions may possibly benefit language acquisition in the DASD. It is currently not known whether the traditional attention-getting strategies naturally employed by Deaf caregivers (Holzrichter and Meier, 2000) are effective in teaching signs to children with ASD; future research is needed to test this hypothesis. Moreover, other complementary strategies such as the use of visual picture schedules should be used to improve communication when signing is difficult in DASD. Finally, it is not clear whether the DASD group in our study would perform significantly better in terms of fine motor praxis compared to hearing children with ASD due to their greater experience with sign language and fingerspelling. However, if this were true, it would be interesting to use the framework of sign language training as an intervention tool to promote praxis and communication in children with ASD.

Limitations and future directions

Since we chose a unique population that was difficult to access, we had a small sample size of only 11 children per group. Although we did not statistically control for the amount of fingerspelling experience, we included only children who were exposed to ASL from birth, thereby controlling for overall sign language exposure. We had more boys than girls in the DASD group and this gender imbalance is recognized in the ASD population (Baio, 2014). In terms of testing, we did not employ a standardized praxis or motor coordination measure. For this reason, we are unable to make judgments on whether there were low-level motor deficits in our control group of deaf children, as noted in past literature (Fellinger et al., 2015). We also had to modify some items during the administration of the ADOS-2 since it is not designed for deaf children.

Conclusion

In this study, we compared praxis performance during fingerspelling and receptive language skills in a novel and unique research population: deaf, native-signing children with and without ASD. Children with ASD made more praxis errors than TD children. Specifically, their movements were slower, more inaccurate or incorrectly sequenced, and involved proximal body parts. The children with ASD also had significantly lower receptive language skills than the TD children. The praxis errors of children with ASD correlated with their receptive language skills and to some extent with their autism severity. Together, these findings emphasize the fine motor dyspraxia observed in DASD within the unique context of fingerspelling.

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Notes

1. As is conventional in the sign language, we refer to people who identify with and belong to the Deaf community with a capital D.
2. Lower Social Communication Questionnaire (SCQ) scores are expected for deaf signing children due to several inapplicable questions, such as the "Response to name" item. We eliminated such items since they would fail to discriminate between deaf children with and without ASD.

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