Relations Between Language and Cognition in Native-Signing Children With Autism Spectrum Disorder

Aaron Shield, Jennie Pyers, Amber Martin, and Helen Tager-Flusberg

Two populations have been found to exhibit delays in theory of mind (ToM): deaf children of hearing parents and children with autism spectrum disorder (ASD). Deaf children exposed to sign from birth by their deaf parents, however, show no such delay, suggesting that early language exposure is key to ToM development. Sign languages also present frequent opportunities with visual perspective-taking (VPT), leading to the question of whether sign exposure could benefit children with ASD. We present the first study of children with ASD exposed to sign from birth by their deaf parents. Seventeen native-signing children with a confirmed ASD diagnosis and a chronological- and mental age-matched control group of 18 typically developing (TD) native-signing deaf children were tested on American Sign Language (ASL) comprehension, two minimally verbal social cognition tasks (ToM and VPT), and one spatial cognition task (mental rotation). The TD children outperformed the children with ASD on ASL comprehension ($p < 0.0001$), ToM ($p = 0.02$), and VPT ($p < 0.01$), but not mental rotation ($p = 0.12$). Language strongly correlated with ToM ($p < 0.01$) and VPT ($p < 0.001$), but not mental rotation ($p = ns$). Native exposure to sign is thus insufficient to overcome the language and social impairments implicated in ASD. Contrary to the hypothesis that sign could provide a scaffold for ToM skills, we find that signing children with ASD are unable to access language so as to gain any potential benefit sign might confer. Our results support a strong link between the development of social cognition and language, regardless of modality, for TD and ASD children. ***Autism Res*** *2016, 00: 000–000*. © 2016 International Society for Autism Research, Wiley Periodicals, Inc.

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Introduction

Two populations of children exhibit delays in the development of theory of mind (ToM), the understanding of mental states: deaf children of hearing parents [Courtin, 2000; Courtin & Melot, 1998; Peterson & Siegal, 1995, 1999; Remmel, Bettger, & Weinberg, 2001; Schick, de Villiers, de Villiers, & Hoffmeister, 2007], and children with autism spectrum disorder (ASD) [Baron-Cohen, Leslie, & Frith, 1985; Happé, 1994]. Unlike deaf children of hearing parents, Deaf[1] children born to Deaf parents who are exposed natively and consistently to a natural sign language show on-target, and sometimes advanced, ToM reasoning [Courtin, 2000; Schick, de Villiers, de Villiers, & Hoffmeister, 2007]. We know little about the development of ToM in deaf children with ASD, even though one in 59 deaf American children carries an ASD diagnosis [Szymanski, Brice, Lam, & Hotto, 2012], a prevalence rate that is slightly higher than that of the general population (1 in 68) [Centers for Disease Control, 2014].

Language has been implicated in the ToM delay in both deaf children of hearing parents and hearing children with ASD, though for different reasons. The ToM delay in deaf children of hearing parents stems from the fact that their hearing impairment leads to delayed exposure to an accessible language, signed or spoken [Gale, de Villiers, de Villiers, & Pyers, 1996; Peterson & Siegal, 1995; Schick, de Villiers, de Villiers, & Hoffmeister, 2007]. Such language delay leads to limitations spanning various aspects of ToM reasoning, not just false-belief understanding, even when minimally verbal measures are used, reducing the language demands of the task (see Pyers & de Villiers [2013] for review). Despite observed delays in false-belief reasoning in early childhood, deaf children of hearing parents eventually master false-belief later in childhood [Wellman, Fang, & Peterson, 2011] or in adulthood [Pyers & Senghas, 2009] and acquire ToM skills, including false-belief understanding, in the same

[1]As is convention, we refer to Deaf children who are immersed in the language and culture of the Deaf community as "Deaf" with a capitalized "d."

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sequence as typically hearing children [Wellman, Fang, & Peterson, 2011].

Like deaf children of hearing parents, children with ASD also exhibit ToM delays, albeit for different reasons [e.g., Baron-Cohen, Leslie, & Frith, 1985; Moran et al., 2011; Schneider, Slaughter, Bayliss, & Dux, 2013]. As in deaf children, ToM ability in children with ASD is related to language ability: higher vocabulary correlates with false-belief performance [Happé, 1995], mastery of embedded clause structures significantly predicts ToM ability [Tager-Flusberg & Joseph, 2005], and mental-state words (e.g., think and believe) are underrepresented in the children's vocabularies [Tager-Flusberg, 2005]. Moreover, ToM ability correlates with the severity of ASD symptoms [Steele, Joseph, & Tager-Flusberg, 2003; Tager-Flusberg, 2003]. Yet, children with ASD show a similar ToM developmental trajectory as typically developing (TD) children [Peterson, Wellman, & Liu, 2005], and most make gains over time [Steele, Joseph, & Tager-Flusberg, 2003].

The interdependency between language and ToM may have its origins in joint attention, which strongly predicts language development [Carpenter, Nagell, & Tomasello, 1998; Mundy & Gomes, 1998; Tomasello & Farrar, 1986]. Children with ASD show less sensitivity to the use of gaze as an indication of referential intent than TD children [Baron-Cohen, Baldwin, & Crowson, 1997]. Conversely, though deaf children of hearing parents are not insensitive to eye gaze [Hao & Su, 2014], their parents spend less time than Deaf parents do in high-quality episodes of sustained joint attention [Gale & Schick, 2009; Spencer & Meadow-Orlans, 1996]. The limited engagement in joint attention for both groups may have cascading negative effects on language acquisition, which in turn delays the time course of ToM development, though with the latter group the impairment originates in the caregiver, while in the former group it originates in the child.

Unlike deaf children of hearing parents, native-signing Deaf children are exposed to sign language from birth, and consequently perform better than deaf children of hearing parents and equivalently to typically hearing children on a range of ToM tasks [Schick, de Villiers, P., de Villiers, J., & Hoffmeister, 2007]. Interestingly, Courtin [2000] and Courtin and Melot [2005] found that these children outperformed a group of age and SES-matched hearing children on false-belief understanding, and suggested that native sign exposure could privilege ToM due to the widespread opportunities for practice with visual perspective-taking (VPT) therein. For example, in order to learn signs, children must first imitate the signs produced by adults, not as they appear from their vantage point, but as their caregivers produce them. Further, descriptions of spatial layouts are typically signed from the signer's perspective, requiring interlocutors to adopt the signer's perspective to interpret the layout correctly [Emmorey, Tversky, & Taylor, 2000; Pyers, Perniss, & Emmorey, 2015]. Consequently, early and frequent exposure to sign language could provide opportunities to engage VPT skills, perhaps enhancing not only VPT skills, but also other aspects of ToM. If so, then we would expect that Deaf native-signing children with ASD, through their exposure to a language rich in spatial and perspective-taking features, could be relatively advantaged in terms of VPT and related ToM abilities compared to what has been reported for hearing children with ASD—provided that they are able to access the language in the first place. Alternatively, if their social deficits are severe enough to limit the acquisition of sign language, then they may never experience the practice with VPT that may accompany normal sign acquisition, and their ToM skills will show the same delays as in nonsigning children with ASD.

Visual perspective-taking is typically classified into two sequential levels of ability. Level 1 VPT includes understanding whether an object is visible to another person, normally acquired by age two [Lempers, Flavell, & Flavell, 1977] and is intact in children with ASD [Baron-Cohen, 1989; Hobson, 1984; Leslie & Frith, 1988]. Level 2 VPT refers to the understanding that objects visible to the self and to another person may appear differently, depending on vantage point, and is typically acquired by age 4–4.5 years [Flavell, Everett, Croft, & Flavell, 1981]. Some studies with individuals with ASD have found evidence of Level 2 VPT deficits [Hamilton, Brindley, & Frith, 2009; Yirmiya, Sigman, & Zacks, 1994], while others have not, leading some to argue that VPT abilities involve a mere spatial calculation, not social understanding of others' mental states, and as such are not part of ToM [Reed & Peterson, 1990; Tan & Harris, 1991].

Whereas VPT reflects the social ability to understand another's visual perspective, mental rotation skills reflect the spatial ability to imagine one's own view of an object from different angles. The task demands of VPT (i.e., putting yourself in someone else's shoes) seem to be easier, even in adult populations, than those involved in mentally rotating objects [Amorim, Isableu, & Jarraya, 2006; Wraga, Creem, & Proffitt, 1999]. However the opposite pattern seems to emerge for children with ASD; researchers are unanimous in their observations that children with ASD have intact, or even enhanced, spatial cognition. Moreover, fluent signers outperform nonsigners on several spatial tasks including spatial memory and mental rotation [Bellugi et al., 1990; Emmorey, Kosslyn, & Bellugi, 1993; Martin, 2009; Wilson, Bettger, Niculae, & Klima, 1997]. Such enhancements in spatial abilities likely derive from experience with the spatial elements of sign languages,

Table 1. Summary of Participant Characteristics

	n (males)	Age (SD)	NVIQ (SD)
ASD	17 (13)	9.62 (2.64)	96.76 (11.49)
TD	18 (8)	9.31 (1.77)	101.56 (10.30)

which require both the signer and the interlocutor to build and maintain a spatial image over the duration of discourse. In this way, sign language experience enhances mental imagery abilities, which in turn enhance mental rotation abilities [Emmorey, Klima, & Hickok, 1998; Emmorey, Kosslyn, & Bellugi, 1993; Martin, Senghas, & Pyers, 2013].

No reports currently exist on the ToM development of deaf children with ASD or the VPT skills of native-signing Deaf children. We fill these gaps with the first report of ToM and VPT skills in a sample of native-signing children with ASD, comparing their performance to that of TD Deaf children with Deaf parents on measures of (a) false-belief understanding, (b) VPT, (c) mental rotation ability, and (d) American Sign Language (ASL) comprehension. Here we present exploratory data from native users of ASL with and without ASD, in order to (a) provide a much-needed profile of language and cognitive development of native-signing children with ASD and (b) investigate the hypothesized benefit in VPT that could arise from native sign language experience. Do native-signing children with ASD show a similar cognitive and linguistic profile as has been reported for hearing, non-signing children with ASD? Or might the visual–manual modality of ASL, with its rich opportunities for perspective-taking, advantage learners?

Method

Participants

We tested children with ASD and TD Deaf children (Table 1), who used ASL as their primary language from birth. All but one were born to Deaf parents; the exception had four Deaf grandparents, and hearing, native-signing parents. Two children with ASD were typically hearing with two Deaf parents.[2] Children with ASD were recruited via a video in ASL posted on social media, and research visits were conducted at the child's home or school. Typically developing children were recruited through schools for the Deaf and were tested in those schools. None of the participants had received a cochlear implant. Because we were recruiting from two rare populations, we tested every child whose family responded to our recruitment notices. All procedures were prospectively reviewed and approved by a duly constituted Institutional Review Board, and the parents of all participating children granted written informed consent.

Autism spectrum disorder group. Seventeen children with ASD are reported here (13 M). With no instruments designed for specific use with deaf children, diagnosing ASD in deaf children is difficult [Mood & Shield, 2014]. We confirmed ASD diagnoses using the Autism Diagnostic Observation Schedule, Second Edition (ADOS-2) [Lord et al., 2012] administered by two ASL-proficient researchers who had attained research reliability on the instrument. Since the ADOS-2 was not designed for deaf children, some tasks could not be performed (e.g., "Response to name," which entails calling a child's name aloud and seeing if the child looks up or responds). Given this limitation of the ADOS-2, for borderline or below threshold scores we relied on the judgment of a native-signing licensed clinical psychologist who was trained in ASD.

Eight additional children (5 M) were excluded from the study for failure to complete any of the measures ($n = 5$) or for inability to confirm diagnosis ($n = 3$) via the ADOS-2 or the clinician's judgment.

Typically developing group. Eighteen typically developing Deaf children (8 M) participated. All children had at least one Deaf parent and had been exposed to ASL from birth. The children were screened for possible ASD using the Social Communication Questionnaire [Rutter, Bailey, & Lord, 2003]. All scored well below the clinical cut-off of 15 ($M = 2.39$; $SD = 2.35$; range = 0–7). No TD children were excluded from the study.

Matching. The two groups were matched for chronological age (Mann–Whitney $U = 138$, $P = 0.63$) and nonverbal intellectual ability (Mann–Whitney $U = 139$, $P = 0.65$, Table 1) as assessed with the Test of Nonverbal Intelligence, Fourth Edition (TONI-4) [Brown, Sherbenou, & Johnsen, 2010].[3]

Measures and Procedures

American Sign Language Receptive Skills Test (ASL-RST) [Enns et al., 2013]. This test measures children's understanding of ASL grammar in phrases and sentences and is appropriate for use with children ages 3–13. A 20-item vocabulary check was administered before the receptive skills test to verify that children knew the signs included in the measure.

[2] We included these two hearing children in the study because the relevant variable here is native exposure to sign language, not deafness.

[3] Pilot work revealed that we would be unable to match the groups on language ability; we would have had to test primarily 3- to 4-year-old TD children to obtain a language-matched sample. Several of our measures were not valid for children under 5 years of age, and in this younger age range we would not have found as many TD children who passed our other cognitive measures (ToM and VPT, for example).

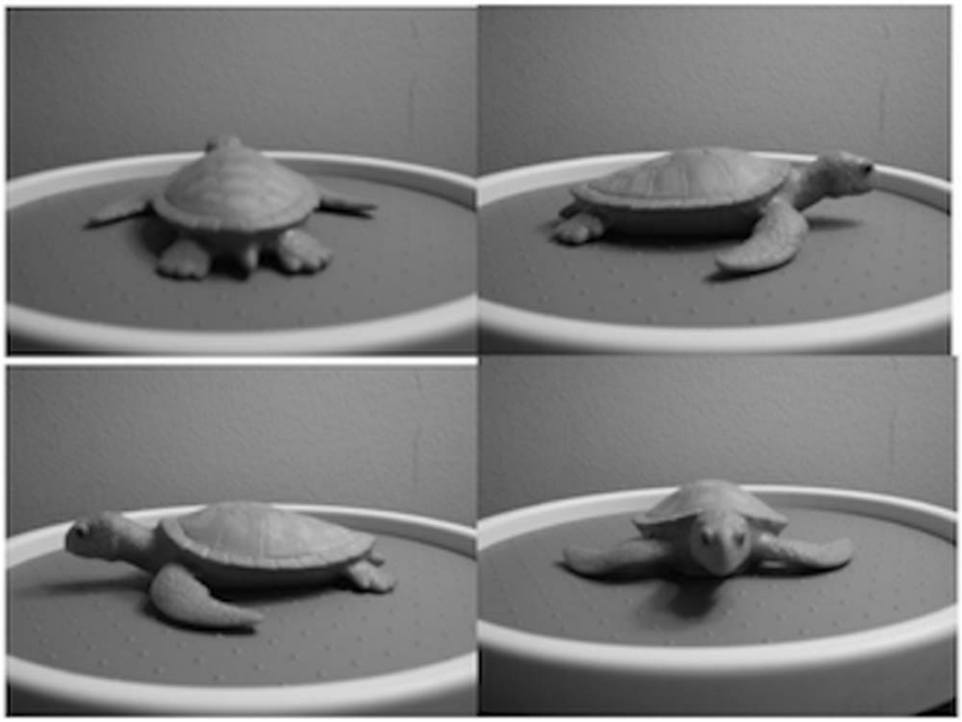

Figure 1. An example of the choices available to children for a given trial on the VPT task.

The ASL-RST was presented on a laptop computer in front of the seated child. For the three practice items and 42 test items, the sign model produced a sentence in ASL on the screen followed by four pictures depicting possible meanings. Children selected the picture that best matched the target sentence. Sentence complexity increased in difficulty as the test progressed; the test was discontinued after five consecutive incorrect responses, in accordance with the manual. Children received feedback after the practice items only (i.e., if an incorrect answer was given, the examiner would sign "no" and repeat the practice item).

We additionally identified five items that required perspective-taking skills in that they represented a spatial layout that required the child to adopt the perspective of the signer to understand correctly; all appeared late in the test.

Minimally verbal false-belief [Pyers & Senghas, 2009]. In this picture completion task, the experimenter set out five individual cards that illustrated a sequence of events. As the experimenter presented each card, he pointed at it and labeled the order of the picture (e.g., "first" and "second"), without narration. The participants were then given two cards that depicted two possible endings and the experimenter signed, "Which one comes next?"[4] then gestured to the participant to move one of the two cards next to the fifth card. The task began with two training trials with feedback designed to confirm that participants understood the picture-completion procedure.

The four test trials depicted false-belief events based on the unseen-displacement task [Wimmer & Perner, 1983], in which an object was moved to a new location out of view of the main character. Participants who selected the picture depicting the character looking for the object in its original location, rather than its new (unobserved) location, were given credit for understanding false-belief. The experimenter nodded and smiled after each test trial, regardless of the child's selection.

Visual perspective-taking. We adapted Reed and Peterson's [1990] Level 2 VPT task to be minimally verbal for deaf children. The experimenter sat across from the child and placed a toy turtle on a turntable in front of the child, then spun the turntable and encouraged the child to do so as well.

Own-perspective trials. The researcher placed a 8½″ × 11″ (21.6 × 28 cm) laminated card containing four images of the turtle on the turntable, from four different angles (Fig. 1) on the table in between the child and the turtle, rotated the turntable so that the turtle's tail or head faced the child, then pointed to each of the four pictures and signed in ASL, "Look! See the turtle? Which one matches?"[5]

[4]In ASL: NEXT WHICH?

[5]In ASL: LOOK! SEE TURTLE? WHICH MATCH?

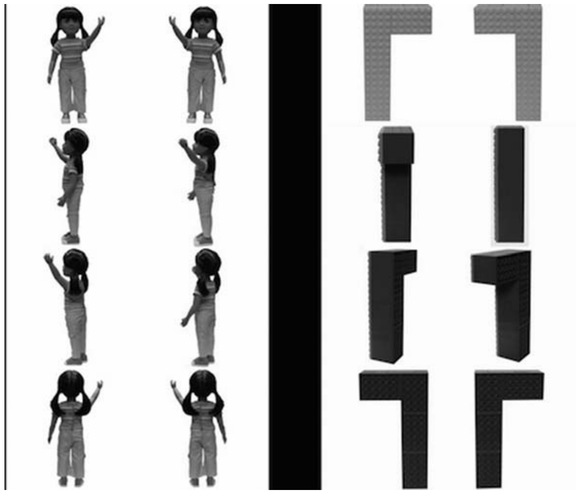

Figure 2. The eight test items on the mental rotation task: the doll and L-block at 0, 90, 120, and 180° of rotation (top to bottom).

After the child pointed to the correct picture, the researcher spun the turntable again so that the opposite end of the turtle faced the child, and placed a new card with the four photos of the turtle in a different configuration, and asked for the matching picture. This procedure was repeated with a second object (a plastic fish) for a total of four trials. A correct trial was one where the child successfully indicated the picture matching his/her view.

Other-perspective trials. The researcher gave the child a new set of pictures in a different configuration, spun the turntable again so that the fish's head or tail was facing the researcher, and signed, "Now, this one is different. I'm looking [at the fish]. Which one do I see?"[6] He pointed at each picture, and then to himself to emphasize that he was looking at the fish and made a puzzled expression, leaning closer to the table and squinting. After the child responded, he repeated the procedure with the other end of the fish (either head or tail). This procedure was then repeated with the turtle. The two front and two back trials were coded as correct if the child successfully indicated the picture matching the point of view of the researcher.

Mental rotation [Martin, Senghas, & Pyers, 2013]. Participants sat in front of a laptop outfitted with a MagicTouch touchscreen overlay and were trained to touch the image on the screen that matched an object placed in front of the laptop. The experimenter simply placed the training object (a triangle-shaped block and a LEGO figure with both hands raised) in front of the laptop and looked expectantly at the participant while two different images, one identical to the object, appeared on the screen. If the child did not respond, the experimented signed TOUCH or TOUCH SAME in the direction of the screen.

Participants then completed two sets of test trials using two object types: a small doll, and an L-shaped block constructed of LEGO bricks. We included these two different objects to explore whether mental rotation abilities were affected by object type. The experimenter held the target object in front of the child, between his body and the laptop without obstructing the screen. Then, two figures (images of the target object shown from a rotated angle) appeared on the screen. One figure was a configural match to the real object (e.g., the doll with its left hand raised) and the other was its mirror image (e.g., the doll with its right hand raised). Children responded by touching the image of the test object that matched the real object. The target object stayed in front of the child throughout the trial set. Children could not touch or turn the real object to match the figures on the screen. For each trial set, children saw the test figures rotated horizontally at 0° (no rotation), 90°, 120°, or 180° (see Fig. 2). Participants completed a total of 32 trials: 16 for the doll (4 trials at each angle of rotation) and 16 for the L-block. A happy face appeared on the screen after a correct response, and a sad face appeared after an incorrect response. Each trial was coded as "correct" if the child touched the matching test figure; credit was given only for the first image touched. We computed the percentage of correct responses.

Results

Language Comprehension

On the vocabulary check, all TD children obtained a perfect score of 20, while the children with ASD had a mean score of 19.3 ($SD = 1.81$, range 13–20). All but one knew at least 18 items; the remaining child was excluded from the analyses.

On the comprehension test, TD children scored significantly higher ($Mdn = 110$) than the children with ASD ($Mdn = 84$), Mann–Whitney $U = 20$, $P < 0.0001$ (Table 2).[7] Seventeen of the eighteen TD children reached the last item on the test, while only five children with ASD did so; the other children were discontinued earlier in the test following five consecutive incorrect responses.

Analysis of specific structures. We compared the performance of the five children with ASD and 17 TD children who completed all the items. These five children with ASD had a significantly lower median raw

[6]In ASL: NOW DIFFERENT. IX-1 LOOK. WHICH SEE IX-1?

[7]We use the more conservative non-parametric tests throughout this paper because assumptions of normality were violated.

Table 2. Mean Group Performance on the ASL Receptive Skills Test

	Vocabulary check (SD)	Comprehension raw score (SD)	Comprehension standard score (SD)
ASD ($n = 17$)	19.29 (1.76)	17.63 (10.68)	86.29 (12.76)
TD ($n = 18$)	20 (0)	33.17 (4.27)	108.72 (6.29)

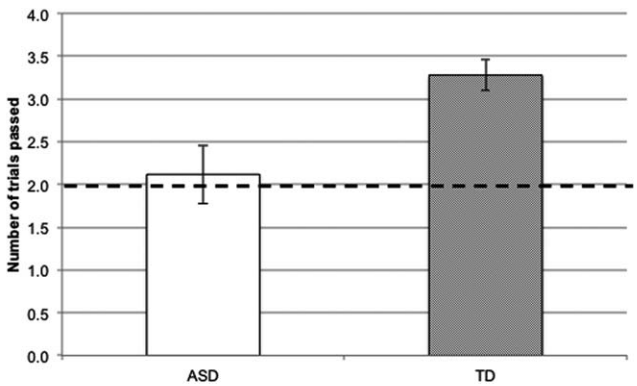

Figure 3. Children with ASD performed at chance on a minimally verbal ToM task, while TD children were significantly better than chance ($P < 0.0001$) and better than the children with ASD ($P = 0.02$).

score of 31 ($M = 29$; $SD = 3.9$, range 22–31) compared to the 17 TD children, who had a median raw score of 35 ($M = 33.5$; $SD = 4.1$, range 26–40; Mann–Whitney $U = 16$, $P = 0.04$).

Of particular interest for our purposes were the perspective-taking items. The overall error rate on these items was high for both groups: 70 of 119 attempted trials were answered incorrectly, an error rate of 58.8%.

False-Belief Understanding

All children completed the minimally verbal false-belief task. All TD children passed both practice trials without the need for feedback, and all but three children with ASD passed both practice trials. Each of the three children with ASD passed one practice trial without feedback and received corrective feedback on the other.

On the four test trials, children with ASD chose the correct ending to the story significantly less frequently (Mdn = 2 items correct, range 0–4) than did the TD children (Mdn = 3 items correct, range 2–4; Mann–Whitney $U = 80$, $P = 0.02$). The ASD mean of 2.12 was not significantly different from chance ($t(16) = 0.34$, $P = 0.73$), unlike the TD mean of 3.28 ($t(17) = 7.2$, $P < 0.0001$ (see Fig. 3).

A score of three or more correct trials is considered above chance performance, and as such, we categorized anyone with a score of three or four as having passed the false-belief test. Significantly more TD children ($n = 15$) than children with ASD ($n = 7$) passed the false-belief test (Fisher's exact test, $P < 0.05$).

Visual Perspective-Taking

All children completed the VPT task, passing all practice ("own-perspective") trials, except for one child with ASD, who passed three of the four trials. Thus, all children showed an ability to report how the objects looked from their perspective. On the test ("other-perspective") trials, children with ASD chose the correct item significantly less frequently (Mdn = 0 correct, range 0–4) than did the TD children (Mdn = 4 correct, range 0–4; Mann–Whitney $U = 70.5$, $P < 0.01$). The ASD mean of 0.88 was not significantly different from chance ($t(16) = 0.32$, $P = 0.75$), unlike the TD mean of 2.83 ($t(17) = 4.47$, $P < 0.0001$). All errors consisted of picking the picture that represented the child's own view (see Fig. 4).

A score of 3 or 4 indicated above chance performance on the VPT task; significantly fewer children with ASD ($n = 3$) performed at above chance rates than the TD children ($n = 12$; Fisher's exact test, $P < 0.01$). Eleven children did not pass this task and did not pass the false-belief task (ASD: $n = 9$; TD: $n = 2$); two children passed the VPT task but not the false-belief task (ASD: $n = 1$; TD: $n = 1$); and seven children who had passed the false-belief task did not pass the VPT task (ASD: $n = 5$; TD: $n = 2$).

Mental Rotation

Three children with ASD were not able to complete the session, leaving eighteen TD children and fourteen children with ASD left to include in the analyses. The data were negatively skewed (−1.19; Shapiro–Wilk's test, $P < 0.01$) and transformations did not correct for nonnormality. As such, we also used nonparametric statistics in the following analyses.

Despite previous reports of sex differences on mental rotation tasks [e.g., Voyer, Voyer, & Bryden, 1995], we observed no sex differences when we considered object types separately (dolls, Mann–Whitney $U = 118$, $P = 0.85$; blocks, Mann–Whitney $U = 122$, $P = 0.97$) or together (Mann–Whitney $U = 111$, $P = 0.65$); we thus combined boys' and girls' scores for subsequent analyses.

Both children with ASD and TD children performed above chance (ASD: $t(13) = 3.93$, $P < 0.001$; TD: $t(17) = 11.02$, $P < 0.0001$; see Figure 5), and the groups did not significantly differ on total accuracy (ASD: Mdn = 24.5; TD children: Mdn = 29; Mann–Whitney $U = 84$, $P = 0.12$). Examining the two object conditions,

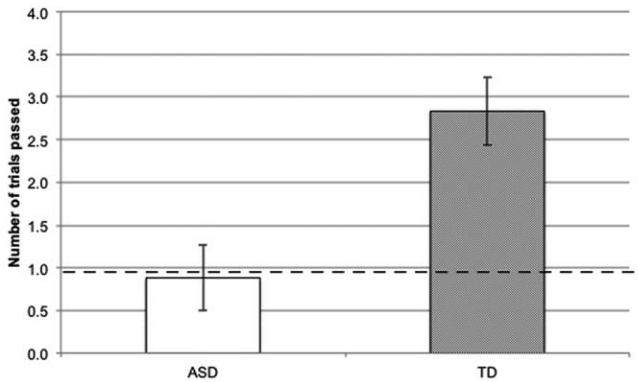

Figure 4. Children with ASD performed at chance on the VPT task, while TD children were significantly better than chance ($P < 0.0001$) and better than the children with ASD ($P < 0.01$).

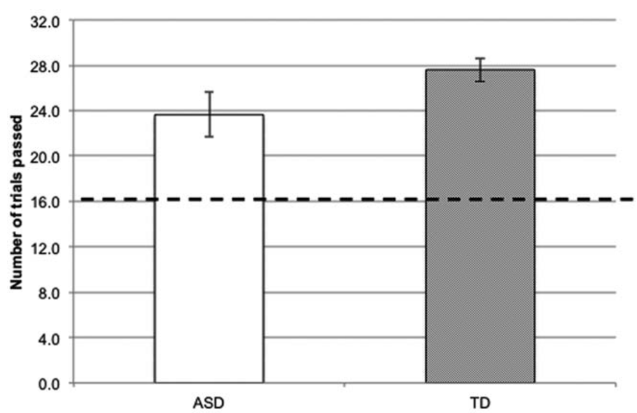

Figure 5. Both ASD and TD children performed significantly above chance on the mental rotation task, and there was no significant difference between the groups.

children with ASD scored 75.4% accuracy for dolls and 71.0% accuracy for blocks. TD children scored an average of 86.8% accuracy for dolls and 85.4% accuracy for blocks. Children with ASD were significantly above chance on both conditions (dolls: $t(13) = 3.86$, $P < 0.001$); blocks: $t(13) = 3.46$, $P < 0.002$), as were TD children (dolls: $t(17) = 9.04$, $P < 0.0001$; blocks: $t(17) = 11.85$, $P < 0.0001$). There were no significant differences between the groups for either condition (dolls: Mann–Whitney $U = 85$, $P = 0.13$; blocks: Mann–Whitney $U = 84.5$, $P = 0.12$).

Children performed more accurately on unrotated (0°) trials than on 120° ($z = -2.71$, $P = 0.007$) and 180° ($z = -2.89$, $P = 0.004$), but not 90° ($z = -1.65$, $P = 0.10$), as shown by Wilcoxon signed-rank tests. The TD group performed above chance on all degrees of rotation (0°: $z = 3.62$, $P < 0.001$; 90°: $z = 3.57$, $P < 0.001$; 120°: $z = 3.54$, $P < 0.001$; 180°: $z = 3.26$, $P = 0.001$); while the ASD group performed above chance on 0° ($z = 3.33$, $P = 0.001$) and 90° ($z = 2.78$, $P = 0.005$); marginally above chance on 120° ($z = 1.83$, $P = 0.067$); and did not perform statistically differently from chance on 180° ($z = 1.34$, $P = 0.178$). Although the ASD group means were lower for all degrees of rotation, no significant differences between the TD and ASD groups were detected for any of the rotation types (0°: Mann–Whitney $U = 103$, $P = 0.39$; 90°: Mann–Whitney $U = 89$, $P = 0.16$; 120°: Mann–Whitney $U = 83$, $P = 0.11$; 180°: Mann–Whitney $U = 86.5$, $P = 0.14$; see Figure 6).

Relationships Among the Tasks

We used Spearman's Rho to detect significant relationships among the various tasks, nonverbal intelligence, and chronological age (Table 3). The strongest relationships were observed among ASL comprehension, ToM, and VPT. By contrast, chronological age was only weakly associated with two of the tasks (VPT and mental rotation), and nonverbal intelligence was not statistically correlated with any of the other variables, though there was a marginally significant association with ASL comprehension ($P = 0.06$). Mental rotation was found to be unrelated to either VPT or language and only weakly related to ToM (see Table 3).

Language-Matched Subsample

We matched the five children with ASD who completed the ASL-RST for language (ASD: $M = 29$, TD: $M = 30.2$; Mann–Whitney $U = 12$, $P = 1.00$) and nonverbal intelligence (ASD: $M = 99.6$, TD: $M = 97.6$; Mann–Whitney $U = 5$, $P = 0.14$) to five of the TD children to determine if the group differences observed could be attributed to the overall lower language of the ASD group. There was no difference between the matched samples on any of the tasks (ToM: Mann–Whitney $U = 12.5$, $P = 0.92$; VPT: Mann–Whitney $U = 9.5$, $P = 0.60$; mental rotation: Mann–Whitney $U = 7.5$, $P = 0.35$; Table 4), suggesting that language deficits in ASD are at the heart of the overall group differences observed in VPT and ToM, even when these constructs are assessed with minimal language.

Finally, we considered whether there was a relationship between performance on the minimally verbal VPT task and performance on the perspective-taking items on the ASL-RST, as would be predicted by Courtin [2000]. Three children with ASD and eleven TD children attained a perfect score on the VPT task, yet only one child in the entire sample received a perfect score on all five perspective-taking items on the ASL-RST. Indeed, of the 14 children who obtained a perfect score on the VPT task, the average error rate on the ASL perspective-taking items was still 58.6%, essentially identical to the overall error rate of 58.8%. There was no significant relationship between performance on the perspective-taking items on the ASL-RST and the VPT task (r_s (20) = 0.28, $P = 0.21$).

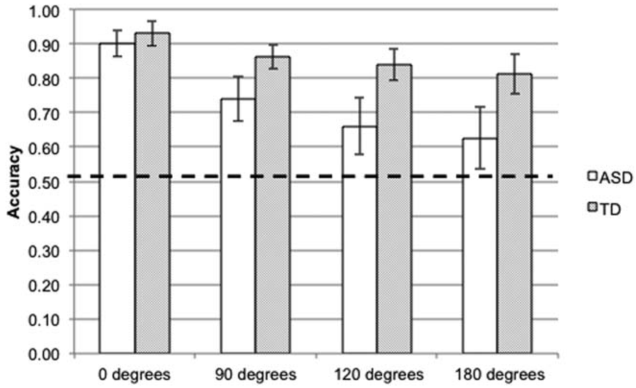

Figure 6. Autism spectrum disorder and TD performance on the mental rotation task, by degree of rotation.

Table 3. Spearman's Rho Correlations Among Variables for All Participants

	1	2	3	4	5
Age	—				
Language	0.17	—			
ToM	0.28	0.45**	—		
VPT	0.35*	0.64***	0.50**	—	
Mental rotation[a]	0.37*	0.22	0.40*	0.23	—
Nonverbal IQ	0.02	0.32†	0.22	0.17	0.29

[a] Correlations between mental rotation and the other tasks were based on the 32 children (14 with ASD and 18 TD) who completed the mental rotation task. All other correlations are based on a total of 35 children (17 with ASD and 18 TD).

†$P < 0.10$; *$P < 0.05$; **$P < 0.01$; ***$P < 0.001$.

Table 4. Language-Matched Subsample (n = 5 Participants in Each Group)

Measure	ASD	TD
TONI-4 standard score	99.6	97.6
Age (years)	11.25	10.25
ASL-RST raw score (of 42)	29	30.2
VPT (4 test items)	2.0	2.8
ToM (4 test items)	3.6	3.6
MR (32 items)	27.4	28.8
ASL-VPT items (5 items)	1.8	1.6

Discussion

With a population of children with ASD who have been exposed to sign language from birth, we replicated the robust finding of a ToM deficit in children with ASD [e.g., Baron-Cohen, Leslie, & Frith, 1985; Happé, 1995]. Compared to mental- and chronological-age-matched TD deaf children, signing children with ASD showed evidence of significant language, ToM, and VPT deficits, but relatively intact mental rotation skills, even when they were tested with minimally verbal measures requiring no expressive language. Despite native ASL exposure from birth, signing children with ASD struggled with both language and ToM in the same way that nonsigning typically hearing children with ASD do. Thus, we found no definitive evidence that the speculated advantage in VPT possibly afforded by early sign language experience [e.g., Courtin, 2000] could be leveraged by children with ASD to overcome ToM deficits.

Two reasons may explain the lack of observed effect of sign language experience on VPT and ToM in children with ASD. First, the social deficits associated with ASD may prevent some children with ASD from gaining full access to the language, thus limiting their experience with the additional perspective-taking that the use of sign language may offer [e.g., Courtin, 2000]. But if and possibly when they do gain access to ASL, they may leverage these perspective-taking opportunities; in our sample, such children were on par with their TD peers in terms of VPT and ToM. However, we cautiously interpret this finding without a comparison to typically hearing children with and without ASD. Indeed we cannot say at this point if sign language exposure leads to a kind of bootstrapping effect, with signing children (both TD and with ASD) showing a kind of advantage over nonsigning children. The native-signing children were not at ceiling on our task, and one-third of the sample did not perform above chance. Further, everyone performed poorly on the language items that required VPT, even children who were at ceiling on the minimally verbal VPT task performed poorly on the items that tap one of the elements of the language that Courtin [2000] argued could support a VPT advantage. Thus, we have to consider that Courtin's [2000] proposal collapses under further scrutiny; children with ASD may not leverage the VPT advantage suggested to be associated with sign language experience because there may in fact be none. Indeed, a recent unpublished study finds no difference between ASL signers and nonsigners on an adult measure of VPT [Secora & Emmorey, 2015].

In other domains of spatial reasoning, however, deaf, signing children show superior performance on spatial tasks compared to age-matched hearing children in a variety of domains, including spatial arrangement and manipulation, mental rotation, and spatial analysis of dynamic displays [Bellugi et al., 1990; Fok & Bellugi, 1986]. These tasks, including mental rotation, are likely mediated by the enhanced abilities to imagine objects [Emmorey, Kosslyn, & Bellugi, 1993] which may be distinct from adopting the perspective of others. Regardless, native sign language experience leads to advantages in spatial cognition. And future work should compare deaf, signing children with and without ASD to age- and IQ-matched hearing children with and without ASD, to identify whether the VPT advantage is present.

The VPT deficit observed in signing children with ASD supports the account that VPT is linked to ToM abilities [Hamilton, Brindley, & Frith, 2009] and is bolstered by the observed statistically equivalent performance of TD and ASD children on our mental rotation task. VPT deficits in children with ASD should be attributed to the social-cognitive skills needed to understand others' viewpoints, not to difficulties with spatial cognition. The children with ASD in our study had intact mental rotation skills, but no advantage over TD children, as some have found [e.g., Muth, Hönekopp, & Falter, 2014; Shah & Frith, 1993; Stevenson & Gernsbacher, 2013]. This lack of advantage, however, may also reflect enhanced mental rotation abilities of our native-signing TD children [Bellugi et al., 1990] rather than a limitation of our sample of native-signing children with ASD relative to nonsigning children with ASD. Children with ASD are able to generate mental images and mentally rotate them, which is one avenue to solving VPT. However, they appear not to apply this skill to the VPT task. We believe that this is due to an under-developed understanding of the mental states of others—not only their thoughts and feelings, but also what they see.

Crucially we replicated the observed relationship between language and ToM development [e.g., De Villiers & Pyers, 2002; Tager-Flusberg & Joseph, 2005]. ASL comprehension scores positively correlated with performance on ToM and VPT tasks, and children with ASD whose language was on par with TD children performed as well as TD children on both. Researchers disagree on which aspects of language support ToM (see Milligan, Astington, & Dack [2007] for a review), and our measure of language cannot distinguish among these alternatives. Experimental training studies with TD children suggest that language plays a fundamental role in fostering children's understanding of the differing mental states and visual perspectives of others [Hale & Tager-Flusberg, 2003; Lohmann & Tomasello, 2003], and the pattern of our results is consistent with these findings.

On the other hand, VPT skills may uniquely shape the acquisition of a visual-spatial language like ASL. ASD and TD children alike, even those children who had a perfect score on the VPT cognitive task, demonstrated poor comprehension (as indicated by a 59% error rate) of ASL structures in which perspective-taking is required. They performed more poorly on these items than on any other grammatical category tested, not mastering these structures until late childhood. Thus linguistic structures with embedded VPT are challenging, late acquired even by typical learners with native exposure to ASL, and may await mature cognitive skills, such as VPT, to master.

For children with ASD acquiring sign, VPT difficulties may result not only in language comprehension problems (as tested in this study), but also in unique production errors: native-signing children with ASD have been shown to reverse the direction of their palm while signing, such that their sign production more closely matches what they see when another person generates that sign rather than how the sign is typically produced, an error pattern that is suggestive of a VPT deficit [Shield & Meier, 2012]. Thus, while impaired VPT abilities appear to be characteristic of ASD, the linguistic manifestations of such a deficit may be different for signed versus spoken languages.

While our results clearly show that native exposure to a signed language does not eclipse the linguistic and cognitive deficits entailed in ASD, it is still unclear whether those children who have sufficient access to ASL then obtain a benefit relative to spoken language. Only a direct comparison between hearing children with ASD and native-signing Deaf children with ASD can determine what role, if any, sign exposure may play in attenuating social deficits. Given that the linguistic structures that Courtin [2000] suggested should support ToM and VPT reasoning in signing children are acquired well after they succeed on these tasks, we think it is unlikely that sign language experience plays a role in the initial development of these aspects of social cognition. It is possible, however, that exposure to, practice with, and eventual mastery of such structures could lead to enhanced performance on both social and spatial cognition tasks much later in development. Lifelong exposure to sign could also aid the development of other social skills known to be impaired in ASD, such as making eye contact, understanding facial expressions, and engaging in episodes of joint attention. Research of this sort will be a powerful tool for better understanding the complex interplay of social and linguistic skills in the development of children with and without ASD.

Taken together, these findings contribute to our understanding of the language and cognitive profile of native-signing children with ASD. In conjunction with previous work on ToM and language development in typically hearing children with and without ASD, this study highlights the pervasive social-cognitive deficits associated with ASD, regardless of type of language exposure. Our findings reinforce the strong relationship between language development, signed or spoken, and the development of ToM. Native sign exposure is important for deaf children to develop a mature understanding of others, but it alone is insufficient to overcome the challenges associated with ASD.

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Conflict of Interest

No conflicts to declare.

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