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Auditory Deprivation Does Not Impair Executive Function, But Language Deprivation Might: Evidence From a Parent-Report Measure in Deaf Native Signing Children

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Deaf children are often described as having difficulty with executive function (EF), often manifesting in behavioral problems. Some researchers view these problems as a consequence of auditory deprivation; however, the behavioral problems observed in previous studies may not be due to deafness but to some other factor, such as lack of early language exposure. Here, we distinguish these accounts by using the BRIEF EF parent report questionnaire to test for behavioral problems in a group of Deaf children from Deaf families, who have a history of auditory *but not language* deprivation. For these children, the auditory deprivation hypothesis predicts behavioral impairments; the language deprivation hypothesis predicts no group differences in behavioral control. Results indicated that scores among the Deaf native signers (n = 42) were age-appropriate and similar to scores among the typically developing hearing sample (n = 45). These findings are most consistent with the language deprivation hypothesis, and provide a foundation for continued research on outcomes of children with early exposure to sign language.

Success in the classroom and in the broader world depends on more than mere knowledge; it also depends on a constellation of cognitive skills that guide both learning and behavior, known as executive function (EF). While the particular inventory of EFs may be described differently by different investigators (see Diamond, 2013, for a three-factor model), there is broad consensus that two central components of EF are behavioral regulation and metacognition. Behavioral regulation enables goal-directed behavior, especially in the face of competing desires, emotions, or impulses that might interfere with achieving the goal. The ability to switch to a new goal when appropriate is another important aspect of behavioral regulation. Metacognition is what allows children and adults to effectively manage their cognitive resources. Its subdomains include the ability to retain and manipulate information in memory, think ahead to solve problems, and maintain focus through controlled attention. Children are not born with adult-like EF skills; rather, they develop as the child matures. Early EF abilities strongly predict later EF abilities (Eigsti et al., 2006; Mischel, Shoda, & Peake, 1988; Mischel et al., 2011); delay or disturbance in the development of execution function has negative consequences for both academic and social outcomes (Blair & Razza, 2007; Bull, Espy, & Wiebe, 2008; Gathercole, Pickering, Knight, & Stegmann, 2004; Henry, Messer, & Nash, 2014; McClelland et al., 2007; Miller, Müller, Giesbrecht, Carpendale, & Kerns, 2013; Moffitt et al., 2011; Ponitz, McClelland, Matthews, & Morrison, 2009; Shoda, Mischel, & Peake, 1990; Welsh, Nix, Blair, Bierman, & Nelson, 2010).

Children with hearing loss are frequently observed to be at risk for problems in EF. As far back as Pintner and Paterson (1917), researchers have noticed that deaf children generally do not perform as well as their hearing peers on a variety of tasks that would

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now fall under the umbrella of EF. These differences between deaf and hearing children have been reported throughout the 20th century (Lesser & Easser, 1972; Myklebust, 1960; Quittner, Smith, Osberger, Mitchell, & Katz, 1994; Reivich & Rothrock, 1972; Smith, Quittner, Osberger, & Miyamoto, 1998) and have survived into the 21st century, despite the advent of newborn hearing screening and improved hearing technologies such as cochlear implants (Barker et al., 2009; Beer, Kronenberger, & Pisoni, 2011; Beer et al., 2014; Castellanos et al., 2014; Conway, Pisoni, Anaya, Karpicke, & Henning, 2011; Dammeyer, 2010; Figueras, Edwards, & Langdon, 2008; Harris et al., 2013; Hintermair, 2013; Horn, Davis, Pisoni, and Miyamoto, 2005; Jiménez-Romero, 2015; Kronenberger, Beer, Castellanos, Pisoni, & Miyamoto, 2014; Kronenberger, Pisoni, Henning, & Colson, 2013; Pisoni, Conway, Kronenberger, Henning, & Anaya, 2010; Quittner, Leibach, & Marciel, 2004, Remine, Care, & Brown, 2008).

Measuring the complex domain of EF is a challenging task. First, researchers must choose from a diverse array of standardized and experimental assessments. A second challenge is to determine whether differences detected under experimental conditions translate meaningfully to real-world contexts. In response to these challenges, Gioia, Isquith, Guy, and Kenworthy (2000) developed the Behavioral Rating Inventory of Executive Function, or "BRIEF." The BRIEF is an 86-item questionnaire designed to quantify the prevalence of concerning behaviors that relate to different aspects of EF. It is designed for use with children between the ages of 5 and 18 (preschool and adult versions are also available). Separate forms are designed for parents and teachers. In either case, an adult who knows the child well indicates whether certain behaviors were never, sometimes, or often a problem over the past 6 months. The BRIEF yields scores at three levels. At the broadest level, the Global Executive Composite (GEC) score reflects a child's overall EF development. It, in turn, is composed of two sub-parts: a Behavioral Regulation Index (BRI) and a Metacognition Index (MI). Each of these is composed of several subscales, as listed in Figure 1. We will refer to the GEC, BRI, and MI as "summary indices," and to the eight basic scales as "subscales." Raw scores on each subscale and index are converted to standardized T scores based on normative data from a large (n = 1,419) and representative sample of typically developing children, stratified by age and sex. T scores have a mean of 50 and standard deviation (SD) of 10; higher scores indicate increased incidence of problematic behavior.

Next, we summarize several recent studies that have used the BRIEF to investigate EF in deaf children. Although there is always a risk in relying on a single instrument as a dependent measure (especially a subjective checklist), the BRIEF is both widely used and well-validated, and presents the advantage of examining behavior "in real life". In line with many previous reports, the following BRIEF-based studies are consistent in finding deficits in EF among deaf children relative to hearing peers and/or test norms.

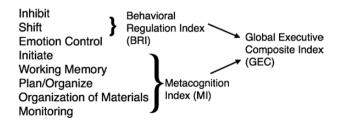


Figure 1. From left: the eight clinical subscales of the BRIEF, which form two summary indices, which combine into a single composite score. Norms for each stage are available.

Pisoni et al. (2010) present parent-report data showing that a group of cochlear implant (CI) users (n = 19) had significantly higher mean T scores than children with typical hearing (n = 30) on all three summary indices and on six of the eight subscales. In addition, the means in the CI group were above the expected value of 50, although whether they were clinically elevated is unknown (statistics were not reported).

Similarly, Beer et al. (2011) present parent-report BRIEF data from 45 children with CIs, whose means were significantly higher than the expected value of 50 on the Inhibit and Working Memory subscales as well as the Behavioral Regulation Index. They also computed the proportion of participants scoring in or above the elevated range (+1 SD; \geq 60), and compared that proportion against the 16% that would be expected to fall in that range in a truly normal distribution. The risk of scoring in the elevated range was higher than chance on all but one subscale and on all three summary indices, ranging from 13% to 31% (tests of statistical significance were not reported).

Kronenberger, Colson, Henning, and Pisoni (2014) administered the BRIEF to 49 CI users and 51 hearing controls aged 7–17.¹ Of the CI users, 92% reported an auditory-oral emphasis in their language training/background/experience. The researchers found significantly higher mean T scores in CI users than in the hearing controls on six of the eight subscales. They did not report any of the three summary indices; however, given that the summary indices are linear combinations of the subscales, it seems likely that the summary indices would also show mean differences. In addition, they found that the CI users were at significantly higher relative risk ratios (risk of T scores \geq 60) on six of the eight subscales than the hearing controls.

Hintermair (2013) collected BRIEF ratings from teachers of 69 deaf students at German schools where deaf students are integrated into a majority hearing classroom ("mainstreaming") and from teachers of 145 students at special schools for the deaf in Germany. Roughly 25% of these participants had CIs. In the absence of German norms, T scores were computed using the American norms. Compared to BRIEF norms, the incidence of clinically significant scores (+1.5 SD; ≥65) was high across the board. The mainstreamed deaf students were at significantly greater than chance risk on four of eight subscales and two of three summary indices. The students at specialized schools for the deaf were at significantly increased risk (relative to chance) on all eight subscales and all three summary indices. These children were at increased risk on several BRIEF subscales relative to those attending mainstream schools; however, the results leave open the question of whether education at special schools leads to more problematic behavior, or whether students with more underlying behavioral problems are more likely to be sent to special schools for the deaf. More relevant for present purposes is the teacher-report finding that 89% of the students were oral-only language users. The remaining 11% were described as using "spoken and sign language"; however, it is unclear whether this means a natural sign language such as DGS,² or sign-supported German. There also may have been a mix; 11% are reported to have deaf parents, but it is not stated whether these are the same 11% who use sign. This suggests that the vast majority of participants in this study, as in the previous three reviewed above, were not exposed to a natural sign language from birth.

In sum, all four studies found clear evidence of EF deficits in deaf children, significant enough to be noticed by parents or teachers, and often approaching or exceeding levels deemed to be clinically significant. These problems were attested in deaf children with and without CIs across a range of ages from preschool through adolescence in different educational settings and cultures, and are consistent with other reports of behavioral problems in CI users based on various other assessment tools such as the Child Behavior Checklist (Achenbach & Rescorla, 2000), Parenting Stress Index (Abidin, 1995) or Strengths and Difficulties Questionnaire (Goodman, 1997), among others (e.g., Barker et al., 2009; Dammeyer, 2010; Jiménez-Romero, 2015). Given the critical role that EF plays in long-term academic and social outcomes, it seems likely that deficits in these skills account for at least some of the difficulties that many deaf children experience in both academic and social development. Therefore, reducing these deficits is expected to result in more positive outcomes.

The first step toward addressing executive dysfunction must be understanding its origins. One prevalent view is that deafness itself has deleterious effects on the development of EF. For example, "... deafness and degraded auditory experience may affect not only speech and language skills, but also other neurocognitive functions" (Beer et al., 2011, p. S89, italics added). Similarly, "... the development of EF is critically dependent on exposure to sequential signals from sensory (particularly auditory) experience" (Kronenberger, Beer, et al., 2014, p. E6, italics added). More recently, Kral, Kronenberger, Pisoni, and O'Donoghue (2016) have argued for viewing deafness as a "connectome disease." This perspective emphasizes that "Loss of hearing has cascading neurological and neurocognitive effects: because no part of the brain works in isolation, loss of a sensory system such as hearing also affects other functions, including higher order neurocognitive tasks" (Kral et al., 2016, p. 614, italics added).

We refer to this view as the "auditory deprivation hypothesis." Proponents of this view emphasize the impact of low-level perceptual (i.e., auditory) experience on the development of higher-level cognitive skills, including EF. Auditory experience has been implicated in the development of other cognitive skills as well; for example, Conway, Pisoni, and Kronenberger's (2009) auditory scaffolding hypothesis attributes deficits in implicit sequence learning to auditory deprivation, and Ulanet, Carson, Mellon, Niparko, and Ouellette (2014) have extended those claims to other sequential processing tasks.

The focus of the current study is on whether auditory deprivation leads to parent-reported problems in behaviors related to EF. If this view is correct, then a logical intervention would be to increase the child's exposure to auditory input through hearing technology. However, the evidentiary basis for this view is complicated by confounds in the studies reviewed above, and indeed in nearly all of the relevant literature. One possible confound is the observation that behavioral outcomes can vary depending on the etiology of deafness, not just its degree. Oberg and Lukomski (2011) report BRIEF data (both parent and teacher ratings, which were highly correlated) from 22 deaf children (age 5-18) that attended a signing school for the deaf. As with the studies reviewed above, they too found significantly elevated mean T scores for the group as a whole. But unlike most previous studies, they conducted a subanalysis to compare children with hereditary and nonhereditary deafness. Their small sample of children with hereditary deafness (n = 5) had normal or better T scores on all subscales and summary indices; the group-level deficits were driven entirely by the group with nonhereditary deafness (n = 17). The results from this small sample are corroborated by Hintermair's (2013) much larger sample (56 genetic vs. 108 unknown), in which teacher ratings revealed that "students with unknown cause [of deafness] have higher [i.e. worse] scores than students with a genetic background" (p. 352). These findings are consistent with Kral et al.'s (2016) connectome perspective insofar as both accounts emphasize how the auditory system is not isolated from the rest of the brain. However, whereas Kral et al. propose that deafness is among the causes of cognitive disturbances, the etiology hypothesis proposes that both deafness

and cognitive disturbances are themselves *consequences* of developmental anomalies caused by genetics, pathogens, medication, etc. We revisit these ideas in the general discussion.

In the present study, we focus primarily on a second confound that is present in the previous research: early language deprivation. Roughly, 95% of deaf children lack exposure to natural human language (spoken or signed) in their earliest months/years of life (Mitchell & Karchmer, 2004). Because these children comprise the bulk of previous research (Beer et al., 2011; Kronenberger, Beer, et al., 2014; Pisoni et al., 2010), it is possible that the observed deficits in EF have less to do with auditory deprivation and more to do with language deprivation. Thus, the relative benefits of hereditary over nonhereditary deafness (Hintermair, 2013; Oberg & Lukomski, 2011) may be a covert effect of having Deaf parents who use sign language at home. Regrettably, neither study provides detailed information about participants' language background.

Several previous researchers have noted a positive correlation between EF and language skills in deaf populations (Figueras et al., 2008; Horn et al., 2005; Houston et al., 2012; Kronenberger, Colson, et al., 2014; Kronenberger, Pisoni, Harris, et al., 2013; Remine et al., 2008). However, the interpretation of this relationship has varied. Some interpretations (e.g., Horn et al., 2005; Houston et al., 2012; Kronenberger, Colson, et al., 2014; Kronenberger, Pisoni, Harris, et al., 2013) are consistent with the auditory deprivation hypothesis; under such a view, weak language skills are caused in part by weak cognitive skills in general, which are in turn attributable, at least in part, to a lack of auditory experience. Other interpretations (e.g., Figueras et al., 2008; Remine et al., 2008) argue that performance on tests of EF may benefit from language skills such as self-talk and covert rehearsal. On this view, weak language skills in deaf populations may explain their poor EF, with influences between language and EF potentially flowing in both directions.

If poor EF and its resulting behavioral consequences are due at least in part to language deprivation rather than to auditory deprivation, then these problems could potentially be averted via early exposure to a natural sign language such as American Sign Language (ASL). This is consistent with Dammeyer's (2010) finding that teacher-rated problems in psychosocial adjustment were related to language skills in deaf children, where good oral or signing skills were associated with reduced risk of behavioral problems. Providing early access to sign language remains controversial (see Napoli et al., 2015, and responses thereto) for both theoretical and practical reasons. Before adjusting the clinical standard of care, it is important to identify empirical evidence that supports any such changes. The present study takes an important step in this direction by studying Deaf children who are exposed to a natural sign language (such as ASL) from birth by their Deaf signing parents. We refer to these children as deaf³ native signers. We analyze the BRIEF scores for Deaf native signers in relation to children with typical hearing and to scale norms/chance. We acknowledge that Deaf native signers constitute only a minority of the population of deaf children (Mitchell & Karchmer, 2004); their inclusion here is motivated primarily by the fact that they afford the opportunity to distinguish between the auditory deprivation hypothesis and the language deprivation hypothesis, which make contrasting predictions where deaf native signers are concerned. The auditory deprivation hypothesis predicts that deaf native signers should be at least as impaired as deaf nonsigners if not more so, given their longer duration of deafness and greater extent of auditory deprivation (because many Deaf native signers do not use hearing technology). Conversely, the language deprivation hypothesis predicts that these children should not be impaired.

The results of the present study address hypotheses regarding the underlying cause of EF problems in deaf children, thereby helping to focus the search for effective solutions for intervention and—ultimately—prevention of these problems.

Method

Participants

Participants were recruited as part of a larger study of cognitive development in deaf and hearing children. Deaf participants (n = 42) came from signing schools and local contacts in Connecticut, Washington, DC, Texas, Minnesota, and Maryland. Hearing participants (n = 45) came from schools and local contacts in Connecticut and California. Children with additional medical diagnoses (e.g., autism, Down syndrome, cerebral palsy, etc.) were excluded. Children with diagnoses of attention deficit hyperactivity disorder (ADHD) or learning disability were considered eligible, because difficulties in EF could plausibly be a cause rather than a consequence of these diagnoses.

Comparison of samples to each other

Demographic information for both groups is listed in Table 1. The deaf and hearing samples did not differ on age (p = .76), sex (p = .31), and which caregiver completed the questionnaire (p = .22). Race and ethnicity were reported by 70% of the deaf sample and 74% of the hearing sample. Based on the reported information, the samples did not differ (p = .66). Socioeconomic status (SES), as measured by the primary caregiver's highest level of education, did not differ between groups (Fisher's exact test, p = .96). There was a nonsignificant trend for hearing children to live in zip codes with a higher median income⁴ [t(81) = 1.64, p = .10]. Meanwhile, the deaf children lived in zip codes that were significantly more urban⁵ [t(81) = 8.25, p < .001]; this is because most of the hearing children came from the rural communities surrounding the University of Connecticut.

Comparison of samples to BRIEF norms for parent form

We compared the present participant groups against the BRIEF's norming sample (n = 1,419, ages 5–18 years) on the dimensions reported in the manual: child's sex, which caregiver completed the form, race/ethnicity, SES, and urban density. There were no significant differences in sex (deaf: p = .56, hearing: p = .40), or

in which caregiver completed the questionnaire (deaf: p = .82, hearing: p = .13).

Statistically comparing measures of race/ethnicity, SES, and urban density against these norms is less straightforward, as different measures were used. The normative sample was 80.5% White, 11.9% African-American, 3.1% Hispanic, 3.8% Asian or Pacific Islander, and 0.5% Native American. The present samples differ from these norms to some degree (deaf: p = .08, hearing: p = .02). African-American participants appear under-represented in both samples, although this may depend in part on how participants reporting more than one racial/ethnic identity were classified. However, the BRIEF manual reports that race/ ethnicity had no significant effect on scores.

The normative sample's SES was measured with the Hollingshead Code, which combines education, occupation, and income. The resulting distribution was approximately normal, centered on middle-middle class, with 3% upper-class and 7.4% lower-class or unassigned. The most direct measure of SES in the present study was parental education. The majority of primary caregivers held a bachelor's degree or more (deaf: 78%, hearing, 79%), suggesting that these families were likely to be at least middle class. Only 6% of parents of the deaf participants and 2% of the hearing participants had a high school diploma or less. Only five children in our sample lived in zip codes where median income exceeded \$100,000, suggesting that few were upper class. (All five of these children were in the deaf group; three were in a single family, and all lived in the environs of Washington, DC, where the cost of living is also high.) Thus, we believe the present samples to be comparable to the normative sample on SES. Furthermore, the BRIEF manual indicates that neither parental education nor SES accounted for a substantial amount of variance on BRIEF scores in the norming sample.

The urban density of the normative sample is described as 26.5% urban, 59% suburban, and 14.5% rural. Unfortunately, no operational definitions of these terms are available, making it difficult to determine how our sample compares. We note also that the manual does not report any analysis of the relationship between urban density and BRIEF scores.

In sum, the deaf and hearing samples in the present study are largely comparable to each other and do not differ from the normative BRIEF sample in any way that is known to impact scores.

Table 1.	Demographic	characteristics	of the	participants
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	Deaf native signers, N = 42	Hearing controls, $N = 45$	F/X^2	р
Mean age: yr; mo	8;03	8;04	0.09	.76
(SD)	(2;03)	(1;10)		
Range	5;01–12;10	5;06–12;11		
Sex (female:male)	26:16	23:22	1.03	.31
Hearing status	Severe or profound congenital deafness	No known hearing impairment	n/a	n/a
Language experience	Exposure to natural sign language from birth; variable speech emphasis at home and school	Exposure to spoken language from birth; mono- lingual: 38/45, bilingual/heritage: 7/45	n/a	n/a
Hearing technologyª	I: 34, II: 6, III: 2	n/a	n/a	n/a
Primary caregiver education level ^b	I: 1, II: 2, III: 6, IV: 8, V: 24	I: 0, II: 1, III: 8, IV: 9, V: 25	0.008	.93
BRIEF completed by	Mother: 37, father: 4, other guardian: 1	Mother: 33, father: 9, other guardian: 3	3.05	.22
Race/ethnicity	White: 26, Black: 1, Asian: 4, native American: 0, unknown/other: 11	White: 26, Black: 0, Asian: 3, native American: 2, unknown/other: 9	0.004	.67

Note. SD, standard deviation.

^aUse of hearing technology: I = none, or hearing aids used less than "sometimes"; II = hearing aids used at least "sometimes"; III = cochlear implants. ^bEducation level: I = less than high school, II = high school or GED, III = some college or associate's degree; IV = bachelors degree; V = some graduate school or advanced degree. Most importantly for present purposes, all deaf children in this sample were exposed from birth to a natural sign language used by at least one Deaf signing parent. (We would have included deaf children with hearing parents who knew ASL before the participating child was born; however, no such children presented themselves.) All deaf children attended special schools for the Deaf; unlike in Hintermair (2013), these schools emphasized a bilingual approach including ASL and English.

Qualified Deaf and hearing children were identified either by direct response from parents who viewed recruitment materials, or by school administrators. Inclusion criteria were verified by email; a more thorough background form was distributed upon enrollment, along with consent documents and the BRIEF questionnaire. We distributed 91 forms and received 87 back (95.6%). (Table 1 lists information for only those children for whom BRIEF data are available.)

Study procedures were approved by the Institutional Review Boards of the University of Connecticut and of the participating schools.

Scoring

Following standard scoring procedures, raw scores for each of the BRIEF's eight subscales and three summary indices were converted to T scores, with a mean of 50 and SD of 10. T scores are normed as a function of a child's sex and age, allowing direct comparisons across children. T scores ≥ 60 are considered elevated, and T scores ≥ 65 are considered clinically significant.

Results

Following the studies reviewed in the introduction, we present two types of analysis. First, we compare the mean T scores for the Deaf participants to those of the hearing participants and to BRIEF norm scores (Figure 2). We then analyze the relative risk of undesirable scores in the Deaf participants relative to the hearing participants and to the rate expected by chance based on the norming sample.

Mean T Scores

Figure 2A shows the mean T scores of Deaf and hearing participants on each of the three summary indices, with higher scores reflecting greater behavioral problems. Figure 2B shows the mean T scores for the three subscales that comprise the Behavioral Regulation Index, while Figure 2C shows the five subscales that comprise the Metacognition Index. Neither group's mean exceeded the expected value of 50 on any subscale or summary index. The hearing group's confidence interval fell below 50 on four subscales and two summary indices, suggesting that we may have tested an unusually well-behaved group of hearing children. Numeric values for mean T scores, standard deviations, and between-group comparisons are given in Table 2.

Following previous studies (Beer et al., 2011; Kronenberger, Beer, et al., 2014; Pisoni et al., 2010), we used separate t-tests to compare the deaf and hearing means; contrary to previous findings, no differences were significant, although the hearing group scored marginally better on two subscales: Working Memory [t(85) = -1.89, p = .06] and Plan/Organize [t(85) = -1.91, p = .06]. These effects do not survive correction for multiple comparisons.

Rates of Elevated Scores

If we focused only on group means, we would risk overlooking potentially important individual differences. Beer et al. (2011) and Kronenberger, Beer, et al. (2014) report the proportion of their samples that fell into the "elevated" range (+1 SD, or T score \geq 60). In a normal distribution, no more than 16% of a sample should score in this range. Table 3 therefore gives the percentage of the sample whose T scores are at or above 60, for both groups. Because the norming distribution is only approximately normal, percentiles are provided as well; we report the percent of the sample scoring at or above the 84th percentile. To determine whether these observations are significant, we calculate relative risk ratios. Relative risk is simply the ratio of observed risk in two groups. Values greater than 1 indicate greater risk in the first (numerator) group, while values less than 1 indicate greater risk in the comparison (denominator) group. Significance is measured by constructing a confidence interval around the ratio; if the confidence interval includes 1, the risk ratio is not significant.

Table 4 presents relative risk ratios for the Deaf native signers (always numerator) in the present study relative to the hearing participants and to chance. The Deaf native signers do show significantly greater risk of elevated scores than the hearing participants on the Inhibit and Working Memory subscales. However, those differences are driven, at least in part, by the surprisingly low rates of elevated scores in the hearing group (Inhibit: 4.4%,

Table 2. Mean T score and SD for each BRIEF subscale and summary index, for the participants in the present study

	Mean T score (SD)			
	Deaf native signers	Hearing controls	t(85)	p^{a}
Inhibit	50 (10.7)	46.9 (7.9)	1.55	.12
Shift	48 (8.6)	49.4 (11.5)	0.67	.50
Emotion Control	46.9 (9.5)	48.1 (10.1)	0.57	.58
Initiate	49.8 (11.2)	47.9 (7.2)	0.95	.34
Working Memory	49.6 (9.9)	46.2 (7.1)	1.89	.06
Plan/Organize	49.4 (9.1)	46.1 (7.3)	1.90	.06
Organization of Materials	48.6 (10.2)	48.7 (8.3)	0.10	.94
Monitor	47.7 (10.6)	45.8 (7.9)	0.99	.32
Behavioral Regulation Index	48 (9.8)	47.7 (9.7)	0.17	.86
Metacognitive Index	48.6 (10)	46.7 (8)	0.96	.34
Global Executive Composite	48.5 (10)	46.7 (7.7)	0.94	.35

Note. SD, standard deviation.

^aUncorrected alpha is .05. When adjusting for eight multiple independent comparisons (one for each subscale), alpha is .006 by both Bonferroi and Sidak corrections.

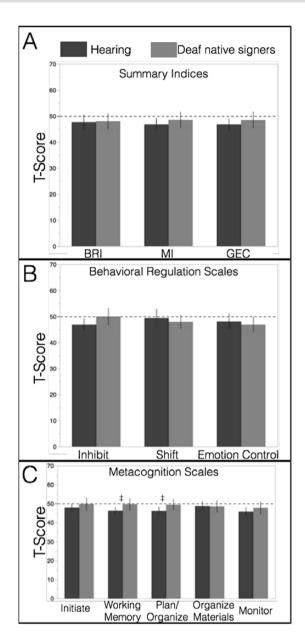


Figure 2. Mean T scores on summary indices and clinical subscales for deaf native signers and hearing controls. Panel A shows the three summary indices: the global executive composite is a linear combination of the Behavioral Regulation Index (BRI) and the Metacognitive Index (MI). Panel B shows the three subscales that comprise the BRI. Panel C shows the five subscales that comprise the MI. Neither group's mean score ever exceeds the expected mean of 50 (dashed line). Error bars represent 95% confidence interval. Double-daggers indicate marginally significant differences between groups, uncorrected for multiple comparisons.

Working Memory: 2.2%). The Deaf native signers did not significantly differ from chance on any summary indices or subscales, including Inhibit and Working Memory. Although the Deaf native signers were at numerically greater than chance risk on these two subscales, they were at numerically *less* risk than would be expected by chance on five of the eight subscales, and on all three summary indices.

Rates of Clinically Significant Scores

Whereas Beer et al. (2011) and Kronenberger, Beer, et al. (2014) report rates of "elevated" scores (+1 SD), Hintermair (2013)

reports rates of "clinically significant" scores (+1.5 SD). Repeating the above analysis with this criterion reveals no significant differences between Deaf native signers and the hearing sample, nor between either sample and chance.

Discussion

Following several previous studies of EF in deaf children, we used the BRIEF to obtain information from parents about their children's executive function. Whereas all previous studies reported significant behavioral problems in deaf children, the Deaf native signers in the present study had mean scores indicative of healthy, normative, age-typical EF. Specifically, their scores did not differ from the typical mean of 50, and they did not have significantly increased rates of either elevated or clinically significant scores relative to what would be expected in a normal distribution.

On the other hand, comparisons of the Deaf native signers to the present hearing controls revealed that the Deaf native signers had greater risk of elevated (but not clinically significant) scores for the Inhibit and Working Memory subscales. The Deaf native signers' mean T score was also marginally higher than the hearing mean for the Working Memory and Plan/Organize subscales. Inhibitory control and working memory are domains in which previous studies of nonsigning deaf children have also noted difficulties (e.g., Figueras et al., 2008; Mitchell & Quittner, 1996; Pisoni & Cleary, 2003). If Deaf native signers are truly impaired on these skills, it would suggest that inhibitory control and working memory are domains where auditory experience does play a direct role. However, the Deaf native signers' rate of elevated scores was not significantly greater than chance on either subscale, nor were their means significantly above the expected T score of 50. Of the 11 Deaf native signers who scored within the elevated range, 5 had T scores of 60: the minimum score that can be considered elevated. The group differences were driven at least in large part by particularly low scores in the hearing participants. Therefore, the present results are equivocal in this regard. Fortunately, a recent study by Marshall et al. (2015) provides additional insight into the impact of auditory deprivation and language deprivation on working memory. Using performance-based measures of nonverbal working memory, they found that deaf children who also experienced a period of language deprivation scored significantly worse than hearing children, but Deaf children with exposure to sign language from birth did not. The data from Marshall et al. (2015) support the hypothesis that language deprivation has a greater adverse impact on working memory than auditory deprivation does.

One concern is that if our recruitment methods yielded a sample of hearing participants whose scores were better than the norming sample, the same might be true of our sample of Deaf native signers. Under this view, the resemblance between the Deaf native signers' T scores and scale norms might be specious, because our sample may not be truly representative of the population of Deaf native signers. We cannot exclude this interpretation on the basis of the current data; however, we note that these are among the first data on BRIEF scores among Deaf native signers, and the largest such sample to date. As such, they represent an important step toward more fully characterizing the population. In addition, the cognitive and behavioral profile that the present scores suggest is thoroughly consistent with a large body of literature documenting typical development in other domains among Deaf native signers, as cited below.

Interestingly, when we look at rates of unusually low (i.e., good) scores (T scores \leq 40), we find no significant differences between the groups, with a numerical trend favoring the Deaf

Table 3. Proportion of sample with T scores \geq 60 (+1 SD)

	Deaf native signers, T scores ≥ 60 (%)	Deaf native signers, percentiles ≥ 84 (%)	Hearing controls, T scores ≥ 60 (%)	Hearing controls, percentiles ≥ 84 (%)	Chanceª (%)
Inhibit	26.2	28.6	4.4	4.4	16
Shift	11.9	11.9	15.6	20.0	16
Emotion Control	7.1	11.9	13.3	13.3	16
Initiate	16.7	21.4	4.4	6.7	16
Working Memory	26.2	26.2	2.2	2.2	16
Plan/Organize	11.9	11.9	4.4	6.7	16
Organization of Materials	9.5	4.8	6.7	6.7	16
Monitor	9.5	9.5	8.9	8.9	16
Behavioral Regulation Index	11.9	11.9	11.1	11.1	16
Metacognitive Index	11.9	14.3	4.4	4.4	16
Global Executive Composite	14.3	9.5	8.9	8.9	16

Note. In a normal distribution, 16% of the sample would be expected to score in this range. Because BRIEF T scores are not perfectly normal, we also list the percentage of the sample that scored above the 84th percentile, which should also not exceed 16%. SD, standard deviation.

^aIn a normal distribution, only 16% of a sample should score more than 1 SD above the mean. Because empirical data are whole-integer counts (i.e., the number of participants scoring more than 1 SD above the mean, or above the 84th percentile of the norming sample), chance can also be based on the nearest whole-integer count to 16% (ASL: 7/42 = 16.67%; Hearing: 7/45 = 15.56%). No results change under this approach.

 Table 4. Relative risk in deaf native signers versus hearing participants and chance

	ASL vs. hear- ing	ASL vs. chance
Inhibit	5.89*	1.57
Shift	0.77	0.71
Emotion Control	0.54	0.43
Initiate	3.75	1.00
Working Memory	11.79*	1.57
Plan/Organize	2.68	0.71
Organization of Materials	1.43	0.57
Monitor	1.07	0.57
Behavioral Regulation Index	1.07	0.71
Metacognitive Index	2.68	0.71
Global Executive Composite	1.61	0.86

Note. Asterisk indicates significance based on 95% confidence interval around risk ratio = 1.

group. It is important to note that clinical instruments such as the BRIEF are designed to measure problematic behavior; the absence of problems does not necessarily mean the presence of excellent behavior. Nevertheless, these results suggest that there may be greater heterogeneity within the deaf population, even within a sample consisting only of Deaf native signers. A full investigation of this finding is beyond the scope of the present study; however, we note that this observation only came to light once we analyzed not only potential deficits but also potential *benefits* associated with deafness (a perspective sometimes known as "Deaf gain," in contrast to "hearing loss"). Future research regarding EF in deaf and hearing populations should consider testing for and reporting strengths as well as weaknesses.

The present findings stand in contrast to the results of all five previous studies that have used the BRIEF to gauge the development of EF in deaf children. Despite typical betweenstudy variation in recruitment/sampling methods, the four American studies (Beer et al., 2011; Kronenberger, Beer, et al., 2014; Oberg & Lukomski, 2011; Pisoni et al., 2010) all reported significantly worse scores in their samples of deaf participants, measured by higher mean scores and/or increased rates of elevated scores, compared to test norms, to chance, and/or to a sample of typically developing hearing children. The German study (Hintermair, 2013) reported similar findings, although the methods in this study were substantially different from the other four. (In addition to comparing students in Germany to norms from the United States, Hintermair used teacher report rather than parent report, and included students with additional comorbid diagnoses, and more mild hearing loss.) The central findings, however, were broadly similar: deaf students had significantly worse scores relative to the BRIEF norms. This effect was especially pronounced for students who attended special (oral) schools for the deaf, but was still present for students attending mainstream schools.

One explanation of the underlying cause of these problems has been auditory deprivation. For example, Kral et al. (2016) argue that congenital deafness should be viewed as a connectome disease, in which early sensory deprivation has cascading effects that extend to higher-order cognitive skills, including EF. This account makes the straightforward prediction that Deaf native signers should be equally at risk of developing problems with EF, because they too experience auditory deprivation. In fact, their predicted risk might be even greater, given that the majority of the Deaf native signers we tested do not regularly use hearing technology. However, this prediction is not supported by the results of the present study. This leads us to seek alternative hypotheses that more successfully account for the data.

One plausible alternative hypothesis is that early language deprivation leads to disturbances in EF; or, conversely, that early language exposure in the current sample of Deaf native signers is a protective factor. Despite their deafness, the present participants had age-appropriate exposure to language, since they were born and raised in homes where a natural sign language was used from birth.⁶ By "natural sign language" we mean a language that arises spontaneously within a community of Deaf users and is acquired and transmitted across generations, evolving its own phonology, morphology, and syntax naturally along the way. This is crucially different from other types of signing systems (e.g., Signed English, Makaton), which often are invented by an individual or group as a way to represent the structure of an existing spoken language on the hands. Even when invented systems borrow lexical items from natural sign languages, they are missing many crucial aspects of natural sign language grammar that are better suited for communicating in

a manual modality (Supalla, 1991). Similarly, signing and speaking at the same time (as is common in Total Communication settings) typically results in signed output that lacks the rich linguistic structure that characterizes natural sign languages (Marmor & Petitto, 1979; Wilbur & Petersen, 1998), with the result that the signed content fails to faithfully convey the message (Tevenal & Villanueva, 2009).

The present results are in alignment with a number of other studies that also find healthy cognitive and psychosocial development among Deaf children with early exposure to sign language, including sustained attention (Dye & Hauser, 2014), theory of mind (Courtin, 2000; Meristo & Hjelmquist, 2009; Meristo et al., 2007; Peterson & Siegal, 1999, 2000; Russell et al., 1998; Schick, De Villiers, De Villiers, & Hoffmeister, 2007; Woolfe, Want, & Siegal, 2002), impulse control (Harris, 1978), IQ (Braden, 1987; Kusché, Greenberg, & Garfield, 1983), working memory (Marshall et al., 2015), and quality of life (Kushalnagar et al., 2011). These references are illustrative examples; an exhaustive review is beyond the scope of this article. The key point is that it has long been observed that when profoundly deaf children have early exposure to sign language input, cognitive and behavioral problems are typically not observed.

We propose two reasons—one practical, one theoretical why early acquisition of sign language might protect deaf children from developing behavioral problems. The practical reason is that children who lack complete command of a language may exhibit undesirable behavior because they are unable to convey their needs, intentions, and desires, and may not understand what other adults or children are asking of them. They also have fewer opportunities for incidental learning by observing communication between others, which would otherwise contribute to their growing understanding of social/cultural standards of behavior. Deaf (and hearing) children who grow up with signing friends and family are not vulnerable to these same problems.

The theoretical reason is inspired by several previous studies that have found that measures of EF are frequently related to measures of spoken language in nonsigning deaf children (Figueras et al., 2008; Kronenberger, Colson, et al., 2014; Kronenberger, Pisoni, Harris, et al., 2013; Luckner & McNeill, 1994; Remine et al., 2008). The majority of these studies acknowledge that language delay, not just auditory deprivation, could play a crucial role. They raise the possibility that during typical development, exposure to the patterns that characterize natural language (e.g., rapid temporal processing, longdistance dependencies, rich hierarchical structure, abstraction from sensory perception to mental representation, etc.) might in fact train neural circuits that are used not only in the service of language processing, but also in nonlinguistic cognitive domains. We agree with this view; however, we hypothesize that sign language would also provide adequate exposure to these types of patterns. A large body of linguistic, psycholinguistic, and neurolinguistic research has firmly established that the only substantive difference between spoken and sign languages is in the sensory signal itself (for reviews, see Sandler & Lillo-Martin, 2006; Emmorey, 1993; Campbell, MacSweeney, & Waters, 2008, respectively). The suggestion that properties of the auditory signal (e.g., its higher temporal resolution relative to vision) is critical to the development of EF (Beer et al., 2011; Conway et al., 2009; Kral et al., 2016; Kronenberger, Beer, et al., 2014; Kronenberger, Colson, et al., 2014; Kronenberger, Pisoni, Harris, et al., 2013; Kronenberger, Pisoni, Henning, et al., 2013) is largely inconsistent with the current findings of intact EF in the Deaf native signers in the present study, with the possible exception of inhibitory control and working memory. An alternative is the

language scaffolding hypothesis, in which the abstract structure of language—spoken or signed—scaffolds the development of other cognitive skills. This view is more consistent with the current data, and merits further investigation.

The potentially reciprocal relationship between language and EF skills deserves further scrutiny. Language skills have been argued to influence performance on EF tasks: self-talk, verbal rehearsal, and other language-based problem solving or self-regulation strategies provide a means of organizing goal-directed behavior and resisting both internal and external distractions (Figueras et al., 2008; Remine et al., 2008; Singer & Bashir, 1999; Vygotsky, 1962). Children with less than full mastery of a native language are therefore less able to deploy these linguistic tools in the service of self-regulation and metacognition. Reports that bilinguals are sometimes advantaged on some EF tasks are also consistent with the idea that enhanced experience with language can enhance aspects of EF (for a review, see Bialystok, Craik, & Luk, 2012; but see also Paap, Johnson, & Sawi, 2015). Currently, it remains unclear whether bilingual advantages in EF generalize to bimodal bilinguals (i.e., those who use both a spoken language and a sign language). Emmorey, Luk, Pyers, and Bialystok (2008) did not find evidence of enhanced inhibitory control in hearing adults who had learned both ASL and English from birth; however, bilingual advantages seem to be least pronounced in healthy adults, and are more easily detected in young children and older adults (Bialystok et al., 2012). Furthermore, the unique affordances of bimodal bilingualism may yield a different cognitive profile than that of unimodal bilinguals. For example, although unimodal bilinguals activate words in both of their languages (see Hall, 2011, for an overview), they must eventually produce a word in only one language, inhibiting the other. However, this constraint is weaker for bimodal bilinguals, who can and often do "code blend": that is, simultaneously produce both a sign and its spoken translation (Emmorey, Borinstein, & Thompson, 2005). Accordingly, Ormel et al. suggest that bimodal bilinguals might show enhancement not in inhibitory control, but in other domains such as auditoryvisual integration. Preliminary findings suggest that this may be the case (Ormel, Giezen, & van Zuilen, 2013; Ormel, Giezen, van Zuilen, & Ng, 2015; Ormel et al., 2016); if these initial reports hold, they would add to the body of evidence suggesting that experience with language, regardless of modality, can impact more general cognitive skills.

The argument for a causal relationship in the other direction (i.e., that EF skills impact language skills) is that there are specific aspects of language structure where controlled processing is necessary: for example, resolving ambiguity by integrating context and inhibiting alternative meanings, overriding a regular past-tense rule to correctly produce irregular verb forms, or switching from an incorrect parse of a garden-path sentence to a correct parse. In children, the evidence is correlational (Ibbotson & Kearvell-White, 2015; Khanna & Boland, 2010; Mazuka, Jincho, & Oishi, 2009; Woodard, Pozzan, & Trueswell, 2016), and thus causality cannot be determined. However, Novick, Hussey, Teubner-Rhodes, Harbison, and Bunting (2014) showed that adults who responded to training on a nonlinguistic working memory task also improved in their ability to recover from syntactic ambiguity in sentence processing. These results provide evidence that EF skills can causally impact language processing, at least in some contexts.

For deaf children acquiring spoken language through a cochlear implant, the relationship between EF and language outcomes has typically been studied with respect to overall ability in some aspect of language (e.g., speech perception, vocabulary, or holistic language measures), rather than specific processes like ambiguity resolution or recovery from garden-path effects, as discussed above in hearing populations. Interestingly, the relationship between EF and language seems to be different for children who use cochlear implants than for hearing children. For example, Kronenberger, Colson, et al. (2014) found that verbal working memory and fluency-speed accounted for more variance in spoken language skills among CI users than among hearing children (matched for age and nonverbal IQ), whereas inhibition-concentration and spatial working memory were stronger predictors of language skills for hearing children than for CI users.

It stands to reason that EF would play a different role for CI users, who acquire spoken language on a delayed and protracted timecourse, typically require explicit instruction/training, and have suboptimal perceptual access to the speech signal. If in fact a lack of exposure to language in early childhood disrupts the development of EF skills, and if EF skills are important for acquiring spoken language through a CI, then the children who are most ready to acquire spoken language through a CI may be those with early exposure to a natural sign language. From this perspective, early sign language exposure would allow the child to develop age-appropriate EF skills, which would in turn facilitate the acquisition and processing of spoken language through a CI. This view is consistent with findings of strong spoken language outcomes among CI users who have been exposed to sign language since birth (Davidson, Lillo-Martin, & Chen Pichler, 2014; Hassanzadeh, 2012). It remains unclear whether deaf children whose hearing families have chosen to learn a sign language show similarly desirable outcomes; current evidence is limited, and the extant evidence is mixed, varying with the particular outcome domain being measured (Allen, 2015; Allen, Letteri, Choi, & Dang, 2014; Dettman, Wall, Constantinescu, & Dowell, 2013; Giezen, 2011; Percy-Smith, Cayé-Thomasen, Breinegaard, & Jensen, 2010; Watkins, Pittman, & Walden, 1998; Wie, Falkenberg, Tvete, & Tomblin, 2007; Yanbay, Hickson, Scarinci, Constantinescu, & Dettman, 2014; Yoshinaga-Itano, Baca, & Sedey, 2010). It may be that improving the amount, timing, and quality of early sign language exposure for such families would yield more consistent or robust results.

In pursuing these questions, future researchers should consider a possibility raised by an anonymous reviewer: that EF skills might be related to communicative interaction in general, rather than language exposure in particular. Because the Deaf native signers in our study had both linguistic input and healthy communicative interaction, we cannot distinguish these factors here. We suspect that the deaf children in previous studies also experienced communicative interactions with their caregivers, but we cannot evaluate their quantity or quality. A study that was able to do so would be a valuable contribution to the literature.

We have argued that early exposure to sign language is a likely explanation for the good performance of the Deaf native signers in the present sample (as well as in similar reports from other measures in previous literature). Nevertheless, we acknowledge that other confounds still remain. In addition to being exposed to sign language from birth, deaf children of Deaf parents may differ from deaf children of hearing parents in ways that may interact with language exposure or that make independent contributions to the development of EF. For example, Corson (1973) points to increased parental acceptance among deaf parents as a potential protective factor. Likewise, Yoshinaga-Itano (2003, 2010) finds that interventions aimed at increasing parental acceptance and understanding of deafness and Deaf culture yield positive outcomes. A related proposal suggests that deaf children who identify as members of the Deaf community have more access to cultural, linguistic, and social capital, compared to deaf children who are not part of the Deaf community (Listman, Rogers, & Hauser, 2011). These researchers focus primarily on psychosocial development among deaf adolescents, but the same factors are highly applicable to cognitive development in younger deaf children. The relative contributions of language exposure and these psychosocial factors remain poorly characterized.

Another possible confound is that Deaf parents might have different standards for what constitutes acceptable behavior than hearing parents. Although we cannot empirically exclude this possibility, we note that nearly 80% of the Deaf parents were college graduates, and as such are likely to have high expectations of their children. In addition, it is common for Deaf parents to also have hearing children (although we did not collect this information in our study); therefore, we do not expect Deaf parents to lack standards of age-appropriate behavior for children with typical hearing.

A remaining confound concerns the biological etiology of deafness. In the present study, 39 of 42 Deaf participants reported hereditary deafness as the cause of hearing loss; the remaining three reported unknown etiology. Previous studies have found that in terms of EF-related behavior problems as measured by the BRIEF, children with hereditary deafness are not at risk (Oberg & Lukomski, 2011), or are at less risk than children with nonhereditary deafness (Hintermair, 2013). In cases of nonhereditary and/or syndromic deafness, the factors that cause prelingual deafness could also compromise other neural systems, leading to cognitive and/or behavioral problems. The present study cannot discriminate the influence of early sign language exposure from that of etiology. Importantly, claims about both etiology and about language exposure are crucially distinct from the claim that hearing loss itself causes cognitive problems. For example, if in fact some third factor causes both hearing loss and cognitive impairment (as in the etiology hypothesis), then there is little reason to suspect that introducing auditory experience will improve cognitive outcomes. And as we argued above, observed correlations between spoken language measures and cognitive measures may attest to the important role that language (i.e., not simply hearing) plays in supporting cognitive development.

One drawback to using the BRIEF is that it is inherently subjective, in that it relies on parents' retrospection about their children's behavior. Such limitations are intrinsic to all checklist-type instruments, and apply to all studies that rely exclusively on subjective measures (e.g., Beer et al., 2011; Hintermair, 2013). The findings from the present study would be bolstered by more objective performance-based measures of EF. This work is currently ongoing in our research group (Hall et al., 2015).

Other future directions include more direct tests of the language deprivation hypothesis. If early exposure to natural sign language confers cognitive benefits, it should be possible to detect these benefits by studying deaf children from hearing families who are exposed to sign language at some point after birth. Although random assignment to a sign language condition is not currently possible, quasi-experimental studies could measure the impact of programs designed to provide early exposure to sign language for deaf children born to nonsigning parents. As noted earlier, only a handful of studies have compared outcomes in children with CIs whose hearing families have chosen to learn a sign language against those of children with CIs whose hearing families have chosen listening and spoken language only. These studies focus on spoken language development rather than cognitive development. Still, several studies that predate cochlear implantation show consistently better psychosocial outcomes in children with early exposure to natural sign language compared to children in programs that emphasize speech or use other forms of manual communication (Preisler, 1999; Preisler & Ahlström, 1997; Preisler, Tvingstedt, & Ahlström, 2002; Watkins et al., 1998). In the context of these previous studies, the present results suggest the potential for dual (and possibly reciprocal) gains in both language and cognitive development if more deaf children are given early exposure to natural sign language. However, this prediction remains to be tested directly.

Conclusions

Several previous studies using the BRIEF have reported evidence of cognitive/behavioral problems in deaf children. One interpretation of these findings is that auditory deprivation perturbs typical development, leading to the observed deficits. However, this interpretation is complicated by a major confound: the deaf children in previous studies lacked not only auditory exposure but language exposure as well. The present study is the first whose design affords the potential to discriminate between these hypotheses, by studying BRIEF scores in Deaf children who have been exposed to a natural sign language from birth, as well as agematched children with typical hearing. Results from parent ratings on the BRIEF found that as a group, deaf children from Deaf families did not differ from predicted norms. The few instances where the Deaf sample appeared to differ from the hearing sample were driven not by significant impairment in the Deaf group but by better-than-normative performance in the hearing group. We therefore conclude that auditory deprivation is not a primary cause of EF problems in deaf children. The present results are consistent with the language deprivation hypothesis, and suggest that early exposure to a natural sign language may play a protective role. More work is necessary to test to causal predictions of this hypothesis, and to rule out other possible interpretations.

Notes

- Kronenberger, Beer, et al. (2014) also administered the preschool version (BRIEF-P) to 24 CI users and 27 hearing controls aged 3–5; to facilitate comparison with the present work, we will focus on the group that used the original version of the questionnaire.
- 2. Deutsche gebärdensprache—the sign language of the German Deaf community.
- 3. We follow the common convention of using lowercase-deaf to describe hearing levels and uppercase-Deaf to describe membership in the Deaf community.
- Median income estimates based on http://www.psc.isr.umich. edu/dis/census/Features/tract2zip/ on November 8, 2015. Zip code information was unavailable for three children.
- 5. Urban density data obtained from http://proximityone. com/zip_urban_rural.htm#table on November 8, 2015.
- 6. In all but two cases, this was ASL. One participant was exposed to a different sign language from birth and learned ASL with his parents over time. Another was exposed to two sign languages from birth.

Conflicts of Interest

No conflicts of interest were reported.

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